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Single Case

Pancreatic Pseudocyst Ruptured due to Acute Intracystic Hemorrhage

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Keywords

Pancreatic pseudocyst · Intracystic hemorrhage · Pancreatic pseudocyst rupture · Intraperitoneal bleeding · Renal cell carcinoma

Abstract

Rupture of pancreatic pseudocyst is one of the rare complications and usually results in high mortality. The present case was a rupture of pancreatic pseudocyst that could be treated by surgical intervention. A 74-year-old man developed abdominal pain, vomiting, and diarrhea, and he was diagnosed with cholecystitis and pneumonia. Three days later, acute pancreatitis occurred and computed tomography (CT) showed slight hemorrhage in the cyst of the pancreatic tail. After another 10 days, CT showed pancreatic cyst ruptured due to intracystic hemorrhage. Endoscopic retrograde cholangiopancreatography revealed leakage of contrast agent from pancreatic tail cyst to enclosed abdominal cavity. His left hypochondrial pain was increasing, and CT showed rupture of the cyst of the pancreatic tail into the peritoneal cavity was increased in 10 days. CT showed also two left renal tumors. Therefore we performed distal pancreatectomy with concomitant resection of transverse colon and left kidney. We histopathologically diagnosed pancreatic pseudocyst ruptured due to intracystic hemorrhage and renal cell carcinoma. Despite postoperative paralytic ileus and fluid collection at pancreatic stump, they improved by conservative management and he could be discharged on postoperative day 29. He has achieved relapse-free survival for 6 months postoperatively.

The mortality of pancreatic pseudocyst rupture is very high if some effective medical interventions cannot be performed. It should be necessary to plan appropriate treatment strategy depending on each patient.

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Introduction

Pancreatic pseudocyst can occur following acute pancreatitis, chronic pancreatitis, or secondary to pancreatic trauma [1]. Histopathologically, pseudocyst presents as a cystic cavity bound to the pancreas by inflammatory tissue and the wall of a pancreatic pseudocyst lacks an epithelial lining. In addition, the cyst contains pancreatic juice or amylase-rich fluid [2]. On rare occasions, it has demonstrated lethal due to the progression of pancreatic pseudocyst by infection, intracystic hemorrhage, or free rupture into the peritoneal cavity. The incidence rate of hemorrhage complication was only 5% of pancreatic pseudocysts, but the mortality rate was more than 40% [1, 3]. Therefore it is necessary to understand the clinical condition of the complication and provide appropriate treatment. In the case of pancreatic pseudocyst rupture, one should treat not only bleeding but also infection and secondary damage due to intraperitoneal exposure of pancreatic juice. Because the life-saving rate by conservative medical management is particularly low, surgical, radiological, or endoscopic intervention are required [4, 5].

We report a rare case of pancreatic pseudocyst ruptured due to intracystic hemorrhage, which could be rescued with surgical intervention and simultaneously resection of renal cell carcinoma.

Case Report

A 74-year-old man presenting with abdominal pain, vomiting, and diarrhea was admitted to a nearby hospital. He did not drink alcohol regularly. On physical examination, his right hypochondrial pain was mildly tender. Laboratory data showed jaundice, high inflammatory reaction, and renal dysfunction: white blood cell count 6,360/ μ L, C-reactive protein 13.51 mg/dL, aspartate aminotransferase 78 IU/L, alanine aminotransferase 211 IU/L, alkaline phosphatase 358 IU/L, γ -glutamyltranspeptidase 440 IU/L, total bilirubin 4.7 mg/dL, serum amylase 54 mg/dL, blood urea nitrogen 45.1 mg/dL, creatinine 2.58 mg/dL. Computed tomography (CT) scan findings of swelling of gallbladder, and bilateral pulmonary infiltrative shadow could lead to the diagnosis of acute cholecystitis and pneumonia. Three days later, serum and urinary amylase levels were elevated (serum amylase 407 mg/dL and urinary amylase 2,592 mg/dL). At that time, CT scan showed high density area in a cyst of pancreatic tail (Fig. 1a), which was considered to be slight intracystic hemorrhage. After another 10 days, he complained about left hypochondrial pain. Because CT scan showed pancreatic cyst rupture with intracystic hemorrhage (Fig. 1b), he was transferred to our hospital on the next day. First CT in our hospital showed size of intracystic hemorrhage was stable (Fig. 1c). We had a policy to make a thorough examination, because there were no worsening of anemia and severe drop in blood pressure. Endoscopic retrograde pancreatography showed a cystic lesion with internal filling defect connecting the main pancreatic duct. Additionally, there was a leakage of contrast medium from the cystic lesion into the abdominal cavity (Fig. 1g). Then, an endoscopic nasopancreatic drainage (ENPD) tube was placed into the main pancreatic duct under the diagnosis of rupture of the pancreatic cyst due to intracystic hem-

orrhage. The pancreatic juice from ENPD was bloody and the cytological finding of the pancreatic juice from ENPD revealed no evidence of malignancy.

Ten days after the insertion of ENPD tube, his left hypochondrial pain was increased. At that time, CT showed the increase in size of the fluid collection besides the cystic lesion (Fig. 1d). In addition, CT showed two left renal tumors (Fig. 1e, f). Finally, he was diagnosed as having ruptured pancreatic cyst with left renal cell carcinoma, and he underwent distal pancreatectomy and left nephrectomy. At operation, because of the mass formed with transverse mesocolon and pancreatic tail hematoma, transverse colon was resected together. Blood loss was 200 g, and we required no perioperative blood transfusion. In the postoperative course, paralytic ileus delayed the beginning of diet. There was fluid collection at pancreatic stump, but because of the absence of infection, it experienced an improvement by conservative management. He could be discharged on postoperative day 29.

The specimen which was resected en bloc with surrounding tissues appeared to be a cystic degeneration of the pancreatic tail with intracystic hematoma surrounded by other pancreatic cysts without hemorrhage (Fig. 2). Histopathological examination revealed outgrowths of fusiform-shaped fibroblast with collagen fiber around hematoma and no epithelial component in the wall of the pancreatic cyst wall (Fig. 3a, b). Then we diagnosed the lesion as pancreatic pseudocyst rupture by intracystic hemorrhage. The left renal tumors measuring 1.8 and 1.5 cm in each maximum diameter were diagnosed as renal clear cell carcinoma because they were immunohistologically positive for CD10 and CAM5.2, and negative for vimentin (Fig. 3c–f). The final pathological stage was stage I according to TNM classification of malignant tumors.

Discussion

Pancreatic pseudocyst often occurs subsequent to acute pancreatitis and chronic pancreatitis and relates to drinking alcohol. The incidence of pancreatic pseudocyst in patients with non-alcoholic drinking is low (5.2%) [6]. In acute pancreatitis, the development of pseudocysts results from the accumulation of enzyme-rich fluid and autodegradation products, and a failure to resorb these accumulations. In chronic pancreatitis, the formation of pseudocysts follows intraductal high pressure and disruption of branched ductules of pancreas by obstructed pancreatic duct [7]. But approximately 40% of pancreatic pseudocysts improve spontaneously with conservative medical management, if presenting symptoms can be controlled [8]. Although rupture of the pseudocyst into abdominal cavity occurs rarely, it frequently leads the patients to lethal condition. The causes of ruptured pancreatic pseudocyst may include abdominal trauma, pancreatitis, infection, intracystic hemorrhage, and diagnostic puncture of cyst. The incidence of intracystic hemorrhage is one of serious complications and has been reported to be 6–10% [9].

Three types of mechanism for bleeding and rupture of pancreatic pseudocyst have been suggested [9–11]. First, severe inflammation and activated lytic enzymes might cause progressive digestion of the elastic component of the vessel wall, with consequent erosion and disruption. Second, pseudocysts might produce erosion of vessels as a consequence of persistent compression, ischemia, and the electrolytic action of enzymatic contact. Third, the inflammatory process and the pseudocyst might cause compression or thrombosis in the portal and splenic vein, leading to localized portal hypertension. The progressive patterns are categorized into three types; only intracystic hemorrhage, bleeding into gastrointestinal tract, and expanding into the abdominal cavity. Intraperitoneal hemorrhage from ruptured

pancreatic pseudocyst was associated with an extremely high mortality rate (35.3%) [12]. Contrast-enhanced CT is an effective and convenient examination for the diagnosis of pancreatic pseudocyst bleeding. In our case, CT of previous hospital showed hemorrhage in pancreatic pseudocyst after acute pancreatitis (Fig. 1a). The cause of intracystic hemorrhage was expected to be erosion and disruption of superficial vessels within the pseudocyst wall by acute pancreatitis. In addition, weakness of pseudocyst wall with intracystic hemorrhage resulted in pancreatic pseudocyst rupture and intraperitoneal bleeding. Although blood effusion was contained in the pancreatic pseudocyst, we could not identify any bleeding points in the pancreatic pseudocyst histopathologically.

The patients with ruptured pancreatic pseudocyst were treated with pancreatectomy, pancreatic cyst-digestive tract anastomosis, and percutaneous drainage. Intraperitoneal bleeding caused by pancreatic pseudocyst ruptured due to intracystic hemorrhage from the pseudocyst wall has been reported in 8 cases [12, 13]. Two patients receiving conservative treatment all died, while 5 (83.3%) of 6 patients who underwent the surgery could be life-saving. It usually requires surgical intervention not only for control of bleeding but also for secondary damage due to exposure of pancreatic juice into the peritoneal cavity and infection of hematoma. On the other hand, although many surgical procedures including exploratory laparotomy had been performed for ruptured pancreatic pseudocyst, they were not always the best strategy. Recent development of interventional radiology with metallic coil resulted in good control of hemorrhage from the artery. In particular, angiographic embolization is available on a case of pseudoaneurysm formation. In case of hemorrhagic shock, if hemostasis is obtained by transcatheter arterial embolization in advance of operative procedure, surgery may be performed more safely. In addition, angiography has also important function in correct localization of bleeding and identification of any unusual arterial anatomy [9]. But emergency operation must often be performed according to the patient's hemodynamic instability. Although we performed endoscopic pancreatic duct drainage through ampulla of Vater in this case, the effect of drainage was poor. Because hematoma in the abdominal cavity had increased in a short span of time, we selected surgical intervention including distal pancreatectomy combined with left nephrectomy in this reported case. The reason why left nephrectomy was performed concomitantly with pancreatic tail was that the adhesion after pancreatectomy could make the nephrectomy difficult.

In conclusion, the mortality of pancreatic pseudocyst rupture is very high if some kind of medical intervention is not performed. It will be necessary to plan appropriate treatment strategy depending on each patient.

Statement of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

The authors have no funding or conflicts of interest to disclose.

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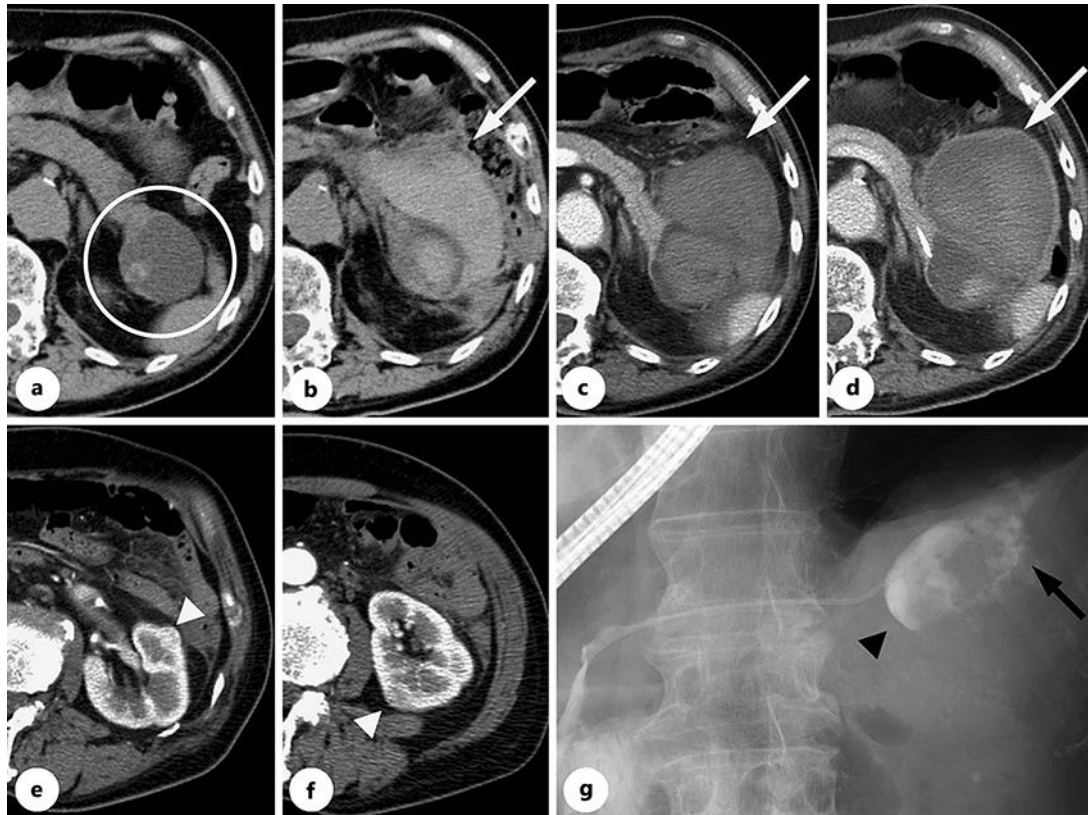


Fig. 1. Preoperative abdominal computed tomography (CT) (a–f) and endoscopic retrograde cholangiopancreatography (g). **a** The slight hematoma is seen within pancreatic tail cyst (circle). **b** Pancreatic tail cyst rupture with intracystic hemorrhage (white arrow) is seen in previous hospital. **c** Pancreatic tail cyst rupture with intracystic hemorrhage (white arrow) in first CT at our hospital is stable. **d** Pancreatic tail cyst rupture into the peritoneal cavity with intracystic hemorrhage is expanded. **e, f** Two tumors in left kidney are seen (white arrowhead). **g** Pancreatic tail cyst with internal filling defect (black arrowhead) is seen. There is leakage outside of pancreatic tail cyst (black arrow). Endoscopic nasopancreatic drainage tube is placed into pancreatic main duct.

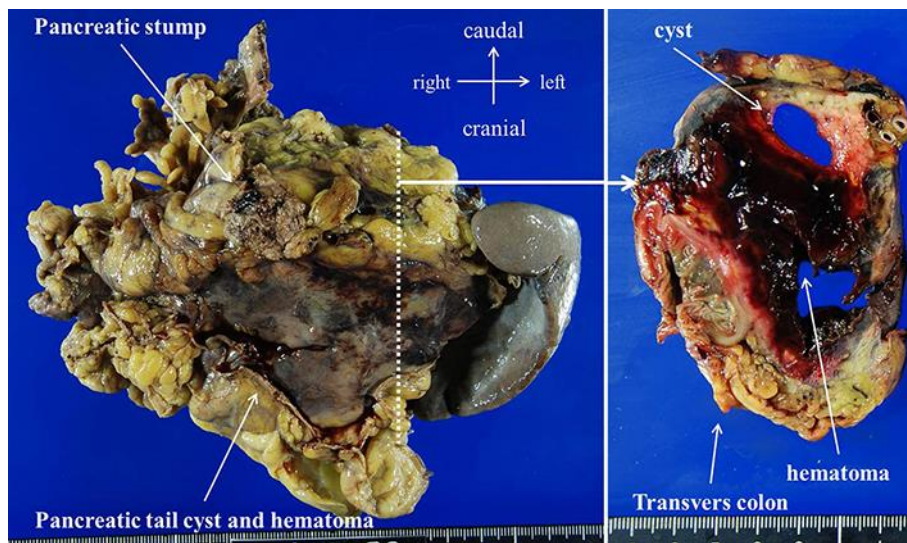


Fig. 2. Macroscopic features of the resected specimen. The resected specimen appeared to be cystic degeneration at pancreatic tail with intracystic hemorrhage and hematoma surrounding pancreatic cyst. Hematoma stuck to transverse mesocolon.

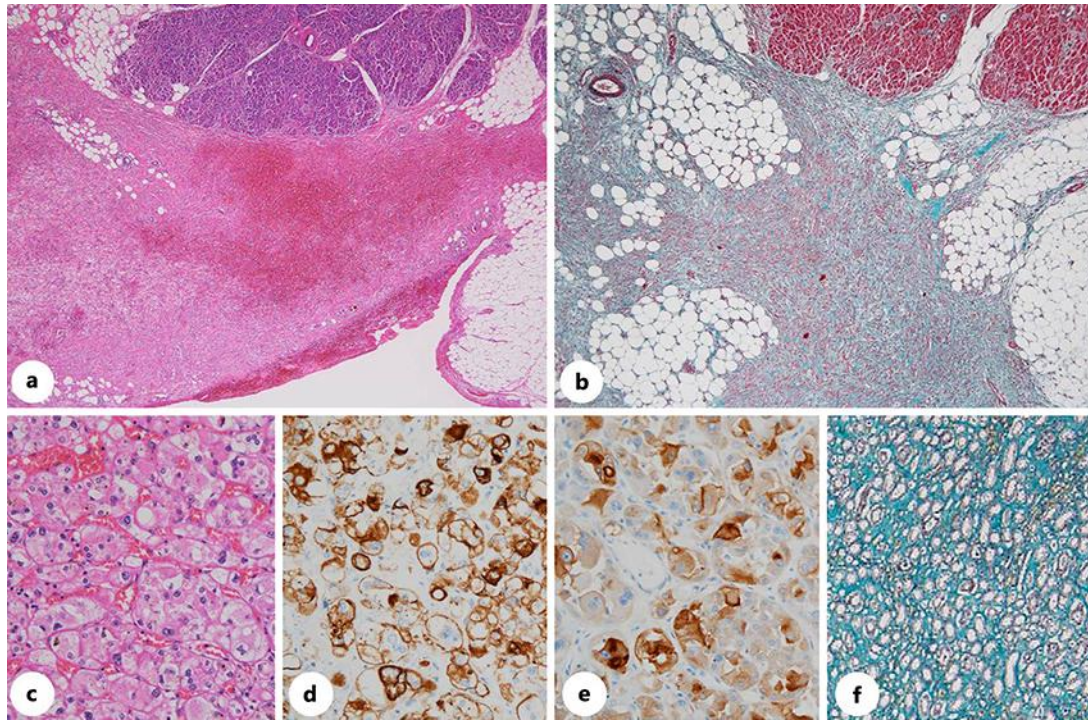


Fig. 3. Histopathological examination findings (**a, b**: pancreas, **c–f**: kidney). **a** Hematoxylin-eosin (HE) staining showed outgrowths of fusiform-shaped fibroblast with collagen fiber around hematoma. $\times 40$. **b** Elastica-Masson Goldner staining showed no epithelial component in pancreatic cyst wall. $\times 40$. **c** Dysplastic cells with granular abundant cytoplasm, large irregular nucleus, and large nucleolus grew proliferously in alveolar configuration. HE. $\times 400$. **d–f** The tumor cells are immunohistologically positive for CAM5.2 (**d**) and CD10 (**e**), and negative for vimentin (**f**). **c–e** $\times 400$. **f** $\times 100$.