

Pancreatico-Pleural Fistula: A Comprehensive Case Series

¹Amey P. Kirtikar, Intern Doctor, Grant Govt. Medical College and Sir JJ Group of Hospitals, Mumbai, Maharashtra, India.

²Sharad B. Ghatge, Associate Professor, Department of Radiology, Grant Govt. Medical College and Sir JJ Group of Hospitals, Mumbai, Maharashtra, India.

³Kirtika Tyagi, Intern Doctor, Grant Govt. Medical College and Sir JJ Group of Hospitals, Mumbai, Maharashtra, India.

⁴Pranay Nagaonkar, Final-year MBBS, Grant Govt. Medical College and Sir JJ Group of Hospitals, Mumbai, Maharashtra, India.

⁵Vedant Wadvalkar, Third-Year MBBS, KJ Somaiya Medical College and Hospital, Mumbai, Maharashtra, India.

Corresponding Author: Amey P. Kirtikar, Intern Doctor, Grant Govt. Medical College and Sir JJ Group of Hospitals, Mumbai, Maharashtra, India.

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Abstract

Pancreaticopleural fistula is one of the rare complications of pancreatitis. The presentation of this condition can range from predominant features of pancreatitis to those of pleural effusion. Due to its resemblance with various chest and abdomen pathologies, clinching the diagnosis is difficult. MRCP (Magnetic Resonance Cholangio pancreaticography) is the most sensitive radiological modality available which can delineate the fistulous tract and confirm the diagnosis. Other modalities, like Contrast-Enhanced CT scans and ultrasonography (USG), can be used to look for collections in the pancreas, and pleural effusion, and to rule out other differential diagnoses. ERCP (Endoscopic Retrograde Cholangio pancreaticography) with stenting is the best definitive management for these cases.

This case series depicts three cases of pancreaticopleural fistula with contrasting clinical features, seen in the backdrop of varying degrees of pancreatitis. Further, it elaborates on the various diagnostic and therapeutic modalities used, noting the outcomes of these patients.

Keywords: Pancreatitis, Cholangiopancreatography Magnetic Resonance, Pleural Effusion

Introduction

Pancreaticopleural fistula (PPF) is an abnormal communication between the pancreas and the pleural cavity. It is one of the rarer complications of pancreatitis, seen in less than 1% of the patients. It is more frequently seen in the background of chronic pancreatitis presenting with recurrent pleural effusion, but can also be seen in acute pancreatitis. Left-sided pleural effusions are twice more likely than right-sided ones ^{1,2}. It occurs as a result of a ruptured pancreatic duct or an extension of a

pancreatic pseudocyst³. It is hypothesized that rupture of the pancreatic duct posteriorly leads to communication with the pleural cavity through the esophageal hiatus in the diaphragm at the T8 level¹⁻³.

There is a great amount of diagnostic difficulty in such cases thus, the diagnosis is usually delayed. Taking into consideration the routine investigations, pleural fluid tap analysis is insightful. High levels of amylase, lipase, and albumin are seen in the pleural fluid with no presence of atypical or malignant cells. The gold standard investigation is a CT abdomen and thorax which shows the changes consistent with the primary pathology in the pancreas. After confirming pancreatitis on a CECT abdomen, MRCP (Magnetic Resonance Cholangio pancreaticography) delineates the presence of a fistulous tract. A diagnostic ERCP (Endoscopic Retrograde Cholangio pancreaticography) can be used for the same purpose⁴.

Symptomatic management of the effusion is done by repeated thoracocentesis and octreotide is given for reducing the pancreatic secretions. Surgical management for definitive treatment is accompanied by a 10% risk of mortality. ERCP procedure can be used for both diagnostic and therapeutic in such patients. A papillary sphincterotomy with pancreatic duct stenting is done for such cases. Any calculi encountered in the pancreatic duct can be removed in this procedure. It has a lower risk of mortality. Thus, ERCP is preferred over conventional surgery¹⁻⁴.

In this case series, we have described three patients with pancreaticopleural fistula who were treated with ERCP stenting and one patient with Video Assisted Thoracoscopic Surgery (VATS) and showed radical improvement in their condition.

Case Report 1

A 46-year-old female presented to a tertiary care hospital with the chief complaint of breathlessness for 10 days, which was acute in onset and gradually progressive. The patient had a history of hospital admission for breathlessness due to a bilateral pleural effusion, a week ago for which pleural fluid tapping was done (2500 mL from left side; 1500 mL from right side). This relieved her symptoms temporarily.

The chest X-ray revealed, bilateral pleural effusion [Figure 1]. A therapeutic thoracocentesis of the pleural fluid was performed in view of the effusion, which led to dramatic improvement in the patient's respiratory drive. The effusion was exudative in nature. Serum Amylase was elevated in the patient.

USG of the gall bladder (Target GB scan) showed distended GB with hyperechoic calculi (largest measuring 16mm). Minimal echogenic sludge was noted within. No changes consistent with cholecystitis were present.

The pancreas on local USG showed a hypoechoic collection measuring 1.3 x 1.0 cm in the proximal part of its body which communicated with the main pancreatic duct. Another hypoechoic collection of 2.3 x 1.2 x 1.5 cm was seen in the lesser sac adjacent to the caudate lobe of the liver.

CECT Abdomen (done 72 hours after admission) showed an ill-defined, hypodense attenuation area in the pancreatic and peripancreatic region measuring approximately 2.2 x 1.7cm encasing the common hepatic artery and abutting origin of the splenic artery and left gastric artery by about 180 degrees [Figure 2]. The splenic artery showed 20-30% luminal occlusion most likely due to thrombosis causing wedge-shaped splenic infarcts [Figure 3]. The spleen measured 11.7 cm. The right lobe of the liver showed hypertrophy measuring 24.4 cm. The gall bladder was distended with hyperdense

sludge. No features of cholecystitis were seen. All these findings raised suspicion for a neoplastic etiology in the pancreas.

However, the tumor markers for pancreatic cancer, CA 19.9 (11.02 IU/L) and CA 125 (15.2 IU/L) were within the normal range.

The patient's condition began deteriorating and thus was maintained on ventilatory support, intravenous fluids, intravenous high-grade antibiotics, and inotropic support in view of shock secondary to septicemia.

The MRCP of the patient was diagnostic of acute necrotizing pancreatitis with walled-off necrosis (T2W hyperintense collection with hypointense rim and debris) in the body of the pancreas. The collection was seen extending cranially forming another collection of 2.0 x 1.1 x 4.4 cm in the sub-diaphragmatic region adjacent to the caudate lobe [Figure 5]. A sinus tract of length 2.7 cm and width 2.8 mm was seen extending up to the right hemidiaphragm with no obviously appreciated connection with the pleural space [Figure 4, 5]. The collection was seen communicating with the posterior aspect of the main pancreatic duct. Moderate peripancreatic fat stranding was also seen.

Other findings on the MRCP, cholelithiasis with sludge, and splenic infarcts were consistent with the CECT done previously. Moderate left-sided and mild right-sided pleural effusion was also seen.

These MRCP findings with a refractory and recurrent bilateral pleural effusion in this patient highly raised the suspicion of a pancreaticopleural fistula. The patient was started on somatostatin analogue – Inj Octreotide 100mcg twice daily.

The pleural fluid tap on culture and sensitivity yielded *Acinetobacter baumannii* which was resistant to all major antibiotics.

Meanwhile, the patient was started on enteral feed via a nasojejunal Freka's tube.

There was a dramatic improvement in the patient's general condition upon starting Inj. Octreotide, with her ICD tube output reducing and finally reaching an impasse at 60-70ml/day for 3 consecutive days.

The patient was advised zero fat diet (via nasojejunal tube), chest physiotherapy, and injection octreotide 100mcg twice daily (to decrease exocrine secretions of the pancreas) and planned for an endoscopic ultrasound with sos ERCP.

Endoscopic ultrasound showed fluid collection in the form of a dilated chain of lake appearance of the pancreas.

A side-viewing endoscope was passed up to the second part of the duodenum. Selective cannulation of the pancreatic duct was carried out with the help of a guide wire. Pancreatic sphincterotomy was carried out and a 5 Fr x 5 cm stent was placed.

The patient improved dramatically over the next 5 days after ERCP stenting. The ICD tube was removed in view of decreased output and the patient was started on oral feed. The patient was discharged and asked to maintain follow-up at the outpatient department.

Case Report 2

A 31-year-old male was referred to a tertiary care hospital with pain in the epigastric and left upper quadrant of the abdomen radiating to the back since the last 20 days which aggravated with consumption of food and decreased on bending forwards. The patient experienced breathlessness since the last 10 days. The patient also had a mild fever which resolved intermittently on taking medications. The patient had a past history of similar episodes. He was a known alcoholic for 9 years.

On a chest radiograph, right-sided hydropneumothorax was seen. An ICD was inserted for the same. A USG of the abdomen and pelvis revealed features suggestive of acute on chronic pancreatitis with chronic right-sided pleural effusion which extended into the subdiaphragmatic space. A mild left-sided pleural effusion with septations was also seen. On CECT abdomen and pelvis, acute on chronic pancreatitis was seen with a 13mm defect was seen in the left hemidiaphragm which was suggestive of a pancreaticopleural fistula. Moderate right-sided hydropneumothorax and mild left-sided pleural effusion were also seen [Figure 6].

MRCP confirmed the findings of the CECT A + P pancreatic protocol, adding to its findings by concluding that the tract was in fact in communication with the posterior aspect of the main pancreatic duct at the region of the head of the pancreas.

The patient was managed conservatively with nasojejunal enteral feeding, intravenous antibiotics and analgesics. EUS followed by ERCP stenting was done as a definitive treatment. A side-viewing endoscope was passed up to the second part of the duodenum. Selective cannulation of the pancreatic duct was carried out with the help of a guide wire. Pancreatic sphincterotomy was carried out and a 5 Fr x 5 cm stent was placed.

The symptoms of the patient resolved considerably over the next week. The nasojejunal tube was removed and the patient was started on a low-fat diet. The ICD was removed once the output reduced to less than 60 ml for 3 consecutive days. Following ICD removal the patient was discharged. The patient is doing well during his 3 monthly follow-ups with no active complaints but a prolonged period of follow up is necessary.

Case Report 3

A 49 year-old-male presented with complaints of right upper abdomen pain which referred to the back since the last 9 days and breathlessness since the last 3 days. The patient is a chronic alcoholic and had a history of similar complaints of pain in the abdomen in the past for which he was managed conservatively. On per abdomen examination, a tender lump of 3 x 2 cm was palpated in the epigastric region. An ultrasound was performed which revealed atrophic pancreas with a pseudocyst and moderate right-sided pleural effusion. Following it up with a CECT abdomen, it was seen that a well-defined hypodense walled-off (wall thickness: 4.5 mm) collection of 2 x 3.1 x 3 cm was seen in the pancreatic body. Superiorly the collection extended along the lesser curvature and was suspected to communicate with the bilateral pleural cavities [Figure 7]. The lesion was seen compressing the 2nd part of the duodenum and infra-hepatic inferior vena cava [Figure 7]. Multiple small intercommunicating collections (3-4 cc) with intra-parenchymal calcifications were seen in the atrophic pancreas. Mild peripancreatic fat stranding was also seen. To confirm the presence of a fistulous tract a MRCP was performed. The presence of a walled-off collection communicating with the main pancreatic duct was confirmed. Two tracts were seen arising from the collection – one of which communicated with the right pleural cavity and the other was blind-ended [Figure 8]. A pseudocyst was seen in the pancreatic head which communicated with the main pancreatic duct. The rest of the pancreas showed calcific foci and a dilated main pancreatic duct suggestive of – chronic pancreatitis. The patient underwent ERCP stenting as definitive treatment and condition of patient improved after that.

Case Report 4

A 12 year-old-male patient came with the chief complaints of difficulty in breathing since the last 6 days,

abdominal pain since the last 5 days and chest pain since the last 5 days. The patient was diagnosed and treated as a case of left-side pleural effusion. A USG-guided therapeutic pleural tapping was done for the multiloculated effusion. On routine microscopy of the tapped pleural fluid, 15% neutrophils and 85% lymphocytes were found. A CT Pulmonary angiography was performed, which revealed no abnormalities. To know the cause of pain in the abdomen and persistent effusion, a CECT of the abdomen and pelvis was performed. It showed – a few ill-defined hypodense collections in the body of the pancreas and in the peripancreatic region. A fistulous tract 5cm long and 5.1mm wide was seen arising from the pancreatic collections and transversing postero-superiorly along the left crus of the diaphragm through the aortic hiatus communicating with the left pleural space. It also showed collapse consolidation of the left lung with mediastinal shift. This is followed by an MRCP to delineate the tract further. It revealed – a T2W hyperintense fistulous tract arising from the hyperintense 1 cc peri-pancreatic collection, communicating with the left pleural space through the aortic hiatus as seen in the CECT. Mild splenomegaly with splenic vein thrombosis was also noted. The patient was posted for decortication and adhesiolysis through Video Assisted Thoracoscopic Surgery (VATS) after which an ICD was inserted. The patient was intubated and managed in the critical care unit (CCU). The ICD was removed nine days after the surgery and then the patient was discharged.

Discussion

Pancreaticopleural fistula is seen in 0.4% of patients with pancreatitis and 4.5% in those with pancreatic pseudocysts⁵. Cases with no known history of pancreatitis have also been reported in the literature ². The clinical presentation of it ranges from symptoms of

pancreatitis predominantly to that of pleural effusion. This contrasting feature was seen in the above-mentioned cases, where the first patient showed more symptoms of pleural effusion whereas the second, third, and last patients had complaints related to both pancreatitis and pleural effusion. Thus, in the first patient primary lung pathologies were ruled out first.

According to Norman Oneil Machado ¹, 77% of the patients developed a left-sided pleural effusion. In a case series by Alhad R Dhebri, and Nicholas Ferran ⁴, they have reported a case with right-sided pleural effusion. In the second case reported by us, a right-sided hydropneumothorax with mild left-sided effusion was seen. A similar presentation was seen in the case reported by Ahmad Ramahi, Kanana Mohammad Aburayyan, Tamer S Said Ahmed, Vyas Rohit, and Mohammad ⁶. But, in the third case, the patient only had a right-sided hydropneumothorax. 69 to 77% of patients presenting with PPF have pseudocysts. In the first cases in our case series, PPF was seen in a background of acute necrotizing pancreatitis in contrast to the third case where the PPF was superimposed over chronic pancreatitis.

A close differential diagnosis to pancreaticopleural fistula is acute pancreatitis with non-communicating pleural effusion. The two can be differentiated based on serum amylase levels which do not rise significantly in pancreaticopleural fistula without acute pancreatitis. MRCP, ERCP, and CECT have sensitivities of 80%, 78%, and 47% respectively, for diagnosing PPF ^{7,8}. CECT Abdomen and pelvis do not always delineate the fistulous tract. But, in the second case on a CECT mentioned by us a 13mm long fistulous tract was visualized [Figure 6].

Medical therapy with thoracocentesis is the first-line management in PPF. The success rate of medical therapy ranges between 31% to 65% in contrast to a success rate

of 94% by surgical interventions as mentioned in a review of literature by King JC, Reber HA, Shiraga S, and Hines OJ⁹.

ERCP is the best available definitive management as it is both diagnostic and therapeutic. A papillary sphincterotomy is first performed which also relieves the obstruction in patients with potential sphincter of Oddi dysfunction. Any stones seen in the main pancreatic duct can be addressed during the same^{1,10}. Conventional surgical intervention is considered in patients not responding even after an ERCP. But, it carries a 10% risk of mortality¹. Another drawback of ERCP is the risk of developing iatrogenic pancreatitis which is anywhere between 2-10% in non-selected patients and 40% in high-risk patients¹¹.

However, the last patient in this case series did not undergo an ERCP for PPF. He was managed by decortication and adhesiolysis under VATS. A similar line of management was carried out for a case reported by Wies Vanderbruggen, Vicky Dhooghe, Bart Bracke, Vera Hartman, Dirk Ysebaert, Paul Van Schil et al¹².

Conclusion

Pancreaticopleural fistula remains a rare yet significant complication of pancreatitis, with varied clinical presentations ranging from isolated pleural effusion to combined pancreatic and pulmonary symptoms. Early recognition supported by imaging modalities such as CECT, MRCP and ERCP is crucial for diagnosis. While conservative management may suffice in selected cases, endoscopic or surgical interventions are often required for definitive treatment. Our series highlights the importance of various imaging modalities and individualized management based on the patient's presentation and response to therapy.

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Legend Figures

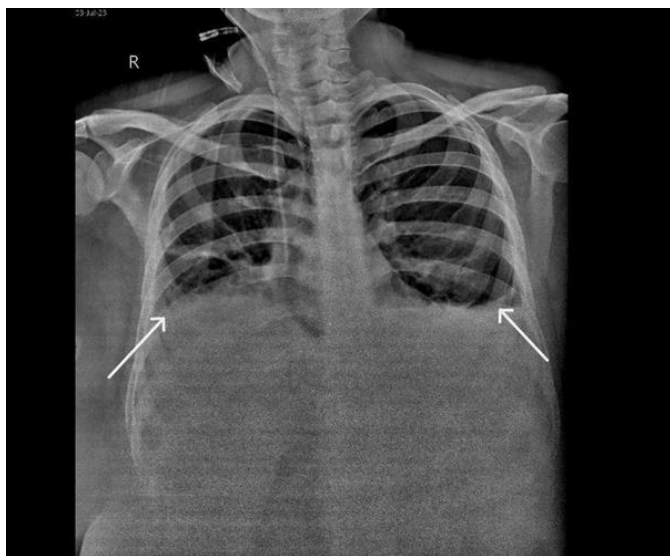


Figure 1: A 46-year-old female presented with the complaint of breathlessness subsequently diagnosed as pancreaticopleural effusion – Plain Chest radiograph in posteroanterior view (i) Bilateral

white arrows showing blunting of bilateral costophrenic angles due to bilateral pleural effusion

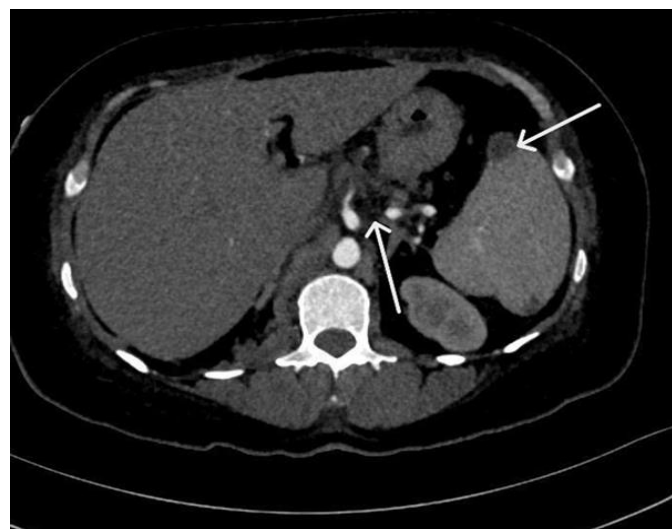


Figure 2: A 46-year-old female presented with the complaint of breathlessness subsequently diagnosed as pancreaticopleural effusion – Contrast Enhanced CT scan arterial phase axial section (i) left white arrow showing hypodense pancreatic collection encasing left gastric artery (ii) right white arrow showing splenic infarct



Figure 3: A 46-year-old female presented with the complaint of breathlessness subsequently diagnosed as pancreaticopleural effusion – Contrast Enhanced CT scan arterial phase axial section (i) white arrow showing splenic artery thrombosis

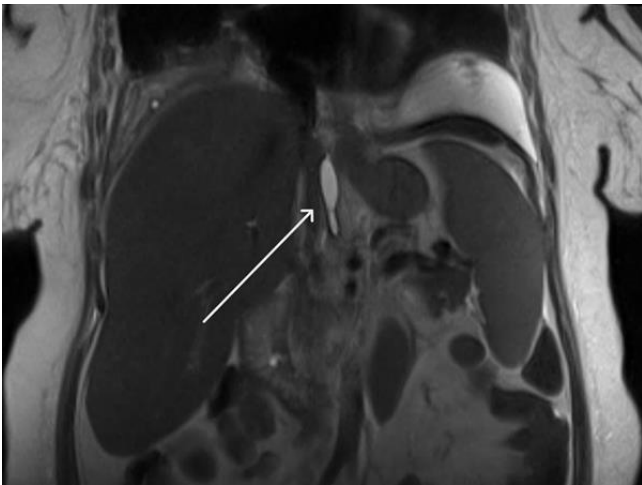


Figure 4: A 46-year-old female presented with the complaint of breathlessness subsequently diagnosed as pancreaticopleural effusion – T2W MR coronal view (i) white arrow showing hyperintense collection extending cranially in the subdiaphragmatic space

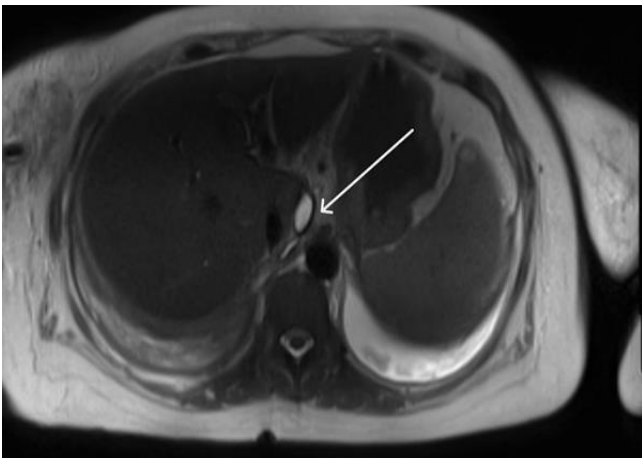


Figure 5: A 46-year-old female presented with the complaint of breathlessness subsequently diagnosed as pancreaticopleural effusion – T2W MR axial view (i) white arrow showing collection extending posterosuperiorly adjacent to the caudate lobe



Figure 6: A 31-year-old male with the complaints of pain in abdomen and breathlessness later diagnosed as chronic pancreatitis with a pancreaticopleural fistula – Contrast Enhanced CT scan venous phase coronal section (i) white arrow showing hypodense defect crossing left hemidiaphragm



Figure 7: A 49 year-old-male presented with complaints of right upper abdomen pain further diagnosed with chronic pancreatitis with pseudocyst and pancreaticopleural fistula - Contrast Enhanced CT scan arterial phase coronal section (i) Hypodense lesion seen compressing 2nd part of duodenum (white arrow)

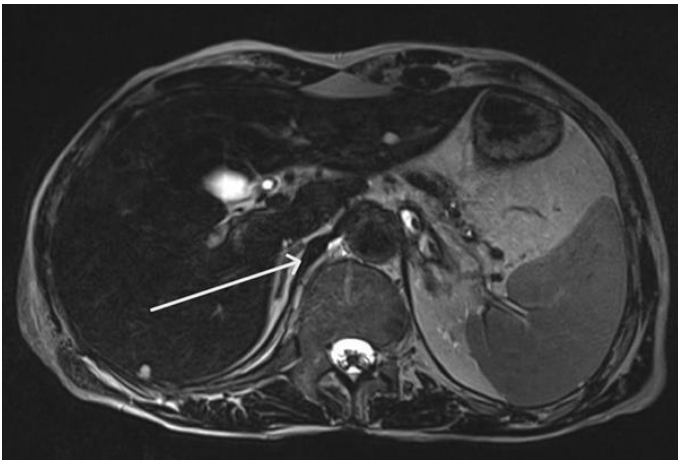


Figure 8: A 49 year-old-male presented with complaints of right upper abdomen pain further diagnosed with chronic pancreatitis with pseudocyst and pancreaticopleural fistula – MRCP (i) white arrow showing pancreaticopleural fistula communicating with right pleural space

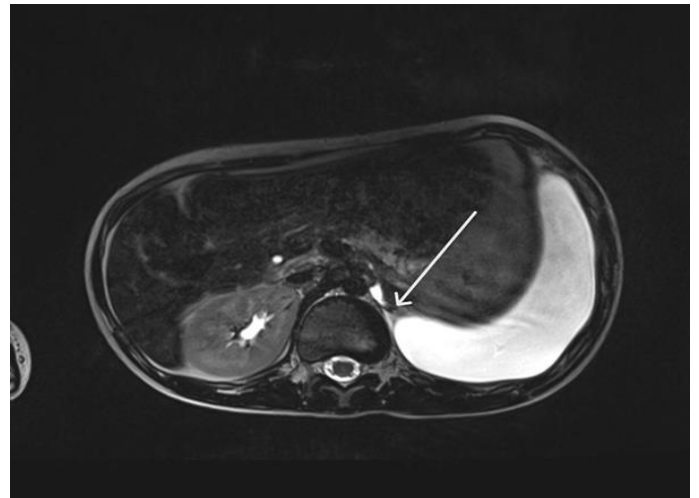


Figure 10: A 12 year-old-male patient came with the chief complaints of difficulty in breathing, pain in abdomen and chest pain later diagnosed as pancreaticopleural fistula - T2W MRCP (i) white arrow showing hyperintense pancreaticopleural fistula seen communicating with the left pleural space



Figure 9: A 12 year-old-male patient came with the chief complaints of difficulty in breathing, pain in abdomen and chest pain later diagnosed as pancreaticopleural fistula - Contrast Enhanced CT scan arterial phase axial section (i) white arrow pointing to Hypodense PPF seen communicating with the left pleural space