



## Pancreaticopleural Fistula –A Rare Complication of Pancreatitis. A Case Report and Review of Literature

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### Abstract

Pancreaticopleural fistula is a rare complication of pancreatitis mostly chronic pancreatitis. It may occur following trauma, surgery of pancreas or adjacent organ. It usually affects middle aged man with chronic alcohol use and attempts to recur on continued alcohol use. Clinical feature to diagnose the entity is not very classical or specific. Initial management include conservative with intercostal drain (ICD) and antisecretory octreotide therapy to more invasive ERCP stenting of main pancreatic duct (MPD/PD). In severe cases surgery too may be considered.

Here we present a case of pancreaticopleural fistula with recurrence following chronic pancreatitis and ongoing ethanol use. He responded to conservative management on both the occasion.

As it is extremely rare entity and also not very common to response to octreotide only so effectively, we would like to share this in a case report section.

**Keywords:** Pancreaticopleural Fistula; Pancreatitis; ICD

### Introduction

Pancreaticopleural fistula has been recognized as an extremely rare clinical entity. It arises as a complication mostly of chronic pancreatitis and also following acute attack due to ductal leak and formation of internal fistulous tract between chest and abdomen. First case reports were published in late 1960 [1]. Pancreatic ascites and pancreaticopleural fistula both of them are recognized as an internal pancreatic fistula and they share underlying common pathogenesis. Pathogenesis includes disruption of main pancreatic duct, resulting in leakage of pancreatic fluid. It usually occurs in adult male and with history of chronic alcoholism. Other causes of pancreaticopleural fistula reported are traumatic rupture or surgical disruption of PD [2]. The thoracic collection may in the form of pleural effusion, pancreatic or mediastinal pseudocyst. It is characterized by massive pleural effusion and there is tendency to recur following conservative treatment particularly with ongoing alcohol intake. Long back it was treated just as nil per mouth and drainage. Since the discovery and use of octreotide as an antisecretory agent has been documented, octreotide has been used in treating this condition, and although the results varies there are reports show-

ing its effectiveness in decreasing pancreatic ductal output and reducing time for closure time of the fistula tract. Nowadays It can be initially treated conservatively with thoracocentesis and octreotide but and ERCP sphincterotomy ± stenting has proven its efficacy in closure of the fistula tract effectively. Surgery can be recommended in only severe refractory cases. Conservative management with antisecretory (octreotide) and stenting may achieve closure in 31-45% while surgery will lead to healing in approximately 80-90% of cases, but mortality is high and may reach up to 10% [3,4].

### Case Report

A 46 years' male, nondiabetic, non hypertensive and smoker with history of chronic alcohol intake, presented to us with dyspnoea on climbing stairs for 10 to 15 days along with chest heaviness and pain in the left chest for 6 days. He did not have any history of cough, fever, haemoptysis, pain abdomen, sweating, anorexia or weight loss. He had history of recurrent pancreatitis of about 4 attacks in the past 2 years, for which he was managed conservatively at his locality. Recent attack of pancreatitis happened a month back for which he was managed at his locality with 4- 5 days of hospitalization.

On evaluation he was pale, tachypnoeic, but had otherwise stable haemodynamics, BMI was 18 kg/m<sup>2</sup>. On auscultations there was dull note on percussion on the left hemithorax. It was substantiated by chest x ray- which showed massive left pleural effusion (Figure 1). Aspiration of pleural fluid about a liter was done on the same day. Pleural fluid was hemorrhagic with high amylase-7805u/l (<150u/l normal), high lipase-2396u/l, LDH- 1075u/l, with cell count 220/cmm, neutrophil 80%, ADA- 39u/l, glucose 70. Cytology ruled out any malignancy and suggestive of acute inflammation, culture did not grow any pathological organism. Laboratory parameters revealed: Hb-10gm%, TLC -8110/cmm, ESR-60mm/hr, Serum amylase, lipase were 188 and 108 u/l respectively. USG abdomen revealed bulky pancreas with heterogeneous echo texture and dilated PD, with no significant peripancreatic collection and left moderate pleural effusion. An ICD was placed on the 2<sup>nd</sup> day due to recurrence of symptoms. CT upper abdomen on the 4<sup>th</sup> day of admission revealed features of acute on chronic pancreatitis (resolving) with irregularly dilated PD, mild collection in peripancreatic area. Internal pancreatic fistula tract seen connecting MPD at body through esophageal hiatus to left lower chest and a small tract towards right chest (Figure 2). We started octreotide 100µg thrice daily subcutaneously along with ICD drain. ICD was removed on 10<sup>th</sup> day as pleural drain gradually decreased to less than 50 ml for 72 hrs. He improved symptomatically too. We gave him the option for ERCP sphincterotomy and PD stenting, but he denied, hence we discharged him on 12<sup>th</sup> day without ICD but he was advised to continue octreotide. He was counselled that he would require octreotide for long duration may be 2-6 months which will add cost issues, compliance and if he do not respond ERCP sphincterotomy and stenting of PD. would be needed. He was asked to abstain from alcohol. Unfortunately, he discontinued octreotide after 1 month and lost follow up with us but at the same time he continued alcohol daily. He came back to us with recurrent pleural effusion but at right side after 2 months (CT abdomen done previously showed a small right sided fistulous tract too). He was managed conservatively in the same previous way (with ICD and Octreotide). An USG abdomen was repeated which suggests features of acute on chronic pancreatitis and evidence of fistula tract from MPD to right lower chest. He was advised ERCP stenting, but again he refused and opted for conservative management as before with octreotide and ICD. He was dyspnoic and an ICD was put in his right lower chest. Gradually his drain output reduced over 7 days and after some clinical improvement he insisted us for discharge on octreotide. Hence he was discharged on request after ICD removal with advice to continue octreotide for at least 3 months otherwise to undergo ERCP at the earliest.



**Figure 1:** Chest x ray- massive left pleural effusion.



**Figure 2:** CT abdomen showing fistulous tract.

He came on follow up this time after 3 weeks. otherwise remained well, and gained weight. Haemoglobin improved to 12gm%. He discontinued alcohol for some time. Octreotide was stopped after 3 months and counselled him that it may recur and in that case he would only require an ERCP stenting.

On follow up for 1 year he was well, but again started small quantity alcohol drinking. He was counselled by our psychologist and he had good family support. Everything failed to refrain him from his drinking habit and after 2 years he developed cirrhosis with ascites which was revealed on USG when he complained about his limb swelling and abdominal swelling for a month. Bilirubin was normal except altered albumin globulin ratio. Platelet

and. Ptime was normal. AFP was 3.2. viral serology was negative. He was non diabetic. Child Pugh score was 7/15. He was advised diuretics. UGI scopy showed large varix 3 columns, he was advised EVL which he did not agree. Hence we put him on NSBB. Later he lost to follow up. On FU he never had any recurrence of pancreatitis or pleural effusion (Figure 3).



**Figure 3:** Repeat chest x ray - on 11th day.

After about 18 months he died following a major upper GI bleeding and sepsis.

## Discussion

Pleural effusion due to pleuropancreatic fistula is a rare entity, accounting for <1% of cases [5]. It is seen in 3-7% patients with pancreatitis [6]. It is less common than pancreatic ascites. More common on left side (76%) [2], and usually recurrent, but may occur on right also 19% of cases, or bilateral in 14%. Combined incidence of pancreaticopleural fistula and pancreatic ascites is seen in 4%-7% cases of chronic pancreatitis, and 6-14% patients with pseudocyst.

It develops as a consequence of ductal leak, or incompletely formed or ruptured pseudocyst [2]. The fistulous tract passes either through aortic or esophageal hiatus or directly transdiaphragmatically. In pancreatic ascites pancreatic duct disruption occurs anteriorly, and here it disrupts retroperitoneally.

Mostly seen in middle aged man between 40-50 years with chronic alcoholism and pancreatitis [3]. 50% patients do not have h/o pancreatitis. Trauma accounts for < 0.5% cases [7] Pancreatic pseudocyst may be noted in 69-77% cases. Dyspnoea is usual presenting symptoms. Clinical manifestations often misleading with cough, fever, dyspnoea, chest pain, rarely they c/o abdominal pain typical of pancreatitis. Diagnosis often delayed due to absence of specific symptoms, The average duration of symptoms 5-6 weeks before diagnosis. Pleural effusion may be associated with ascites (20% cases) and pericarditis (4%) [8]. Major complication is superinfection which increases morbidity. Other causes of pleural effusion with high amylase has to be excluded like- esophageal rupture, malignancy of lung, rectum, breast, female reproductive system, leukemia, lymphoma, trauma, surgery, pneumonia, hydronephrosis, tuberculosis, liver cirrhosis [9].

High index of suspicion is needed to establish the diagnosis. It should be distinguished from small self-limiting reactive pleural effusion in pancreatitis. It has high amylase (normal <150u/l), and high albumin content (>3g/dl) [2] which is not seen in reactive pleural effusion.

Chest x-ray is a simple first line investigation [4]. Pleural fluid analysis will indicate high amylase, lipase, and absence of TB, malignancy. CT chest and abdomen are the gold standard for diagnosis and it can demonstrate the fistulous tract as well as the condition of the pancreas [2,3]. MRCP also helpful to diagnose pancreatitis and fistulous tract demonstration and is a noninvasive alternative to ERCP [5,6].

The diagnosis may be confirmed with CT scan, MRCP or an ERCP, but as ERCP is an invasive test and carries risks and complications so noninvasive imaging is nowadays used for demonstration. ERCP is reserved for therapeutic drainage. Sometimes ERCP fails to detect the leakage point [10], particularly where site of ductal disruption is distal. ERCP leads to diagnosis in 80% of cases and demonstrates fistulous tract in 59-74% [4,11].

In the past there was a high failure rate with conservative management by keeping nil per mouth and chest drainage and the patient would invariably require surgery. Success rate of conservative management has increased following use of octreotide, an antisecretory agent, and stenting [2,11]. Also it can be offered as an initial management protocol and ERCP and surgery can be reserved for refractory, severe and recurrent cases. The available treatment modalities currently followed are

Medical management- with octreotide and chest drainage;

Aim of medical treatment is to reduce secretion and reduce stimulation of pancreatic secretion [11,12]. In the past failure rates with medical management were high due to no ICD and short term (2-3 weeks) use for octreotide and patients often failed to respond and required surgical drainage [13]. Surgical success was higher but mortality was also high of 10%. Nowadays due to long term use of octreotide 2.5-6 months and chest drainage for 6-24 days, conservative results have improved [12]. Usual dose of octreotide varied in different studies (starting from 50 µg s/c TID initially and titrated according to fistula output, to maximum dose 250 µg s/c TID) [11]. Octreotide reduces fistulous output and decreases time to fistula closure.

ERCP- has revolutionized the concept of nonsurgical management with papillary sphincterotomy, dilatation of stenosis of PD. In cases of stenting failure only papillotomy may also help in such scenarios. Surgical management carries increased mortality although success rates are higher. It is usually reserved for complicated cases with recurrence and ERCP failure.

In our case we had a middle aged man of chronic alcoholism with recurrent attacks of pancreatitis with underlying chronic pancreatitis. He had pancreaticopleural fistula which responded to initial conservative medical management with antisecretory octreotide and ICD. He continued alcohol and the fistula recurred. He

had two fistulous tract one at left and one small at at right chest. Although he was advised for an ERCP but he did not agree, hence we had to restart octreotide for the second time for at least 3 months. He meanwhile stopped drinking for some duration. Later on follow up for 2 years he did not have any recurrence of the fistula. although he developed alcoholic liver disease which subsequently decompensated with ascites, bled and died of the complications of cirrhosis.

### Conclusion

Pancreatopleural fistula is a rare complication of pancreatitis. It can be managed conservatively or by ERCP sphincterotomy and PD stenting. In recurrent cases surgery may be an option. Few literatures support the use of octreotide but some other did not show any significant results. In our patient he had responded to octreotide for 3 months in 2 such episodes, initially combined with ICD. This case report revisits the rare complication of pancreatitis and its management.

Octreotide was effective in our case with no further recurrence without any adverse effects. Although long term study is required to establish its beneficial effects.

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