Pancreatico-Pleural Fistula with Right Sided Pancreatic Pleural Effusion in an Adult Male - Report of a Rare Case

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Abstract: Pancreatic Pleural Effusion is a rare complication of pancreatitis and is defined as significant fluid accumulation in the pleural space with high Amylase content. Pancreatico-Pleural Fistula (PPF) is a rare complication seen in patients with Pancreatitis. It requires a high index of clinical suspicion as patients typically present with pulmonary symptoms related to the pleural effusion rather than Pancreatitis. It has been predominantly (90%) seen in alcoholic middle-aged men, usually presenting with dyspnea and left-sided pleural effusion. Rarely it can be right-sided or bilateral. The diagnosis should be suspected if a patient presents with pleural effusion in a setting of pancreatitis or alcohol intake. The significantly raised amylase in the pleural fluid offers an vital clue to the diagnosis. Magnetic resonance cholangiopancreatography (MRCP) can aid in visualizing the fistula. We present a case of massive right pleural effusion secondary to a PPF due to Pancreatitis.

Keywords: Pancreatic pleural effusion, Pancreatico-pleural fistula

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I. Introduction

Pancreaticopleural fistula (PPF) is a rare condition that occurs in approximately 0.4% of patients with pancreatitis¹. It is an uncommon severe complication of chronic and acute pancreatitis or after a traumatic disruption of the pancreatic duct² It is much more commonly seen in patients with chronic pancreatitis (90%) than acute pancreaticis³ Due to chronic pancreatic inflammation, an abnormal connection (fistula) is formed between the pleural space and the pancreatic duct leading to pancreatic secretion drainage into the pleura, causing pleural effusion that is high in amylase. Pancreatic pleural effusions account for less than 1% of all cases of pleural effusion ⁴ A massive pleural effusion is the most common radiologic finding, and it can be left-sided (76%), right-sided (19%), or bilateral(14%)⁵. It poses a diagnostic challenge. Patients mostly present with pulmonary symptoms related to the pleural effusion rather than to pancreaticing to delay in diagnosis⁶ Therapeutic options for PPF are limited. Magnetic resonance cholangiopancreatography (MRCP) and Endoscopic Retrograde Cholangio pancreatography (ERCP) play a vital role if a discrete leak is evident.

II. Case Report

We report a rare case of a 36 years old male alcoholic presenting with breathing difficulty, cough, right-sided chest pain and abdominal pain for one week. On physical examination, blood pressure 120/80 mm Hg, heart rate of 90 beats per minute, respiratory rate of 20breaths/minute, temperature 37.6°C, oxygen saturation 97% with room air were seen. Heart examination showed a regular rate and rhythm with normal S1 and S2. Examination of lungs showed decreased breath sounds on auscultation and stony dull note on percussion of the right side of the chest. Abdominal examination was significant for epigastric and right hypochondriac tenderness. Other systems' examination was unremarkable.

III. Investigations

Serum amylase of 580 IU/L, serum lipase of 2026 IU/L, serum creatinine 0.9mg/dl, BUN 23 mg/dL and total bilirubin 0.9 mg/dL. Chest X-ray (Fig: 1) showed a white-out of the right hemithorax suggestive of the pleural collection. Ultrasound Chest and Abdomen showed Gross right pleural effusion with underlying lung collapse. Computed Tomography (CT) of the chest and abdomen showed a massive right pleural effusion (Fig: 2). Diagnostic thoracocentesis was done, and evaluation of pleural fluid revealed cola-colored, lymphocyte predominant exudates with Adenosine Deaminase (ADA) of 32U/L and high Amylase levels (47860 IU/L). MRI Abdomen with MRCP confirmed the diagnosis of peripancreatic cysts with chronic pancreatitis along with gross right pleural effusion (Fig: 3). The patient was conservatively treated with Octreotide and therapeutic thoracocentesis.



FIGURE 1: Chest X-ray shows large right side pleural effusion.

IV. Discussion

Pleural effusion in pancreatic disease occurs due to two mechanisms. The first is reactionary pleural effusion due to pancreatitis, which is usually small and left-sided (may be bilateral). It is characterized by an average amylase level (< 1000 U/L) and low protein concentration (< 3 g/dl)⁷. This type of effusion is seen in acute pancreatitis and resolves spontaneously, with the recovery of the disease. The second type of pleural effusion in patients with pancreatitis is usually large, left-sided, recurrent, and has a high level of amylase (> 1000 U/L) and proteins (> 3 g/dl). This type of effusion is seen in both chronic and recurrent pancreatitis. In this type of effusion, fluid accumulates in the pleural cavity due to a fistulous communication between either a pancreatic duct or a pseudocyst and the pleura⁸.

Our patient has the second type of Pancreatic Pleural effusion.



Fig 2: Chest CT showing right pleural effusion.

Clinical features are often variable, but overall, pulmonary symptoms are more common than abdominal symptoms. According to Ali *et al.*, ⁹ the most common symptoms are dyspnea in 65%, cough in 27%, and chest pain in 23%. Abdominal pain has been reported in 29% of the cases. Clinically, a massive pleural effusion with a high fluid amylase level and increased protein concentration in a patient with pancreatitis suggest a diagnosis of PPF. Other causes of elevated amylase in the pleural fluid are acute pancreatitis, pneumonia, liver cirrhosis, pulmonary tuberculosis, esophageal perforation, female reproductive tract malignancy, lung cancer, metastatic carcinoma, lymphoma, and leukemia. Our study patient showed all the above signs and symptoms.



Fig 3: MRCP shows findings consistent with peripancreatic cyst

A direct demonstration of this fistulous communication is difficult. CT scan is beneficial in determining the size of the effusion and also reveals changes in pancreatitis. A CT scan (33-47%) may demonstrate the fistulous tract. MRCP and ERCP are considered the investigation of choice for suspected PPF. We were able to get MRCP for our study patient, but ERCP was not available.

Therapeutic options for PPF include medical treatment, endoscopic management, and surgery. The aim of medical treatment is to reduce pancreatic exocrine secretions. Somatostatin analogs are most commonly used along with thoracentesis and/ or tube thoracostomy, which encourage the apposition of pleural surfaces. Medical treatment is usually attempted for two to three weeks. Octreotide administration, along with ERCP and stenting, has been used for a longer period (2.5 to 6 months)¹¹. Octreotide is given in an initial dosage of 50 μ gs, administered subcutaneously three times a day, and the dose is titrated based upon the fistula output. The maximal dose employed is 250 μ g, three times daily¹². It is reported that octreotide significantly reduces the fistula output and decreases the time to fistula closure. The reported success rate of medical management is 30-60%¹³.

We treated this patient with Octreotide and repeated thoracentesis. The institutional Gastroenterologist advised us to proceed with the course. The patient is stable now, and we are keeping him under observation.

V. Conclusion

Pleural effusion secondary to Pancreatico-pleural Fistula is challenging to diagnose and, at times, difficult to treat. A high index of suspicion with early pleural fluid amylase testing and imaging will avoid delay in diagnosis and associated morbidity and mortality. A simple test like pleural fluid Amylase can be done so that further invasive procedures can be avoided. MRCP and ERCP if available confirm the diagnosis.

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