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Recurrent Pleural Effusion Secondary to a Pancreatic-Pleural Fistula Treated Endoscopically

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 44
Final Diagnosis: Pancreatic pleural fistula
Symptoms: Short of breath
Medication: —
Clinical Procedure: ERCP
Specialty: Gastroenterology and Hepatology

Objective: Unusual clinical course





Background: Pancreatic-pleural fistula (PPF) is an uncommon complication of pancreatitis. Pleural effusions secondary to PPF are caused by fistulization of pancreatic secretions to the thorax derived from the rupture or leakage of a pseudocyst.

Case Report: We describe the case of a 44-year-old male with recurrent right-sided pleural effusions and alcoholic pancreatitis who presented with epigastric pain and shortness of breath. Pleural fluid analysis revealed an amylase of 7002 U/l. MRCP showed segmental narrowing and stricture of the proximal main pancreatic duct and an area of walled-off necrosis. The fistula was managed endoscopically with ERCP and placement of a plastic stent into the pancreatic duct. The pleural effusion resolved and subsequent examinations showed no evidence of recurrence.

Conclusions: The diagnosis of PPF is challenging. Endoscopic treatment of PPF can be a safe and effective approach.

MeSH Keywords: Pancreatic Fistula • Pancreatitis • Pleural Effusion

Full-text PDF: <http://www.amjcaserep.com/abstract/index/idArt/903925>

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Background

A pancreatic-pleural fistula (PPF) is an uncommon complication of pancreatitis [1]. In contrast to pleural effusions seen in pancreatitis that are usually clinically insignificant, a PPF can produce recurrent and large volume effusions [2]. A PPF is formed by leakage or rupture of a pancreatic pseudocyst or pancreatic duct (PD) disruption [3]. If there is posterior communication of these structures to the retroperitoneum, the pancreatic secretions may form a fistula to the pleural cavity through the aortic or esophageal orifice producing an effusion [4]. We present the case of a patient who presented with a recurrent pleural effusion due to a PPF who was successfully managed with endoscopic treatment.

Case Report

A 44-year-old male presented with acute onset of epigastric pain radiating to the back, exacerbated by movement and associated with nausea, non-bloody emesis, and shortness of breath. He had a past medical history of recurrent alcoholic pancreatitis, multiple right-sided pleural effusions, AIDS, hepatitis C infection, and hypertension. He was taking azithromycin, fluconazole, pantoprazole, and sulfamethoxazole/trimethoprim. Social history was significant for cocaine and intravenous drug use as well as heavy alcohol use for 10 years.

On physical examination, the patient was afebrile, heart rate 118 beats per minute, blood pressure 115/70 mmHg, respiratory rate 18 per minute, and oxygen saturation 94% on 2 liters by nasal cannula. Exam revealed a dry oral mucosa, right hemithorax with absent breathing sounds, dullness to percussion from the base to the apex, and an unremarkable left hemithorax. The abdomen was soft, mildly distended, with generalized tenderness with epigastric predominance, and absent bowel sounds, and the rectal examination was normal.

Initial laboratory workup showed leukopenia, normocytic anemia, and lipase of >7500 U/l. A chest X-ray showed a right pleural effusion occupying more than 90% of the right hemithorax, producing almost complete atelectasis of the right lung (Figure 1). A magnetic resonance cholangiopancreatography (MRCP) of the abdomen with contrast revealed sequelae of prior necrotizing and chronic pancreatitis, segmental narrowing and possible stricture of the proximal main PD, a small area of necrosis in the uncinata process which appeared to communicate with the PD, and a 12.8×8.1×9 cm area of walled-off necrosis in the lesser sac extending along the portal vein to the porta hepatis (Figure 2).

The patient was started on intravenous fluid resuscitation, pain control, and bowel rest. At the same time, a therapeutic

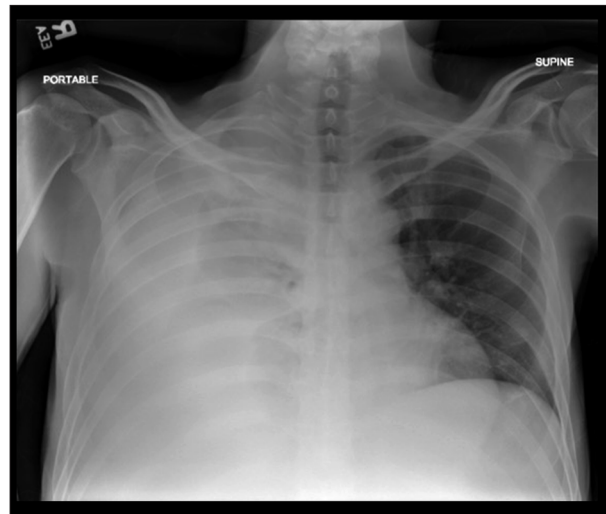


Figure 1. Chest X-ray on admission showing large right pleural effusion.

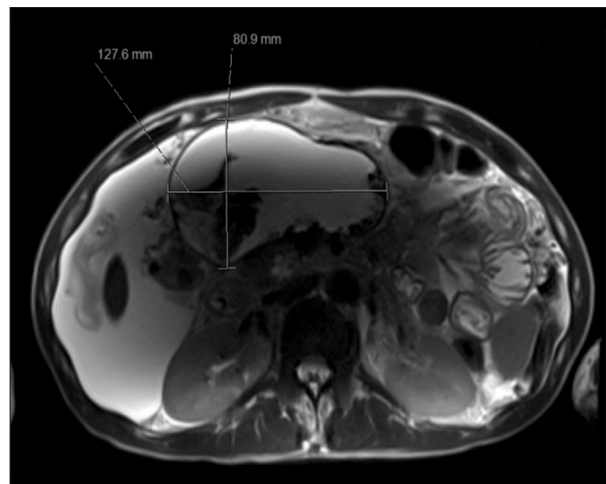


Figure 2. MRCP showing a 12.8×8.1×9 cm area of walled-off necrosis in the pancreas.

thoracentesis was performed on the right hemithorax, and analysis revealed an exudative effusion with amylase of 7002 U/l. An endoscopic retrograde cholangiopancreatography (ERCP) was performed based on the MRCP findings and for the concern of a pancreatic-pleural fistula. A pseudocyst in the ventral PD in the head of the pancreas, changes in the PD consistent with chronic pancreatitis, and a partial obstruction in the PD in the genu of the pancreas were identified. One 5 Fr by 7 cm plastic stent with a full external pigtail and a single internal flap was placed into the ventral PD bridging the cystic area and the stricture. Clear fluid and diminutive stones flowed through the stent (Figure 3).

A few days after the procedure, the abdominal pain and right pleural effusion resolved (Figure 4) and the patient was discharged asymptomatic. Four months after this event, the patient

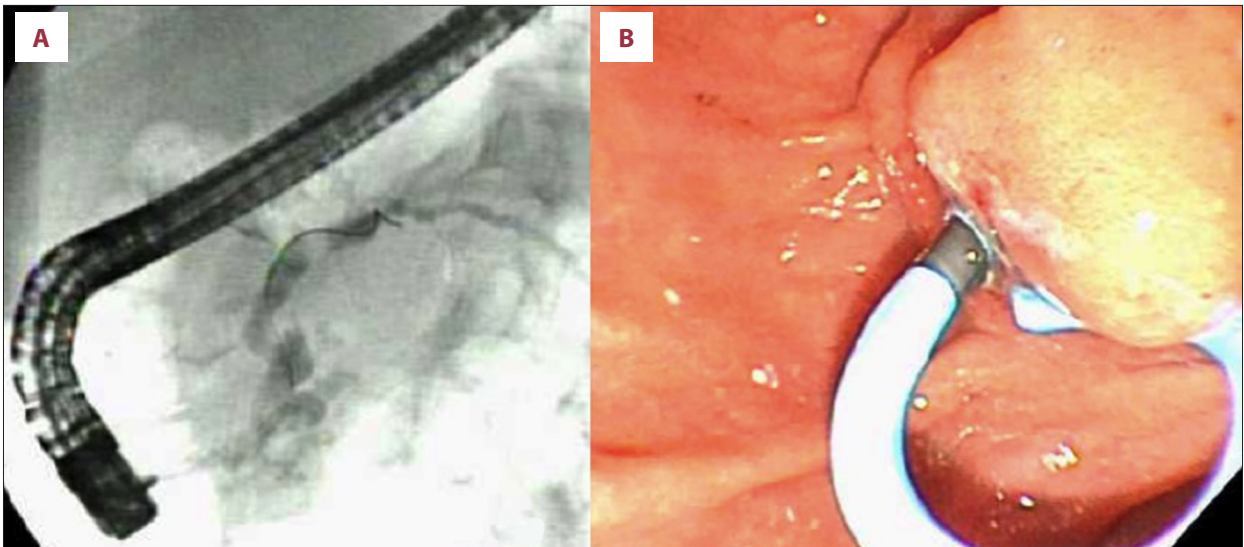


Figure 3. ERCP showed: (A) partial obstruction in the pancreatic duct, (B) one 5 Fr by 7 cm plastic stent was placed into the ventral pancreatic duct bridging the cystic area and the stricture.

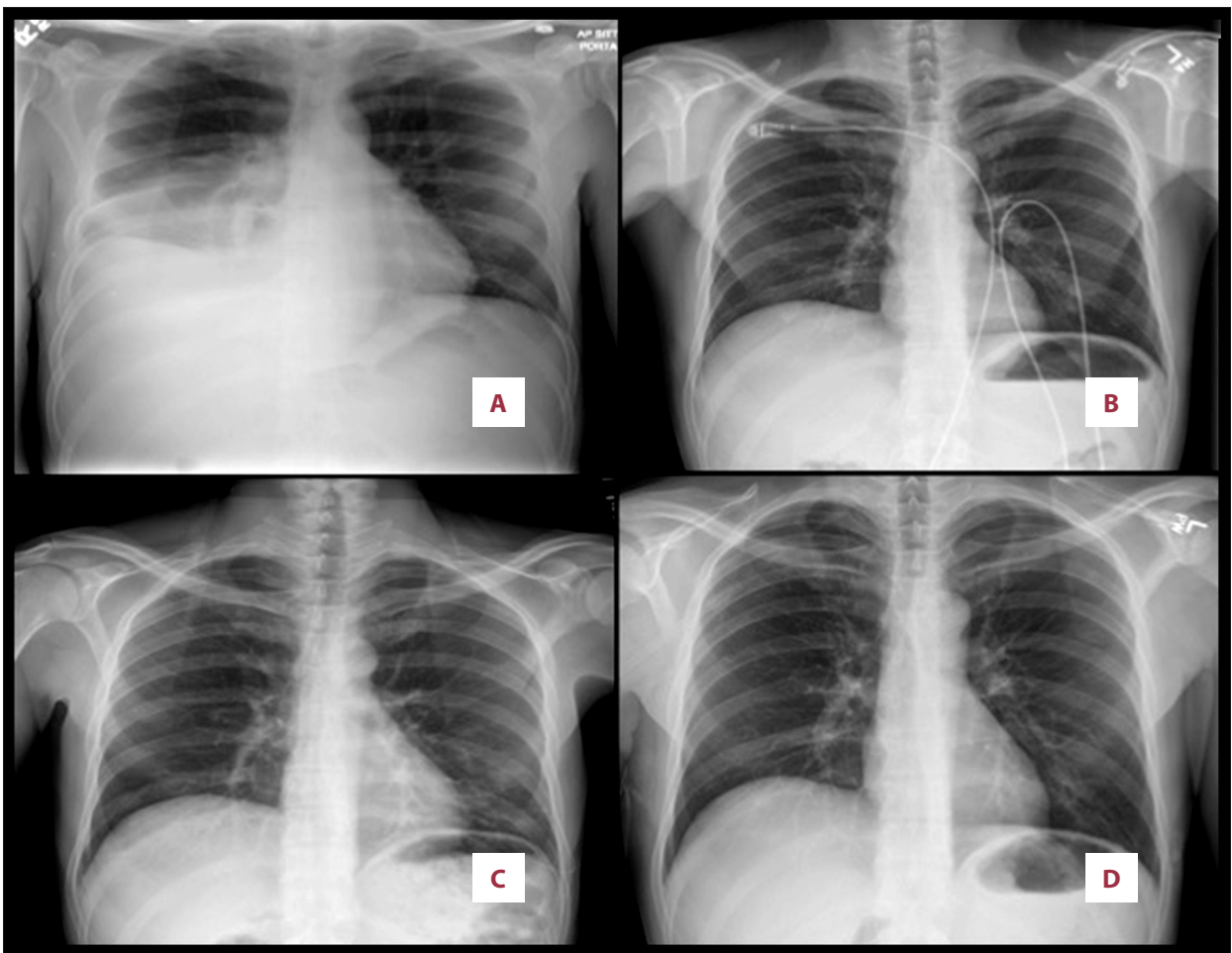


Figure 4. Chest X-ray follow-up at: (A) 2 weeks, (B) 4 months, (C) 6 months, and (D) 18 months shows resolution of the pleural effusion.

presented with abdominal pain secondary to partial obstruction of the pancreatic stent. This was exchanged with a 7 Fr by 7 cm plastic stent without complications. Follow-up examination at 2 weeks, and 4, 6, and 18 months revealed no evidence of recurrent pleural effusion (Figure 4).

Discussion

We present the case of a patient with a PPF to highlight the potential role of endoscopic management of this entity. Right hemithorax effusions are not typical, as up to 76% of the cases present with only left hemithorax involvement [5]. Diagnosis is usually delayed due to the absence of abdominal symptoms, as patients usually present with shortness of breath, cough, and chest pain [6] secondary to pleural effusions, which could be large and recurrent if the PPF is not treated. The first step in the approach to a PPF is the determination of the pleural fluid amylase level. Although there is no cutoff level to establish a diagnosis, pleural amylase is usually > 1000 U/L, with levels of >50 000 U/L highly suggestive of a PPF [1]. Once an elevated amylase pleural level is confirmed, the next step is obtaining abdominal imaging with a MRCP. This modality is superior to abdominal computed tomography in visualizing the pancreatic parenchyma and ducts, pancreatic fluid collections, pseudocysts, and, occasionally, the fistula site, which could be extremely useful to determine the optimal therapeutic intervention [7].

Since PPF is uncommon, the management remains controversial, as there are no clinical studies that compare the available therapeutic options. Medical management with bowel rest and

somatostatin is successful in only 30–60% of the cases, with a 15% and 12% rate of recurrence and mortality, respectively [8]. If after 2 or 3 weeks of initiation of medical treatment there is no resolution, an endoscopic intervention should be attempted [1]. Endoscopic treatment aims to reduce the pancreatic-duodenal pressure gradient within the PD or pseudocyst by creating a pathway of least resistance into the duodenum [7]. This is achieved by placement of a transpapillary PD stent with sphincterotomy of the major papilla and pseudocyst drainage in patients with partial PD disruption or stricture [9]. If this is not achieved with conventional ERCP, EUS-guided rendezvous ERCP may be another option [10]. Finally, surgical intervention is reserved for patients with failure of endoscopic management or for those who have complete PD disruption or severe stricture [11].

Endoscopic treatment for PPF, along with other modalities, has been described in the past [1,4,6,9,12]. It has been used in conjunction with somatostatin [4, 12] and complemented with chest tube placement for drainage of large pleural effusions [1,4,6], nasopancreatic drainage [12], and surgical treatment if the latter fail to resolve the fistula [1]. In our case, we were able to resolve the recurrent large pleural effusions secondary to the PPF with the placement of a PD stent without need for complementary therapies.

Conclusions

In conclusion, endoscopic treatment in patients with the presence of a pancreatic pseudocyst and a partial disruption of the PD, as in our case, can be a safe and effective approach.

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