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CASE REPORT

A case of pancreatic pseudocysts accompanied by infection, pseudoaneurysm ruptures, and pseudocystocolonic fistulae

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Abstract

Pancreatic pseudocysts (PPs) can be accompanied by infection, pseudoaneurysm ruptures, and fistulae to other organs, which can be fatal without appropriate treatment. Herein, we present the case of an 82-year-old man with PPs accompanied by infection, pseudoaneurysm rupture, and pseudocystocolonic fistula that were managed via multidisciplinary treatment. Computed tomography (CT) revealed two inflamed PPs, one each in the pancreatic head and tail. He was, therefore, diagnosed with infectious PPs. The pancreatic head PP shrunk on endoscopic nasopancreatic drainage (ENPD), but the pancreatic tail PP did not. Endoscopic ultrasound (EUS)-guided transluminal drainage was performed to treat the pancreatic tail PP; his symptoms improved. However, he vomited blood at 14 day post-drainage. Angiography revealed pseudoaneurysm rupture in a left gastric artery branch. After successful angioembolization, he developed hematochezia 2 days later. We suspected re-bleeding of the pseudoaneurysm. The bleeding stopped spontaneously, but CT and radiography revealed the presence of a pseudocystocolonic fistula. Careful follow-up was performed, and he has not had any symptoms at 9 month post-discharge. We managed PP-related complications via ENPD, EUS-guided transluminal drainage, angioembolization, and careful follow-up. Infection, pseudoaneurysm rupture, and pseudocystocolonic fistula are rare, but can occur simultaneously. Therefore, clinicians should consider these complications when treating patients with PPs.

Keywords Pancreatic pseudocyst · Infection · Pseudocystocolonic fistula · Pseudoaneurysm

Introduction

Pancreatic pseudocysts (PPs) are peripancreatic fluid collections surrounded by a well-defined wall and contain no solid component [1]. The incidence of PPs is 5–15% in acute pancreatitis and 20–40% in chronic pancreatitis [2]. PP symptom development is related to the location and size of the PPs. PP symptoms include abdominal pain, early satiety with respect to eating, nausea/vomiting, jaundice, and weight loss [3]. The small PPs are spontaneously resolved, but the large PPs, especially those accompanied by chronic pancreatitis, are less likely to resolve spontaneously, causing secondary complications, such as infection, pseudoaneurysm ruptures, and fistulae between the pancreas and other organs [4, 5]. Although we, sometimes, experience each of these complications, the frequency of complicated cases, wherein several complications simultaneously occur, is rare.

Since these complications can be fatal, optimal treatment should be administered when such complications occur. However, we lack strong evidence regarding the optimal treatments for PPs and their complications, because randomized-controlled studies on these conditions might be unreliable given their rarity. Therefore, the accumulation of individual and complicated cases is still important to bridge this gap in knowledge. Herein, we report a case of PPs successively accompanied by an infection, a pseudoaneurysm rupture, and a pseudocystocolonic fistula.

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Case report

An 82-year-old man presented with fever and left back pain, and was admitted to our hospital. He began to experience fever and left back pain 7 days prior to admission. He drank approximately 60 g of alcohol daily and had a history of alcohol-induced acute pancreatitis. Physical examinations revealed slight fever but no abdominal pain. The results of the laboratory tests on admission were as follows: white blood cell count, 13,900/mm^3; C-reactive protein level, 22.1 mg/dL; amylase level, 743 U/L, and lipase level, 553 U/L. Upon admission, contrast-enhanced computed tomography (CT) was performed, revealing two PPs, one each in the pancreatic head and tail, surrounded with inflamed tissue (Fig. 1a, white arrow and Fig. 1b, white arrow head); he was, therefore, diagnosed with infectious PPs. A pancreatic calculus was also detected on CT, implying the presence of chronic pancreatitis. Upon the discovery of the infectious PPs, an antibiotic (meropenem, 1.5 g/day) was administered; however, the inflammation persisted (Fig. 2). Therefore, we performed endoscopic retrograde pancreatography, which showed that the PP in the pancreatic tail was not connected to the main pancreatic duct (MPD), but the PP in the pancreatic head was (Fig. 3a). Endoscopic nasopancreatic drainage (ENPD) using a 5-Fr nasal pancreatic drainage tube was performed to drain the PP in the pancreatic head at 14 day post-admission (Fig. 3b), and the dose of meropenem was increased to 3.0 g/day at 18 days post-admission. Although the PP in the pancreatic head shrunk as a result of the drainage (Fig. 3c, white arrow), the PP in the pancreatic tail did not and inflammation persisted. Therefore, endoscopic ultrasound (EUS)-guided transluminal drainage was performed at 28 day post-admission to treat the PP in the pancreatic tail. EUS examination did not detect necrotic tissue in the PP of the pancreatic tail (Fig. 4a). A 7-Fr double pig-tail plastic stent and 5-Fr nasocystic tube were inserted in the pancreatic tail PP from the stomach (Fig. 4b). ENPD fell out during EUS-guided transluminal drainage. Since a culture of the pancreatic tail PP content showed the presence of methicillin-resistant *Staphylococcus aureus*, vancomycin (1.5 g/day) was administered to treat the infection. Subsequently, the PP in the pancreatic tail shrunk, and the infection was completely cured.

The clinical course appeared favorable after the drainage of the pancreatic tail PP, but the patient suddenly vomited blood at 42 day post-admission. Although the stomach contained coagulated blood, there were no ulcers in the stomach (Fig. 5a). Contrast-enhanced CT showed coagulated blood in the stomach (Fig. 5b, white arrow) and the PP in the pancreatic tail (Fig. 5b, whited dotted circle), but no extravasation was detected and we could not detect the cause of bleeding by CT. Angiography was then performed to identify the bleeding site and stop the hemorrhage. Celiac arteriography revealed a pseudoaneurysm in a branch of the left gastric artery (Fig. 5c). The patient was then diagnosed with pseudoaneurysm rupture. Angioembolization was successfully performed for pseudoaneurysm rupture, and blood flow into the pseudoaneurysm completely disappeared (Fig. 5d). However, at 44 day post-admission, hematochezia occurred, which we suspected was the result of re-bleeding from the pseudoaneurysm which we had previously embolized. A CT scan of the pancreas revealed no extravasation, implying spontaneous hemostasis and a pseudocystocolonic fistula that was not present at the initial presentation (Fig. 6a). Angiography was not performed at this time, because hematochezia had ceased, implying the cessation of re-bleeding. A radiograph of the pancreatic tail pseudocyst, recorded from the nasocystic tube, confirmed the presence of a pseudocystocolonic fistula, which was thought to have formed when re-bleeding occurred (Fig. 6b). We recommended interventional therapies for the pseudocystocolonic fistula, but the patient rejected to undergo additional therapies, such as surgery or endoscopic closure of the fistulae. He was discharged

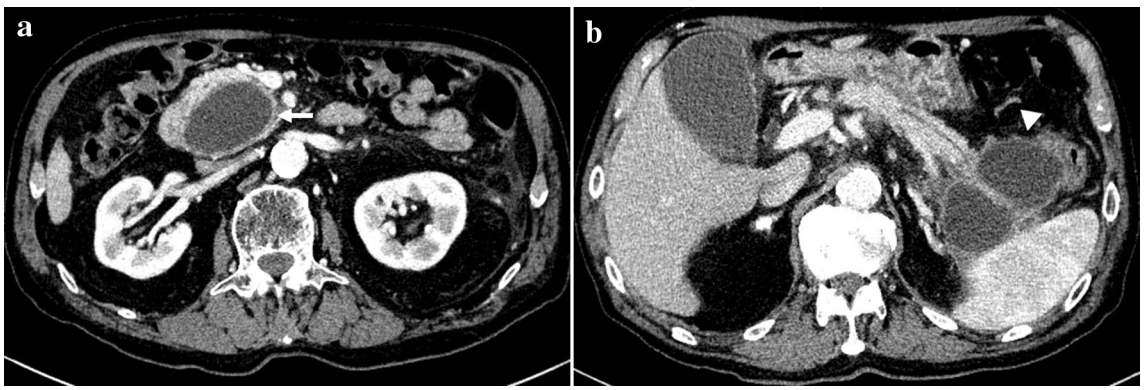

Fig. 1 Contrast-enhanced computed tomography of the patient's pancreas on admission. **a** A pancreatic pseudocyst in the pancreatic head. **b** A pancreatic pseudocyst in the pancreatic tail

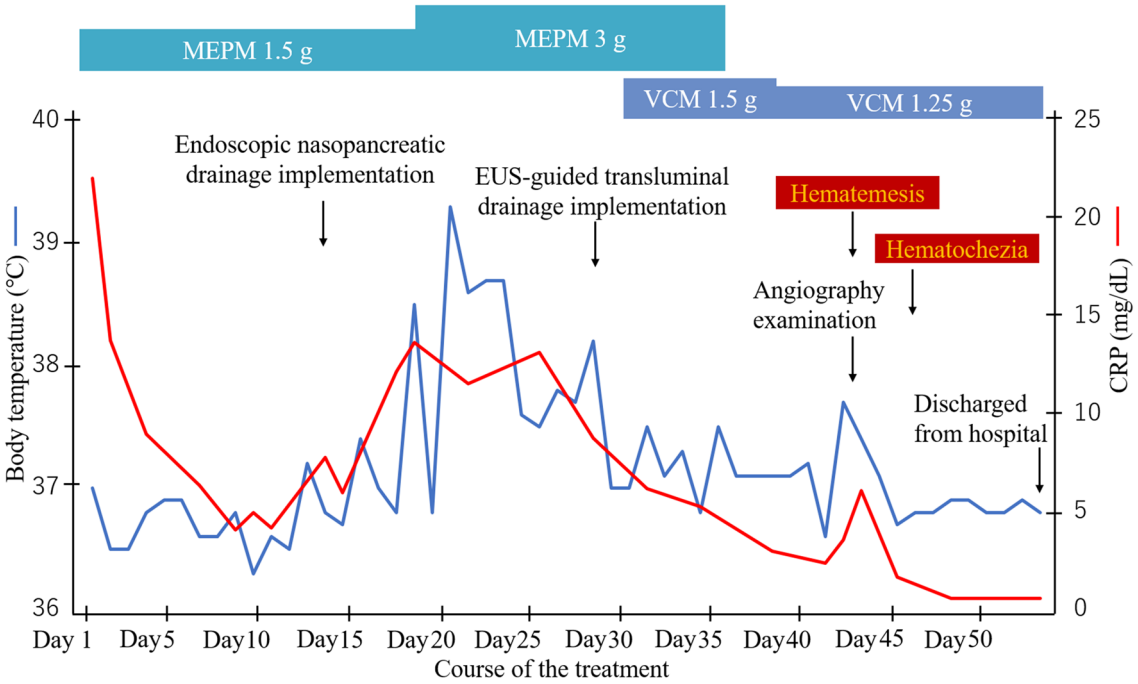

Fig. 2 Clinical course of the patient as represented by the line graph. The blue line shows the course of body temperature and the red line shows the course of the CRP levels. Inflammation improved promptly after EUS-guided transluminal drainage. Hematemesis occurred at 42 day post-admission and angioembolization was performed. 2 days after the procedure, hematochezia occurred, which stopped spontaneously. He was discharged at 54 day post-admission. *CRP* C-reactive protein, *EUS* endoscopic ultrasound, *MEPM* meropenem, *VCM* vancomycin

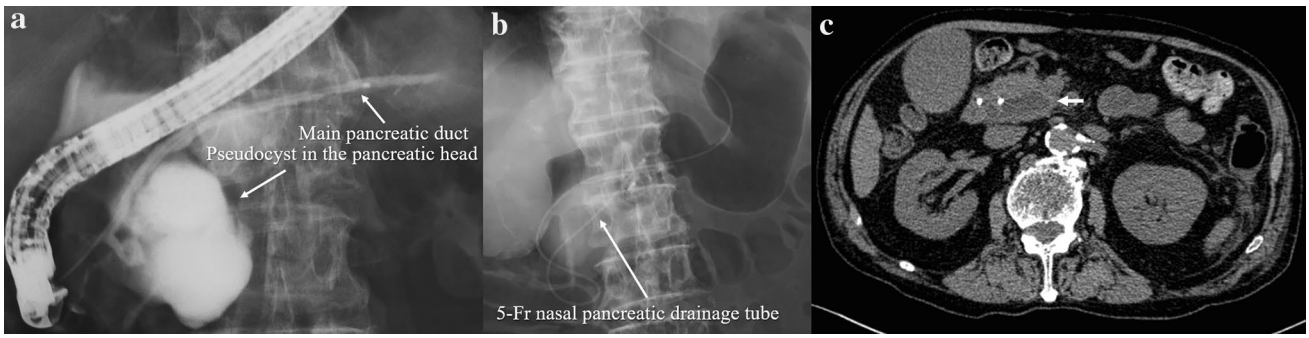

Fig. 3 Endoscopic retrograde pancreatography implementation. **a** Pancreatography showing the main pancreatic duct and the pancreatic pseudocyst in the pancreatic head. **b** A 5-Fr nasal pancreatic drainage tube was placed in the main pancreatic duct to drain the pancreatic pseudocyst in the pancreatic head. **c** Computed tomography scan showing a shrunk PP in the pancreatic head (white arrow)

at 54 day post-admission and has not had any pseudocystocolonic fistula-related symptoms at 9 month post-discharge.

Discussion

PPs are encapsulated well-defined collections of fluid that lack necrotic tissue and contain pancreatic fluid [1]. Some PPs can result in pain, discomfort, and secondary complications, such as infection, fistulae between the pancreas and adjacent organs, biliary obstruction, and pseudoaneurysm ruptures [6]. Interventional treatments are typically administered when these symptoms and complications develop. In our case, the infectious PPs, which first required treatment via endoscopic drainage, were followed by the development of a pseudoaneurysm rupture and pseudocystocolonic fistula. Such a complicated case is rare and requires multidisciplinary treatment.

For infected PPs, drainage is needed to control infection. There are several drainage methods that can be applied,

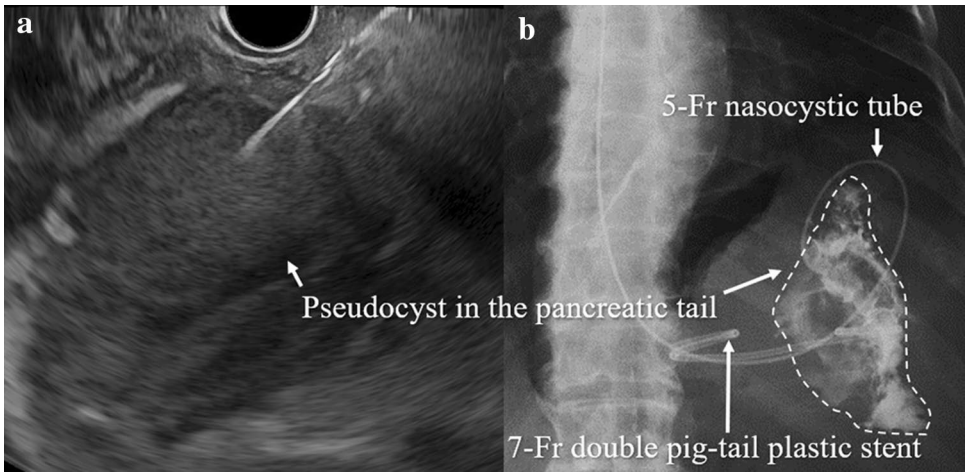

Fig. 4 Endoscopic ultrasound-guided transluminal drainage results. **a** An endoscopic ultrasound image showing no necrotic components in the pseudocyst present in the pancreatic tail. **b** A radiographic image of the retention of urografin in the pancreatic pseudocyst in the pancreatic tail (white dot circle). A 7-Fr double pig-tail plastic stent and 5-Fr nasocystic tube were inserted in the pseudocyst in the pancreatic tail from the stomach

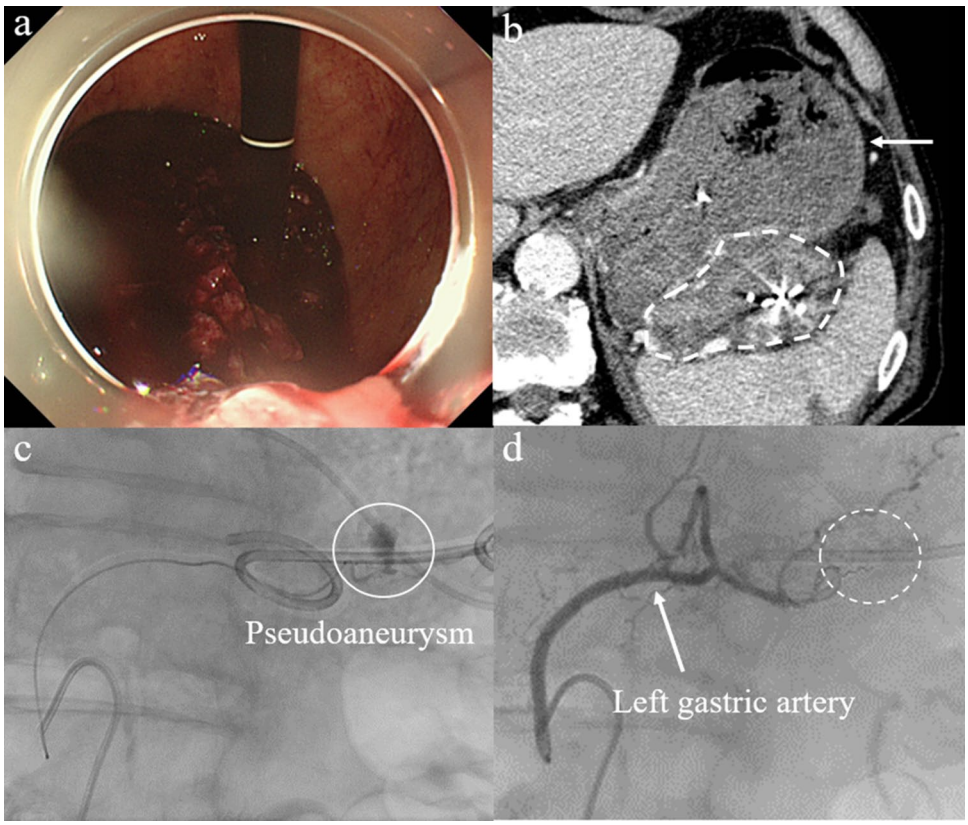

Fig. 5 Upper gastrointestinal endoscopy and angiography results. **a** An endoscopic image of the patient's stomach filled with coagulated blood. **b** Computed tomography scan showing coagulated blood in the stomach (white arrow) and PP in the pancreatic tail (white dotted circle). **c** An angiographic image of the pseudoaneurysm in a branch of the left gastric artery (white circle). **d** An angiographic image showing that blood did not flow into the pseudoaneurysm after angioembolization (white dotted circle)

including surgical drainage, percutaneous drainage, conventional endoscopic transluminal drainage, and EUS-guided transluminal drainage. Surgical and percutaneous drainage are traditional effective treatments for infected PPs, and their treatment success rates (i.e., cyst resolution rates) are equivalent to those of endoscopic drainage (surgery, 90.9–93.8% vs. endoscopy, 84.6%) [7]. However, endoscopic drainage, especially EUS-guided drainage, is preferable, because endoscopic techniques have become more effective and less invasive over the years [8–11]. EUS-guided transluminal drainage can identify and avoid vascular structures between cysts and the gastric lumen, thereby providing a more accurate assessment of the PP content and location of the non-bulging pseudocysts than the conventional endoscopic drainage [12]. The treatment success rates of EUS-guided transluminal drainage ranges from 80 to 100%, and the complication rate ranges from 5 to 16% [8–10, 13–15].

Pseudoaneurysm rupture is a rare but life-threatening complication of PPs. The incidence of pseudoaneurysm ruptures in PP cases ranges from 4 to 10%, and the associated

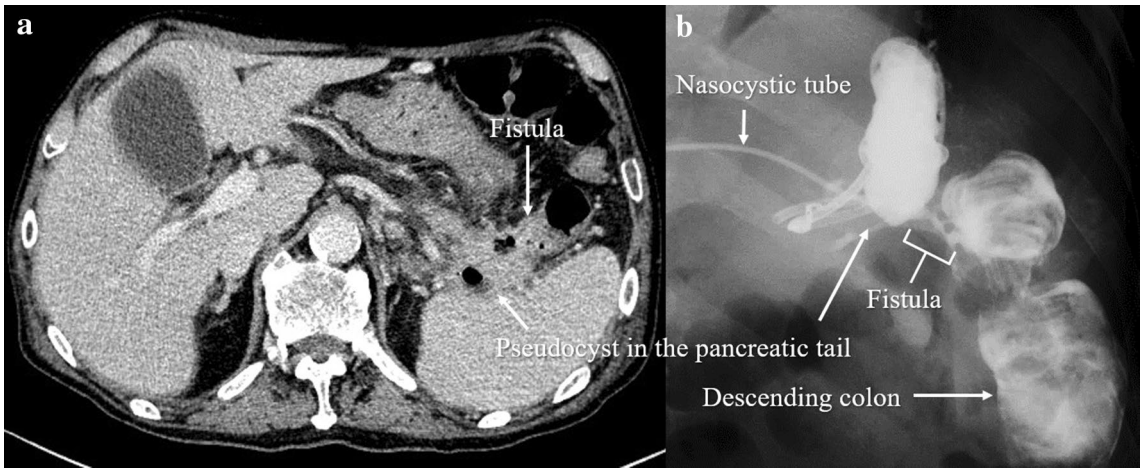

Fig. 6 Contrast-enhanced computed tomography and radiography results after hematochezia. **a** A contrast-enhanced computed tomography image showing the shrunken pseudocyst in the pancreatic tail, along with the pseudocystocolonic fistula. No extravasation was detected. **b** Radiography image showing the pseudocystocolonic fistula

mortality rate may be as high as 90% without appropriate treatment [16]. There are two treatment options for pseudoaneurysm ruptures, namely, angioembolization and surgery. The success rate of angioembolization is 80% for bleeding pseudoaneurysm ruptures around the pancreatic head and 50% for those around the splenic artery. The complication and mortality rates of angioembolization are 17.4 and 6%, respectively [17]. Since the initial surgery for pseudoaneurysm ruptures is associated with high motility rates in emergency situations, surgery should only be considered when angioembolization fails [16, 18].

Pseudocystocolonic fistulae are also rare and serious complications of PPs. The spontaneous closure of pseudocystocolonic fistulae cannot be expected for most cases. Pseudocystocolonic fistulae have mortality rates that range between 17 and 67%, and can be the cause of persistent infection, sepsis, and bleeding [19–22]. Therefore, the surgical or endoscopic treatment of fistulous tracts is recommended once pseudocystocolonic fistulae form. Although surgery is the traditional treatment method for pseudocystocolonic fistulae, less invasive endoscopic techniques and devices, including ligation bands, endoloops, fibrin glue, hemoclips, or a combination of these therapies, have been reported to be of use [23, 24]. Moreover, the usefulness of over-the-scope clips for fistula closure has also been reported recently [25]. While surgery or endoscopic treatment was performed in reported cases, neither surgery nor endoscopic treatment was performed in our case, since the patient did not want further interventional treatment. Fortunately, the patient has not had any detrimental pseudocystocolonic fistula-related events to date. Our case is important, since it shows the long-term natural course of pseudocystocolonic fistula without treatment, which is not well understood.

In conclusion, we successfully managed the complications of PPs in our case via a multidisciplinary approach. As described in the previous reports, endoscopic drainage was effective for infectious PP and angioembolization was effective for pseudoaneurysm rupture. Although surgery or endoscopic therapy is recommended for pseudocystocolonic fistulae, careful follow-up may be a treatment option in patients without pseudocystocolonic fistula-related symptoms. Complications of PPs can occur simultaneously and can be fatal if not treated appropriately. Therefore, clinicians should keep these complications in mind when treating patients with PPs and should promptly choose the optimal treatment appropriate for the patients' condition. We believe that our case report provides further evidence regarding the optimal interventional methods for similar cases.

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Compliance with ethical standards

Conflict of interest None of the authors have a conflict of interest.

Human/animal rights All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008(5).

Ethics approval Ethical approval was not required.

Informed consent Written informed consent was obtained from the patient.

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