

Massive Right-sided Pleural Effusion due to Pancreaticopleural Fistula: A Rare Case Report

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ABSTRACT

Pancreaticopleural Fistula (PPF), a rare occurrence, is characterised by an abnormal connection between pleural and peritoneal cavity. It forms usually due to the rupture of pancreatic pseudocyst. Pancreatic pseudocyst is formed due to the repeated episodes of acute or chronic pancreatitis. Most pancreatic pathologies, such as acute and chronic pancreatitis, cause clinically insignificant effusions that resolve with disease control, whereas PPF causes massive effusion and is thus rarely considered as a cause of massive pleural effusion due to its low incidence. PPF are most common in middle-aged alcoholic males and typically present as massive recurrent left-sided pleural effusions. In the present case, a 50-year-old male presented to the Department of Respiratory Medicine with the complaint of right-sided chest pain, progressive breathlessness, abdominal pain and loss of appetite. Patient was labourer by occupation, chronic alcoholic, non diabetic. Contrast Enhanced Computed Tomography (CECT) thorax with abdomen showed massive right-sided pleural effusion and pancreatitis. A diagnosis of PPF was confirmed as its cause. Pleural fluid analysis for elevated amylase, confirms the diagnosis and investigations such as CECT thorax and abdomen, Endoscopic Retrograde Cholangiopancreatography (ERCP), or Magnetic Resonance Cholangiopancreatography (MRCP) may be used to establish the fistulous communication between the pancreas and the pleural cavity. PPF should always be considered as a differential diagnosis while finding a cause for right-sided pleural effusion.

Keywords: Alcoholic, Contrast enhanced computed tomography, Pleural tapping, Recurrent pancreatitis

CASE REPORT

A 50-year-old Hindu, married male, labourer by occupation came to the Department of Respiratory Medicine with a chief complaint of right-sided chest pain, progressive breathlessness, abdominal pain and loss of appetite in the last four months. Abdominal pain was dull aching to burning type, more over the epigastric region, radiating to back and was associated with nausea. Pain aggravated on bending forwards.

Patient also visited to the family physician for similar complaint of chest pain and dyspnoea, two months back where chest X-ray was done, which suggested moderate right-sided pleural effusion [Table/Fig-1]. Patient was started on empirical antitubercular treatment, but the symptoms of the patient did not resolve even after two months and was referred to the department. Patient was a chronic alcoholic since last 20 years with a frequency of around 200-300mL per day and was admitted for acute pancreatitis before three years for which he was treated symptomatically. Patient was non diabetic and non hypertensive. There was no relevant family history.



[Table/Fig-1]: Chest X-ray posteroanterior view showing right-sided moderate pleural effusion (yellow arrow).

On examination patient was conscious, co-operative and well oriented to time place and person, had a heart rate of 110 beats per minute, respiratory rate was 20/min, temperature was 98.1°F and oxygen saturation was 95% at room air. On systemic examination, respiratory movements were reduced over the right-side and trachea shifted towards left-side. On percussion there was presence of stony dull note over the right hemithorax. On auscultation the breath sounds and vocal resonance were decreased over the right hemithorax.

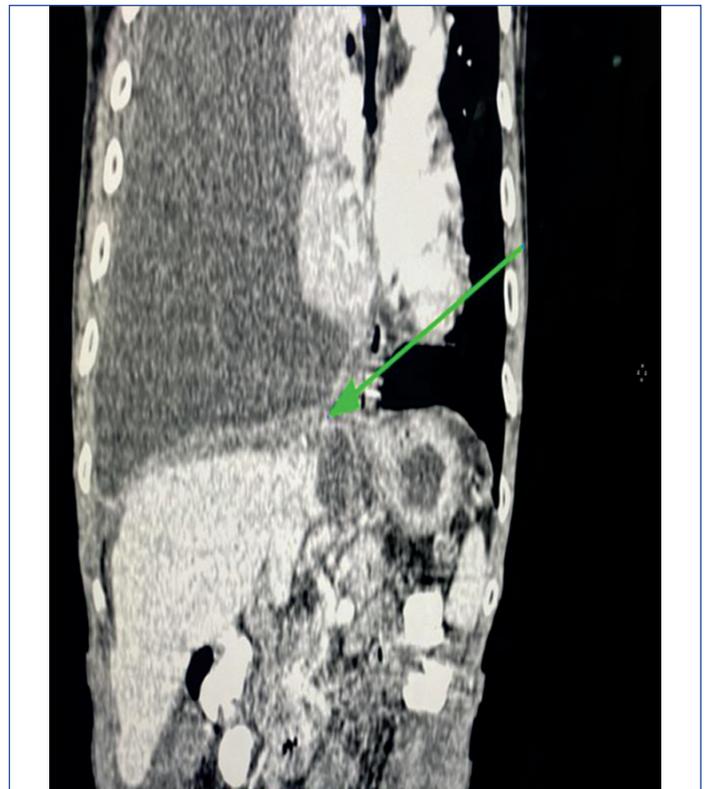
Patients' routine blood investigations were carried out which revealed haemoglobin was 8.8 gm/dL, total white blood cell count was 14,000 cells/cumm, serum creatinine was 0.6 mg/dL, serum total bilirubin was 0.6 mg/dL and random blood sugar was 107 mg/dL. Patients' serum amylase was 514 U/L and serum lipase was 526 U/L.

Patients' chest X-ray PA view showed massive right-sided pleural effusion with shifting of trachea to the opposite side [Table/Fig-2]. Ultrasonography (USG) Thorax was suggestive of right-sided pleural effusion of approximately 2000 mL in amount [Table/Fig-3]. Thoracocentesis was performed from right-sided 5th intercostal space and 1100 mL of pleural fluid was drained. The pleural fluid tapped was brownish black in colour [Table/Fig-4] with normal glucose and pH value. Pleural fluid adenosine deaminase was 15 U/L. Pleural fluid Cartridge Based Nucleic Acid Amplification Test (CBNAAT) did not detect the *Mycobacterial Tuberculi*. The pleural fluid amylase was 2683 U/L and pleural fluid lipase was 3250 U/L. Thus, the differential diagnosis thought of to be was reactionary fluid secondary to pancreatitis, trauma, pancreatic malignancy, pulmonary malignancy, oesophageal rupture or Pancreaticopleural Fistula (PPF).

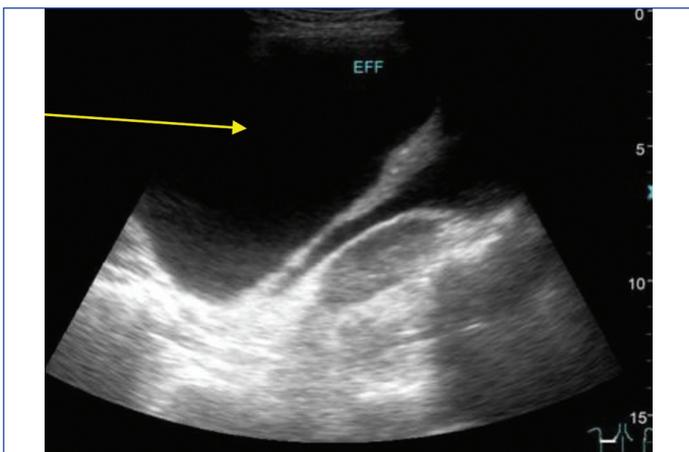
Contrast-enhanced Computed Tomography (CECT) thorax revealed a massive pleural effusion on the right-side, as well as the collapse of the underlying lung. CECT Abdomen revealed a subtle peripheral enhancing cystic lesion of size 4.4x4.1x7.5 cm involving the epigastric region. It reached up to the body of the pancreas inferiorly, indicating



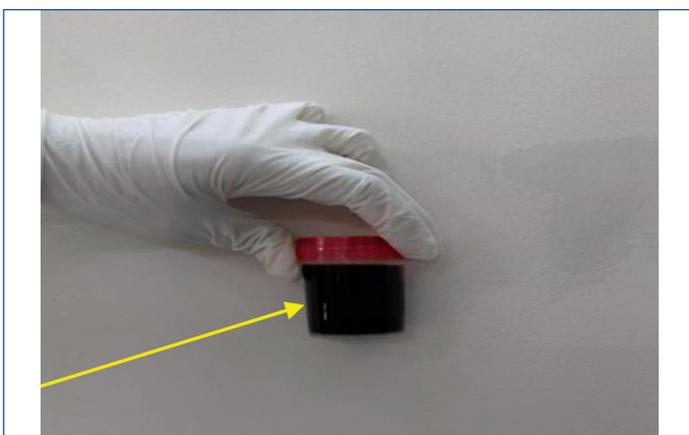
[Table/Fig-2]: Chest X-ray PA view showing massive right-sided pleural effusion (yellow arrow).



[Table/Fig-5]: Contrast-enhanced computed tomography thorax with abdomen pelvis showing fistulous tract between head of pancreas and pleural cavity (green arrow).

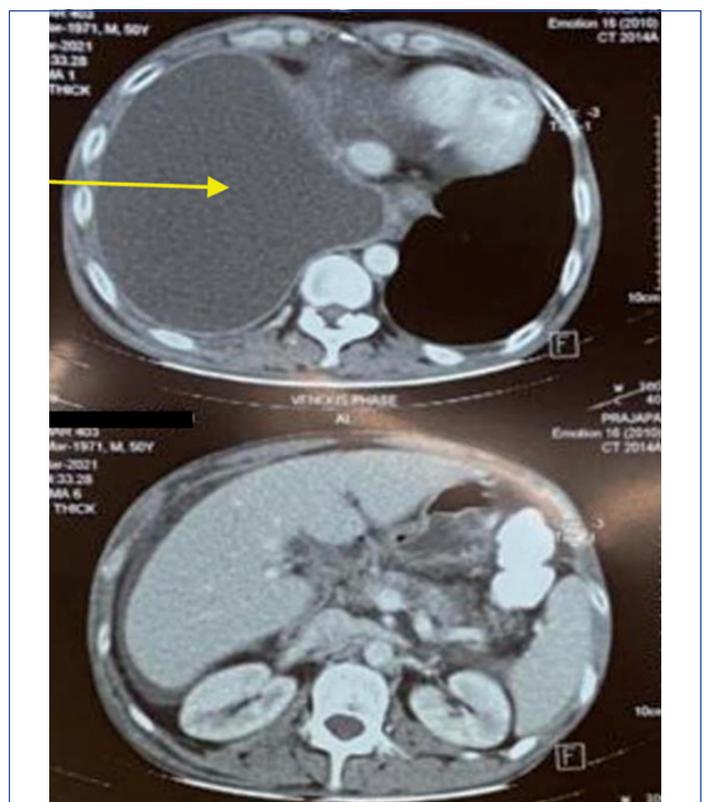


[Table/Fig-3]: Ultrasonography thorax showing pleural effusion (yellow arrow).



[Table/Fig-4]: Showing brownish black pleural fluid (yellow arrow).

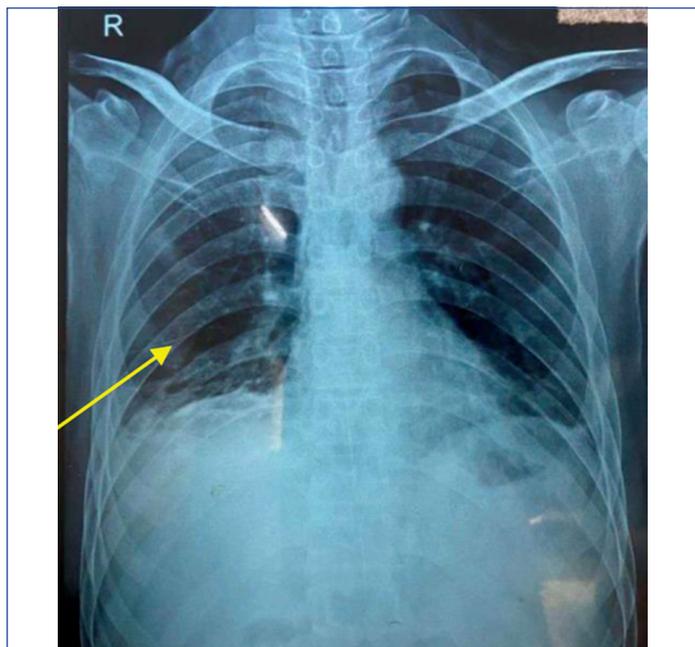
that it originated in the pancreas. It communicated superiorly via a small fistulous track connecting the cystic lesion and the diaphragmatic pleura, which extended into a large peripherally enhancing collection in the right pleural cavity. The distal main pancreatic duct appeared dilated and measured 6 mm [Table/Fig-5,6]. Thus, the diagnosis of presence of PPF was made.



[Table/Fig-6]: Contrast-enhanced computed tomography thorax with abdomen pelvis showing pancreatic changes (yellow arrow) and fluid in right pleural cavity.

Patient was referred to gastrosurgeon for an opinion and was started on injection ceftriaxone 1 gm i.v. every 12 hourly and injection metronidazole 500 mg i.v., every eight hours for 14 days. Patient was given injection octreotide 100 mcq subcutaneously every eight hourly for 10 days and total parental nutrition. Pleural effusion was drained over three days through thoracocentesis up to 1100 mL per day as the patient denied for ICD insertion or any other invasive procedure. With the advancement of the treatment over the period of two weeks patients complain of dyspnoea regressed and

abdominal symptoms also decreased in intensity. Follow-up X-rays showed decreased in the amount of pleural fluid which got cleared completely after two weeks [Table/Fig-7].



[Table/Fig-7]: Follow-up X-ray showing decreased right pleural cavity fluid (yellow arrow).

DISCUSSION

The PPF is an abnormal connection formed between pleural cavity and pancreatic duct. Pancreatic secretions tend to ascend to the pleural cavity forming pleural effusions [1]. PPFs lead to pleural effusion that is extremely rare. Pleural effusion occurs in 0.4% of chronic pancreatitis patients and accounts for approximately 1% of total pleural effusion cases [2,3]. Pleural effusions can occur because of the reactive changes with pancreatitis but in such cases, they are more common on left-side and are self-limiting. Pleural effusions occur in up to 3-7% of cases with pancreatitis. Pleural effusions related to PPF are recurrent, also more common on left-side but rarely they can occur over right-side or can even be bilateral [1]. In the present case, the effusion was seen on right-side which was a rarer presentation.

Suspicion of PPF can occur by presence of high levels of amylase in pleural effusion usually above 1000 U/L. Amylase rich pleural effusions are also seen in cases of malignancy which can be either primary (mesothelioma) or metastatic (especially adenocarcinoma and oesophageal rupture, lymphoma, leukaemia, liver cirrhosis, hydronephrosis and pulmonary tuberculosis but are usually not very high and thus were thought to be as the differential diagnosis [4]. In this case also the pleural fluid amylase was 2683 U/L, but CECT thorax showed the presence of PPF giving the final diagnosis.

In the cases with moderately elevated amylase levels, imaging findings are helpful in diagnosis. Following this, the next step is confirmation of

the presence of the fistula, for which most sensitive imaging modality is Magnetic Resonance Cholangiopancreatography (MRCP), followed by Endoscopic Retrograde Cholangiopancreatography (ERCP) and CT scan [5]. CT has the advantage of being non invasive and the capability of providing three-dimensional images [6]. In the above described case, diagnosis of presence of PPF was also made on CECT Thorax as patient denied any invasive procedure proving CECT Thorax to be diagnostic modality for this case.

Kaur H et al., saw a similar case of massive pleural effusion secondary to PPE. In their case effusion was seen over right side and PPE was diagnosed on CECT Thorax and Abdomen. The patient was managed conservatively and patient showed improvement on follow-up [7].

Treatment of PPF includes conservatively with stent placement and/or octreotide or surgically. ERCP is essential for diagnosis and management of it. Conservative treatment has shown its success in around 31-45% whereas success rates with surgery have been 80-90% but has a high mortality. As the patient denied for any invasive procedure, patient was managed conservatively [8].

In a similar case seen in Egypt, a MRCP was diagnostic for the PPF. The patient was managed by stent placement in the main pancreatic duct through ERCP. ERCP was diagnostic as well as therapeutic as it showed the duct morphology also showing its necessity in a case of PPF. But it has a drawback of being invasive procedure [9].

CONCLUSION(S)

The PPF are a rare occurrence and right-sided pleural effusions because of them are even rarer, proving to be a diagnostic challenge because predominant symptoms are related to chest rather than abdomen. Thus, PPF as differential should be kept in patients with habit of chronic alcoholism and whenever in suspicion pleural fluid amylase and CECT thorax with abdomen should be carried out.

REFERENCES

- [1] Tauseef A, Nandakumar S, Vu L, Chimpiri AR, Tierney WM. Pancreaticopleural fistula. *Pancreas*. 2009;38:e26-31.
- [2] Tay CM, Chang SKY. Diagnosis and management of pancreaticopleural fistula. *Singapore Med J* [Internet]. 2013;54(4):190-94. Available from: <http://dx.doi.org/10.11622/smedj.2013071>.
- [3] Dhebri AR, Ferran N. Nonsurgical management of pancreaticopleural fistula. *JOP*. 2005;6(2):152-61.
- [4] Villena V, Pérez V, Pozo F, López-Encuentra A, Echave-Sustaeta V, Arenas J, Martín Escribano Amylase levels in pleural effusions: A consecutive unselected series of 841 patients chest. *Chest*. 2002;121:470-74.
- [5] Singh S, Yakubov M, Arya M. The unusual case of dyspnea: A pancreaticopleural fistula. *Clin Case Rep*. 2018;6:1020-22.
- [6] Chan EEH, Shelat VG. Pancreaticopleural fistula causing massive right hydrothorax and respiratory failure. *Case Rep Surg* [Internet]. 2016 [cited 2022 Oct 29];2016:8294056.
- [7] Kaur H, Singh D, Kajal NC, Garg D. A case report of pancreatico pleural fistula presenting as recurrent right pleural effusion. *Arch Pulmonol Respir Care*. 2021;7(1):15-17. Doi: 10.17352/aprc.000069.
- [8] Safadi BY. Marks pancreatic pleural fistula: The role of ERCP in diagnosis and treatment gastrointestinal. *Gastrointest Endosc*. 2000;51:213-15.
- [9] Bediwy AS. Pancreatico-pleural fistula: A rare cause of massive right-sided pleural effusion. *Egyptian Journal of Chest Diseases and Tuberculosis*. 2015;64(1):149-51. ISSN 0422-7638.

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