

Pancreaticopleural and pancreaticomediastinal fistula extending to the cervical region, with dysphagia as initial symptom

A case report

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Abstract

Rationale: Pancreaticopleural and pancreaticomediastinal fistulas are rare complications of pancreatitis. They are often misdiagnosed and there are no strict guidelines of treatment. In this study, we present a brief report of a combined pancreaticopleural and pancreaticomediastinal fistula extending to the cervical region, causing dysphagia and cervical swelling as initial symptoms.

Patient concerns: A 36-year-old female with history of alcohol abuse and pancreatitis presented progressing dysphagia and mild dyspnea on admission.

Diagnosis: Chest X-ray and chest and abdominal computed tomography scan (CT) indicated pancreaticopleural fistula combined with pancreaticomediastinal fistula, a diagnosis confirmed by high amylase levels in pleural fluid.

Interventions: Conservative treatment was administered and ERCP was performed but pancreatic duct stenting was impossible. The patient presented rapid anterior cervical swelling with progressing dysphagia and dyspnea. CT showed fistula penetration to the cervical region. The patient underwent urgent surgery and pancreaticojejunal anastomosis was performed.

Outcomes: The surgery led to recovery. Six months later, the patient reported good health and weight gain.

Lessons: Coexistence of pancreaticopleural and pancreaticomediastinal fistula with cervical penetration is an extremely rare pancreatitis complication. It presents with dysphagia and anterior cervical swelling as initial symptoms. It is important to consider this complication in all patients with history of pancreatitis, presenting with dysphagia.

Abbreviations: CRP = C-reactive protein, CT = computed tomography, ERCP = endoscopic retrograde cholangiopancreatography, PLT = platelet count, PT = prothrombin time, WBC = white blood cell count.

Keywords: dysphagia, pancreaticomediastinal fistula, pancreaticopleural fistula, pancreatitis

1. Introduction

Pancreatitis, either acute or chronic, may lead to a leakage in the posterior wall of pancreatic duct, either directly from the duct or due to a ruptured pseudocyst. This causes pancreaticopleural and/or pancreaticomediastinal fistulas, rare complications affecting <1% of patients with chronic pancreatitis.^[1,2] The patients

are predominantly middle-aged men with a history of alcohol abuse. They usually suffer from massive and recurrent pleural effusion, with high fluid amylase levels, which may appear on either side with predominance of the left side.^[1] Many cases of pancreaticopleural fistulas and a few pancreatic pseudocysts penetrating to the mediastinum were described with complications such as mediastinitis, pneumonia, hemothorax, and heart tamponade.^[3–5] The main symptoms are dyspnea, dysphagia, and chest pain.^[6] Dysphagia rarely occurs as the presenting symptom and is more common in mediastinal fistulas.^[7,8]

This is a paper describing an extremely rare case of combined pancreaticopleural and pancreaticomediastinal fistula in a female patient with dysphagia and cervical edema as initial symptoms.

2. Case presentation

A 36-year-old female was admitted to our hospital, complaining of progressing dysphagia and mild dyspnea. Her medical history revealed acute alcoholic pancreatitis 7 years prior, followed by chronic pancreatitis with repeated exacerbations. The patient admitted to be a current smoker. She also had a history of duodenal ulcers, right ovarian cyst, and Wernicke's encephalopathy.

On physical examination, there was dullness on percussion in the left mid and lower lung as well as bronchial breathing in these areas. The abdomen was soft with epigastric tenderness on the left side. There were no palpable lymph nodes on the neck.

Editor: N/A.

Ethical approval: The patient gave her informed consent for the publication.

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

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Medicine (2019) 98:5(e14233)

Received: 4 October 2018 / Received in final form: 25 December 2018 /

Accepted: 1 January 2019

<http://dx.doi.org/10.1097/MD.0000000000014233>

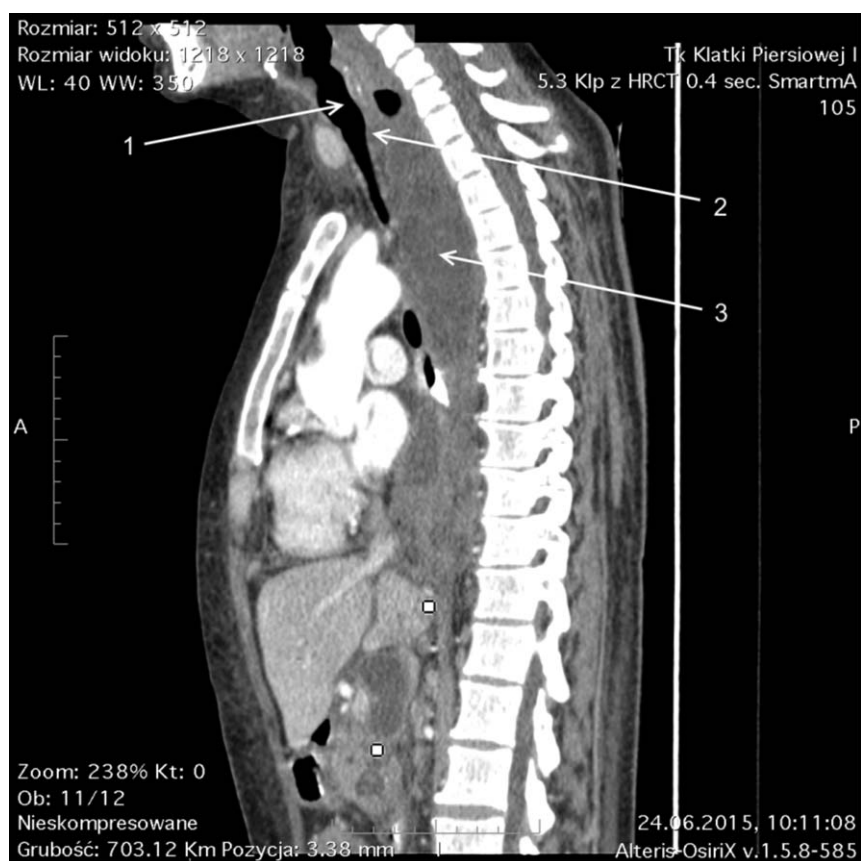


Figure 1. Chest CT scan, sagittal reconstruction. (1) Trachea. (2) Compressed oesophagus. (3) Large volume of fluid in the pleural cavity and in the mediastinum.

ECG showed sinus rhythm with a rate of 114 bpm. Laboratory tests revealed hemoglobin decreased to 8.80 g/dL (normal 11.5–15.0 g/dL), white blood cell count (WBC) was elevated to $16.09 \times 10^3/\mu\text{l}$ (normal $4.0\text{--}10.0 \times 10^3/\mu\text{l}$), platelet count (PLT) was elevated to $715 \times 10^3/\mu\text{l}$ (normal $130\text{--}400 \times 10^3/\mu\text{l}$), and prothrombin time (PT) was elevated to 15.30 s (normal 9.4–12.5 s). Biochemical blood tests revealed elevated amylase levels (604 U/l; normal <90 U/l) and lipase levels as high as 441 U/l (normal 21–67 U/l) with C reactive protein (CRP) 134.77 mg/l (normal <5 mg/l). Other parameters were normal or with insignificant changes.

Chest radiogram was performed showing large volume of fluid in the left pleural space. Pleurocentesis resulted in 2700 ml of serosanguinous fluid with amylase levels at 5128 U/l. Abdominal and chest computed tomography (CT) showed large volume of fluid in both pleural cavities and in the mediastinum, reaching above the thyroid gland with the esophagus translocated to the right side, its lumen narrowed from the thyroid level to the level of tracheal bifurcation. Pleural and mediastinal effusion showed connection to the pancreatic fistula (Figs. 1 and 2). In the pancreatic head with uneven borders, an irregular collection of fluid and calcifications was found, the pancreatic duct was widened to 6 mm, and a pseudocyst was shown in the proximity of pancreatic head, with a diameter of 26 mm (Fig. 3).

The imaging diagnostics along with high amylase levels in pleural fluid and history of chronic pancreatitis confirmed the diagnosis of pancreaticopleural and pancreaticomediastinal fistula as the reasons of dysphagia and dyspnea.

Pharmacological treatment was administered (fluids and jelly orally, fluids parenterally, analgesics, antibiotics, anticoagulant, proton pump inhibitor, vitamin K supplementation, and chest drainage) and endoscopic retrograde cholangiopancreatography (ERCP) was performed. However, the pancreatic duct was not visible after contrast infusion and in spite of precut papillotomy, the instrument could not be inserted further.

Next attempt of endoscopic treatment was planned, but patient's condition worsened with progressing dysphagia, dyspnea, and chest pain. Tender swelling of the neck was observed. Hemoglobin dropped to 5.9 g/dl, WBC was elevated to $21 \times 10^3/\mu\text{l}$, and CRP was 224 mg/l.

The patient was prepped for urgent surgery. A cholecystectomy was performed, sequesters were removed from the mesentery (Fig. 4), and Roux-en-Y pancreaticojejunal anastomosis was performed. A T-tube was inserted into the pancreatic duct and led out by Witzel's method.

After the surgery, the patient was treated for 6 days in the intensive care unit due to respiratory insufficiency (parenteral nutrition, antibiotics, analog-sedation, mechanical ventilation). A chest radiogram on the 4th postoperative day showed only small amount of pleural fluid bilaterally. After the patient returned to our ward, the rest of her recovery was uneventful. The patient was discharged on the 12th postoperative day. Chest CT performed 2 months after surgery revealed small residual amount of fluid in the right pleural cavity. The patient reported good general feeling and weight gain of 5 kg during her visit in the surgical clinic 6 months after surgery.



Figure 2. Chest CT scan, frontal reconstruction. (1) Trachea. (2) Pleural effusion. (3) Branching pancreatic fistula.



Figure 3. Abdominal CT. Collection of fluid in the pancreatic head (1). Widened main pancreatic duct (2).

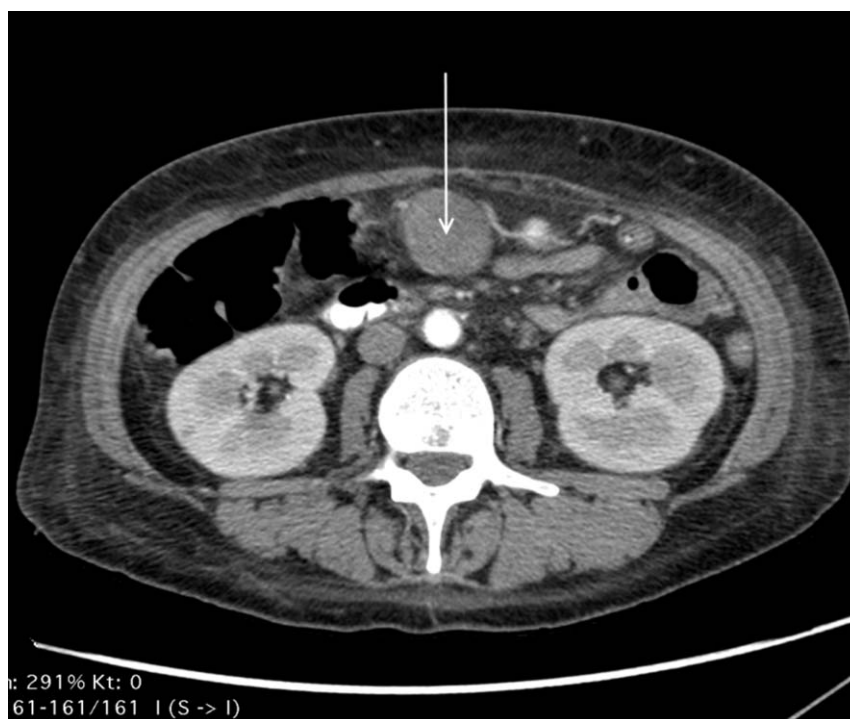


Figure 4. Abdominal CT. Necrosis in the mesentery (arrow).

3. Discussion

The most unusual features of this case include concomitant pancreaticopleural and pancreaticomediastinal fistulas with cervical infiltration causing severe dysphagia and fast progressing anterior cervical swelling as the presenting symptoms. We have found only a handful of reports of similar pancreatic fistulas with cervical extensions.^[9–12] Another case of pancreatic fluid localized in the cervical region described by Rokach was associated with ectopic pancreatic tissue.^[13]

There are no strict guidelines concerning treatment of pancreaticopleural and pancreaticomediastinal fistulas. It is generally agreed that if conservative treatment with octreotide does not give good results in 3 weeks, endoscopic therapy or surgery should be performed.^[11,14] Medical treatment is rather encouraged for cases with mild pancreatic duct dilatation. In more advanced cases like this one, it may cause unnecessary delay in invasive treatment and lead to more complications and prolonged hospital stay.^[15,16] Magnetic resonance cholangiopancreatography is advised as a noninvasive and accurate way to visualize the fistula.^[11,17,18] In this case, CT provided the diagnosis, and ERCP was the first choice not only as a means of visualization but also as a treatment method. Unfortunately, it failed in this case: the pancreatic duct was not visible due to its obstruction and there was no possibility of stent insertion. Therefore, surgery was inevitable in order to obtain decompression of the pancreatic duct and healing of the pancreaticopleural fistulas.^[19,20] In some cases of less complicated pancreatic fistulas, endoscopic or laparoscopic internal drainage is also effective and safe.^[8,9,20]

Some reports describe cases of unnecessary thoracic surgery performed before the right diagnosis led to abdominal procedure and recovery,^[21,22] which indicates the need to emphasize that in

all patients with a history of pancreatitis, recurrent pleural effusion and dysphagia differential diagnosis should include pancreaticopleural and pancreaticomediastinal fistula.

4. Conclusion

Pancreaticopleural and pancreaticomediastinal fistulas are rare complications, which have to be considered in patients with history of acute and chronic pancreatitis. If the fluid reaches the mediastinum, the patient may present dysphagia as initial symptom and cervical involvement may present with rapidly progressing neck swelling. If the endoscopic treatment is unsuccessful, surgery is the best option.

Author contributions

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