Case Report

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A Rare Cause of Recurrent Pleural Effusion- Pancreaticopleural Fistula

Internal Medicine Section

NAFEES AHMAD KHAN[1], HUMA FIRDAUS[2], ARUSHI VERMA[3], MEHTAB AHMAD[4], MOHAMMAD SHAMEEM[5]

ABSTRACT

Pancreaticopleural fistula is one of the rarest complication of acute pancreatitis, chronic pancreatitis and pancreatic pseudocyst. Diagnosis requires a high index of clinical suspicion in alcoholic patients presenting with pancreatitis and associated recurrent pleural effusion. Diagnosis is delayed as patients usually present with predominant pulmonary symptoms of dyspnea and chest pain. Hereby authors presented a case, where a 50-year-old chronic alcoholic male presented with chief complains of shortness of breath and chest pain. Based on the chest radiograph, Computed Tomography (CT) scan and pleural fluid analysis diagnosis of pancreaticopleural fistula was made. The patient was managed with intercoastal tube drainage and symptomatic treatment for other complains. Medical management, endoscopic and surgical interventions bring out a good prognosis in pancreaticopleural fistula.

Keywords: Chest pain, Chronic alcoholic, Chronic pancreatitis, Pancreatic pseudocyst, Shortness of breath

CASE REPORT

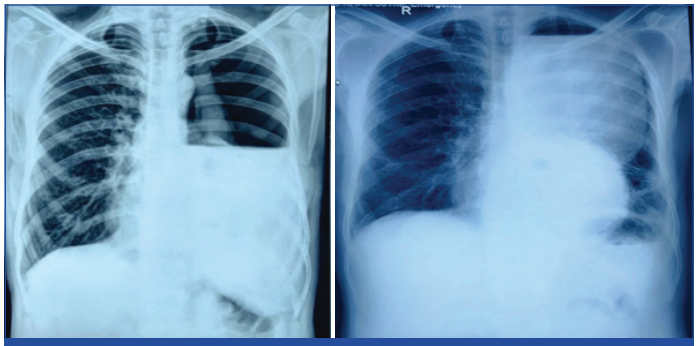
A 50-year-old male presented to the Emergency Department with complains of left-sided chest pain and shortness of breath since last two months, which had increased in the last five days. Appetite and sleep of the patient had decreased in the past two months. Patient had no history of diabetes mellitus, tuberculosis or hypertension. Chest pain was mild in intensity dull aching and diffuse in character not radiating to other sites. He did not complain of any abdominal symptoms. He used to take some oral medication for chest pain from a local practitioner, on and off, for the last two months. His shortness of breath was gradual in onset and progressive. It used to be aggravated on lying down supine and was relieved in recumbent position with head end elevated. He went to some local physician with these complains where chest X-ray was done and diagnosis of left hydropneumothorax was made. A chest tube was placed on left side, four days before presenting to the present hospital [Table/Fig-1]. The patient was chronic alcoholic for last 15 years and used to take around 60 mL of local alcoholic drink daily.

Two litres of brownish turbid fluid was drained post intercoastal tube insertion. No fluid investigations were sent at that time for diagnosing tuberculosis, however, antitubercular treatment was started. After four days of intercostal tube insertion, the tube got accidently removed and he presented with the complains of chest pain and shortness of breath.

The patient was admitted for further evaluation. The patient looked sick. His blood pressure on presentation was 80/60 mmHg, and respiratory rate was 40/minute. On auscultation, breath sounds were reduced on left hemithorax, and it was dull on percussion. A repeat chest X-ray was done, which showed a well-defined homogenous opacity with air fluid level in left lung with sparing of left costophrenic angle [Table/Fig-2].

Blood investigations of the patient were done, and were found to be within normal limits. Findings have been summarised in [Table/Fig-3].

Ultrasound (USG) thorax was done which showed fluid in pleural space posteriorly in interscapular region. Diagnostic and therapeutic aspiration was done from the site. A 200 mL of brownish pleural fluid was aspirated on the day of admission, and the patient was relieved of chest pain and shortness of breath. He was started on broad spectrum antibiotic ceftriaxone-sulbactum along with metronidazole. Noradrenaline was started for management of low blood pressure Pleural fluid investigation was done, fluid was transudative, pleural fluid amylase and lipase were raised. Pyogenic culture of pleural fluid showed pseudomonas species, sensitive to amikacin and colistin. The results have been summarised in [Table/Fig-4].



[Table/Fig-1]: Showing air fluid level in left hemithorax.
[Table/Fig-2]: Showing well-defined homogenous opacity with air fluid level with sparing of left costophrenic angle. (Images from left to right)

Parameters	Results
Total leucocyte count	22,500/cumm
Haemoglobin	9.8 gm/dL
Platelets	3,59,000/cumm
Blood urea nitrogen	10 mg/dL
Serum creatinine	0.55 mg/dL
Serum sodium	138 meq/L
Serum potassium	4.1 meq/L
Total serum protein	4.5 gm/dL
Albumin/globulin ratio	1.3
Total bilirubin	0.7 gm/100 mL
Human immunodeficiency virus, Hepatitis B surface antigen, Hepatitis C virus	Negative

[Table/Fig-3]: Blood investigation reports.

Parameters	Values
Protein	2.3 gm/dL
Sugar	57 gm/dL
Cells	505/cumm
Adenosine deaminase	28.9
Acid fast bacilli stain	Negative
Pyogenic culture	Pseudomonas species

Antibiotic sensitivity	Amikacin and colistin
Amylase	352 IU/L
Lipase	275 IU/L

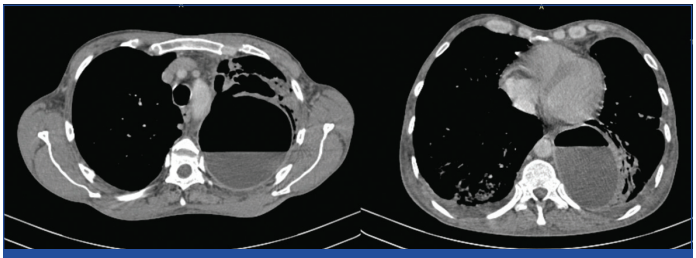
[Table/Fig-4]: Pleural fluid examination findings.

The patient again started complaining of shortness of breath. Repeat USG thorax was done, and around 200 mL fluid was aspirated from interscapular region on day 2 of admission and patient was relieved of shortness of breath following aspiration of fluid.

The patient was started on antibiotics according to sensitivity report, three days after admission. Antitubercular treatment was stopped, since reports were not suggestive of tubercular effusion. Next day repeat chest X-ray showed similar findings as the previous one homogenous opacity with air fluid level with sparing of left costophrenic angle [Table/Fig-2]. However, again in the morning of day 3 patient had similar complains of shortness of breath and chest pain and there was refilling of pleural fluid as seen in USG thorax. In view of recurrent filling of the pleural cavity a CT scan thorax was planned to find the cause of rapid refilling of pleural fluid.

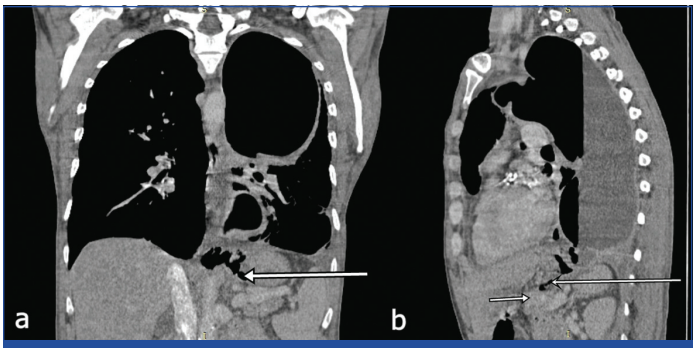
Contrast enhanced CT scan of thorax with abdomen showed atrophic pancreas with multiple parenchymal and intraductal calcific foci in head and tail region with dilated main pancreatic duct (3.4 mm). Hypodense collection in peripancreatic region which appeared to be extending into left pleural cavity forming a large collection with air-fluid level through a focal defect in medical part of left hemidiaphragm (~0.87 cm).

Axial contrast enhanced CT images of the thorax demonstrated a large irregular but well-defined walled cavity with air fluid level, and loculation extending from left lung base to the apex. There were focal areas of eroded walls and adjacent small consolidations and infective nodules [Table/Fig-5].



[Table/Fig-5]: Axial contrast enhanced CT scan showing a large irregular but well defined cavity with air fluid level, loculations and small consolidations and infective nodules.

Coronal and sagittal contrast enhanced CT images of the lower thorax and abdomen demonstrated a thin irregular fistulous tract with foci of air extending from the thorax to the abdomen upto the body of pancreas along the left crux of diaphragm (shown with arrows), reliably demonstrating the communication between the pleural and likely branch pancreatic duct [Table/Fig-6a,b].



[Table/Fig-6a,b]: Coronal and sagittal contrast enhanced CT scan of lower thorax and abdomen showing a thin irregular fistulous tract with foci of air extending from thorax to abdomen upto the body of pancreas along the left crux of diaphragm.

After this CT scan report, amylase and lipase level of pleural fluid was tested to confirm diagnosis of pancreaticopleural communication. Amylase was raised 3.5 times normal lab value (352 IU/L) and lipase was raised 2.5 times normal lab value (275 IU/L).

So, on the basis of clinical history, laboratory and radiological findings diagnosis of complicated chronic pancreatitis with pseudopancreatic cyst formation with pancreaticopleural fistula extending into left pleural cavity was made. However, the patient was very sick and died on day 6 of admission before any intervention for the defect could be done.

DISCUSSION

Incidence of pancreaticopleural fistula in acute pancreatitis, chronic pancreatitis and pancreatic pseudocyst are 1% , 0.4% and 4.5 %, respectively [1]. It is formed primarily of granular and fibrous tissue and lacks epithelial lining hence called pseudocyst and is filled with pancreatic secretions [1]. Alcohol is the most common etiological agent causing chronic pancreatitis leading to pancreaticopleural fistula formation [2]. Others causes of pancreaticopleural fistula formation are gallstone pancreatitis, trauma, congenital anomalies of pancreatic duct or idiopathic pancreatitis [3]. Treatment of pancreaticopleural fistula can be medical, endoscopic or surgical.

The clinical presentation is often misleading as presenting symptoms are usually associated with significant pleural effusion and consist of dyspnea, cough, chest pain, fever and septicaemia [3,4]. Pulmonary symptoms are more common than abdominal symptoms and are usually the presenting symptom with dyspnoea being the most common [5].

As symptoms are mainly pulmonary, the diagnosis is usually delayed with the average time of diagnosis is five weeks [6]. Diagnosis can be established via thoracentesis with pleural fluid analysis demonstrating exudative effusion with high amylase level [6,7]. There is no cut-off point for amylase level. However, it is usually significantly elevated with a mean amylase level above 10,000 U/L [1,7]. Of note, serum amylase has no role in the diagnosis [8]. Elevated levels of pleural fluid amylase are also found to be associated with other conditions such as pancreatitis, pulmonary malignancy (adenocarcinoma, mesothelioma) or esophageal rupture [9].

Pancreaticopleural fistula can be visualised by CT scan but overall sensitivity is low [10]. The investigation of choice now-a-days is Magnetic Resonance Cholangiopancreatography (MRCP) because it is a non invasive procedure as opposed to Endoscopic Retrograde Cholangiopancreatography (ERCP) and it can visualise structures beyond the strictures [7].

Treatment options for pancreaticopleural fistula are medical, endoscopic and surgical [1,6,7]. Medical treatment with octreotide accompanied by total parenteral nutrition has been found to be successful in 31-65% of patients [11]. Octreotide is given to decrease pancreatic secretions.

Intercostal drainage insertion can be done to drain recurrent pleural effusion. However, medical management is a long process and increases duration of hospital stay. Other complications of medical management include malnutrition, catheter infection and sepsis [6]. Endoscopic treatment with ERCP works by the following mechanism [12,13]:

1. Mechanically block abnormal pancreatic duct connection with pleural space.
2. To keep pancreatic duct open so that pancreatic secretions go downward to duodenum through path of lower resistance escaping the abnormal pleural connection with higher resistance.

Complications include anatomical disruption of pancreatic duct and recurrent fluid accumulation [12,13]. However, when both medical and endoscopic treatment fails it is an indication for surgical management. With advanced techniques it is associated with quicker recovery, better outcome and low morbidity and mortality than medical or endoscopic treatment [7].

Francisco E et al., [2], Mahmoud M et al., [14], Hirosawa T et al., [15] and Kaur H et al., [16] young alcoholic patients who presented with chief complains of dyspnea and chest pain. Upon investigation patients were found to have pancreaticopleural fistula with pleural effusion. The authors followed varied management modalities-surgical intervention, conservative medical management, conservative and endoscopic management, and conservative medical management. Acute pancreatitis, chronic pancreatitis and pancreatic pseudocyst can all lead to formation of pancreaticopleural fistula. Pancreatic pseudocyst formation is more common in chronic and alcohol related pancreatitis as compared to acute and non alcohol related pancreatitis [17].

The communication between the thoracic structures and pancreatic duct can be through pseudocysts which may be incompletely formed or ruptured or through thin linear fistulous tracts through the esophageal or aortic diaphragmatic orifice or less commonly transdiaphragmatically. The result is large pleural effusions, unilateral or bilateral, mediastinal fluid collections or pseudocysts [18]. Depending upon site of disruption of pancreatic duct it can either disrupt posteriorly forming a fistula between pancreatic duct and pleural space leading to pleural effusion usually on left side which can be massive and recurrent but can be either bilateral or only on right side. If disruption occurs anteriorly it leads to pancreatic ascites with collection of amylase and lipase rich fluid in the peritoneum [3,4].

CONCLUSION(S)

Pancreaticopleural fistula is a rare complication of acute pancreatitis, chronic pancreatitis and pancreatic pseudocyst. A high index of clinical suspicion is necessary for early diagnosis and intervention as presenting symptoms are predominantly pulmonary and delay in diagnosis and intervention can adversely affect the outcome.

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PARTICULARS OF CONTRIBUTORS:

1. Assistant Professor, Department of TB and Respiratory Diseases, Jawaharlal Nehru Medical College, Aligarh, Uttar Pradesh, India.
2. Senior Resident, Department of TB and Respiratory Diseases, Jawaharlal Nehru Medical College, Aligarh, Uttar Pradesh, India.
3. Junior Resident, Department of TB and Respiratory Diseases, Jawaharlal Nehru Medical College, Aligarh, Uttar Pradesh, India.
4. Assistant Professor, Department of Radiodiagnosis, Jawaharlal Nehru Medical College, Aligarh, Uttar Pradesh, India.
5. Professor, Department of TB and Respiratory Diseases, Jawaharlal Nehru Medical College, Aligarh, Uttar Pradesh, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Huma Firdaus,
Senior Resident, Department of TB and Respiratory Diseases, Jawaharlal Nehru Medical College, AMU, Aligarh, Uttar Pradesh, India.
E-mail: huma2107@gmail.com

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