CASE REPORT

Left pleural effusion caused by pancreaticopleural fistula with a pancreatic pseudocyst

Takanobu Hirosawa,[1] Taro Shimizu,[1] Takuya Isegawa,[2] Mayo Tanabe[3]

[1]Department of Diagnostic and Generalist Medicine, Dokkyo Medical University, Tochigi, Japan
[2]Division of Nephrology and Endocrinology, The University of Tokyo, Tokyo, Japan
[3]Showa University Koto Toyosu Hospital, Digestive Diseases Center, Tokyo, Japan

Correspondence to
Dr Takanobu Hirosawa,
t.hirosawa1983@gmail.com

Accepted 7 August 2016

SUMMARY

Pancreaticopleural fistula is an uncommon complication of chronic pancreatitis. The authors described a case of a man with medical history of alcohol-related chronic pancreatitis, presented with dyspnoea. The roentgenogram showed a massive left pleural effusion. Additional work-up revealed a pancreaticopleural fistula and amylase-rich pleural effusion. His respiratory state improved after the insertion of chest drainage tube. During his admission, conservative and endoscopic therapy was required for the treatment of his complication of mediastinal abscess and arterial aneurysm in the pancreatic pseudocyst.

BACKGROUND

Pancreaticopleural fistula (PPF) is an uncommon complication of chronic pancreatitis. It presents as a massive pleural effusion. The fistula is commonly associated with alcohol-related chronic pancreatitis. The available management of PPF includes: medical therapy, endoscopic and surgical intervention. This case is important because his medical history and previous symptom of chest pain suggested the rupture of the cystic lesion. Furthermore, a clinician can suspect the presence of PPF, when there is a high level of amylase in the pleural fluid on analysis.

CASE PRESENTATION

A 58-year-old man presented with left side chest pain that started a week ago. He could not drink since that day. His pain improved spontaneously; thereafter, he developed progressive exertional dyspnoea. He had a history of alcohol-related chronic pancreatitis and occasional pancreatic pseudocyst.

He was a heavy drinker of alcohol (3 cups of Awamori daily, which is a local alcohol drink in Okinawa, average alcohol content 25%), and had a smoking history of 38 pack-years. His medication and family history were unremarkable.

His vital signs at the time of presentation were as follows: blood pressure of 170/112 mm Hg, heart rate of 120 bpm, regular, temperature 96.8°F, respiratory rate of 30 breaths/min and arterial oxygen saturation of 87% under room air. His jugular vein was distended and his trachea was deviated to right side. Auscultation of the lung revealed reduced left breath sound.

INVESTIGATIONS

A chest roentgenogram revealed decreased permeability in the left lung field and the deviated trachea (figure 1). A 12-lead ECG showed low voltage.

A contrast-enhanced CT scan of the chest and abdomen revealed massive left pleural effusion, causing compressive collapse of most of the left lung, displacement of the mediastinum to the right and pancreatic cystic lesion (figure 2).

Laboratory examination showed normal complete blood count and liver function test, and showed mild elevated amylase level (326 IU/L, normal range 44–132 IU/L) and erythrocyte sedimentation rate (13 mm/hour, normal range <10 mm/hour).

Thoracentesis fluid on left side was black in colour, and fluid analysis showed lactate dehydrogenase level 757 IU/L, total protein level 2.6 g/dL (26 g/L), glucose level 103 mg/dL (5.7 mmol/L) and haemoglobin 1.5 g/dL (15 g/L), consistent with an exudative pleural effusion. Additional test revealed elevated pleural amylase (10 649 IU/L). The cytology of pleural effusion was negative. All culture and stain for bacteria and mycobacteria were negative.

TREATMENT

The patient was managed conservatively with chest drainage. Endoscopic retrograde cholangiopancreatography (ERCP) was planned. The pancreatogram showed extravasation of contrast above the diaphragm (figure 3). A 5 French 5 cm plastic stent

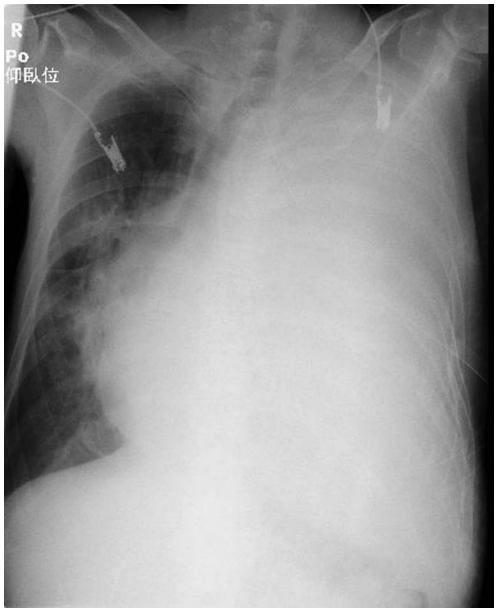

Figure 1 Chest roentgenogram showing decreased permeability in the left lung field and the deviated trachea.

To cite: Hirosawa T, Shimizu T, Isegawa T, *et al*. *BMJ Case Rep* Published online: [*please include* Day Month Year] doi:10.1136/bcr-2016-217175

Figure 2 Chest CT scan revealing pancreatic cystic lesion (arrows).

was placed in the pancreatic duct. On the 12th day of admission, after checking remission of pleural effusion, his chest tube was removed. On the 17th day of admission, he developed fever, and a repeat CT scan revealed mediastinum abscess formation (figure 4), and arterial aneurysm in the pancreatic pseudocyst (figure 5). A broad-spectrum antibiotic therapy was started. An emergency angiogram was performed for the aneurysm, and success for embolism for branch of left gastric artery. On the 19th day of admission, a 7 French endoscopic nasopancreatic drainage (ENPD) tube was placed in pancreatic pseudocyst. A repeat CT showed remission of mediastinum abscess. Repeat ERCP was undertaken, his ENPD tube was removed and, inserted 7 French 12 cm pancreatic stent.

OUTCOME AND FOLLOW-UP

After rehabilitation, he was discharged home. Outpatient follow-up were planned, but failed as the patient was lost to follow-up. Unfortunately he could not stop drinking alcohol, and dropped out of medical follow-up. After 2 years, he died because of unknown cause.

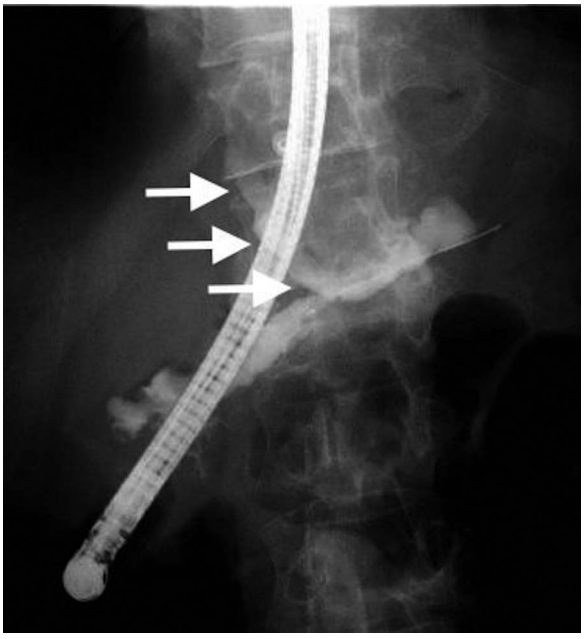

Figure 3 Pancreatogram showing extravasation of contrast above the diaphragm.

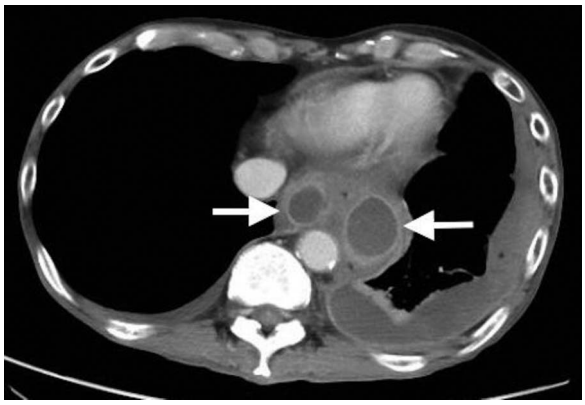

Figure 4 Chest CT scan revealing mediastinum abscess formation (arrows).

DISCUSSION

PPF is a rare clinical complication of pancreatitis.[1] Its incidence is estimated at 0.4% in patients with pancreatitis, and 4.5% in those presenting with a pancreatic pseudocyst.[2] The most common cause of pancreatitis leading to PPF is alcohol abuse, as in this case.[3]

The most common presenting symptoms of PPF were dyspnoea (65–76%), as seen in this case. Cough (27%), chest pain (23%) and fever are also seen with a lesser frequency.[4] From his history of illness, 7 days before admission, left-sided chest pain would be caused by the rupture of the pancreatic pseudocyst. Thereafter, he could not drink alcohol which he consumed on daily basis. Unintentional cessation of drinking alcohol by a heavy drinker may suggest his or her severe health problem. Increased pleural effusion would lead to progressive dyspnoea.

In this case, through intuitive diagnostic process, namely system 1 diagnostic process, we initially suspected aortic dissection based on his clinical presentation. However, this patient had a history of chronic pancreatic pseudocyst. Based on this, with an episode of sudden onset chest pain, we intuitively could have thought of pancreatic pseudocyst rupture. As our reflection from diagnostic errors, this case implies the importance of listing non-thoracic aetiologies as differential diagnoses even if the patient's symptom is located over the thoracic region. Another clinical implication/take home message is that rupture of the pancreatic pseudocyst can be a diagnosis in sudden onset of chest/abdominal pain in alcoholic patients.

Figure 5 Abdominal CT scan showing arterial aneurysm in the pancreatic pseudocyst (arrows).

Black pleural effusions are extremely rare. Aside from PPF, it has been reported to be associated with *Aspergillus niger* infection, *Rhizopus oryzae* infection, metastatic melanoma, haemorrhage, haemolysis and other causes (charcoal-containing empyema).[5]

The diagnosis of pleural effusion secondary to PPF is made based on the characteristics of pleural effusion with high fluid amylase level as well as imaging indicating a pseudocyst connecting with the thoracic space.

Patient's perspective

During the admission, I tried to stop drinking after discharge.

Learning points

- Unintentional cessation of drinking alcohol suggests severe problems for a heavy drinker.
- Amylase-rich pleural effusion would be black in colour.
- Remarkable elevation of pleural amylase is key for the diagnosis of pancreaticopleural fistula.

Available medical treatment options include chest drainage, nasojejunal tube, pancreatic replacement therapy and a somatostatin analogue. Endoscopic management includes ERCP with or without pancreatic stenting and nasopancreatic drainage followed by stenting. When medical and endoscopic management fail, surgical intervention is considered.[3] Our patient suffered several complications after the admission; mediastinum abscess formation and arterial aneurysm in the pancreatic pseudocyst. Fortunately, medical treatment and endoscopic management was effective and successfully cured the patient.

Twitter Follow Takuya Isegawa at @Takuya Isegawa

Contributors All authors conceived the paper. TH mainly wrote the paper. TS, TI and MT supervised the paper. All authors participated in writing of the paper, and TS approved the final version.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1 Burgess NA, Moore HE, Williams JO, *et al*. A review of pancreaticopleural fistula in pancreatitis and its management. *HPB Surg* 1992;5:79–86.
2 Wypych K, Serafin Z, Gałązka P, *et al*. Pancreaticopleural fistulas of different origin: report of two cases and a review of literature. *Pol J Radiol* 2011;76:56–60.
3 Ali T, Srinivasan N, Le V, *et al*. Pancreaticopleural fistula. *Pancreas* 2009;38:e26–31.
4 Aswani Y, Hira P. Pancreaticopleural fistula: a review. *JOP* 2015;16:90–4.
5 Saraya T, Light RW, Takizawa H, *et al*. Black pleural effusion. *Am J Med* 2013;126:641.e1–6.