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Case Report

A rare case of pancreato-pleural fistula with left sided massive pleural effusion

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ABSTRACT

Pancreaticopleural fistula is a very rare complication of acute and chronic pancreatitis. High index of suspicion is needed to diagnose in patients with alcohol induced pancreatitis, presenting with recurrent or persistent pleural effusion. Patient typically presented with pulmonary symptoms, rather than abdominal complaints, leading to delay in the diagnosis. Here we present a case of a known chronic pancreatitis who presented to us with massive left sided pleural effusion. Blood coloured pleural fluid analysis showed lipase and amylase levels in hundred thousand and ten thousand ranges. Diagnosis was made by CECT abdomen and confirmed with MRCP. Patient was treated with first line medical management of thoracocentesis and ERCP with pancreatic duct stenting.

Keywords: Pancreaticopleural fistula, Pleural effusion, Pancreatitis

INTRODUCTION

Pancreaticopleural fistula is very rare and a serious complication of chronic pancreatitis, with an incident rate of 0.4% in patients with pancreatitis.[1] It is characterized by massive pleural effusion, which could be recurrent or persistent.

Literature suggests, control of the pancreatic secretion with octreotide, and closure of the fistula, improves the outcome. Here, we present a case of pancreaticopleural fistula, who presented with a massive left sided hemorrhagic pleural effusion, who was managed with pancreatic duct stenting. Predominant respiratory system findings, and hemorrhagic nature of the pleural fluid, posed a challenge to the clinical diagnosis, which is being discussed here.

CASE REPORT

35-year-old male, chronic alcoholic for 15 years, with past history of recurrent pancreatitis, pancreatic diabetes, presented with complaints progressive breathlessness and right sided chest pain for 2 weeks. On physical examination, patient was appearing malnourished, tachypneic, with oxygen saturation of 96% at room air, stony dull note to percussion, with absent breath sounds, on the entire left side of the chest. Cardiac auscultation revealed normal heart sounds. ECG showed sinus tachycardia, and cardiac biomarkers were not raised. Epigastric tenderness was observed on abdominal examination. Examination of other systems was unremarkable. Chest radiography revealed massive left sided pleural effusion (Figure 1).

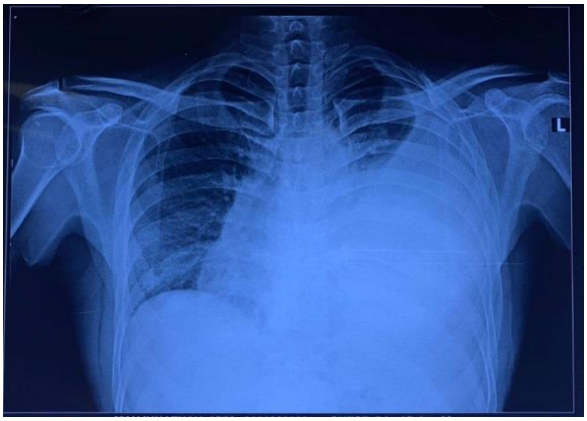

Figure 1: Before the first thoracocentesis, showing massive left sided pleural effusion.

Emergency thoracocentesis was done, approximately 1 litre of bloody fluid (Figure 2) was tapped from the left pleural space and sent for analysis which revealed an exudative pattern, with pleural fluid LDH- 704 IU/l (serum LDH- 136 IU/l), total protein- 4.8 g/dl (serum total protein- 6.3 g/dl) lipase level of 1,86,440 IU/l, amylase level of 22,362 IU/l with the presence of RBCs. Pleural fluid for cartridge based nucleic acid amplification test (CBNAAT) tested negative for *Mycobacterium tuberculosis*. Cytological evaluation of the fluid did not reveal malignant cells, CA-19-9 levels were normal and cultures were negative for aerobic, anaerobic and fungal organisms. Serum amylase and lipase levels were 403 IU/l, 1564 IU/l, respectively.

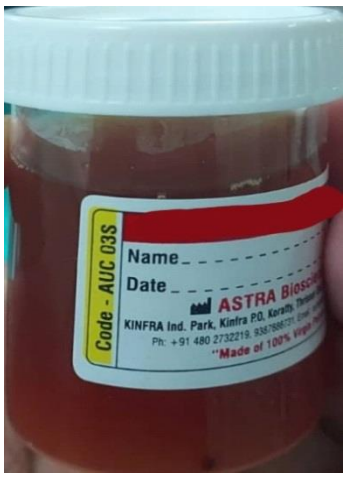

Figure 2: Blood coloured pleural fluid, aspirate.

Pancreatitis can sometimes provoke a reactive pleural effusion, but a pancreatic pleural effusion is distinguished by the high amylase content (typically greater than 1000 units/l) of the pleural fluid, observed in this patient, with recurrent development of left sided pleural effusion (Figure 3). With the working diagnosis of necrotizing pancreatitis with pancreaticopleural fistula. Plain CT of the chest and abdomen (Figure 4) showed moderate left sided pleural effusion with bulky pancreas and few parenchymal calcifications. CECT (contrast enhanced computed tomography) of the chest and abdomen (Figure 5) showed thin hypodense peripherally enhancing thin-walled collection tracking from the distal body of the main pancreatic duct to the posterior mediastinum, with communication to the left pleural cavity and another thin hypodense peripherally enhancing collection noted in the anterior mediastinum, pericardiac region abutting the right atrium with all these features suggestive of pancreatico pleural fistula.

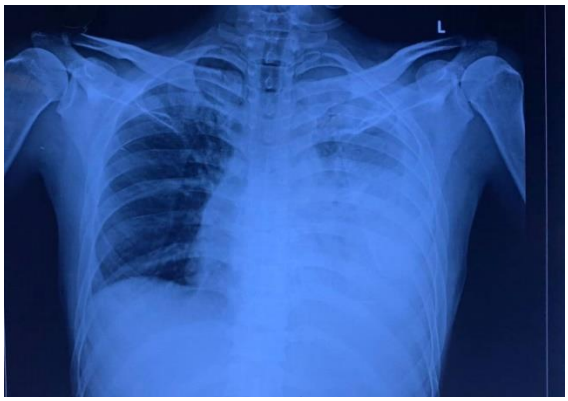

Figure 3: Repeated chest X-ray with recurrent left sided pleural effusion.

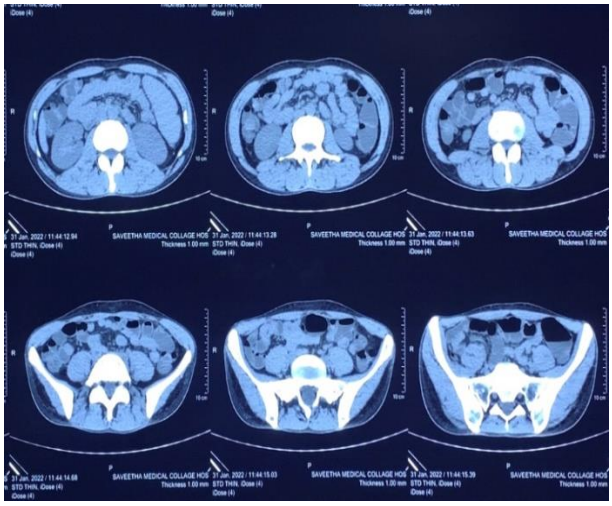

Figure 4: CT abdomen showing bulky pancreas and few parenchymal calcifications.

He was given bowel rest, hydration with IV Fluids was done. MRCP was done to get a better anatomical imaging of the pancreatico pleaural fistulous tract, to rule out the presence of strictures (Figure 6), showed- A linear T2/ flair hyperintensity of maximum thickness 12 mm extending from the body of pancreas superiorly into the mediastinum with extension posteriorly to visualized pericardium abutting the esophagus and another T2/ flair hyperintense focus of size 13×34 mm seen immediately on the right lateral aspect of aforementioned linear hyperintensity- fluid collection. Patient was taken up for endoscopic retrograde cholangiopancreatography,

selective pancreatic duct cannulation done. Pancreatogram done, revealed a passage of contrast outwards at mid- pancreatic duct and the cannula exchanged with bow sphincterotome. Sphincterotome was performed to divert the fluid leak. A 5 Fr×5 cm single pigtail pancreatic duct stent was placed in the pancreatic duct. X-ray erect abdomen was done to confirm the position of the stent (Figure 7). Post procedure chest X-ray was repeated (Figure 8), patient had complete resolution of pleural effusion, was symptomatically better, and was discharged and is being followed up periodically.

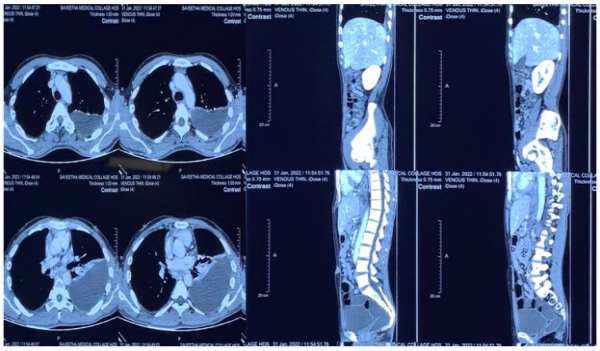

Figure 5: CECT chest and abdomen showing thin hypodense peripherally enhancing thin-walled collection tracking from the distal body of the main pancreatic duct to the posterior mediastinum, with communication to the left pleural cavity. Thin hypodense peripherally enhancing collection noted in the anterior mediastinum, pericardiac region abutting the right atrium.

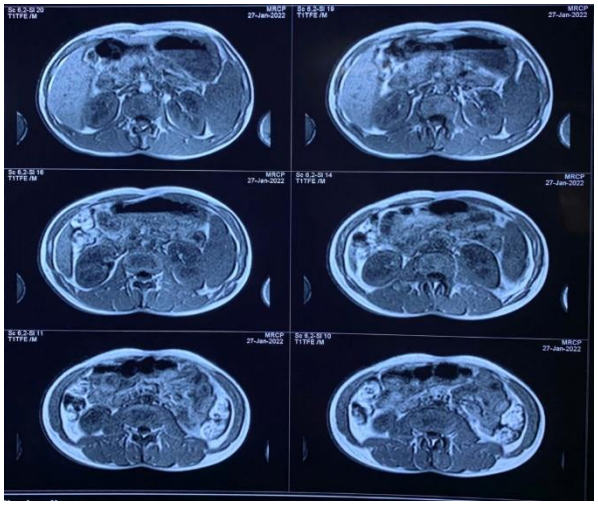

Figure 6: MRCP showing, a linear T2/flair hyperintensity of maximum thickness 12 mm extending from the body of pancreas superiorly into the mediastinum with extension posteriorly to visualized pericardium abutting the esophagus AND another T2/ flair hyperintense focus of size 13×34 mm seen immediately on the right lateral aspect of aforementioned linear hyperintensity- fluid collection.

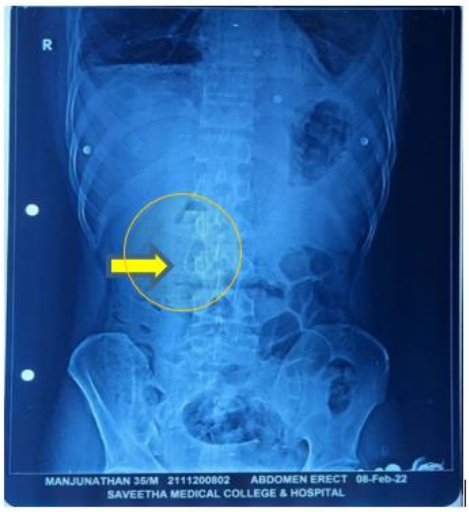

Figure 7: X-ray erect abdomen post ERCP, to confirm the position of the stent.

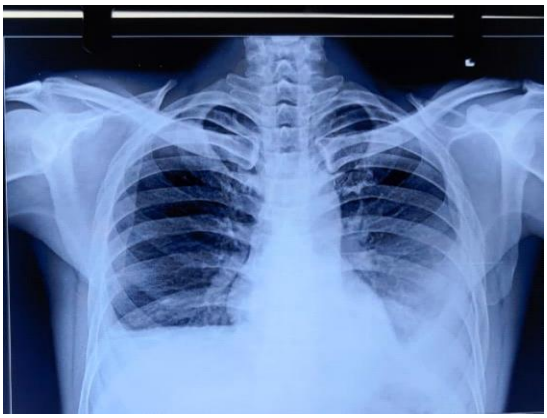

Figure 8: Chest X-ray after ERCP, with complete resolution of the left sided pleural effusion.

DISCUSSION

Pancreaticopleural fistula is a very rare complication of chronic pancreatitis, if so, seen in majority of the patients aged 40 to 50 years with chronic alcohol consumption.[3] It is an abnormal collection of pancreatic secretions contained within a non-epithelial wall, primarily composed of granular and fibrous tissue.[4] The various etiologies for its formation include alcohol related chronic pancreatitis (most common), pancreatitis due to gall stones, idiopathic pancreatitis, trauma or congenital pancreatic duct anomalies.[5] Pleural effusion as a presentation of pancreaticopleural fistula is very rare, with an incidence rate of 0.4 to 1% cases.[6] Pleural effusions are predominantly left sided, however right sided and bilateral effusions occur in 19% and 14% of the patients, respectively, clinical presentation is often misleading as the presenting symptoms are usually associated with pleural effusion in the form of cough, dyspnea and chest pain, rarely do patients present with abdominal pain as a chief complaint typical of pancreatitis.[7] The underlying pathogenesis is that, the

recurrent, chronic inflammation of the pancreas eventually leads to the disruption of the pancreatic duct, posteriorly, the pancreatic secretions tract via the retroperitoneal space and via the path of least resistance reaches the pleural cavity, presenting as recurrent pleural effusion, ascites will be seen in an anterior disruption of the pancreatic duct .Studies have also reported posterior disruption of the pancreatic duct dissecting into the mediastinum through aortic or esophageal hiatus.[8-10]

In our case, the fistulous tract was extending from the body of pancreas superiorly into the mediastinum with extension posteriorly to visualized pericardium abutting the esophagus, esophagogram was performed to rule out the passage of fistulous tract via the aorta or esophageal hiatus and its absence was confirmed. Pleural effusion due to PPF needs to be distinguished from the small reactive self-limiting left sided effusion that occurs in 3-17% patients with acute pancreatitis.[10] Whereas, pleural effusion in PPF is large and recurrent despite repeated thoracocentesis, and with a characteristic pleural fluid amylase level >50,000 IU/l.[11] Other causes of amylase rich pleural effusion include parapneumonic effusion, pulmonary tuberculosis, esophageal rupture and malignancies like leukemia, lymphoma.[12]

Our patient presented with massive left sided pleural effusion, which on thoracocentesis revealed bloody pleural fluid with a probable suspicion of malignant disease, trauma, or pulmonary embolization, the pleural fluid amylase and lipase were highly elevated with hundred thousand as range and was negative for tuberculous origin, negative for malignant cell cytology, which narrowed the probability to pancreatico pleural fistula. Next step was to confirm the diagnosis, the most sensitive imaging modality is MRCP with a sensitivity of 80% and helps in giving a detailed anatomy of the area, followed by ERCP with a sensitivity of 78% and a benefit of being both a diagnostic and therapeutic tool, with potential complications like pancreatitis, ductal perforation and bleeding.[13] In this case CT Contrast of the chest and abdomen was performed which confirmed the diagnosis with the presence of chronic pancreatitis changes with dilatation of the distal main pancreatic duct and fistulous tract extending cranially via the posterior mediastinum up to the bifurcation of the pulmonary artery with another tract in the anterior mediastinum, pericardiac region abutting the right atrium.

Treatment

The treatment of PPF involves a multidisciplinary approach, management options can be medical, endoscopic, or surgical depending on the patient and presentation. in our case the medical management comprised of bowel rest with intravenous hydration, thoracocentesis, which was done twice, and tube thoracostomy was planned if recurrent pleural effusion wasn't resolving, as there was no further development of effusion tube thoracostomy was deferred. Superinfection is a major complication and was efficiently avoided with appropriate antibiotics. ERCP and stent placement have revolutionized the concept of non-surgical management.[14] Indications for surgery are- failure of conservative treatment and ERCP. Endoscopic management with ERCP stenting to the pancreatic duct was thought of to be the best option to the patient. ERCP stenting has 2 goals-first, to mechanically block the abnormal connection of the pancreatic duct with pleura, second goal is to keep the pancreatic duct open so that the pancreatic secretions can pass downstream the duct which is of low resistance to the duodenum, thus avoiding the abnormal fistulous connection with the pleura which was of higher resistance.[15]

On performing ERCP ductal stenting there is a faster transition to feeding and reduction of recovery time, as both a diagnostic and therapeutic tool has reduced the hospital stay and mortality rate compared to surgical management.[16] Therefore, ERCP was performed, showed contrast leak at the mid pancreatic duct, a 5 Fr×5 cm plastic stent was placed in the main pancreatic duct crossing the fistulous tract, and a biliary and pancreatic sphincterotomy was performed to divert the fluid leak. Follow up chest X-ray showed complete resolution of the pleural effusion, patient improved clinically. Post procedure, patient had no complications, hence was discharged and is on routine follow up.

CONCLUSION

Pancreaticopleural fistula is a very rare complication of pancreatitis observed in patients with chronic alcohol consumption. The diagnosis of pancreaticopleural fistula requires high index of suspicion, and is identifiable with good history, examination, key laboratory findings of very high pleural fluid amylase, lipase levels and imaging modalities to demonstrate the fistulous tract. Treatment involves multidisciplinary approach. Choosing the mode of management as medical, endoscopic or surgical is case dependent. Endoscopic intervention in the form of ERCP with pancreatic stenting has emerged as first line therapy for PPF, with recent advances, and due to its less invasiveness, endoscopic interventions are preferred over surgical therapy.

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