

Case report

Chronic asymptomatic pseudocyst with sludge aggregates masquerading as mucinous cystic neoplasm of the pancreas

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Abstract: Pseudocyst of the pancreas is sometimes difficult to distinguish from mucinous cystic neoplasm of the pancreas. A 37-year-old asymptomatic Japanese man was diagnosed with hypertension. He had a 20-years history of habitual drinking of alcohol, but no history of pancreatitis or abdominal trauma. During examinations to ascertain the cause of hypertension, ultrasonography and computed tomography incidentally demonstrated a huge cyst in the head of the pancreas. Laboratory data were within normal limits, including serum levels of amylase, carcinoembryonic antigen, and carbohydrate antigen 19-9. Imaging studies showed a huge unilocular cyst, measuring 7 cm, in the head-to-body of the pancreas, and two small unilocular cysts, measuring 1.4 and 1.5 cm, in the tail and head of the pancreas, respectively. A mural nodule was suspected in the largest cyst. Endoscopic retrograde cholangiopancreatography demonstrated communication of the main pancreatic duct with the two small cysts in the head and tail of the pancreas but not with the huge cyst. There were no ductal changes suggesting chronic pancreatitis. Laparotomy was performed under the tentative diagnosis of potentially malignant mucinous cystic neoplasms of the pancreas. However, inflammatory adhesion was dense around the pancreas and the mural nodule suspected preoperatively was found to be sludge aggregates in a pseudocyst. The diagnosis of an intraoperative frozen section of the cyst wall was pseudocyst of the pancreas. Cystojejunostomy was performed. We report this case because the preoperative diagnosis was mucinous cystic neoplasm of the pancreas, but the diagnosis changed with careful intraoperative examinations, to pseudocyst of the

pancreas. We discuss the differential diagnosis of the two conditions.

Key words: mucinous cystic neoplasm of the pancreas, pseudocyst of the pancreas

Introduction

In adults, 70% of cystic lesions of the pancreas are pseudocysts, while only 10%–15% are neoplastic cysts.¹ However, pseudocyst of the pancreas is sometimes difficult to differentiate from mucinous cystic neoplasm of the pancreas, despite recent progress in diagnostic imaging modalities. According to Warshaw et al.,¹ one-third of the cystic neoplasms of the pancreas were originally diagnosed as pseudocysts, and 40% of mucinous cystic neoplasms were misdiagnosed as pseudocysts of the pancreas. Several authors have reported imaging characteristics that differentiate the two conditions.^{1–4} Lewandrowski et al.^{5,6} have also reported that various contents of the cyst were useful to differentiate between the two diseases. With the recent progress in diagnostic imaging techniques, cystic lesions including chronic pseudocysts and mucinous cystic neoplasms, have been detected more frequently in asymptomatic patients. Precise preoperative differentiation is difficult or even impossible, especially in such asymptomatic patients. From a clinical point of view, the differentiation of the two diseases is crucial. When the condition is an inflammatory process, operation is not indicated in most instances. On the other hand, when the disease is mucinous cystic neoplasm of the pancreas, the lesion should be resected because of apparent or latent malignancy. We report a patient in whom the preoperative diagnosis was mucinous cystic neoplasm of the pancreas and the diagnosis of pseudocyst was first made during operation.

Case report

A 37-year-old asymptomatic Japanese man diagnosed with hypertension was admitted to a nearby hospital for examinations to ascertain the cause of the hypertension. Ultrasonography (US) and computed tomography (CT) incidentally displayed a huge cystic lesion in the head of the pancreas. The patient was referred to us for further evaluation of the cystic tumor. He had a 20-years history of habitual drinking of alcohol, but no history of pancreatitis or abdominal trauma.

On admission, no mass was palpable in the abdomen. Laboratory data were all within normal limits, including serum amylase (68 U/l), carcinoembryonic antigen (CEA, 3.9 ng/ml), and carbohydrate antigen (CA19-9, 8.6 U/l). US showed a cystic mass, 7 cm in diameter, in the head-to-body of the pancreas with a papillary projection therein (Fig. 1). Endoscopic Ultrasonography (EUS) also demonstrated a cystic mass, inside which a hyperechoic lesion was identified. CT revealed two cystic masses in the pancreas (Fig. 2). One was a huge cyst, 7 cm in diameter, located in the head-to-body. The other was a small cyst, measuring 1.4 cm, in the pancreatic tail. In addition to these two cysts, magnetic resonance cholangiopancreatography (MRCP) demonstrated one more small cyst, measuring 1.5 cm, in the head of the pancreas (Fig. 3). The largest cyst in the pancreatic head was unilocular, had a mural nodule, and caused ventral deviation of the pancreas, suggesting a diagnosis of mucinous cystadenoma or mucinous cystadenocarcinoma. Endoscopic retro-

grade cholangiopancreatography (ERCP) demonstrated communication of the main pancreatic duct with the two small cystic lesions in the head and tail of the pancreas but not with the huge cyst in the head (Fig. 4). There were no ductal alterations suggesting chronic pancreatitis. Cytological examination of the pancreatic juice aspirated at ERCP under secretin stimulation was negative for malignant cells. Celiac angiography showed a large avascular mass in the pancreatic head.

At laparotomy, marked inflammatory adhesion was noted around the pancreas. Aspiration of the cyst contents yielded brown turbid fluid which contained abundant inflammatory cells, no epithelial cells, and

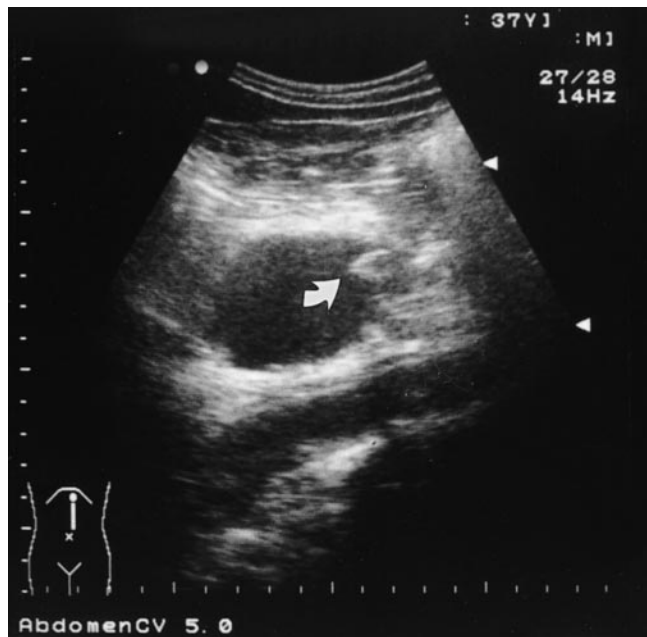


Fig. 1. Ultrasonography shows a papillary projection in the cyst in the head-to-body region of the pancreas (arrow)



Fig. 2. Computed tomography shows two cystic masses, one in the head and one in the tail of the pancreas, measuring 7 and 1.4 cm, respectively

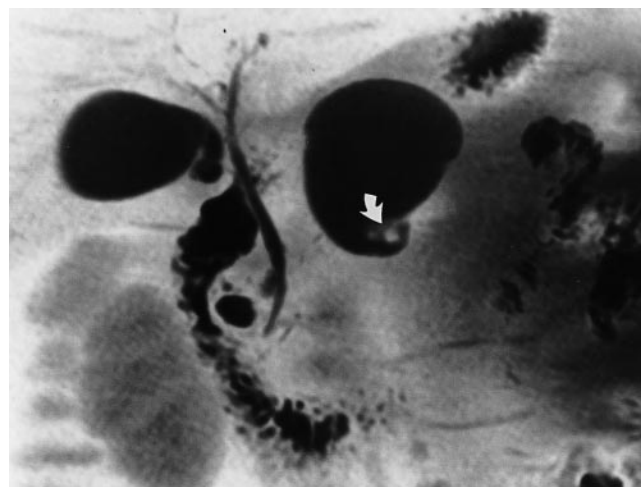


Fig. 3. Magnetic resonance cholangiopancreatography shows three unilocular cysts. The largest cyst in the head of the pancreas, measuring 7 cm, had a solid component (arrow) in its caudal part

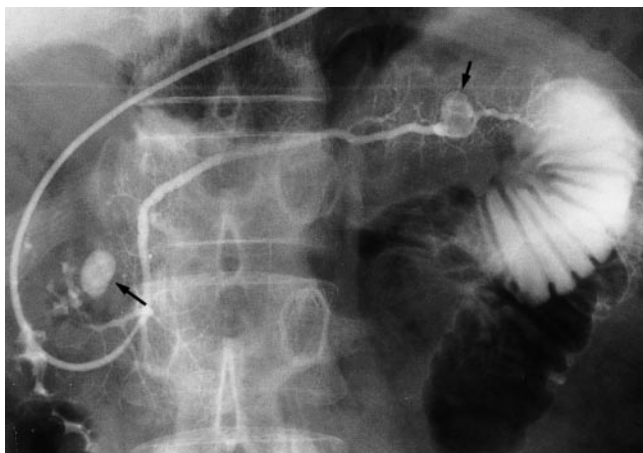


Fig. 4. Endoscopic retrograde cholangiopancreatography shows two cystic lesions communicating with the ductal system. The slight cranial deviation of the main pancreatic duct in the head-to-body region of the pancreas is caused by the identified huge cystic mass



Fig. 5. Macroscopic view of sludge aggregates in the pseudocyst

no malignant cells. The amylase level in the fluid was 60 100 U/l, CEA was 9.0 ng/ml, and CA19-9, 53 657 U/ml. A 5-cm incision was made on the wall of the cyst to take specimens of the mural nodule and the cyst wall for intraoperative frozen section diagnosis. The mural nodule detected on US proved to be dirty sludge aggregates attached to the cyst wall (Fig. 5). The internal surface of the cyst was covered with yellowish inflammatory exudate. Specimens from several areas of the cyst wall were composed of fibrous tissue with inflammatory infiltrates and lacked lining epithelium. These findings were consistent with the diagnosis of a pseudocyst of the pancreas. Cystojejunostomy was performed. The two other cystic lesions in the head and tail of the pancreas could not be detected by

intraoperative ultrasonography. The patient had an uneventful postoperative recovery and was discharged 1 month after the operation. He was alive and well 6 months after the operation.

Discussion

Several clinical features have been reported to differentiate a pseudocyst and a mucinous cystic neoplasm of the pancreas.^{2,3} Patients with mucinous cystic neoplasm of the pancreas are usually women in the fourth or fifth decade of life. Those with a pseudocyst usually have a history of acute pancreatitis or they have identifiable antecedent factors such as alcoholism, gallstones, or trauma. Moreover, serum amylase level is elevated in 50%–75% of patients with a pseudocyst.² However, the patient we have described was completely asymptomatic and lacked any of these factors or findings. This made differentiation of the two conditions difficult.

Some radiological findings have been reported to be useful for the differential diagnosis of the two conditions.^{1–4} Pseudocysts usually present with a solitary unilocular cyst on US, CT, and magnetic resonance imaging (MRI).^{1–3} They do not have septa or mural nodules in the cyst. Multicystic structures with solid components and septa are suggestive of neoplastic cysts. Nishihara et al.⁴ reported that on T1-weighted MRI, mucinous cystic tumors containing gelatinous mucin showed higher signal intensity than pseudocysts containing watery fluid. On ERCP, about 60% of patients with pseudocysts have a communication with the pancreatic ductal system, while mucinous cystic neoplasms of the pancreas are often separate from the pancreatic ductal system and have no ductal abnormalities characteristic of chronic pancreatitis.^{1–3} On angiography, pseudocysts are usually hypovascular and show deviation of vessels, while mucinous cystic neoplasms of the pancreas sometimes show hypervascularity and vascular encasement.^{1,2} In the present patient, preoperative imaging showed a huge unilocular cyst with a mural nodule and no pancreatic changes suggestive of chronic pancreatitis. The tentative preoperative diagnosis of potentially malignant mucinous cystic neoplasm was made. The mural nodules, however, were intraoperatively found to be sludge aggregates in the pseudocyst.

Analysis of cystic fluid has also been reported to be helpful in distinguishing pseudocysts from cystic neoplasms of the pancreas. The fluid in the pseudocysts had high amylase activity, a low CEA level, and low fluid viscosity, and cytologic examination of the fluid was negative for malignancy.^{6,7} The combination of these findings enables us to make a more accurate

diagnosis. In this patient, intraoperative aspiration of the cystic fluid showed that the amylase level in the cystic fluid was 60100U/l; CEA 9.0ng/ml; and Ca19-9 53657U/ml. The CEA and CA19-9 levels seemed to be high enough to suggest a malignant potential of the present cyst. However, the CEA and CA19-9 levels in the cystic contents are not conclusive in the differential diagnosis of benign and malignant conditions.⁷ We should also be careful when performing puncture of pancreatic cysts, because there are potential risks of peritonitis, pancreatic juice leakage, and tumor seeding after aspiration of neoplastic cysts.⁷ Recently, molecular biological findings, such as *K ras* codon 12 point mutation, *p53* mutation, and telomerase activity, have been reported to be useful for diagnosing malignancies. However, in the present patient, we did not measure telomerase activity in the pancreatic juice and cystic fluid.

The intraoperative gross appearance and biopsy findings of the cyst wall are important. When biopsy reveals fibrous tissues without epithelial lining cells, a pseudocyst should be presumed.^{2,3} However, the absence of an epithelial lining in a limited biopsy specimen does not always rule out cystic neoplasms, because the epithelium of cystic tumors is occasionally denuded, and the diagnosis of the biopsy specimens can be misleading.^{2,3,8} Therefore, multiple biopsies from different areas are necessary for an accurate intraoperative diagnosis.

In the asymptomatic patient, described here the presence of a mural nodule and the absence of ductal changes suggestive of chronic pancreatitis made the

preoperative diagnosis more puzzling. The preoperative diagnosis was a mucinous cystic neoplasm of the pancreas and the diagnosis of a pseudocyst was first made intraoperatively. We should carefully examine such a patient intraoperatively as well as preoperatively so that we select an adequate operative procedure and avoid unnecessary surgery.

References

1. Warshaw AL, Compton CC, Lewandrowski K, et al. Cystic tumors of the pancreas. *Ann Surg* 1990;212:432–445.
2. Warshaw AL, Rutledge PL. Cystic tumors mistaken for pancreatic pseudocysts. *Ann Surg* 1987;205:393–398.
3. Lumsden A, Bradley EL III. Pseudocyst or cystic neoplasm? Differential diagnosis and initial management of cystic pancreatic lesions. *Hepatogastroenterology* 1989;36:462–466.
4. Nishihara K, Kawabata A, Ueno T, et al. The differential diagnosis of pancreatic cysts by MR imaging. *Hepatogastroenterology* 1996;43:714–720.
5. Lewandrowski K, Southern J, Pins M, et al. Cyst fluid analysis in the differential diagnosis of pancreatic cysts: A comparison of pseudocysts, serous cystadenomas, mucinous cystic neoplasms, and mucinous cystadenocarcinoma. *Ann Surg* 1993;217:41–47.
6. Lewandrowski KB, Warshaw AL, Compton CC, et al. Variability in cyst carcinoembryonic antigen level, fluid viscosity, amylase content, and cytologic findings among multiple loculi of a pancreatic mucinous cystic neoplasm. *Am J Clin Pathol* 1993;100:425–427.
7. Sand JA, Hyoty MK, Mattila J, et al. Clinical assessment compared with cyst fluid analysis in the differential diagnosis of cystic lesions in the pancreas. *Surgery* 1996;119:275–280.
8. Compagno J, Oertel JE. Mucinous cystic neoplasms of the pancreas with overt and latent malignancy (cystadenocarcinoma and cystadenoma). A clinicopathologic study of 41 cases. *Am J Clin Pathol* 1978;69:573–580.