Three cases of mediastinal pancreatic pseudocysts

Eiji Matsusue¹, Yoshio Fujihara¹, Kazunori Maeda², Masaru Okamoto³, Atsushi Yanagitani², Kiwamu Tanaka², Kazuhiko Nakamura¹ and Toshihide Ogawa⁴

Abstract
A rare complication of acute or chronic pancreatitis is the formation of a mediastinal pancreatic pseudocyst (MPP), which is caused by tracking of pancreatic fluids through anatomical openings of the diaphragm into the mediastinum. Herein, we report the imaging characteristics of three cases of this condition. Our results revealed three features in common: (i) the connection between the mediastinum and the pancreatic cystic lesion; (ii) the presence of pleural effusions; and (iii) imaging findings consistent with chronic pancreatitis, such as pancreatic atrophy and calcifications and dilatation and/or stricture of main pancreatic duct (MPD). Serial diameter changes of the MPD and of the adjacent pseudocysts were necessary for the determination of the therapeutic strategy used in each case.

Keywords
Pancreatic pseudocyst, mediastinal extension, thoracopancreatic fistula, management strategy, computed tomography (CT)

Introduction
Pancreatic pseudocysts are often caused by acute or chronic pancreatitis. Mediastinal pancreatic pseudocysts (MPPs) are rare complications of pancreatitis (1,2). An inflammatory disruption of the pancreatic duct leads to leakage of pancreatic secretions. Anterior ductal disruption often results in pancreatic ascites, whereas posterior ductal disruption results in thoracopancreatic fistula, which can be classified into MPP, pancreatobronchial fistula, or pancreatico-pericardial fistula (3). Therefore, MPPs result from retroperitoneal extension of pancreatic fluids through the anatomical openings of the diaphragm into the mediastinum (1,4). MPPs may present with a triad of chest pain, dysphagia, and dyspnea (4), although their diagnosis is sometimes difficult to make, especially in cases without abdominal pain.

A definitive diagnosis can be made by imaging modalities, such as computed tomography (CT) or magnetic resonance imaging (MRI), showing cystic lesions extending from the pancreas into the mediastinum (3,5–9). Management of MPPs depends upon their size, quantity, location, and relationship to adjacent anatomical structures, as well as the presence of communication of the cyst with the pancreatic duct (10,11). Furthermore, draining pancreatic fluid, which reduces pressure within the pseudocysts and pancreatic duct, is also important in managing patients with MPPs (4,10).

We present three cases of MPPs and describe their clinical settings and imaging features.

¹Department of Radiology, Tottori Prefectural Central Hospital, Tottori, Japan
²Department of Gastroenterology, Tottori Prefectural Central Hospital, Tottori, Japan
³Department of General Medicine, Tottori Prefectural Central Hospital, Tottori, Japan
⁴Division of Radiology, Department of Pathophysiological Therapeutic Science, Tottori University, Tottori, Japan

Corresponding author:
Eiji Matsusue, Department of Radiology, Tottori Prefectural Central Hospital, 730 Ezu, Tottori, Tottori 680-0901, Japan.
Email: matsusuee@pref.tottori.jp
Case reports

Case 1

A 58-year-old man presented with 3 weeks of dysphagia, which was experienced predominantly with solid foods. He had lost 3 kg in the last month. He had no past medical history. He had been a heavy drinker (1 L of beer per day) and a heavy smoker (20 cigarettes per day for 35 years). Laboratory tests showed the following: 162 U/L serum amylase (normal value <37–125 U/L), 160 IU/L serum lipase (normal value <6–53 U/L), 172 mg/dl blood glucose, 8040/µL white blood cells, and 10.61 mg/dl C-reactive protein (CRP). Tumor markers (SCC, CEA, and CA19-9) were negative.

Upper gastrointestinal endoscopy showed mild narrowing of the lower esophagus. On chest and abdominal CT, cystic lesions extended from the abdomen into the mediastinum along with the esophagus and aorta, presumably via the esophageal and aortic hiatus. One of the mediastinal cysts also extended into the retrocrural space. Bilateral pleural effusions were seen. One of the abdominal cysts was connected to the pancreatic body. The main pancreatic duct (MPD) in the pancreatic tail was slightly dilated. A calcification was seen in the body of the pancreas (Fig. 1A–C). A laparoscopic lymph node biopsy was performed due to the possibility of necrotic lymph node metastases. There were no malignant cells in the specimens. Conservative antibiotic therapy was administered. Two months’ symptom after onset, repeat chest and abdominal CT images showed that the size of each cystic lesion had diminished. The patient was then diagnosed with pancreatic pseudocysts caused by chronic pancreatitis. Eight months after symptom onset, he returned with complaints of upper left abdominal pain. An abdominal CT indicated that the size of the cyst connected with the pancreatic body had increased (Fig. 1D). CT-guided percutaneous drainage of the pancreatic cyst was performed and the size of the cyst was reduced. There was no increase in the size of the cyst upon further follow-up CT. He currently receives regular outpatient treatment.

![Pancreatic pseudocysts in Case 1. (A) Axial enhanced chest CT at symptom onset. Cystic lesions (arrows) extending from the abdomen into the mediastinum along with the esophagus and aorta. Bilateral pleural effusions are seen. The esophagus (arrowhead in white) is located behind the cystic lesion (short arrow). (B) Coronal enhanced lower chest and upper abdominal CT at symptom onset. The connection between the mediastinal and abdominal cystic lesions (short arrows) is well identified on the coronal image. One of the cysts (arrowhead in black) is connected to the pancreas body (arrowhead in white). (C) Axial enhanced upper abdominal CT at symptom onset. The pancreatic cyst with the black arrowhead is the same as the cyst with the black arrowhead noted above (B). A calcification is seen in the pancreas body. The main pancreatic duct (MPD) of the pancreas tail is slightly dilated (arrow). The cystic lesion (long arrow) extends into the retrocrural space. (D) Axial enhanced upper abdominal CT obtained 8 months after symptom onset. The size of the pancreatic cyst is enlarged (arrowhead in black).](image-url)
Case 2

A 65-year-old man presented with 3 months of abdominal fullness along with 3 days of continuous fever of approximately 38°C. He had no past medical history. He had been a heavy drinker (1.5 L of beer per day). Laboratory tests showed the following: 1259 U/L serum amylase, 819 IU/L serum lipase, 2.5 g/dl serum albumin, 9180/µL white blood cells, and 24.9 mg/dl CRP. Tumor markers (CEA and CA19-9) were negative. Upper gastrointestinal endoscopy did not show any remarkable findings. Chest and abdominal CT showed fluid collections with cystic lesions extending from the abdomen into the mediastinum along the esophagus via the esophageal hiatus. The connection between the mediastinal and abdominal lesions was well identified on a sagittal image (Fig. 2A). Bilateral pleural effusions were seen. Abdominal CT and MRI showed a small cyst and a slightly dilated MPD in the pancreatic body in addition to the peripancreatic cystic lesions (Fig. 2B). The diagnosis of pancreatic pseudocyst was made. Under conservative antibiotic therapy, the mediastinal fluid collection and cystic lesions disappeared, although the peripancreatic cystic lesions were still present. Therefore, CT-guided percutaneous drainage of the dorsal cyst of the pancreatic body was performed along with the placement of a pigtail drainage catheter. Furthermore, an MPD stricture was seen in the pancreatic body on endoscopic retrograde cholangiopancreatography (ERCP). Consequently, a tube stent (7 Fr) was endoscopically placed in the MPD (Fig. 2C). Three months later, amylase and lipase levels had normalized and the tube stent was removed. Eight months later, an abdominal CT indicated that both the cyst and the adjacent MPD were again slightly enlarged (Fig. 2D). Finally, elective pancreaticojejunostomy of the body and tail of the pancreas was performed in another hospital.

Case 3

A 67-year-old man presented with 1 month of abdominal fullness and appetite loss and with several days of cough, sputum, and dyspnea. The patient had acute pancreatitis 3 years ago. He was also operated on for left lung cancer (stage IIIA) 2 years ago. He had been a heavy drinker (1 L of beer per day) and a heavy smoker.

![Fig. 2. Pancreatic pseudocysts in Case 2. (A) Sagittal enhanced chest and upper abdominal CT. Cystic lesions (arrows) extending into the mediastinum along the esophagus (arrowheads in white). The connection between the mediastinal and abdominal cystic lesions (arrows) is well identified on the sagittal image. (B) Axial upper abdominal fat-suppressed T2-weighted image at symptom onset. Cystic lesions (arrows) are seen in the peripancreatic region. A small cyst is seen in the pancreas body (arrowhead in black). The adjacent MPD is slightly dilated (arrowhead in white). (C) Axial unenhanced upper abdominal CT obtained one month after symptom onset. A tube stent (arrowhead in white) is placed in the MPD. A pigtail drainage catheter (arrow) is placed in the dorsal cyst of the pancreas body. (D) Axial enhanced upper abdominal CT obtained 1 year after symptom onset. Both the cyst (arrowhead in black) and the adjacent MPD (arrowhead in white) are enlarged again. The dorsal cyst of the pancreas body has disappeared (arrow).]
(30 cigarettes per day for 45 years). Laboratory tests showed the following: 816 U/L serum amylase, 338 IU/L serum lipase, 11.8 g/dl hemoglobin, 8000/μL white blood cells, and 15.39 mg/dl CRP. Tumor markers (CEA and CA19-9) were negative. On chest and abdominal CT, fluid collections with cystic lesions extending from the abdomen into the mediastinum along the esophagus and inferior vena cava, presumably via the esophageal hiatus and inferior vena cava hiatus, were noted. Bilateral pleural effusions, pericardial effusions, and ascites were also seen. The connection between the cysts (arrows) around the liver (asterisk) and the esophagus (arrowheads in white) and the pancreas head with multiple cysts (short arrows) is well identified on the sagittal image. (B) Sagittal enhanced lower chest and upper abdominal CT at symptom onset. The connection between the cysts (arrows) around the liver (asterisk) and the esophagus (arrowheads in white) and the pancreas head with multiple cysts (short arrows) is well identified on the sagittal image. (C) Axial enhanced upper abdominal CT at symptom onset. The pancreas head with multiple cysts (short arrow) and calcification (long arrow) is seen. MPD dilatation (arrowhead in white) is seen in the pancreas body and tail. MPD stricture (arrowhead in black) is suspected in the pancreas head. (D) ERCP obtained 1 week after symptom onset. Dilatation and irregularity of the side branches (short arrows in white) and an MPD stricture (arrowhead in black) are seen in the pancreatic head region.

Endoscopic pancreatic ductal drainage was considered challenging. Elective pancreaticojejunostomy was performed in another hospital.

Discussion

Pancreatic pseudocysts are a major complication resulting from chronic or acute pancreatitis. Eighty percent of pancreatic pseudocysts are located within the head and body of the pancreas, and 20% are extrapancreatic and located in areas such as the pleura, mediastinum, liver, spleen, and pelvis (12). MPPs belong to the category of thoracopancreatic fistula and occur as a result of posterior rupture of the pancreatic duct into the retroperitoneal space. Pancreatic fluids extend upward through anatomical openings of the diaphragm into the mediastinum. The fluids typically travel through the esophageal and aortic hiatus into the posterior mediastinum (1,4). If the diaphragm is penetrated through the inferior vena cava hiatus or the foramen of Morgagni, the fluid extends into the middle and anterior mediastinum, respectively (13).
Related symptoms may include chest pain, dysphagia, dyspnea secondary to an accompanying pleural effusion, and weight loss secondary to dysphagia. A history of pancreatitis or alcoholism may help in the diagnostic workup, although the above symptoms are rarely helpful as most mediastinal masses will present with similar symptoms due to compression of adjacent structures (10). In our three cases, the following symptoms were seen: dysphagia and weight loss in case 1, abdominal fullness and fever in case 2, and abdominal fullness, appetite loss, cough, sputum production, and dyspnea in case 3. Therefore, it was difficult to suspect pancreatitis in all of our cases according to their symptoms at their initial presentations, although all of them were middle-aged men and heavy drinkers.

The definitive diagnosis is based on imaging studies showing cystic lesions extending from the pancreas into the mediastinum. CT is most commonly used to establish the diagnosis, and it also helps to depict pancreatitis and the connection between the mediastinal cystic structures and the pancreas (3–8). MRI and magnetic resonance cholangiopancreatography (MRCP) provide further details of pancreatic ductal anatomy (3,6,9,10). Recent advances in endoscopic techniques, such as endoscopic ultrasound (EUS), are helpful in evaluating the mediastinal mass and cysts and also play an important therapeutic role by allowing EUS-guided aspiration and drainage of the cysts (14). In all of our cases, chest and abdominal CT showed mediastinal cystic lesions connected with pancreatic or peripancreatic cystic lesions via anatomical openings in the diaphragm. The abdominal cystic lesions connected with the posterior mediastinum via the esophageal hiatus in all cases, connected with the posterior mediastinum via the aortic hiatus and retrocrural space in case 1 and connected with the middle mediastinum via the inferior vena cava hiatus in case 3. Bilateral pleural effusions were also seen in all cases. Pancreatic atrophy or focal swelling, pancreatic calcifications, dilatation, and/or stricture of the MPD and pancreatic pseudocysts, whose findings are key findings of chronic pancreatitis, were seen in all cases on abdominal CT. The connection between mediastinal and abdominal lesions and various extension routes of pancreatic fluids through the diaphragm into the mediastinum were well identified on axial thin slice images and on coronal and/or sagittal reconstructed images. Furthermore, MRI and MRCP provided further details of pancreatic ductal dilatation and/or stricture.

Management of MPPs depends on their size, quantity, location, relationship with adjacent anatomical structures, the presence of infection or hemorrhage, the presence of communication of the cyst with the pancreatic duct, and symptom severity (10,11). Spontaneous regression is rare but has been reported to occur with complete abstinence from alcohol and with the use of total parenteral nutrition (15). Successful resolution of the pseudocysts after medical therapy, such as octreotide and bromhexine hydrochloride, has also been reported (16,17). Draining pancreatic fluid and transpapillary stenting of the pancreatic duct are useful treatments that reduce the high pressures associated with pseudocysts and pancreatic duct narrowing (4,10). CT-guided percutaneous drainage or EUS-guided drainage is widely performed for draining pancreatic fluid. ERCP with transpapillary stenting of the pancreatic duct can be used when the pseudocyst communicates with the pancreatic duct (4,10,18–20). Surgical treatment should be performed for patients with failure of the abovementioned less-invasive therapies, as well as patients with bleeding, infection, pseudocyst rupture, and acute life-threatening presentations. Surgical options include internal drainage, external drainage, cyst-enterostomy, or pancreatic resection (21,22). In our three cases, CT and MRI were useful imaging modalities for evaluating the MPD diameter, the site of stenosis, the length of stricture, and the size and location of the pseudocysts. The use of these modalities allowed accurate evaluations of the sites of the pancreatic ductal stricture and of the serial changes in these lesions, which were necessary for the determination of the therapeutic strategy used in each case.

In conclusion, we present three cases of MPP and describe their clinical settings and radiological imaging features. A definitive diagnosis could be made via imaging modalities, such as CT or MRI, showing cystic lesions extending from the pancreas into the mediastinum. Precise assessments of pancreatic ductal stricture and pseudocysts and sufficient decompression of these lesions are essential in managing patients with MPP.

Declaration of conflicting interests
The author(s) declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding
The author(s) received no financial support for the research, authorship, and/or publication of this article.

References
1. Cameron JL. Chronic pancreatitis ascites and pancreatic pleural effusions. Gastroenterology 1978;74:134–140.
2. Rose EA, Haider M, Yang SK. Mediastinal extension of pancreatic pseudocyst. Am J Gastroenterol 2000;95:3638–3639.
3. Tajima Y, Tsutsumi R, Kuroki T, et al. Evaluation and management of thoracopancreatic fistula. Surgery 2006;140:773–778.
4. Gupta R, Munoz JC, Garg P, et al. Mediastinal pancreatic pseudocyst: a case report and review of the literature. Med Gen Med 2007;9:8.
5. Kirchner SG, Heller RM, Smith CW. Pancreatic pseudocyst of the mediastinum. Radiology 1977;123:600–606.
6. Fulcher AS, Capps GW, Turner MA. Thoracopancreatic fistula: clinical and imaging findings. J Comput Assist Tomogr 1999;23:181–187.
7. Ito H, Matsubara N, Sakai T, et al. Two cases of thoracopancreatic fistula in alcoholic pancreatitis: clinical and CT findings. Radiat Med 2002;20:207–211.
8. Andrén-Sandberg A, Dervenis C. Pancreatic pseudocyst in the 21st century. Part I: classification, pathophysiology, anatomic considerations and treatment. J Pancreas 2004;5:8–24.
9. Tajima Y, Fukuda K, Tsuneoka N, et al. Demonstration of a pancreaticopleural fistula with magnetic resonance cholangiopancreatography. Am J Surg 2004;187:741–742.
10. Ajmera AV, Judge TA. Mediastinal extension of pancreatic pseudocyst: a case with review of topic and management guidelines. Am J Ther 2012;19:e152–156.
11. Zhang AB, Zheng SS. Treatment of pancreatic pseudocysts in line with D'Egidio’s classification. World J Gastroenterol 2005;11:729–732.
12. Wang SJ, Chen JJ, Changchien CS. Sequential invasions of pancreatic pseudocysts in pancreatic tail, hepatic left lobe, caudate lobe, and spleen. Pancreas 1993;8:133–136.
13. Xu H, Zhang X, Christie A, et al. Anatomic pathways of peripancreatic fluid draining to mediastinum in recurrent acute pancreatitis: visible human project and CT study. PLoS One 2013;8:e62025.
14. Bhasin DK, Rana SS, Chaudhary VS, et al. Successful resolution of a mediastinal pseudocyst and pancreatic pleural effusion by endoscopic nasopancreatic drainage. J Pancreas 2005;6:359–364.
15. Frenzer A, Schubarth P, Soucek M, et al. Disappearance of a large mediastinal pseudocyst in a patient with chronic alcoholic pancreatitis after total parenteral nutrition. Eur J Gastroenterol Hepatol 1995;7:369–371.
16. Yasuda H, Ino Y, Igarashi H, et al. A case of pancreatic pleural effusion and mediastinal pancreatic pseudocyst: Management by a somatostatin analogue octreotide. Pancreas 1999;19:410–412.
17. Tsujimoto T, Takano M, Tsuruzono T, et al. Mediastinal pancreatic pseudocyst caused by obstruction of the pancreatic duct was eliminated by bromhexine hydrochloride. Intern Med 2004;43:1034–1038.
18. Brahmabh C, McKinney J, Litchfield J, et al. Mediastinal pancreatic pseudocyst with hemorrhage and left gastric artery pseudoaneurysm, managed with left gastric artery embolization and placement of percutaneous trans-hepatic pseudocyst drainage. Gastroenterol Rep (Oxf) 2014; pii gout084 [Epub ahead of print].
19. Mallavarapu R, Habib TH, Elton E, et al. Resolution of mediastinal pancreatic pseudocysts with transpapillary stent placement. Gastrointest Endosc 2001;53:367–370.
20. Kim DJ, Chung HW, Gham CW, et al. A case of complete resolution of mediastinal pseudocyst and pleural effusion by endoscopic stenting of pancreatic duct. Yonsei Med J 2003;30:727–7317.
21. Gots L, Agocs L, Kostic S. Transdiaphragmatic cystojejunostomy with Roux-en-Y loop for an exclusively mediastinal pancreatic pseudocyst. Scand J Thorac Cardiovasc Surg 1996;30:181–183.
22. Bardia A, Stoikes N, Wilkinson NW. Mediastinal pancreatic pseudocyst with acute airway obstruction. J Gastrointest Surg 2006;10:146–150.