

# Recurrent hemoptysis: An unusual cause and novel management

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## Abstract

We report a rare case a 15 year old boy who presented with recurrent hemoptysis. There was past history of pancreatitis. A CT scan of thorax revealed a small collection in the region of the tail of the pancreas and a tract from it extending across the diaphragm into the posterobasal segment of left lower lobe, suggesting a pancreatico-pleuro-pulmonary fistula. The fistula was embolised by percutaneous injection of glue into the collection and fistula, which resulted in good symptom control.

**Key words:** Glue embolization; hemoptysis; pancreatico-pulmonary fistula; pancreatitis

## Introduction

Complications of pancreatitis have been well-described with fistulous communications with adjacent organs. Here, we would like to present a case of chronic pancreatitis, which presented with recurrent massive hemoptysis. On imaging, a fistulous tract was demonstrated between the pancreas and posterobasal segment of the left lower lobe suggesting a pancreatico-pulmonary fistula, which was the cause of recurrent hemoptysis.

Standard of care in these cases has been total parenteral nutrition (TPN) with Endoscopic retrograde cholangiopancreatography (ERCP)-guided stent placement, nasopancreatic drain, chest tube placement, and in some cases surgery. We describe a minimally invasive procedure wherein we embolized the fistulous tract with glue, which was effective and avoided the disadvantage of long-term stent placement, drains, and TPN.

## Case History

A 15-year-old boy with known chronic pancreatitis presented with hematemesis since 1 year, for which he was evaluated and diagnosed to have gastric ulcer, varices, and was put on medications for the same. He later presented with recurrent hemoptysis for 2 months with approximately 100 ml of fresh blood every day. There was no tuberculosis or contact with tuberculosis in the past. On examination, his vitals were stable and no abnormality was detected on systemic examination.

Initially, possibility of hematemesis mistaken for hemoptysis was considered. However, bronchoscopy revealed blood in the left main bronchus and bronchoalveolar lavage revealed hemosiderophages. Contrast-enhanced computed tomography (CT) showed reticular and ground glass opacities and patchy areas of consolidation in the left lower

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10.4103/0971-3026.184412

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**Cite this article as:** Surya P, Keshava SN, Irodi A, Vyas S, Thangakunam B. Recurrent hemoptysis: An unusual cause and novel management. Indian J Radiol Imaging 2016;26:267-70.

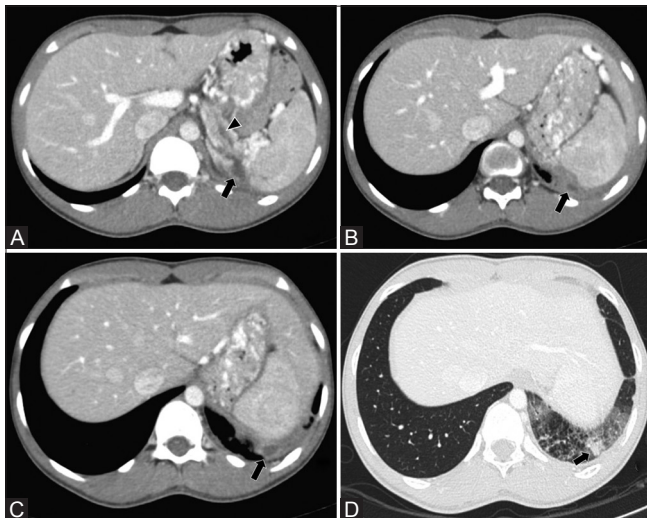
lobe basal segments [Figure 1D]. There was no abnormal vascularity or vascular malformations in the lung.

Atrophic pancreas with mildly dilated main pancreatic duct, in keeping with the known chronic pancreatitis was seen. The mid and distal part of the splenic vein was thrombosed with extensive gastric varices, perisplenic, mesenteric, and omental collaterals. A small collection was seen in the tail of the pancreas measuring 14 × 19 mm, which had communication with the pancreatic duct [Figure 1A]. There was a hypodense peripherally enhancing irregular tract measuring approximately 6 mm in diameter extending superiorly from the collection, across the posterior aspect of the diaphragm into the posterior basal segment of left lower lobe, suggesting a pancreatico-pulmonary fistula [Figure 1B and C]. Minimal left-sided pleural effusion was noted.

T2 SPAIR-T2 Spectral adiabatic inversion recovery sections [Figure 2] through the ROI also showed the collection in relation to the tail of the pancreas and confirmed the presence of the tract, as seen on CT.

Imaging was not able to find any predisposing structural abnormality for his chronic pancreatitis. His biochemical parameters were also not indicating any particular predisposing cause. ERCP was not done.

As the patient had incessant hemoptysis, bronchial artery embolization was attempted, and hypertrophied right intercostobronchial and left bronchial arteries were selectively cannulated and embolized with gelfoam



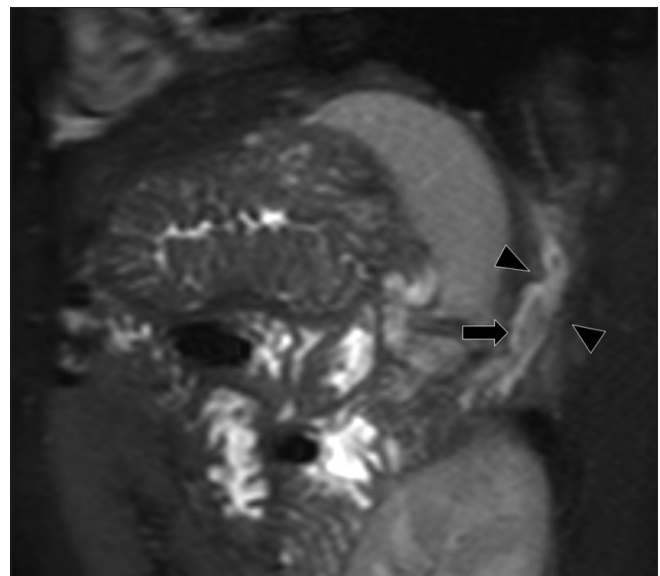
**Figure 1 (A-D):** Axial contrast enhanced computed tomography image showing dilated pancreatic duct (arrowhead in A) and a small collection in relation to the tail of the pancreas (arrow in A). A hypodense tract (arrows in B and C) is seen extending from the collection across the diaphragm into the posterobasal segment of the left lower lobe (arrow in D). Surrounding ground glass opacities are also seen adjacent to the fistula in the left lower lobe

particles. However, there was no significant symptomatic improvement.

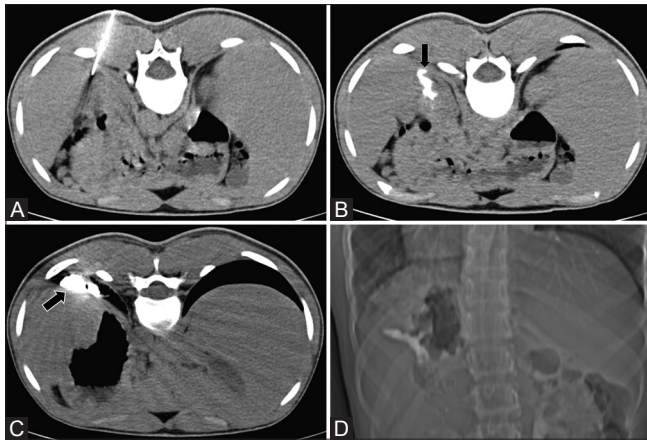
Hemoptysis was considered likely to be caused by pancreatic secretions causing damage to the lung parenchyma. Therefore, total parenteral nutrition (TPN) with Octreotide for prolonged period, pancreatic duct stenting, and surgery were all considered as treatment options. A novel idea of embolizing the fistulous tract with glue was also thought of. All the options were discussed with the patient who opted for embolization of the tract. The procedure was undertaken using CT guidance.

CT guidance was chosen over fluoroscopy to enable accurate needle positioning into the fistulous tract. When this procedure was performed, we did not have a combined facility to perform both in the same room. We believe that a table with both CT and fluoroscopic facility is appropriate for this kind of procedure, which would provide real-time monitoring.

In prone position, under local anesthesia, a 22G 10 cm needle was introduced percutaneously into the tract just above the pancreatic tail [Figure 3A]. 1 ml of 10% dilute nonionic water soluble contrast was injected, which opacified the tract from the pancreatic tail to the left lower lobe across the diaphragm. Approximately 1 ml of 50% hyostacryl glue was injected after clearing the syringe with 5% dextrose. There was good obliteration of the tract on check scan [Figure 3B-D]. An attempt was made to further inject 0.5 ml of 100% glue, but this was not possible, indicating solidification of the glue in the tract. No extension of the



**Figure 2:** T2 SPAIR sagittal section reveals fistula from the abdomen across the diaphragm (arrow heads) into the posterobasal segment of the left lower lobe



**Figure 3 (A-D):** Plain CT axial sections in prone position shows needle position in situ within the fistula (A) prior to glue embolization. Post glue embolization reveals an intact glue cast in the fistula at the distal pancreatic end with no reflux of glue into the proximal pancreas (B) and glue cast opacifying the pulmonary end of the fistula (C). Tomogram (prone position) reveals the glue cast in left hypochondrium (D)

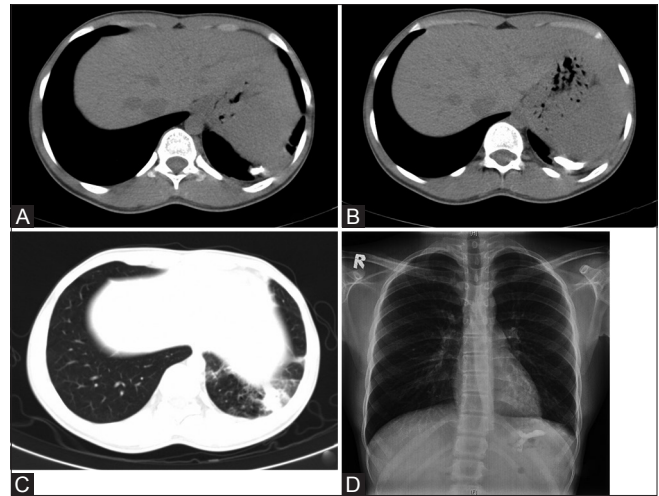
glue cast into the pancreatic duct or into the lung was noted. Particular attention was given to avoid the reflux of glue into the main pancreatic duct by monitoring limited CT slices after injecting glue percutaneously. When dilute contrast was injected and checked on CT, the flow was toward the lung. The total amount of glue injected was also small. There were no procedure related complications.

On follow-up after 1 month, patient had an improved sense of well-being. His hemoglobin had improved from 8 to 12 gm%. His hemoptysis had significantly reduced in frequency and quantity (50–100 ml almost daily prior to the procedure to 4–5 ml of streaky hemoptysis once in few days). A repeat limited CT thorax revealed intact glue cast sealing the fistula [Figure 4]. Residual ground glass opacities and consolidations in the posterobasal segment persisted with mild reduction compared to previous scan.

We now have a 1 year follow-up of the patient after the procedure and the patient is doing fine with no morbidity. He was able to resume his academic activities shortly after the procedure and has been on regular follow-up with no significant further episodes of hemoptysis.

## Discussion

Common complications of pancreatitis are localized to the abdomen such as pseudocyst, abscess, necrosis, and vascular complications. Extraabdominal complications include pleural effusions, mediastinal pseudocyst, and thoracopancreatic fistulas.<sup>[1]</sup> Thoracopancreatic fistula can be to the pleural space, to the bronchial tree, and to the mediastinum or pericardial sac.<sup>[2]</sup> Pancreatico-pleural fistula are common among them and occur in ~0.4% of acute pancreatitis and ~2.3–4.5% of patients presenting with



**Figure 4 (A-D):** Computed tomography axial sections done during 1 month follow-up reveals an intact glue cast in the fistula (A, B). Patchy ground glass densities persist, but have reduced compared to before obtaining the scan. Chest radiograph done during the same period shows an intact glue cast similar to the scan seen in D

pancreatic pseudocyst. Postulated pathogenesis is recurrent inflammation causing a posterior ductal disruption, leading to fluid and secretions extending posteriorly in the retroperitoneum through the path of least resistance into the pleural cavity. More commonly, this occurs through one of the diaphragmatic hiatus and can occasionally occur directly through the diaphragm.<sup>[3]</sup>

In our case, the fistula extended from a small pseudocyst which also had communication with a mildly dilated pancreatic duct. The fistula was seen to terminate in the posterobasal segment of the left lower lobe. Basal segments of the left lower lobe had diffuse ground glass densities believed to be due to inflammation and alveolar damage resulting from pancreatic secretions. There have been few previous reports about pancreatico-bronchial fistulas and only one previous case report of pancreatico-pulmonary fistula.<sup>[4]</sup> Patients with pancreatico-pulmonary fistula can present with hemoptysis, as in our case, due to secretions causing damage and sloughing of alveoli. Only a thin layer of effusion was seen in our case, unlike massive recurrent effusions in pancreaticopleural fistulas.

Diagnosis depends on the demonstration of fistulous tract or detecting amylase in the pleural fluid. CT scans demonstrate the tract in less than 50% of cases of pancreatic-pleural fistula<sup>[5]</sup> compared to 79% in ERCP. Magnetic resonance cholangiopancreatography (MRCP) has also been shown to be equally sensitive in the demonstration of the fistula. MRCP done in our case failed to detect the fistula, which was better seen on contrast enhanced CT thorax and T2W and SPAIR axial MRI images.

There are no definite guidelines for the management of thoracopancreatic fistulas, and most management strategies

come from previous published studies. Medical strategies such TPN with octreotide and chest tube placement have helped in some cases and failed in some cases.<sup>[5]</sup> ERCP-guided stent placement along with TPN has been suggested by some to be efficacious in closing down the fistula.<sup>[5]</sup> Duration of drainage can last between 4–12 weeks.<sup>[6]</sup> Some advocated placement of a nasopancreatic drain, which when suctioned frequently reduced the intraductal pressure and aided in healing.<sup>[5]</sup> Surgery is indicated only when conservative ERCP measures fail.<sup>[6]</sup> Fibrin injection of glue in closing of thoracopancreatic fistula has been reported in previous literature as a successful procedure with no further intervention required.<sup>[7]</sup> We embolized the collection and the fistula by percutaneous glue injection under CT guidance.

The technique of percutaneous glue injection proved to be a simple and an efficacious, minimally invasive technique with no significant associated morbidity as compared to conventional techniques such as long-term TPN and nasogastric tube insertion, requiring a protracted treatment course with associated morbidity.

One limitation of our technique was lack of real-time monitoring by fluoroscopy, which we believe would be a better option for the same.

## Conclusion

Pancreaticopleuro-pulmonary fistula is a rare type of thoracopancreatic fistula, and demonstration of the fistula on CT, MR, MRCP, or ERCP is confirmatory. While pancreaticopleural fistulas present with recurrent

effusions, patients with pancreaticopleuro-pulmonary fistula can present with recurrent hemoptysis. Percutaneous glue injection into the fistula is an effective, minimally invasive technique that can be used to treat these fistulas.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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