

Successful Diagnosis and Treatment of a Pancreaticopleural Fistula in a Patient Presenting with Unusual Empyema and Hemoptysis

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Pancreaticopleural fistula (PPF) is a rare complication in patients with pancreatitis. Its symptoms are similar to those of empyema or pleural effusion; therefore, it is important to consider PPF in the differential diagnosis. Herein, we describe the diagnosis and treatment of PPF in a patient presenting with unusual empyema and delayed hemoptysis.

Key words: 1. Pleural diseases
2. Pleural effusion
3. Empyema
4. Thoracoscopy
5. Video-assisted thoracic surgery
6. Magnetic resonance imaging

Case report

Pancreaticopleural fistula (PPF) is a rare complication in patients with pancreatitis; in cases of PPF, the communication between the pancreas and the pleural cavity causes pancreatic secretions to drain directly into the pleural cavity, rather than into the duodenum. This is a critical issue in patients with acute pancreatitis, but it is more common in those with chronic pancreatitis or pancreatic pseudocysts [1]. As the pleural effusion is amylase-rich, it is not easy to diagnose PPF early, which may delay treatment [2]. Herein, we review a patient with an unusual presentation of empyema and delayed hemoptysis, in whom we diagnosed a PPF that was success-

fully treated via endoscopic retrograde cholangiopancreatography (ERCP).

A 43-year-old man visited an external medical center with chief complaints of right chest and abdominal pain. He was a heavy drinker with a history of recurrent acute pancreatitis. He was transferred to Pusan National University Hospital because of persistent right-side empyema despite the performance of a closed thoracostomy and administration of intravenous antibiotics for 2 weeks. Consistent with recurrent pancreatitis, the patient's initial serum amylase and lipase levels were 590 IU/L and 488.6 U/L, respectively, and conservative therapy including medication had been started based on a consultation with a hepatologist. Computed tomography (CT) re-

Received: September 4, 2018, Revised: October 21, 2018, Accepted: October 22, 2018, Published online: June 5, 2019

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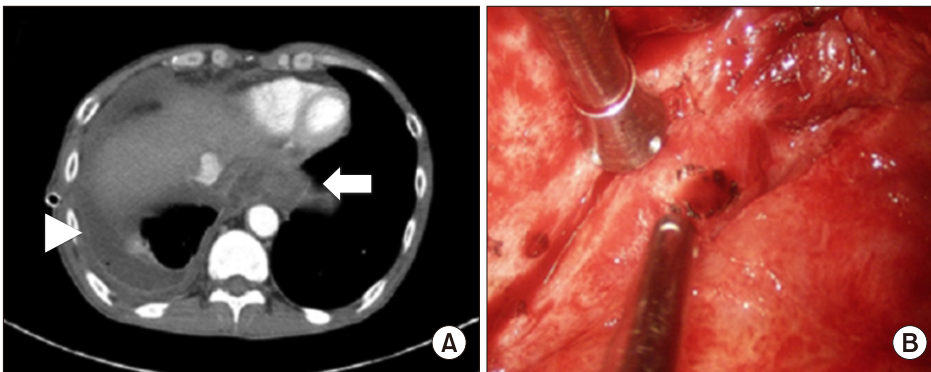


Fig. 1. (A) On the first visit, computed tomography revealed a right pleural effusion with pleural thickening (white arrowhead) and fluid in the periesophageal space (white arrow). (B) Intraoperative findings: thickened visceral pleura and serous fluid in the periesophageal sac.

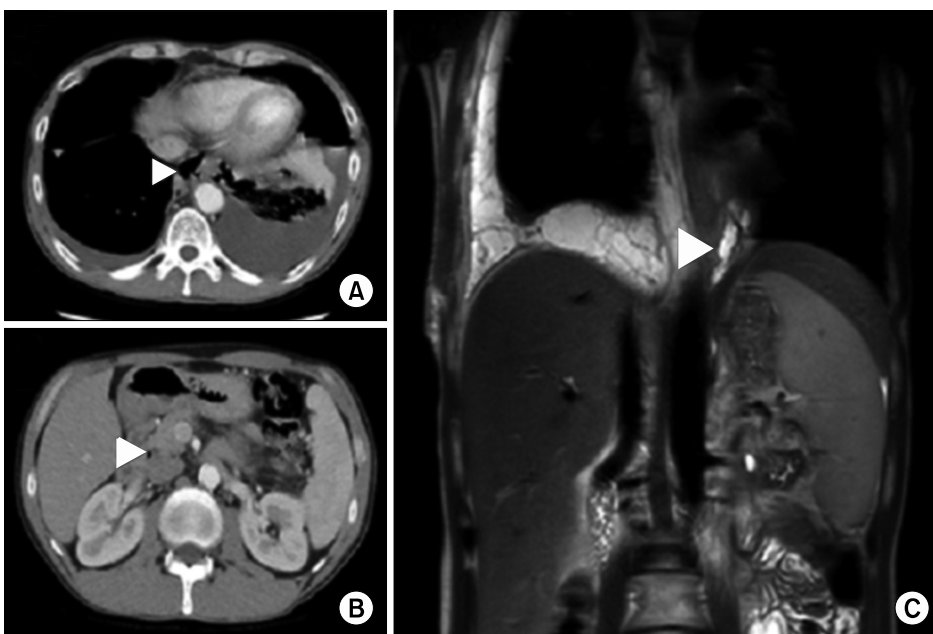


Fig. 2. Chest computed tomography findings on the day of re-admission (A) and several days later (B); and MRI of the pancreas (C). (A) A left pleural effusion and a pneumomediastinum (white arrowhead) revealing a distal esophago-nodal fistula. (B) As air was present in the peripancreatic space (white arrowhead), we scheduled pancreatic MRI. (C) MRI revealed a fluidic connection between the distal pancreatic duct and the left pleura (white arrowhead) accompanied by a disrupted pancreatic duct. MRI, magnetic resonance imaging.

vealed a right-side pleural effusion and fluid in the periesophageal space (Fig. 1A). Under suspicion of empyema or an esophageal perforation, we performed video-assisted exploration of the periesophageal space and pleural decortication. This revealed a thickened visceral pleura and serous fluid in the periesophageal sac (Fig. 1B). Tissue culture of a visceral pleural peel did not yield any microorganisms. However, because the pleural fluid was amylase-rich (1,269.8 IU/L) and a periesophageal sac was evident on CT, we scheduled esophagography, which yielded no evidence of leakage. The chest tube was removed on post-operative day (POD) 12 and the patient was discharged on POD 14.

However, he was re-admitted with hemoptysis 3 weeks later. CT revealed a left pleural effusion and a

pneumomediastinum that we interpreted as a distal esophago-nodal fistula (Fig. 2A). We performed left-side closed thoracostomy, followed by endoscopy and bronchoscopy, but no significant pathology was evident. At that time, the amylase level of the pleural effusion was 5,250 IU/L, and the patient's serum amylase and lipase levels were elevated, at 315.0 IU/L and 185.2 U/L, respectively. Since the hemoptysis had persisted, we performed bronchial artery arteriography, which revealed no evidence of extravasation. As air was apparent in the peripancreatic space (Fig. 2B), magnetic resonance imaging (MRI) of the pancreas was performed on POD 47, revealing a disrupted pancreatic duct and a PPF (Fig. 2C). We performed ERCP and there was no evidence of a fistula. Even though we attempted to insert a stent

into the ampulla, it failed due to acute angulation. However, since the structural sphincter had been corrected after 3 ERCP sessions, the hemoptysis and left pleural effusion resolved and the patient's serum amylase levels fell. The patient was discharged 29 days after his second admission; no recurrence or complication has been noted.

Written informed consents were obtained from patients.

Discussion

PPF is a rare complication of pancreatitis. The typical symptom is recurrent pleural effusion in either or both pleural space(s), but usually on the left [3,4]. Although the pleural fluid is amylase-rich, this may be attributable to pancreatitis, esophageal perforation, or any of several tumors, rendering the diagnosis difficult [3,5,6]. However, when amylase levels are elevated in the pleural fluid, several modalities can be used to diagnose PPF. The sensitivities of CT, ERCP, and magnetic resonance cholangiopancreatography have been reported to be 47%, 78%, and 80%, respectively [7,8].

Our case differed somewhat from other reported cases, because the initial site of fluid accumulation was the right periesophageal space. To rule out empyema with or without esophageal perforation, we first performed a video-assisted exploration. The chest tube was removed on POD 12 because drainage had decreased and the X-ray findings had improved. The patient was discharged on POD 14 without any symptoms. However, at the time of the second admission for persistent hemoptysis, CT revealed peripancreatic air and a left pleural effusion probably caused by obliteration of the right pleura during the previous operation. Pancreatic MRI revealed pancreatic duct obstruction and a PPF. ERCP was used for simultaneous assessment of the disrupted pancreatic duct and duct decompression. After 3 ERCP sessions, the patient's serum amylase levels fell and the hemoptysis (triggered by the amylase-rich pleural effusion) resolved.

Hence, for patients with a history of pancreatitis who exhibit recurrent pleural effusions or other atypical symptoms, pleural fluid amylase levels should be checked and pancreatic MRI performed if a PPF is suspected (Fig. 3). ERCP simultaneously assesses the

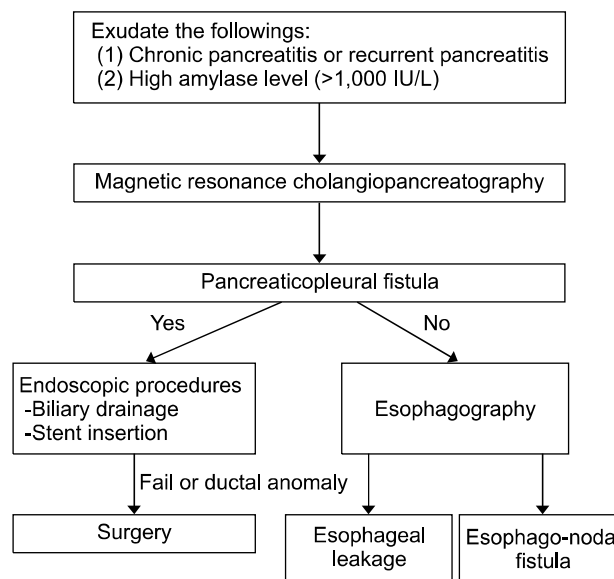


Fig. 3. Pancreaticopleural fistula diagnosis and treatment algorithm.

fistula and decompresses the duct. If endoscopy fails in patients with ductal anomalies, surgery is recommended to prevent septic complications (Fig. 3). Therefore, PPF management should be determined by the ductal anatomy revealed by pancreatic MRI, and clinicians should keep in mind that a PPF closes spontaneously during ERCP.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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Diagnosis of Pancreaticopleural Fistula with Empyema

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