Pancreaticopleural Fistula Presenting as Left Neck Swelling: A Case Report

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Michal Senitko, MD¹ and Jonathan Puchalski, MD, MEd¹

Abstract

Pancreaticopleural fistula is an uncommon complication of pancreatitis. It results from communication of the pancreatic duct to the pleural space. The authors describe a case of a 54-year-old man with a history of alcohol abuse and chronic pancreatitis, who presented with left neck swelling, dysphagia, and dyspnea. Computed tomography imaging revealed right pleural effusion and multiloculated fluid collections in and around the pancreas and retroperitoneum with extension through the posterior mediastinum into the left neck. Analysis of the neck aspirate and the pleural effusion showed very elevated amylase levels. This is the first known case of a pancreaticopleural fistula presenting as neck swelling due to cystic fluid collection.

Keywords

pancreatic pleural effusion, exudative pleural effusion, pleural fluid analysis

Introduction

Pancreaticopleural fistula (PPF) results from communication of the pancreatic duct (PD) to the pleural space. It is an infrequent but known complication of acute pancreatitis with an estimated incidence of 0.4%.¹ More commonly, it results as a complication of chronic pancreatitis and pancreatic pseudocyst. Patients often present with dyspnea, cough, or chest pain, and pleural fluid analysis demonstrates a markedly elevated amylase level.² We present the first known case of a PPF presenting as neck swelling due to cystic fluid collection.

Case Study

A 54-year-old male with a history of alcohol abuse and chronic pancreatitis was referred for an evaluation of progressive left neck swelling, dysphagia, and dyspnea. He denied cough, chest pain, fever, or abdominal pain. Physical examination demonstrated swelling in the left neck and decreased bibasilar breath sounds. He underwent aspiration of a fluid collection in his left neck that revealed serosanguinous, noninfected fluid. Additional studies demonstrated a markedly elevated amylase (>24 000 IU/L) and lipase (>40 000 IU/L) in the neck aspirate.

The patient's neck computed tomography (CT) demonstrated upper thoracic abnormalities for which additional CT imaging ultimately confirmed multiloculated fluid collections in and around the pancreas and retroperitoneum with extension through the posterior mediastinum into the left neck (Figure 1A-D). A right thoracentesis yielded 850 ml of serosanguinous fluid. Analysis confirmed an exudative effusion with an amylase level of 7560 U/L, negative cultures, and negative cytology (Table 1). Given the history, radiographic findings, and findings from the pleural fluid and neck aspirate, PPF was diagnosed.

Management

The patient underwent an endoscopic retrograde cholangiopancreatography (ERCP) that confirmed PD disruption with a cystic fluid collection in the pancreatic tail (Figure 2A). A PD stent was placed proximal to the defect (Figure 2B). An ultrasound-guided right thoracotomy tube was placed due to symptomatic recurrence of the effusion (Figure 3).

Surgery was consulted, and a nonoperative approach was recommended. The patient received postpyloric tube feeds and antibiotics. The chest tube was removed after 8 days due to cessation of fluid reaccumulation. At 2 months, repeat ERCP demonstrated complete resolution of the leak, and follow-up CT confirmed complete resolution of the pleural effusion, mediastinal, and peripancreatic fluid collections (Figure 4).

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Corresponding Author:

Jonathan Puchalski, MD, Yale University School of Medicine, 15 York Street, LCI 100, New Haven, CT 06510, USA. Email: jonathan.puchalski@yale.edu

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¹ Yale University School of Medicine, New Haven, CT, USA

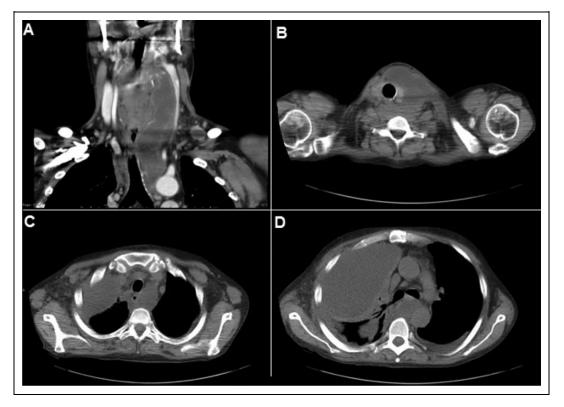


Figure I. A—Computed tomography (CT) of neck: coronal view demonstrates the cystic fluid collection on the left. B, C, D—CT of the chest: axial views demonstrate the cystic fluid collection in the neck, pleural space, and mediastinum.

Table 1. Analysis of Fluid From the Left Neck Aspirate and Right Pleural Effusion.

	WBC, cells/UL	Lymphocytes, %	Granulocytes, %	Tissue Cells, %	RBC, cells/UL	Glucose, mg/dL	Protein, g/dL	LDH, U/L	Amylase, Lipase, U/L	pН
Neck aspirate	2472	43	51	6	40 124	n/a	n/a	33 346	>24 000, >40 000	7.03
Pleural effusion	3000	18	54	28	56 000	56	3.3	6378	7560, n/a	7.12

Abbreviations: LDH, lactate dehydrogenase; n/a, not available; RBC, red blood cells; WBC, white blood cells.

Discussion

Pleural effusions due to PPF are a rare entity characterized by exudative effusions with elevated amylase levels. The pleural effusions are typically large and rapidly reaccumulate.³ They occur more commonly in men and are unilateral in more than 80% of cases, with no laterality preference. Pancreaticopleural fistula is associated with acute and chronic pancreatitis, most commonly alcohol abuse in adults. Other causes include gallstones, trauma, and PD abnormalities.¹ The effusions are characterized by elevated amylase with levels typically >1000 U/L and levels >50 000 U/L being pathognomonic. Lipase and pleural fluid albumin are also typically elevated.

The most common presenting symptoms are dyspnea (65%), abdominal pain (29%), chest pain (23%), and cough (2%).⁴ When a PPF tracks anteriorly, pancreatic ascites

typically results.^{5,6} More commonly, a posterior track forms into the retroperitoneum with subsequent extension into the mediastinum. In these cases, the lack of abdominal symptoms may be explained by decompression of the pancreatic pseudocyst and other anomalies into the chest cavity. As symptoms are very nonspecific, a high index of suspicion is required.

Imaging is often performed to clarify the extent of abdominal and pleural disease or to identify the fistulous track. Computed tomography is a valuable initial diagnostic tool but has a low sensitivity (47%) in a comparison to magnetic resonance cholangiopancreatography (MRCP; 80%).⁴ The ERCP is often used, given its diagnostic sensitivity of 78% and its added value of enabling a concomitant therapeutic approach, such as placement of a pancreatic stent. As noted above, pleural fluid analysis is a cornerstone of the diagnostic evaluation.

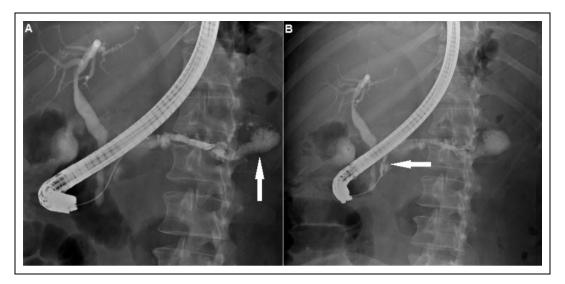


Figure 2. A—Contrast injected during endoscopic retrograde cholangiopancreatography (ERCP) demonstrates a peripancreatic fluid collection (arrow). B—The pancreatic ductal stent was placed successfully (arrow).

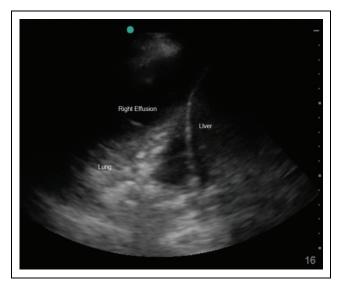


Figure 3. Ultrasound of the right pleural space.

Early surgical intervention is definitive but with advances in medical therapy, often not the first approach in current therapeutic algorithms.⁷ Combined medical therapy may include endoscopic techniques (dilation, stenting, biliary stone retrieval), postpyloric or parenteral nutrition, and octreotide to decrease the need for surgical intervention.^{2,8} Although randomized control trials are lacking, the available literature suggests that ductal anatomy may initially direct the treatment approach.^{4,9}

To our knowledge, this is the first case of a PPF resulting in a right-sided pleural effusion with a contralateral fluid collection into the neck stemming from extension of the fistulous tract beyond the thorax. A multidisciplinary approach is essential in management as patients have medical, nutritional, and, sometimes, psychosocial or addiction challenges. In this case, surgery was obviated by an endoscopic approach that enabled complete resolution of the abdominal, thoracic, and extrathoracic pathology.



Figure 4. Follow up abdominal computed tomography (CT; A, Axial view. B, Coronal view) demonstrates complete resolution of the posterior mediastinal fluid collection, right pleural effusion, and peripancreating fluid collections with pancreatic duct (PD) stent in place (arrow).

Declaration of Conflicting Interests

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Author Biographies

Michal Senitko, MD recently completed his Interventional Pulmonary Fellowship at Yale University School of Medicine, in the division of Pulmonary, Critical Care and Sleep Medicine at Yale New Haven Hospital, New Haven, CT. He is now developing Interventional Pulmonary Program at the University of Mississippi Medical Center, Jackson, MS.

Jonathan Puchalski, MD, MEd is an associate professor of Medicine at Yale University School of Medicine, and is the Director of Interventional Pulmonology in the division of Pulmonary, Critical Care and Sleep Medicine at Yale New Haven Hospital, New Haven, CT.