

Case Report**PANCREATICOPLEURAL FISTULA: REVISITING A RARE COMPLICATION****Niket Mayank Shah**

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Abstract

Pancreatico-pleural fistula [PPF] is a rare complication of acute and chronic pancreatitis. PPF occurs secondary to either pancreatic duct leakage or an incomplete or ruptured pseudocyst. The pleural effusion in pancreatic diseases can be either reactionary or pathological due to the fistula. The triad of previous pancreatitis, disturbance of the pancreatic duct with pleural effusion on imaging and pleural exudate showing high amylase levels has been thought to be diagnostic of PPF. The sensitivity of CT for fistula visualization is 47–63%. Magnetic resonance cholangiopancreatography (MRCP) allows for better visualization and endoscopic retrograde cholangiopancreatography (ERCP) allows therapeutic intervention with sensitivity of 80% and 78% respectively. Management strategy includes initial conservative treatment with or without octreotide infusion and if conservative measures fail, then surgical options [Fistula division with pancreatic ductal drainage/resection] can provide cure.

Keywords: PANCREATICOPLEURAL FISTULA; PLEURAL EFFUSION; CHRONIC PANCREATITIS

INTRODUCTION

Pancreatic fistula is a rare complication of acute pancreatitis, chronic pancreatitis and pancreatic trauma. It develops because of pancreatic ductal disruption or rupture of a pseudocyst. Pancreatic ascites develops due to free secretion of pancreatic juice into peritoneal cavity. Its challenging to diagnose this rare complication and even more challenging to treat it adequately. Left pleura is the most common location for pancreatic fistula followed by right pleura and both pleural cavities [6]. Such exudative pleural effusions can cause chest pain and dyspnea, like any emergent thoracic pathology e.g., aortic dissection; as a result, this diagnosis is challenging to identify. Pancreaticopleural fistula occurs in 0.4% of patients with acute pancreatitis, in 4.5% of patients with pancreatic pseudocyst and in 20-30% cases of untreated chronic pancreatitis. The pleural effusion associated with pancreaticopleural fistula is typically refractory to drainage and tends to accumulate rapidly. Thoracentesis with measurement of amylase in pleural fluid is the most important diagnostic procedure followed by MRCP or CT for the anatomy of fistulous tract. The management is based on three

lines of treatment: the first is the conservative medical management which includes suspension of the oral intake, initiation of total parenteral nutrition and administration of somatostatin analogues; the second line is based on endoscopic management by placing a stent in the pancreatic duct, and the third line is the surgical option, consisting of surgical resection (distal pancreatectomy) and enteric shunts (pancreatojejunostomy or cystojejunostomy). Here we present 2 cases of pancreato-pleural fistula.

Case 1

A 37 years young male presented with history of cough with expectoration for 5 months and dyspnea on mild exertion for 3 months which gradually worsened over last 4 weeks. Patient was diagnosed to have chronic calcific pancreatitis for 5 years and he was on oral pancreatic enzyme supplementations and oral analgesics for pain relief. Patient was not a smoker and not an alcoholic. Patient was hemodynamically stable and was admitted under thoracic unit. His vital parameters were stable and was maintaining room air saturation 95%. Blood evaluation was unremarkable. Serum amylase and serum lipase were well within normal limits. His chest x-ray [Figure 1] was suggestive of left sided pleural effusion. IV contrast enhanced CT chest and abdomen [Figure 2 and 3] showed atrophic pancreas with intra-pancreatic calcification, dilated main pancreatic duct [maximum diameter 6 mm] throughout pancreas and left sided pleural effusion. There was a fistulous tract of size 11mm diameter and 15mm length between pancreatic body and left pleural cavity. Left intercostal drainage tube was inserted and the fluid analysis showed high amylase value [25000 U/L]. Referral was sought of department of surgical gastroenterology for management of pancreatico-pleural fistula. After thorough counseling about risks and benefits of intervention with family members of the patient, patient was planned for laparotomy and division of communication between pancreas and pleural cavity. Patient underwent elective laparotomy. Intraoperatively fistulous tract was seen arising from body of pancreas [Figure 4]. Hence division of fistulous communication was done and Roux-en-Y fistulo-jejunostomy with externalization of anastomotic stent was performed [Figure 5]. Post operative period was uneventful. On post operative day 3, ICD drain fluid amylase was found to be undetected. Patient recovered well. ICD was removed on post operative day 4 and was discharged on day 7 with an anastomotic external stent

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Received: June 20, 2022**Published:** July 01, 2022

Citation: Niket Mayank Shah. PANCREATICOPLEURAL FISTULA: REVISITING A RARE COMPLICATION. Int Clin Img and Med Rew. 2022; 2(3): 1079.

which was removed after 6 weeks. Patient is doing well on 5 years of follow up.

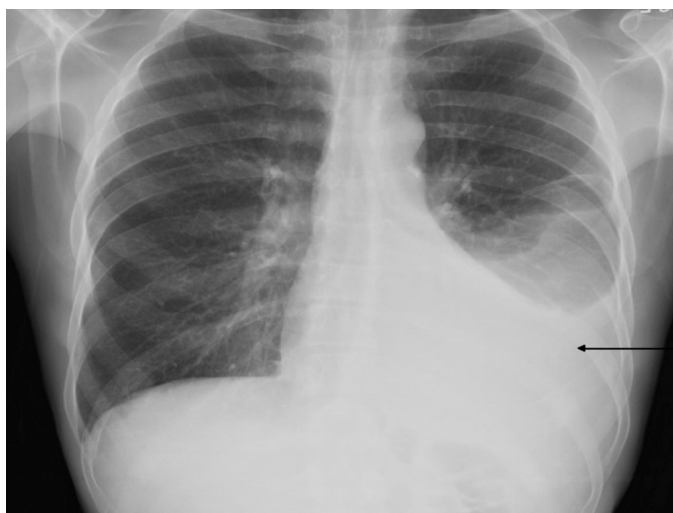


Figure 1: Chest X-ray PA view suggestive of left sided pleural effusion



Figure 2: CT Chest showing right sided pleural effusion



Figure 3: CT abdomen showing calcification in head of pancreas

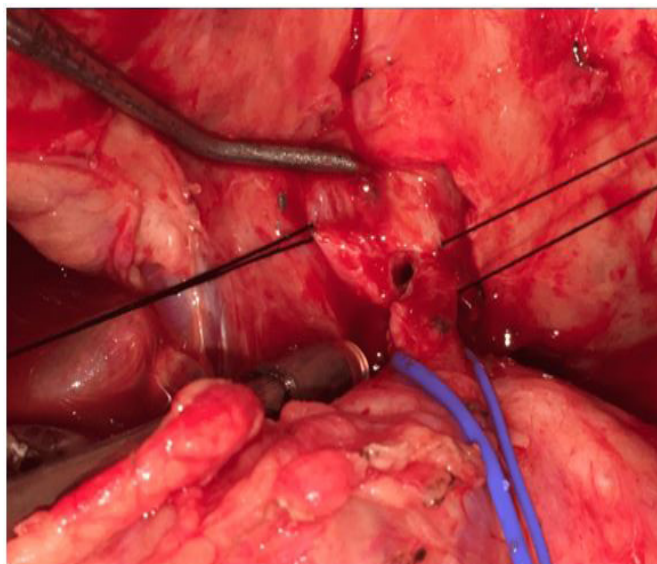


Figure 4: Intra-operative picture showing fistulous tract between pancreas and right thoracic cavity

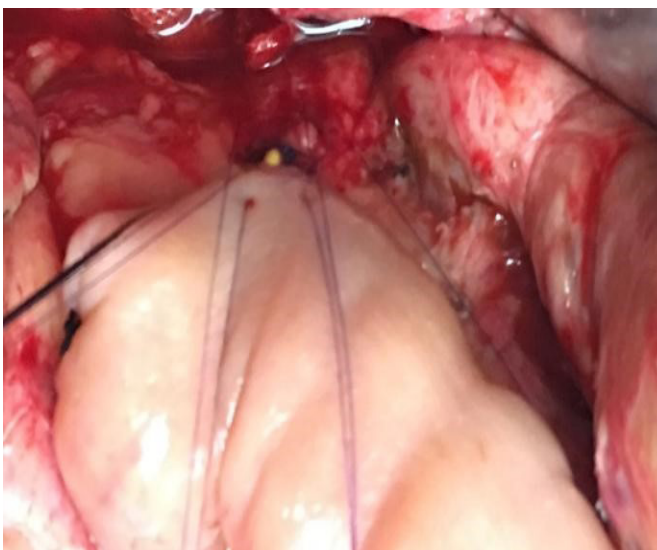


Figure 5: Intra-operative picture showing fistula-jejunostomy

Case 2

A 35 years young male patient presented with right sided chest pain for 1 month and dyspnea for 1 week. Patient had history of recurrent upper abdominal pain for past 2 years. He was diagnosed to have chronic calcific pancreatitis and was managed conservatively by oral analgesics and pancreatic enzyme supplementation. Patient was a chronic smoker for 10 years and he used to consume alcohol daily for 7 years. Patient was admitted at local hospital where chest X-ray [Figure 6] was suggestive of right sided pleural effusion and hence right sided intercostal drainage tube was placed and he was referred to our center. Patient had stable vital parameters and normal room air saturation on admission. On blood evaluation, patient had severe hypoalbuminemia [1.6 gm/dL] with raised serum amylase [370 U/L] and raised serum lipase [158 U/L]. Rest of the laboratory tests were normal. IV contrast enhanced CT chest and abdomen [Figure 7, 8, 9] showed Calcifications in head of pancreas, dilated MPD [Diameter 9 mm], peri-pancreatic region collection without MPD communication and right sided intra-thoracic fistulous extension [16mm length and 5mm diameter] from pancreatic body causing right pleural effusion with collapse of underlying lung parenchyma suggestive of chronic calcific pancreatitis with pancreatico-pleural fistula. Drain fluid amylase was 7522 U/L. After

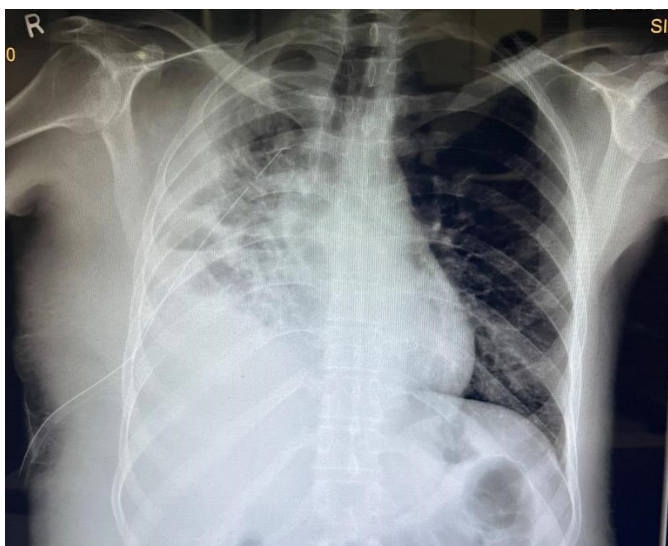


Figure 6 : Chest X-ray PA view showing left sided pleural effusion with ICD in situ

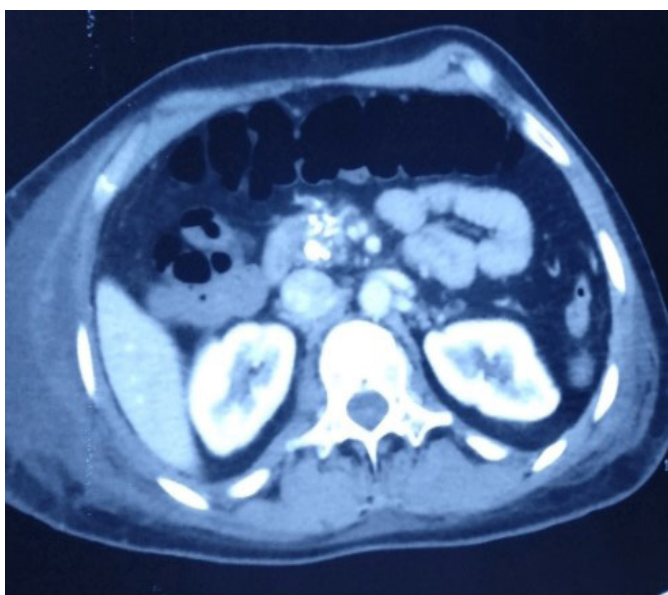


Figure 7: CT abdomen showing calcifications in head of pancreas

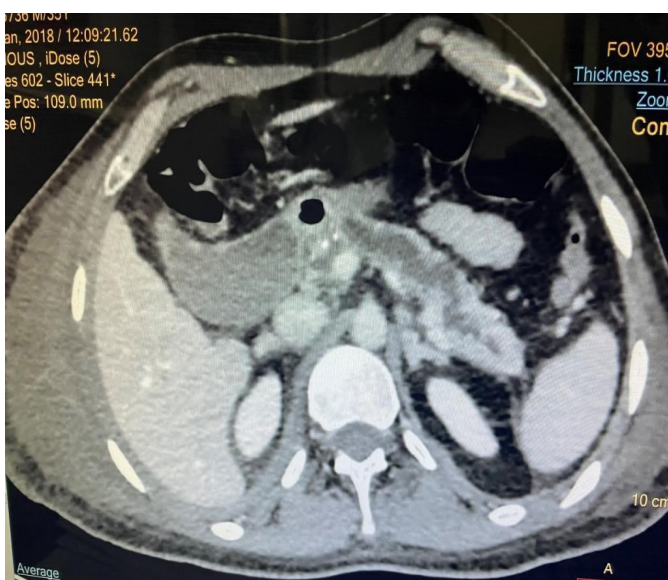


Figure 8: CT Abdomen showing dilated main pancreatic duct

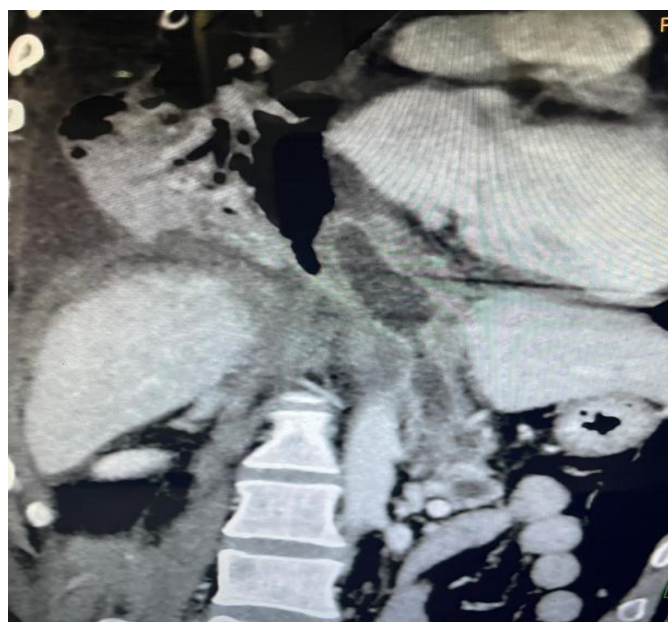


Figure 9: CT Abdomen showing fistulous tract between pancreas and left pleural cavity

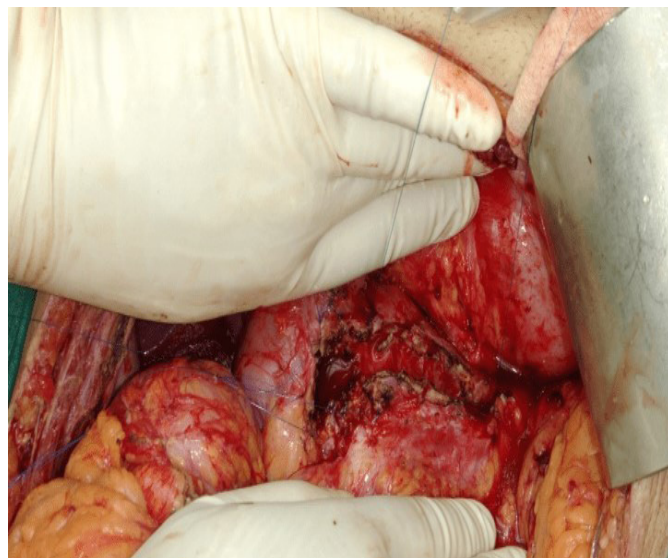


Figure 10: Intra-operative picture of dilated main pancreatic duct being prepared for pancreatico-jejunostomy

detailed counseling, patient underwent frey's procedure [Figure 10], drainage of peri-pancreatic fluid collection and division of fistulous tract. Post operative period was uneventful and patient recovered well. ICD tube was removed on day 5 once drain fluid analysis showed undetectable amylase level and patient was discharged on post operative day 8. Patient is doing well on 4 years of follow up.

DISCUSSION

The main pathology underlying these fistulae is a persistent chronic inflammatory process of the pancreas, most often attributed to alcoholic pancreatitis. PPF occurs secondary to either pancreatic duct leakage or an incomplete or ruptured pseudocyst, with pancreatic enzymes eroding the fascial planes posteriorly. These enzymes erode into the mediastinum via the esophageal or aortic hiatus, resulting into fistula or pseudocyst communicating with the pleural cavity upon rupture. An anterior pancreatic disruption is more likely to lead to ascites but can also cause a PPF. A hydrothorax more commonly develops on the left but right or bilateral hydrothoraces have been documented. The pleural effusion in pancreatic diseases can be either reactionary

or pathological due to the fistula. Reactionary effusions are usually small, left-sided (or may be bilateral), with amylase levels < 1000 U/L and low protein content < 3 g/dl, mostly seen in acute pancreatitis and resolve spontaneously. Effusions due to fistulae are usually large, commonly left-sided, with amylase levels >1000 u/l and high protein content > 3 g/dl, mainly seen in pseudocysts and chronic pancreatitis and require intervention for control. A significantly raised pleural fluid amylase is the most important and confirmatory diagnostic test. Acute pancreatitis, pneumonia, malignancy, oesophageal perforation and pulmonary tuberculosis are also associated with raised pleural fluid amylase levels. There is no documented threshold amylase levels in the literature but a level above 50 000 IU/L, is highly suggestive of PPF. Both our patients presented with chest symptoms with high pleural fluid amylase levels i.e. 25,000 U/L and 7522 U/L respectively against a background of chronic pancreatitis suggesting the diagnosis of PPF. The most common symptom of PPF is dyspnoea (65–76%) followed by abdominal pain, cough, chest pain, and fever [17]. These symptoms can be confused for other acute thoracic pathology, making a diagnosis and treatment challenging. Pre-disposing factors include male sex, fifth decade of life, and chronic pancreatitis secondary to alcohol, trauma, and choledocholithiasis.

The triad of previous pancreatitis, disturbance of the pancreatic duct with pleural effusion on imaging, and pleural exudate showing high amylase levels has been thought to be diagnostic of a pancreatopleural fistula.

Amylase rich pleural fluid has been documented to be black in color, prompting the consideration of diagnosis of PPF.

Diagnostic work up includes a chest radiograph followed by a CT scan to evaluate for pancreatitis and associated complications; however, the sensitivity of CT visualization is 47–63%. Magnetic resonance cholangiopancreatography (MRCP) and endoscopic retrograde cholangiopancreatography (ERCP) allow for advanced visualization and therapeutic intervention respectively with sensitivity of 80% and 78% respectively. MRI with MRCP has been found to be superior to ERCP because it provides an image of the whole pancreas as well as a possible pseudocyst. It is also able to delineate the duct of Wirsung distal to the site of obstruction.

There are no specific guidelines or approach for PF management because the data is limited. Conservative management with hemodynamic monitoring has resulted in resolution in 30–60% cases. Conservative treatment includes complete bowel rest, total parenteral nutrition, and broad-spectrum antibiotics. Octreotide has been used to decrease pancreatic fistula output and reduce the closing time. Both therapeutic and diagnostic, thoracentesis or tube thoracostomy can measure amylase and drain symptomatic pleural effusions.

Medical management is usually with octreotide alone used at an initial dosage of 50 mcg TDS increasing to 250 mcg TDS. A long course of treatment for about 2 to 3 weeks is usually required and there is a low success rate of between 30 and 65% as well as needing a longer hospital stay. It is mostly useful in patients without any ductal disruption or obstruction. In the setting of failed medical management or stricture, an ERCP with stent placement or sphincterotomy is warranted.

ERCP can identify the presence of a ductal stricture and the site of ductal disruption. Pancreatic stent placement provides a low resis-

tance path for the flow of pancreatic secretions and allows mechanical closure of the site of the fistula. The duration of therapy and success rate is highly variable. A strategy of repeated ERCP every 6–8 weeks to assess fistula closure has been used by some authors. Conservative management can be successful in patients with pancreatic ductal single stricture with just pancreatic stent placement, whereas patients of multiple strictures, complete duct disruption, large cysts or impacted ductal calculi are managed with surgery.

Due to inflammatory reaction around pancreas, retroperitoneal space, large vessels and surrounding organs, surgical interventions can result in severe complications including death.

In the early stage of diagnosis, it is advisable to minimize the initial surgery and total excision of the fistula may not be initially possible. The first intervention may involve drainage followed by staged surgeries when the acute inflammation resolves.

Goals of management are to restore normal anatomy and decreasing pancreatic secretions leakage, which often requires definitive surgical treatment.

Successful medical conservative management can provide resolution of fistula in 30–60% with a 15% risk of recurrence. Recommended duration of continuing conservative management is 2–3 weeks. Endoscopic therapy with stent placement in the pancreatic duct is used to restore ductal anatomic continuity; it is used in cases of disruption of the pancreatic duct in the body and distal stenosis to the disruption site.

CONCLUSION

A pancreatopleural fistula is a rare complication and a challenging diagnosis in patients with chronic pancreatitis and requires thorough evaluation in patients presenting with recurrent respiratory symptoms in the background of pancreatitis. Significantly raised pleural fluid amylase levels with MRCP to delineate the pancreatic ductal anatomy are useful in making the diagnosis and deciding the further management. Conservative medical management by somatostatin analogues and endoscopic stenting have a low success rate and surgical intervention often offers the best chance of cure.

Reference

1. Kaye MD. Pleuropulmonary complications of pancreatitis. *Thorax*. 1968 May;23(3):297–306. doi: 10.1136/thx.23.3.297. PMID: 4872925; PMCID: PMC471792.
2. Norman Oneil Machado, “Pancreatopleural Fistula: Revisited”, *Diagnostic and Therapeutic Endoscopy*, vol.2012, Article ID 815476, 5 pages, 2012, <https://doi.org/10.1155/2012/815476>
3. Kiewiet J, J, S, Moret M, Blok W, L, Gerhards M, F, de Wit L, T: Two Patients with Chronic Pancreatitis Complicated by a Pancreatopleural Fistula. *Case Rep Gastroenterol* 2009;3:36–42. doi: 10.1159/000210442
4. Adel Salah Bediwy, Pancreatopleural fistula: A rare cause of massive right-sided pleural effusion, *Egyptian Journal of Chest Diseases and Tuberculosis*, Volume 64, Issue 1, 2015, Pages 149–151, ISSN 0422–7638, <https://doi.org/10.1016/j.ejcdt.2014.08.004>.
5. Chawla, Gopal & Chauhan, Nishant & Dutt, Naveen & Yadav, Taruna & Jain, Priyank. (2019). Pancreatic pleural effusion masquerading as right sided tubercular pleural effusion. *Monaldi Ar-*

- chives for Chest Disease. 89. 10.4081/monaldi.2019.1125.
6. King JC, Reber HA, Shiraga S, Hines OJ. Pancreatic-pleural fistula is best managed by early operative intervention. *Surgery*. 2010 Jan;147(1):154-9. doi: 10.1016/j.surg.2009.03.024. Epub 2009 Jun 9. PMID: 19744435.
 7. Aswani Y, Hira P. Pancreaticopleural fistula: a review. *JOP*. 2015 Jan 31;16(1):90-4. doi: 10.6092/1590-8577/2915. PMID: 25640793.
 8. Ali T, Srinivasan N, Le V, Chimpiri AR, Tierney WM. Pancreaticopleural fistula. *Pancreas*. 2009 Jan;38(1):e26-31. doi: 10.1097/MPA.0b013e3181870ad5. PMID: 19106743.
 9. Francisco E, Mendes M, Vale S, Ferreira J. Pancreaticopleural fistula: an unusual complication of pancreatitis. *BMJ Case Rep*. 2015 Feb 12;2015:bcr2014208814. doi: 10.1136/bcr-2014-208814. PMID: 25678619; PMCID: PMC4330436.
 10. Hirose T, Shimizu T, Isegawa T, et al; Left pleural effusion caused by pancreaticopleural fistula with a pancreatic pseudocyst; *BMJ Case Reports* 2016;2016:bcr2016217175.
 11. Aswani Y, Hira P. Pancreaticopleural fistula: a review. *JOP*. 2015 Jan 31;16(1):90-4. doi: 10.6092/1590-8577/2915. PMID: 25640793.
 12. Tay CM, Chang SK. Diagnosis and management of pancreaticopleural fistula. *Singapore Med J*. 2013 Apr;54(4):190-4. doi: 10.11622/smedj.2013071. PMID: 23624444.
 13. Olakowski M, Mieczkowska-Palacz H, Olakowska E, Lampe P. Surgical management of pancreaticopleural fistulas. *Acta Chir Belg*. 2009 Nov-Dec;109(6):735-40. doi: 10.1080/00015458.2009.11680526. PMID: 20184058.
 14. Chebli JM, Gaburri PD, de Souza AF, Ornellas AT, Martins Junior EV, Chebli LA, Felga GE, Pinto JR. Internal pancreatic fistulas: proposal of a management algorithm based on a case series analysis. *J Clin Gastroenterol*. 2004 Oct;38(9):795-800. doi: 10.1097/01.mcj.0000139051.74801.43. PMID: 15365408.
 15. Aswani Y, Hira P. Pancreaticopleural fistula: a review. *JOP*. 2015 Jan 31;16(1):90-4. doi: 10.6092/1590-8577/2915. PMID: 25640793.
 16. Hirose T, Shimizu T, Isegawa T, et al; Left pleural effusion caused by pancreaticopleural fistula with a pancreatic pseudocyst; *BMJ Case Reports* 2016;2016:bcr2016217175.
 17. Ali T, Srinivasan N, Le V, Chimpiri AR, Tierney WM. Pancreaticopleural fistula. *Pancreas*. 2009 Jan;38(1):e26-31. doi: 10.1097/MPA.0b013e3181870ad5. PMID: 19106743.
 18. Yuki Ong & Vishal G Shelat (2021) Ranson score to stratify severity in Acute Pancreatitis remains valid – Old is gold, *Expert Review of Gastroenterology & Hepatology*, 15:8, 865-877, DOI: 10.1080/17474124.2021.1924058
 19. Ramahi A, Aburayyan KM, Said Ahmed TS, Rohit V, Taleb M. Pancreaticopleural Fistula: A Rare Presentation and a Rare Complication. *Cureus*. 2019 Jun 24;11(6):e4984. doi: 10.7759/cureus.4984. PMID: 31501720; PMCID: PMC6721877.
 20. Singh S, Yakubov M, Arya M. The unusual case of dyspnea: a pancreaticopleural fistula. *Clin Case Rep*. 2018 Apr 10;6(6):1020-1022. doi: 10.1002/ccr3.1434. PMID: 29881555; PMCID: PMC5986032.
 21. Ali T, Srinivasan N, Le V, Chimpiri AR, Tierney WM. Pancreaticopleural fistula. *Pancreas*. 2009 Jan;38(1):e26-31. doi: 10.1097/MPA.0b013e3181870ad5. PMID: 19106743.
 22. Akahane T, Kuriyama S, Matsumoto M, Kikuchi E, Kikukawa M, Yoshiji H, Masui K, Fukui H. Pancreatic pleural effusion with a pancreaticopleural fistula diagnosed by magnetic resonance cholangiopancreatography and cured by somatostatin analogue treatment. *Abdom Imaging*. 2003 Jan-Feb;28(1):92-5. doi: 10.1007/s00261-001-0144-2. PMID: 12483394.