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Case Report

AN UNUSUAL CASE OF RESPIRATORY DISTRESS IN THE EMERGENCY DEPARETMENT – UNCOVERING A RARE COMPLICATION OF A COMMON DISEASE

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ABSTRACT
Although pleural diseases in cases of pancreatitis are well recognized, haemorrhagic pleural effusions are less common and most commonly are on the left side. A right sided massive haemorrhagic pleural effusion is a rare finding and needs further investigation to find out the cause. Here we describe a case with a recent history of Necrotizing Pancreatitis who presented with massive right sided pleural effusion which was picked on Physical examination and Chest Radiography in the emergency. Percutaneous needle aspiration revealed frank blood in the pleural cavity. A diagnosis of Pancreatico-Pleural Fistula was pursued after ruling out other common causes and was subsequently confirmed using biochemistries, radiological modalities, invasive study and most importantly through clinical judgement. Therapeutic interventions in form of chest tube drainage and pancreatic duct stenting were done and the patient responded drastically. He was discharged in good health and was reviewed on a later date for stent removal. Due to high index of suspicion and prompt interventions, patient avoided undergoing major abdominal surgery which adds to the morbidity and mortality in such aggressive disease.

Keywords: *Pancreatico-Pleural Fistula*

INTRODUCTION
Pancreatico-pleural fistula is one of the rare complications of pancreatitis. It is commonly associated with chronic pancreatitis but it has also been described in cases of acute pancreatitis. Small exudative pleural effusions on either side are found which are rich in amylase. Hemothorax is rarest presentation of a Pancreatico-pleural fistula and results in hemodynamic collapse if large enough. Prompt recognition is necessary as sometimes patient may require extensive surgery for repair of the fistulous tract. Also, in a country like India, a large hemothorax from pancreatic disease needs to be differentiated from other common causes like malignancy, tuberculosis and other infections. Hence this case represents a diagnostic challenge to the internist or emergency residents and urges them to look beyond the obvious.

CASES
We describe a 31 year old male with history of alcoholism from last 10-12 years. The patients had recovered from acute necrotizing pancreatitis which he suffered 8 weeks ago.
Despite strong medical advice against it, patient resumed consuming alcohol with frequent binging episodes. Patient was brought to our emergency department with an episode of acute onset severe epigastric pain which had started 72 hours ago, followed by a rapid onset of respiratory distress from last 24 hours.
On initial examination in the emergency department, the patient appeared pale with normal consciousness and orientation. His vitals revealed tachycardia (118 beats/ min), tachypnea (32breaths/min), Hypotension (80/50 mm Hg) and a normal body temperature with cool peripheries. His pulse oxymetry showed a saturation of 84% on room air.
Examination of his respiratory system revealed a left ward shift of the trachea. On the right side there were reduced tactile fremitus, dull percussion note over the entire hemi thorax, absent breath sounds and reduced vocal resonance. Cardiac examination showed a left ward shifted apical impulse with otherwise

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normal auscultatory findings. There was marked tenderness in the epigastric region without any apparent organomegaly. Patient was further managed in the medical ICU.

Initial resuscitative measures included multiple intravenous crystalloid infusionsand oxygen therapy. His shock responded to fluid administration. Later in course, vasopressor agent was added to maintain mean arterial pressure within normal limits. The initial chest radiograph (Figure 1) showed a diffusely whiteout right hemi-thorax with shifting of mediastinal structures to the opposite side.

Diagnostic percutaneous needle aspiration with a wide bore needle revealed frank pleural haemorrhage in the right pleural cavity (Figure 2).

Therapeutic pleural tap followed with removal of 500 ml of pleural fluid which resulted in transient partial relief of his respiratory distress. An intercostal drain was put in the right pleural cavity on the next day.

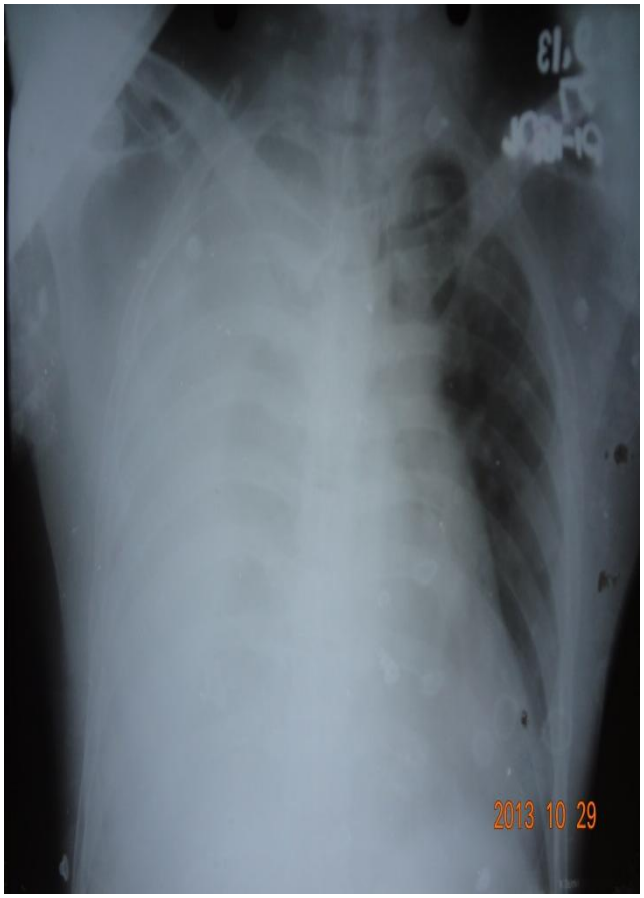

Figure 1: Chest radiograph showing gross pleural effusion on the right side

Figure 2: Haemorrhagic pleural fluid in a container bedside

His haematological and biochemical parameters were as follows:

Hemoglobin - 6.5 g/dl, Hematocrit- 27%, Total leucocyte count- 14,000/ per cu. mm.

Total serum protein/Albumin- 4.9 and 2.2 g/dl respectively.

Bilirubin (*Total/Direct/Indirect*) - 1.1/0.28/0.82 mg/dl

AST / ALT - 17/10 IU/ml

Blood Urea – 21 mg/dl; Serum Creatinine – 1.0 mg/dl

Serum Calcium(*Total*)- 8.6 mg/dl

Sr. Amylase/Lipase- 101/1592 IU/L

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Pleural fluid Studies:
Protein/Sugar- 4.9 g/dl and 44 mg/dl respectively
Total cell count- 1800 cells/ mm^3
Differential count- Polymorphs- 60%, Lymphocytes - 35%, Mesothelial cells- 5%, Hematocrit – 15%
Pleural fluid Gram's stain did not reveal any organism.
Pleural fluid cultures- Sterile.
Pleural fluid AFB stain – Negative
These finding were consistent with acute pancreatitis and pleural cavity haemorrhage and warranted further evaluation for presence of a Pancreatico-pleural fistula.
Computed tomography of the abdomen was carried out with intravenous contrast to look for pancreas and related complications. There was marked destruction in the peri pancreatic region with walled off necrosis. Right sided pleural effusion was also noted. No delineation of the fistulous tract could be made at the time of scan (Figure 3)

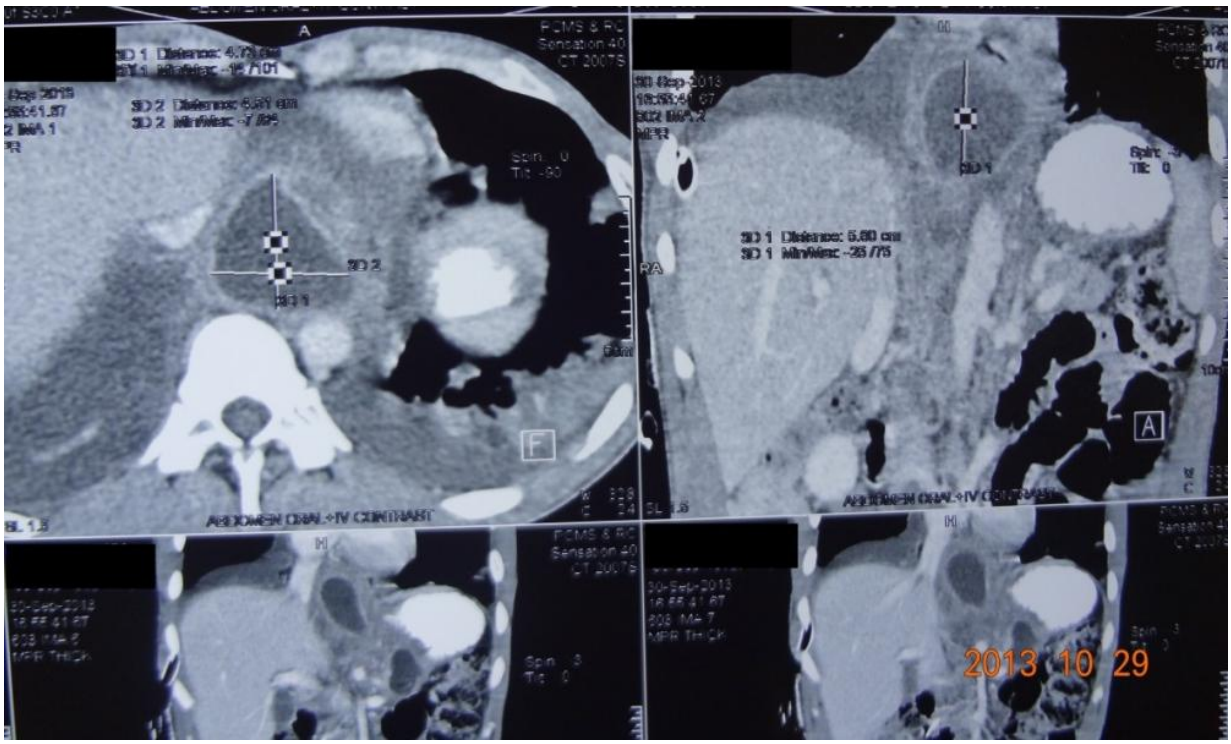

Figure 3: CT scan of abdomen consistent with peripancreatic fluid collection and a right pleural effusion

This was followed by an emergent ERCP to establish the exact path of the fistulous tract and also for stenting of the pancreatic duct to assist in drainage of the necrotic fluid collection around the pancreas.
ERCP did not reveal the fistulous tract. This was followed by papillotomy and insertion of a 5 F plastic pancreatic duct stent (Figure 4). The procedure was uneventful and patient made drastic recovery in next 48 hours. His pleural fluid accumulation was reduced from 500 ml/ day to less than 100 ml/ day along with change in quality from frankly haemorrhagic to blood stained transudative. Repeat pleural fluid amylase levels were 201 IU/ ml which pointed towards cessation of the pleural fluid leak. He was given multiple blood transfusions as well before and after the procedure which resulted in rise of the peripheral hematocrit.

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Figure 4: Post ERCP radiography showing reduction of pleural fluid collection and a radio-opaque pancreatic duct stent in-situ

Abbreviations- ERCP – Endoscopic retrograde cholangio-pancreaticography, g/dl- grams per decilitre, IU/ml – International units per millilitres, mg/dl - milligrams per decilitre, PPF – Pancreatico pleural fistula)

Subsequently, the patient showed improvement in his vitals and the chest drain was removed. He was observed for two days in the ward for any deterioration. He was later discharged in stable condition and was advised to return after 14 days for removal of the pancreatic duct stent which was successfully carried out on. He remains healthy till date and has joined rehabilitation program to aid abstinence from alcohol.

DISCUSSION

Pleural effusions are seen in up to 53% patients with pancreatitis. Majority of them are small effusions which are rich in amylase. Left sided effusion is thrice as more common as compared to right side and in some cases the effusion can be bilateral. They are often picked up on plain chest radiographs (McKenna, 1977).

Pancreatico-pleural fistula (PPF) is one of the rare complications of pancreatitis and can pose as a decoy in diagnosing pancreatic pathology as most of the cases present with respiratory distress or large or recurrent haemorrhagic pleural effusions. Males in their mid-Forties with history of chronic alcoholism and several previous episodes of pancreatitis form the classical description of patients who tend to develop PPF. Hence most of the cases are seen in association with chronic rather than acute pancreatitis (Kilic *et al.,* 2008; Vyas, 2009; Tay *et al.,* 2013).

Regarding the pathogenesis of these complications, possible theories include: A) Transport of amylase rich fluid from the blood across the capillaries that supply the pleural space. Due to development of a concentration gradient across the capillary membrane, there is diffusion of fluid into the pleural space. B) Direct perforation or formation of actual fistula between the abdominal and thoracic cavity is possible and has been demonstrated at necropsy. Burrowing pseudocycts can follow this course and leak into the pleural space. C) Implication of the lymphatic plexus in and around the diaphragm that connects the abdominal cavity with the mediastinum and sub-pleural space and subsequent leakage of the enzyme rich fluid into pleural space due to increased permeability (Kaye, 1968). Diagnosis of PPF requires demonstration of inflammation in the pancreas and high pleural fluid amylase levels. Although there is no cut off for the level of pleural fluid amylase, a level of >200 IU/L is considered to be amylase rich. An

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alternative diagnosis should be sought in the absence of pancreatic disease as high amylase levels can be seen in variety of conditions, most common being pulmonary neoplasms, tuberculosis, para pneumonic effusion /empyema, liver cirrhosis and other benign conditions. In cases of PPF, the pleural fluid amylase would be significantly elevated (>1000 IU/L) with mean levels above 10,000 IU/L (Tay *et al.,* 2013). Once the suspicion of PPF is generated, several imaging modalities are available for visualisation of the fistulous tract among which CT, ERCP and MRCP are the most widely accepted ones with sensitivity of 47%, 78% and 80% respectively. Initial attempts of visualizing the fistulous tract were made by injecting the contrast medium into the pleural space. ERPC was widely used afterwards for a long period and was advantageous in regards that it allowed direct visualisation of the ampulla and stenting of the pancreatic duct during the procedure. But due to the invasive nature and inability to visualize the tract beyond a stricture, the use of ERCP as the first investigation is discouraged. As such, despite its lack of therapeutic options, MRCP is considered the imaging study of choice for PPF due to its superiority over CT in identifying a fistula in the pancreatic region and its non-invasiveness as compared to ERCP (Keiwiet *et al.,* 2009). The management of PPF can be either conservative, which includes medical and endoscopic procedures, or surgical exploration and repair of the fistulous tract. Medical management includes administration of Octreotide and total parenteral nutrition. Chest tube drainage can be continued up to a period of 25 days but attempt should be made to remove the drain as soon as possible. Endoscopic management includes small ampulla sphincterotomy and placement of a pancreatic duct stent to allow drainage of necrotic amylase rich fluid into the duodenum and facilitating closure of the fistulous tract. The success rate of medical management is estimated to be 31%-65% (Dhebri, 2005).

Surgical treatment is used as a last resort when the medical management has failed. However, unnecessarily delaying the surgical procedure when strongly indicated can result in greater morbidity and mortality (Wronski *et al.,* 2011).

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