PANCREATICOPLEURAL FISTULA: A RARE COMPLICATION OF CHRONIC PANCREATITIS PRESENTING WITH MASSIVE PLEURAL EFFUSION

Subbiah Senthil Nathan ¹, Gunasekaran Nallusamy ^{2*}, Priyadarshini Varadaraj ³ and Keesari Sai Sandeep Reddy ⁴

^{1,2,3,4} Post Graduate, Department of Internal Medicine Saveetha Medical and Hospital, Chennai, India. *Corresponding Author Email: drguna59@gmail.com

DOI: 10.5281/zenodo.12649704

Abstract

Pancreaticopleural fistula, a rare and serious complication of chronic pancreatitis, presents diagnostic challenges due to its atypical presentation. Here, we report a case of a patient with known chronic pancreatitis who presented with massive left-sided pleural effusion, highlighting the importance of considering this rare complication in patients with pulmonary symptoms. Blood-colored pleural fluid analysis revealed significantly elevated lipase and amylase levels in the hundred thousand and ten thousand ranges, confirming the pancreatic origin of the effusion. Diagnosis was further supported by imaging modalities, including computed tomography (CT) and magnetic resonance cholangiopancreatography (MRCP). Management involved thoracocentesis for symptom relief and endoscopic pancreatic duct stenting via Endoscopic Retrograde Cholangiopancreatography (ERCP) to address the underlying fistula. This case underscores the need for heightened clinical suspicion and multimodal diagnostic approaches in identifying pancreaticopleural fistula, facilitating timely intervention and optimal patient outcomes.

INTRODUCTION

Pancreaticopleural fistulas (PPFs)-related pleural effusions are exceedingly uncommon; they account for roughly 1% of pleural effusion cases and occur in 0.4% of individuals with chronic pancreatitis [1, 2]. PPFs can be acute or chronic pancreatitis, and they usually result from rupture of the primary pancreatic duct or a pancreatic pseudocyst.

They can also come from trauma, albeit this is less common. These are primarily observed in male patients with prolonged alcohol use, according to demographics [3, 4]. An anterior tract forms from the pancreas into the anterior retroperitoneum as a result of inflammation brought on by exposure to pancreatic digesting enzymes. This leads to fluid buildup and subsequent contact with the pleural cavity [5]. Therefore, the typical patient presentation of dyspnea and other respiratory problems frequently complicates diagnosis [1].

Pleural effusions caused by PPFs typically reoccur and show positive results for lipase and amylase [4]. Exudative pleural fluid, varying in appearance from straw-colored to serosanguinous, is commonly shown during thoracentesis [6, 7]. Less than one-third occur on the right side, with the left side experiencing them more frequently [3].

Rarely, bilateral pleural effusions caused by PPFs may manifest [8]. The exceedingly unusual development of a trapped lung may result from the retention of fluid in these pleural effusions if they are not drained over an extended length of time [9].

Diagnostics like magnetic resonance cholangiopancreatography (MRCP) or endoscopic retrograde cholangiopancreatography (ERCP) are used for anatomic mapping once clinical suspicion strongly suggests a PPF. ERCP may also be used therapeutically to place a stent at the site of duct disruption [4, 9]. In addition to its mechanical function of blocking the fistula to stop recurrence of pleural effusion, the stent may also widen the strictures of the pancreatic duct [2, 4].

Case Presentation

A 35-year-old male, a chronic alcoholic for 15 years with a history of recurrent pancreatitis and pancreatic diabetes, presented with progressive breathlessness and right-sided chest pain over the past 2 weeks. Upon physical examination, the patient appeared malnourished and tachypneic, with oxygen saturation at 96% on room air.

Stony dullness was noted upon percussion, along with absent breath sounds on the entire left side of the chest. Cardiac auscultation revealed normal heart sounds, and ECG showed sinus tachycardia with normal cardiac biomarkers. Epigastric tenderness was observed upon abdominal examination, while examination of other systems was unremarkable. Chest radiography revealed a massive left-sided pleural effusion.

Emergency thoracocentesis was performed, yielding approximately 1 liter of bloody fluid from the left pleural space. Analysis of the pleural fluid revealed an exudative pattern, with notable findings including a pleural fluid LDH of 704 IU/L (compared to serum LDH of 136 IU/L), total protein of 4.8g/dl (compared to serum total protein of 6.3g/dl), lipase level of 186,440 IU/L, and amylase level of 22,362 IU/L, along with the presence of RBCs.

Further investigations, including cartridge-based nucleic acid amplification test (CBNAAT) for Mycobacterium tuberculosis, cytological evaluation, and cultures, were negative for infectious or malignant etiologies. Serum amylase and lipase levels were also elevated.

Plain CT of the chest and abdomen revealed moderate left-sided pleural effusion with a bulky pancreas and parenchymal calcifications. On CECT abdomen axial section, a dilated and tortuous main pancreatic duct (MPD) distal to the proximal body, with a tiny exophytic thin-walled peripherally hypodense component/collection arising from the body of the distal pancreatic MPD, was observed (see Figure 1).

Another axial section revealed a hypodense collection at the distal MPD tracking cranially into the posterior mediastinum (see Figure 2).

Additionally, on sagittal section, a hypodense collection seen from the distal MPD was likely communicating/draining to the left pleural cavity (see Figure 3). These imaging findings provided crucial anatomical details guiding the management and further monitoring of the patient's condition.

A diagnosis of Necrotizing Pancreatitis with Pancreatico pleural Fistula was established. The patient underwent bowel rest and hydration with IV fluids. Post-procedure chest X-ray revealed complete resolution of the pleural effusion. The patient showed symptomatic improvement and was discharged, with periodic follow-up scheduled.



Figure 1: CECT Abdomen (Axial section)

Blue arrow shows Main Pacreatic Duct (MPD) distal to the proximal body is dilated \sim measuring – 4 mm and tortuous with tiny exophytic thin walled peripherally hypodense component / collection seen arising from the body of the distal pancreatic MPD



Figure 2: CECT Abdomen (Axial section)

Arrow shows hypodense collection at distal MPD tracking cranially into the posterior mediastinum.



Figure 3: CECT Abdomen (Sagittal section)

Arrow shows hypodense collection seen from distal MPD is likely communicating / draining to the left pleural cavity.

DISCUSSION

Less than 1% of individuals with acute pancreatitis, 0.4% of patients with chronic pancreatitis, and 4.5% of patients with pseudocysts experience pancreaticopleural fistula [1, 10-12]. The most common cause of fistula formation is chronic pancreatitis, often alcohol-induced, leading to the development of pancreatic pseudocysts [10-16]. Trauma, idiopathic pancreatitis, gallstone pancreatitis, or congenital abnormalities of the pancreatic duct can also contribute to PPF development. This abnormal connection can occur through the direct transdiaphragmatic connection of the pancreatic duct to the pleural space or the rupture of a posterior pancreatic pseudocyst into the retroperitoneum, allowing pancreatic secretions to ascend to the pleural space through the esophageal or aortic hiatus [1, 12, 15]. Reactive pleural effusion associated with pancreatitis, typically left-sided and self-limiting, should be differentiated from PPF-related pleural effusion. Ascites is a symptom of pancreaticperitoneal fistula, developing when the pancreatic pseudocyst ruptures anteriorly. Rarely, contact with the pericardium, esophagus, or bronchial tree may occur, with the latter presenting as pericardial effusion [1, 10]. Pancreaticopleural fistulas (PPFs) typically arise from chronic pancreatitis, either due to the disruption of pancreatic ducts or the formation of pseudocysts [15]. Fistulization can occur anteriorly into the peritoneum, forming a pancreaticoperitoneal fistula, or posteriorly into the retroperitoneum, resulting in an ascending pancreaticopleural fistula. Chemical irritation, sympathetic nervous system involvement, or spread via abdominal lymphatics are proposed mechanisms for the formation of pleural effusions [12, 13].

Patients with PPFs often present with respiratory symptoms such as breathlessness and chest pain, along with signs of pleural effusion on physical examination. Diagnosis is confirmed by thoracocentesis, revealing pleural fluid with significantly elevated amylase and lipase levels. Imaging modalities like CT and MRI provide detailed anatomical information, showing the fistulous tract extending from the pancreas to the pleural cavitv. Treatment options include conservative management with thoracocentesis and somatostatin analogues, as well as surgical interventions such as pancreatic resection and pseudocyst drainage in cases of recurrence or failure. ERCP remains the gold standard for demonstrating pancreaticopleural fistulas, allowing for both diagnostic and therapeutic interventions [1, 10, 17, 18]. This highlights the efficacy of endoscopic management in treating PPF and reducing hospital stay and mortality rates compared to surgical interventions. However, it's important to note that complications associated with endoscopic procedures and the long-term recurrence rate of PPF should be considered in the overall management strategy. Therefore, a thorough evaluation of the patient's condition and close followup are essential for optimal outcomes.

CONCLUSION

In conclusion, pancreaticopleural fistula (PPF) represents a rare yet significant complication of pancreatitis, particularly in individuals with a history of chronic alcohol consumption. A prompt and accurate diagnosis of PPF necessitates a high index of suspicion, thorough history-taking, meticulous physical examination, and key laboratory investigations revealing markedly elevated pleural fluid amylase and lipase levels. Additionally, imaging modalities play a crucial role in visualizing the fistulous tract. The management of PPF requires a multidisciplinary approach, with treatment modalities ranging from medical to endoscopic or surgical interventions, depending on

the individual patient's presentation and underlying factors. In recent years, endoscopic intervention, particularly through endoscopic retrograde cholangiopancreatography (ERCP) with pancreatic stenting, has emerged as the first-line therapy for PPF, owing to its effectiveness and minimally invasive nature. These advancements underscore the importance of continued research and innovation in improving the management and outcomes of this rare complication. Overall, a comprehensive understanding of PPF, along with a tailored approach to treatment, is essential for optimizing patient care and outcomes.

References

- 1) Tay CM, Chang SK: Diagnosis and management of pancreaticopleural fistula. Singapore Med J. 2013, 54:190-4. 10.11622/smedj.2013071.
- 2) Dhebri AR, Ferran N: Nonsurgical management of pancreaticopleural fistula. JOP. 2005, 6:152-61. 10.7759/cureus.4984
- 3) Bediwy AS: Pancreatico-pleural fistula: a rare cause of massive right-sided pleural effusion. Egypt J Chest Dis Tuberculosis. 2015, 64:149-51.
- 4) Aswani Y, Hira P: Pancreaticopleural fistula: a review. JOP. 2015, 16:90-4.
- 5) Sut M, Gray R, Ramachandran M, Diamond T: Pancreaticopleural fistula: a rare complication of ERCP-induced pancreatitis. Ulster Med J. 2009, 78:185-186. 10.1002
- 6) Chan EE, Shelat VG. Pancreaticopleural fistula causing massive right hydrothorax and respiratory failure: Case Rep Surg. 2016:8294056. 10.1155
- 7) El-Beialy H, Fernandez I: Unusual case of persistent unilateral pleural effusion secondary to pancreaticopleural fistula. Int J Surg Case Rep. 2012, 3:435-6. 10.1016
- 8) Ramahi A, Aburayyan K, Said Ahmed TS, Rohit V, Taleb M: Pancreaticopleural fistula: a rare presentation and a rare complication. Cureus. 2019, 11:4984. 10.7759
- Wronski M, Slodkowski M, Cebulski W, Moronczyk D, Krasnodebski IW: Optimizing management of pancreaticopleural fistulas. World J Gastroenterol. 2011, 17:4696-703. 10.3748/wjg.v17.i42.4696
- 10) Cazzo E, Apodaca-Rueda M, Gestic MA, et al.: Management of pancreaticopleural fistulas secondary to chronic pancreatitis. Arq Bras Cir Dig. 2017, 30:225-228.
- 11) Altasan T, Aljehani Y, Almalki A, Algamdi S, Talag A, Alkattan K: Pancreaticopleural fistula: an overlooked entity. Asian Cardiovasc Thorac Ann. 2013, 22:98-101.
- 12) Mihai C, Floria M, Vulpoi R, Nichita L, Cijevschi Prelipcean C,: Pancreatico-pleural fistula from diagnosis to management. A case report. J Gastrointestin Liver Dis. 2018, 27:465-469. 10.15403
- 13) Williams SG, Bhupalan A, Zureikat N, Thuluvath PJ, Santis G, Theodorou N, Westaby D: Pleural effusions associated with pancreaticopleural fistula. Thorax. 1993, 48:867-68.
- 14) Tombroff M, Loicq A, De Koster JP, Engleholm L, Govaerts JP: Pleural effusion with pancreaticopleural fistula. BMJ. 1973, 1:330-1.
- 15) Wakefield S, Tutty B, Britton J: Pancreaticopleural fistula: a rare complication of chronic pancreatitis. Postgrad Med J. 1996, 72:115-6.
- 16) Galluzzo A, Iannicelli E, Marignani M, Delle Fave G, David V: A pancreatico-pleural fistula diagnosed with magnetic resonance cholangiopancreatography. JOP. J Pancreas (Online. 2008, 9:654-57.
- 17) Fulcher AS, Turner MA: MR pancreatography: a useful tool for evaluating pancreatic disorders. Radiographics. 1999, 19:5-24.
- 18) Kiewiet JJS, Moret M, Blok WL, Gerhards MF, de Wit LT: Two patients with chronic pancreatitis complicated by a pancreaticopleural fistula. Case Rep Gastroenterol. 2009, 3:36-42.