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Case report

Serous cystadenoma of the pancreas associated with obstructive jaundice

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We herein report a case of pancreatic serous cystadenoma in a patient who presented with jaundice, and we provide a review of the literature. A 53-year-old man was admitted with complaints of jaundice and weight loss. With a preoperative diagnosis of pancreatic serous cystadenoma with obstructive jaundice, he underwent pylorus-preserving pancreatoduodenectomy. A cystic tumor partially protruding into the bile duct was observed in the pancreatic head. Histology verified serous cystadenoma of the pancreas. Histologically, no atypia was proven in the epithelium. There have been only eight case reports dealing with serous cystadenoma of the pancreas with obstructive jaundice. Although serous cystadenoma of the pancreas has essentially a benign nature, pylorus-preserving pancreatoduodenectomy is the treatment of choice when available to avoid the recurrence of obstructive jaundice.

Key words: serous cystadenoma, pancreas, neoplasm, cystic tumor, obstructive jaundice

Introduction

Serous cystadenoma is a benign tumor of the pancreas, and is sometimes diagnosed at the time of general health checks, without any symptoms being shown. We herein report a case of serous cystadenoma of the pancreas in a patient who presented with obstructive jaundice that required differential diagnosis from duct cell carcinoma. There have been only eight cases of pancreatic serous cystadenoma complicated by obstructive jaundice reported in the literature.

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Case report

A 53-year-old man, who had suffered from jaundice and weight loss since mid-August 2000, was admitted to our department on September 4, 2000 because of elevated serum biliary enzyme levels and a mass 3 cm in diameter in the pancreatic head, detected by extracorporeal ultrasonography (US). His body height, weight, temperature, blood pressure, and pulse rate were 165 cm, 60 kg, 36.9°C, 120/80 mmHg, and 60 beats/min, respectively. His skin and conjunctiva were jaundiced. There was no mass or tenderness in the abdomen.

The laboratory tests on admission showed abnormalities as follows: aspartate aminotransferase (AST), 205 IU/l; alanine aminotransferase (ALT), 301 IU/l; lactic dehydrogenase (LDH), 482 IU/l; alkaline phosphatase (ALP), 736 IU/l; gamma guarosine triphosphate (GTP), 826 IU/l; total bilirubin, 9.3 mg/dl; and carbohydrate antigen (CA) 19-9, 87.8 U/ml.

US revealed a well-demarcated hypoechoic mass in the pancreatic head; the mass was 35 mm in diameter, and there was smooth dilatation, up to 10 mm in diameter, of the upstream pancreatic duct (Fig. 1). The lesion appeared as a solid mass with a cystic change 2 cm in diameter in its periphery. On magnetic resonance cholangiopancreatography (MRCP), the suprapancreatic biliary tree showed dilatation and the gallbladder was enlarged (Fig. 2). At the site of biliary obstruction, there was a multilocular cystic mass in the pancreatic head with a dilated proximal main pancreatic duct. The mass showed a low signal intensity multilocular cystic lesion on a T1 weighted image (WI) and high intensity on a T2 WI by magnetic resonance imaging (MRI). The cystic cavities had various intensities. Computed tomography (CT) visualized a mass 35 mm in diameter in the pancreatic head, which turned out to be a multilocular cyst following enhancement of the septum after the administration of contrast medium. There were small foci of calcification in the lesion (Fig. 3). No

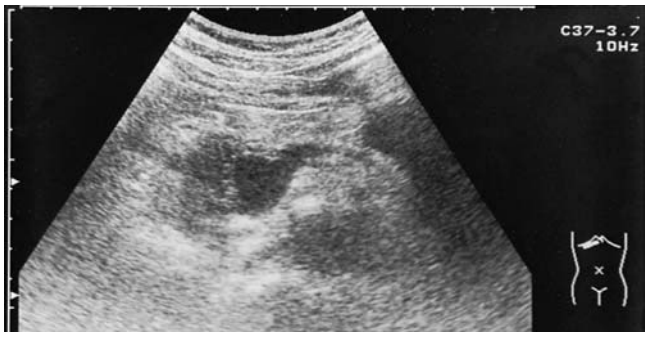


Fig. 1. Transabdominal ultrasonography revealed a mass with cystic components in the pancreatic head

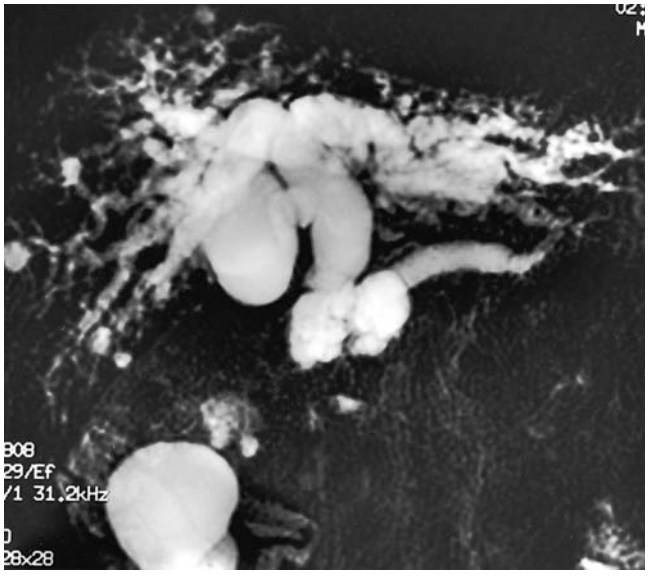


Fig. 2. Magnetic resonance cholangiopancreatography (MRCP) demonstrated a dilated biliary tree and proximal pancreatic duct. They were obstructed by a cystic mass in the pancreatic head

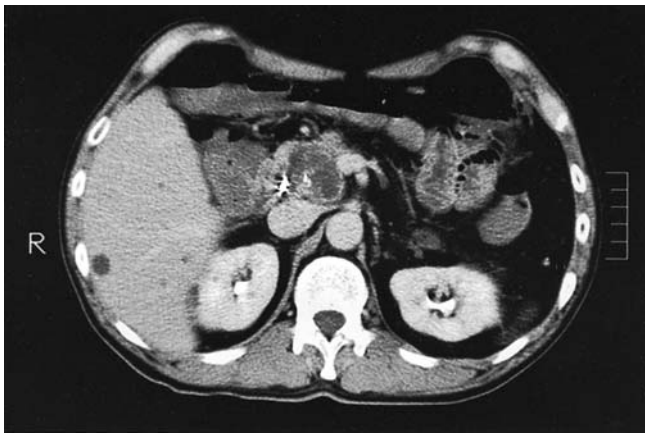


Fig. 3. Contrast enhanced computed tomography (CT). A multilocular cystic mass with calcification is seen in the pancreatic head

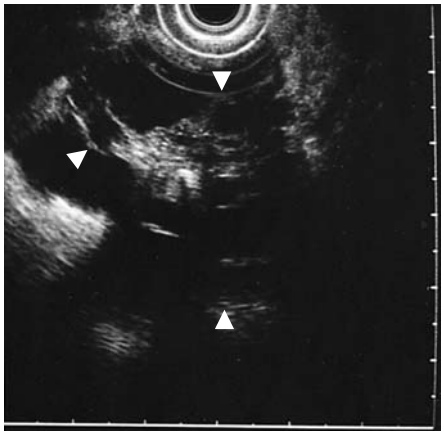


Fig. 4. Endoscopic ultrasonography (EUS) visualized a multilocular cystic mass with cysts of various sizes in the head of the pancreas (*arrowheads*)

solid mass was apparent in the portion with biliary stenosis. Endoscopic ultrasonography (EUS), by transduodenal scanning, also demonstrated a multilocular cystic mass, most of which consisted of fine cysts showing a solid mass-like appearance and some larger ones at the periphery (Fig. 4). These findings were highly suggestive of serous cystadenoma. The bile duct was obstructed at the level of the upper margin of the pancreas and contained sludge. Endoscopic retrograde cholangiopancreatography (ERCP) revealed a smooth stenosis of the lower bile duct, 4cm in length, and stenting was performed (Fig. 5a). Pancreatography visualized unilateral compression and localized stenosis of the main pancreatic duct in the head, with dilatation of the proximal duct (Fig. 5b). Near the stenosis, there were two cysts about 10mm in size that had communication with the main pancreatic duct. These were diagnosed as retention cysts. Endoscopically, no enlargement of the papilla of Vater, patulous orifice, or secretion of mucin from the papilla was observed. Intraductal ultrasonography (IDUS) of the bile duct and the pancreatic duct revealed no solid masses at the sites of stenosis, and only multiple cystic changes were depicted (Fig. 6). Superselective gastroduodenal arteriography showed an avascular area concordant with the cystic lesion, without tumor staining or arterial encasement.

Pylorus-preserving pancreatoduodenectomy was performed on October 5, with a preoperative diagnosis of pancreatic serous cystadenoma associated with retention cysts. At surgery, a cystic mass, about 4cm in diameter, showing an expansive growth surrounding the bile

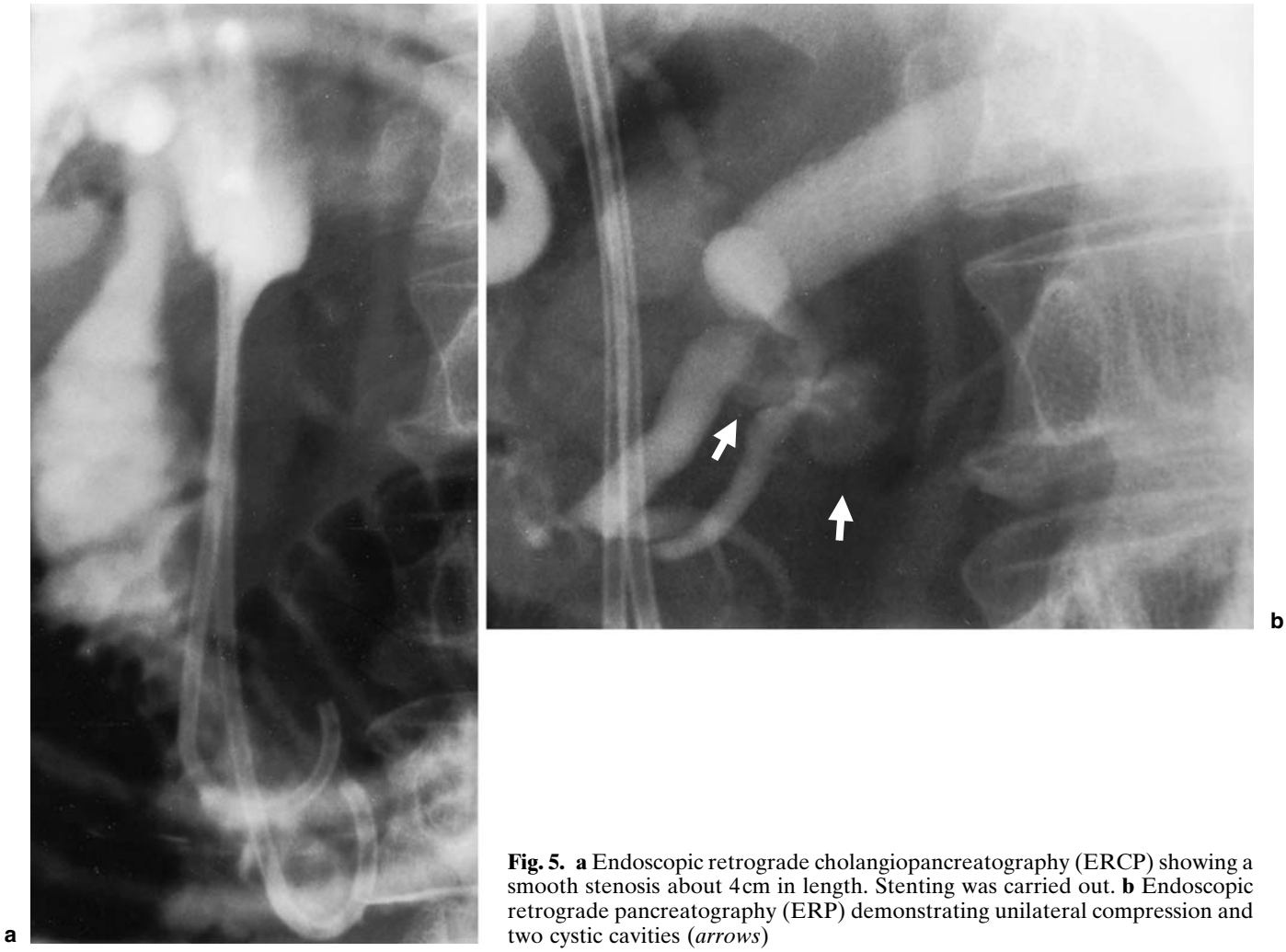


Fig. 5. **a** Endoscopic retrograde cholangiopancreatography (ERCP) showing a smooth stenosis about 4 cm in length. Stenting was carried out. **b** Endoscopic retrograde pancreatography (ERP) demonstrating unilateral compression and two cystic cavities (*arrows*)

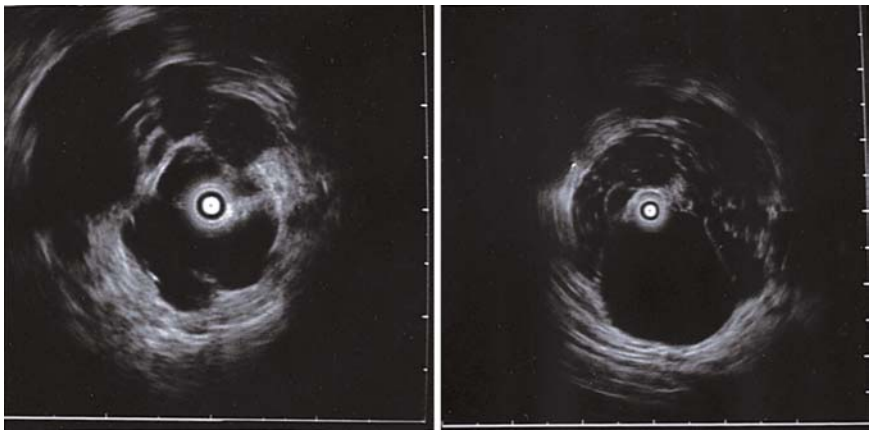
duct, was identified in the pancreatic head. The resected mass, 35 × 33 × 27 mm in size, had a whitish cut surface and was composed of multiple fine cysts forming a honeycomb structure (Fig. 7a,b). Histologically, the multilocular cyst was lined with single-layered cuboidal cells without atypia (Fig. 8a,b). No tumor infiltration into the interstitium or vessels was proven. The stenotic portions of the bile duct and pancreatic duct were surrounded by the cysts and manifested slight fibrosis and inflammatory change. Based on these findings, a diagnosis of serous cystadenoma was made. Among the cysts, a few were lined by columnar epithelium showing periodic acid-Schiff (PAS)-positive staining, and these were diagnosed as retention cysts.

Discussion

Serous cystadenoma of the pancreas is considered to have no malignant potential. Therefore, the necessity for treatment and the indications for surgery are controversial. Thus far, nine cases that were malignant, which is the only absolute indication for surgery, have been reported in the literature since the report of George et al. in 1989.[1–9] In all cases, histology did not verify adenocarcinoma, and the diagnosis of serous cystadenocarcinoma was made based on findings of invasion and metastasis.

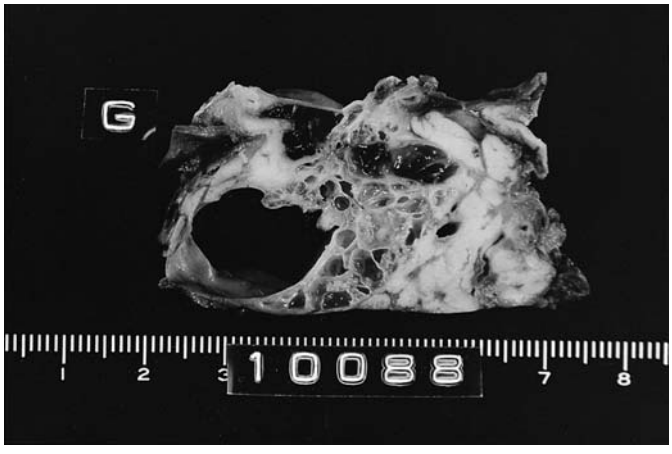
Abdominal pain, mass, and general health check are the most frequent clues leading to the diagnosis,[10] and only nine cases, associated with obstructive jaundice, including the case reported here, were reported in the literature in the period 1989 to 2000.[2,11–17] It should be kept in mind that serous cystadenoma of the pancreas can cause obstructive jaundice.

The bile duct was obstructed by mechanical compression of the cysts in all nine patients and there was no patient with invasion of the bile duct by the tumor. Hirata et al.[13] speculated that the configuration of the lesion and the bile duct in the pancreatic head affected



a b

Fig. 6a,b. Intraductal ultrasonography (IDUS) at the sites of stenosis **a** in the bile duct and **b** in the pancreatic duct. The stenoses were caused by cystic changes alone, and no solid mass was observed



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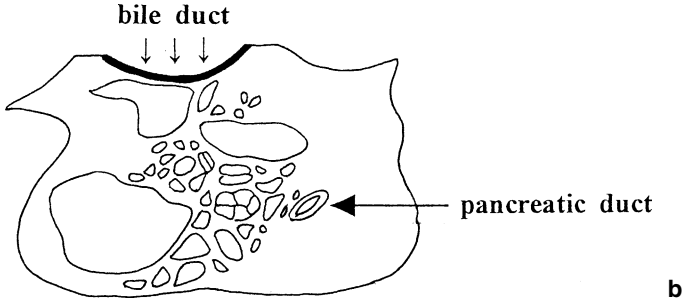
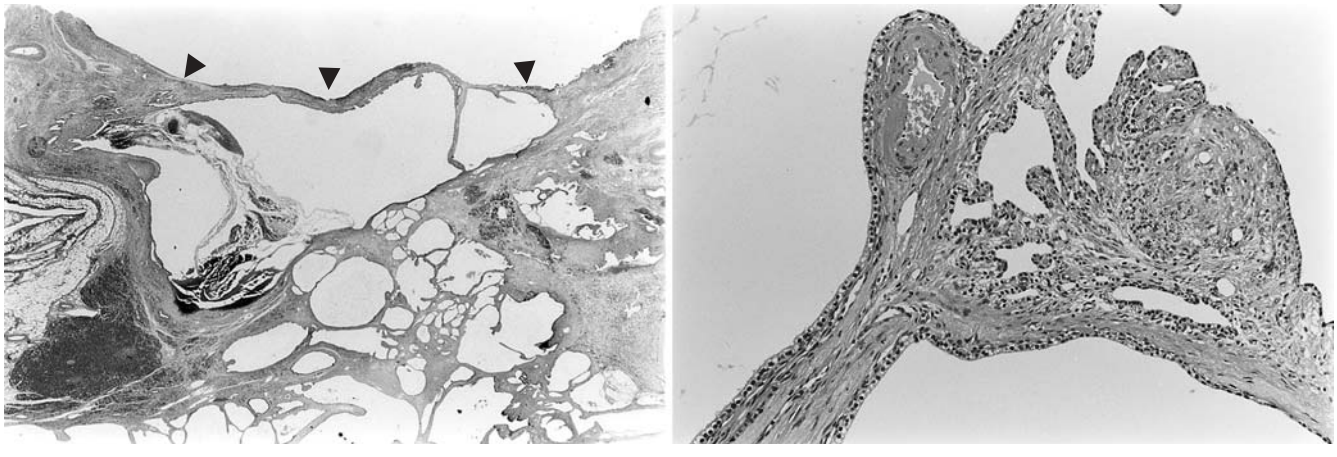


b

Fig. 7. **a** Macroscopic view of the cut surface of the resected specimen; **b** schema



a b

Fig. 8a,b. The multilocular cyst was lined with single-layered cuboidal cells without atypia. No invasion of the bile duct (*arrowheads*) by tumor cells was seen. **a** H&E, ×2.5; **b** H&E, ×50

Table 1. Patients with pancreatic serous cystadenoma associated with obstructive jaundice

Case no. Year	Author	Sex	Age (years)	Location	Size (cm)
1979	Kishi et al.[11]	F	54	Ph	6 × 4 × 3
1987	Rin[12]	M	68	Ph	4.5 × 4
1988	Hirata et al.[13]	F	71	Ph	4 × 4 × 4
1989	Iwata et al.[14]	M	61	Ph	2.5 × 3 × 3
1991	Kamei et al.[2]	F	72	Ph	10 × 10 × 8
1995	Laurent et al.[15]	F	34	Ph	5 × 3
1996	Baba et al.[16]	F	36	Ph-Pt	?
2000	Maekawa et al.[17]	M	62	Ph	3.5 × 2.7 × 1.7
2001	Our patient	M	53	Ph	3.5 × 3.3 × 2.7

Ph, Pancreatic head; Pt, pancreatic tail

the manifestation of obstructive jaundice, regardless of the size of the lesion. As for the ERCP images in the patient reported here, the stenosis of the bile duct was smooth and long, and the pancreatic duct was compressed unilaterally. These findings are different from those of pancreatic cancer. Communication with the ductal system is seldom seen in patients with pancreatic serous cystadenoma.[18] In our patient, the cysts that had communication with the main pancreatic duct visualized by ERCP were considered to be coexistent retention cysts just next to the tumor, and this was confirmed histologically.

Generally, it is considered to be easy to make a diagnosis of serous cystadenoma of the pancreas because of the characteristic findings by diagnostic imaging. US visualizes the lesion as a solid mass. EUS, however, is able to depict the internal structure having a characteristic honeycomb appearance, due to the use of higher-frequency ultrasound. When a tumor is composed of densely arranged fine cysts, it is visualized as a hyperechoic mass by US, and when the cystic components are larger, the lesion is visualized as a multilocular hypoechoic mass. These findings are affected by the size and density of the cystic component, and the amount of interstitial connective tissue.[19] In our patient, the central portion of the tumor was visualized as a hyperechoic solid mass reflecting the aggregation of fine cystic components by US, and larger cystic cavities were demonstrated at its periphery. CT and MRI were useful for recognition of the general image of the lesion, but it was not possible to evaluate the cause of biliary obstruction by these modalities. IDUS clarified the cause of biliary obstruction and stenosis of the pancreatic duct as compression by the cystic lesion alone, which had a characteristic honeycomb appearance. This modality is very effective for the assessment of the internal structure of cystic lesions of the pancreas near the main pancreatic duct.[20,21] Differentiation from mucinous cystadenoma and cystadenocarcinoma is necessary and important when evaluating cystic tumors of the pancreas. In our patient, the lesion was not globular and was not accompanied by any solid components. Endoscopic ultrasonography and intraductal ultrasonography play major roles in the differential diagnosis of cystic changes of the pancreas because of their high resolution in visualizing cystic structures.

Among the nine patients reported in the literature who showed obstructive jaundice (Table 1), there was a correct preoperative diagnosis in only two, and the other patients underwent surgery with a preoperative diagnosis of pancreatic cancer. This is a typical phenomenon indicating the difficulty of making a precise diagnosis preoperatively in patients with obstructive jaundice. The proven absence of a solid component in the smooth wall of the cystic lesion was regarded as the rationale for the diagnosis of a benign tumor complicated by jaundice.

What is the best treatment for patients with serous cystadenoma of the pancreas associated with obstructive jaundice? Enucleation of the tumor or biliary reconstruction alone, without removal of the mass, entails the risk of recurrent jaundice with the development of residual tumors. Pylorus-preserving pancreatoduodenectomy, therefore, may be the treatment of choice when available.

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