Case Report
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Pancreatico-pleural Fistula in a 9-Year-Old Child

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Abstract

Pancreatico-pleural fistula (PPF) is a rare clinical entity which is even rarer in children. The report describes a 9-year-old girl developing a PPF as a complication of Acute on Chronic Pancreatitis

Keywords: Pancreatico Pleural fistula in children

1. Introduction

Pleural Effusion (PE) is a common problem across all age groups, Tuberculosis being the commonest cause in India. We present a case of 9-year-old girl child developing PE due to PPF. PPF is very rare in children. Most of the cases in adults are seen among alcoholic males.

2. Case Report

A 9-year-old girl presented in the casualty of our hospital in a very moribund condition withfever and breathlessness since3 days prior to admission. Patient's relatives gave history of pain in upper abdomen radiating to the back,anorexia, weight loss and episodes of vomiting since 20 days.

Patient had similar episodes of radiating abdominal pain with vomiting one year back which was treated conservatively. Patient belonged to lower socioeconomic strata from tribal belt. There was no significant family history.

Patient was in severe respiratory distress, her respiratory rate was more than 60 per minute, pulse was around 120 per minute and low volume. On auscultation, air entry was significantly reduced on Left hemithorax and she had tenderness and guarding in upper abdomen. She had to be intubated and put on ventilator.

Blood Investigations showed leucocytosis and had low haemoglobin of 8 gm. Pancreatic enzymes were done in view of history of pain in abdomen radiating to the back and guarding in upper abdomen. They were instead of came significantly raised, Serum Amylase was (2021IU) and Lipase was (1230IU). X-Ray chest showed massive left pleural effusion (PE) for which an intercostal drain was inserted which drained around 1.5 L of serous fluid (Figure 1). Routine pleural fluid analysis was exudative in nature.

After stabilizing the patient, an Ultrasound of abdomen was done which showed prominent Common Bile Duct (CBD) of 10mm diameter and a 9mm stone in it. Gall Bladder (GB) was normal. Pancreas was oedematous with

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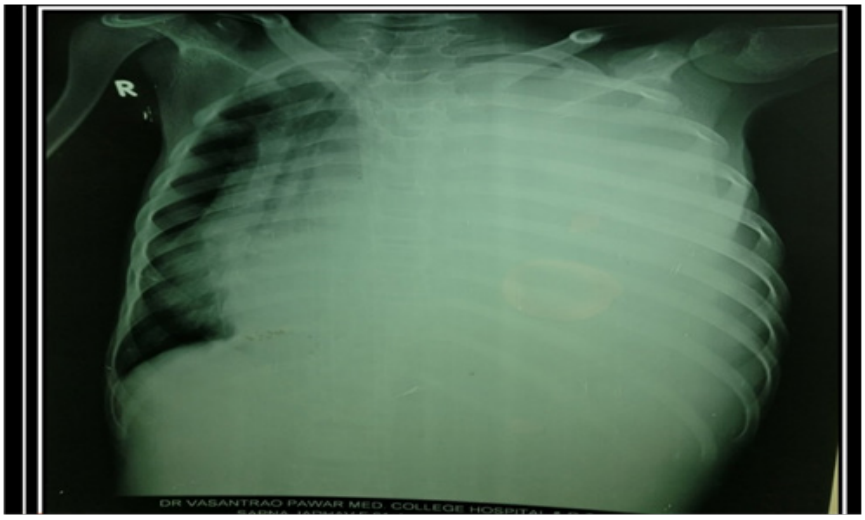

Figure 1. Pre procedure Xray-Chest PA view.

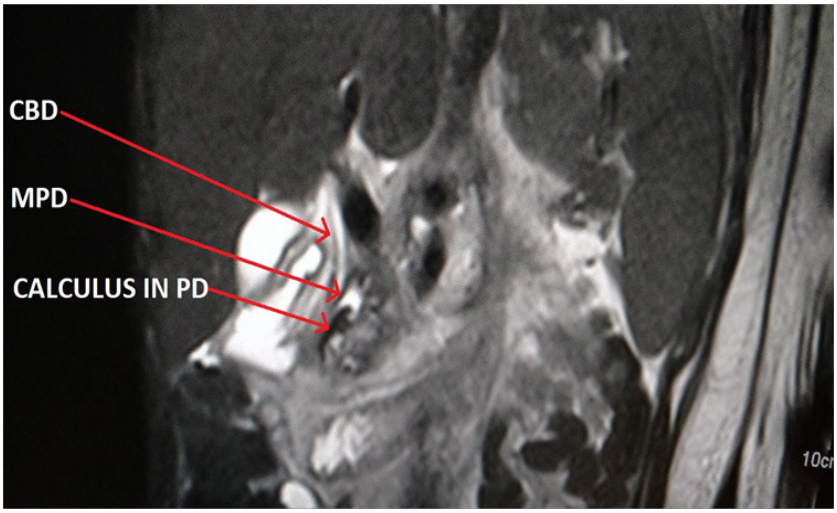

Figure 2. MRCP image.

peri-pancreatic fluid collection and there was gross free fluid in the left pleural cavity. Hence provisional diagnosis of acute gall stone pancreatitis with pleural effusion was made. Patient was put on Higher Antibiotics and Inj. Octreotide. She continued to drain around 1L of pleural fluid daily and had become quite debilitated.

After 4 days of mechanical ventilation, she could be weaned off ventilator but continued to need nasal Oxygen and drain about 1L of pleural fluid. She underwent Contrast CT Abdomen which showed oedematous pancreas with calcifications in its parenchyma and dilated Main Pancreatic Duct (MPD). It also showed normal CBD and GB. Hence decision to perform MRCP was taken.

At MRCP, T2 weighted images showed Hypointense, elongated calculus measuring 20mm x 7mm within the MPD near its insertion into the duodenum. MPD was dilated, 8mm in diameter in its entire course. Pancreas was diffusely oedematous with peri-pancreatic fluid collection and fat stranding. There were no stones in the biliary tree (Figure 2).

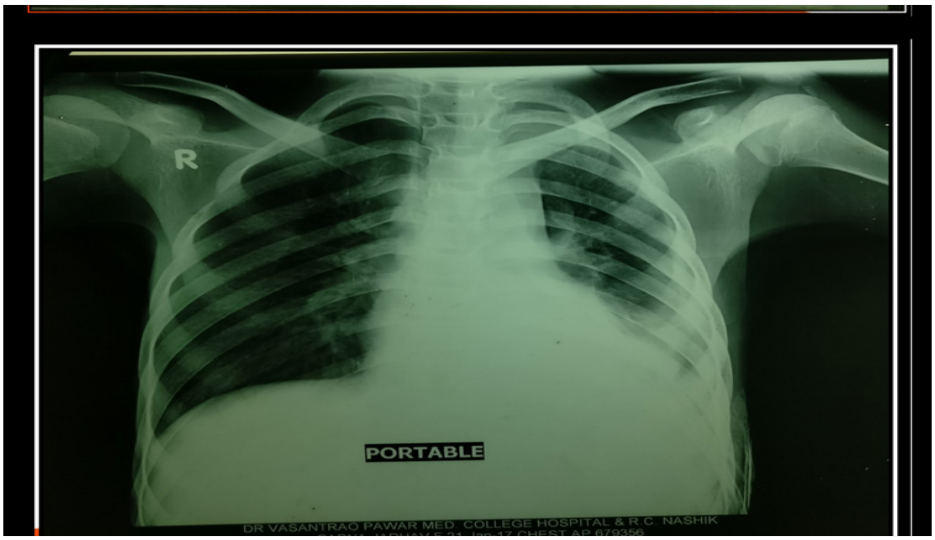

Figure 3. Post ERCP and Pancreatic duct stenting.

At this stage pleural fluid amylase was asked for, which came 44,000 IU. Genetic Testing for etiology of chronic pancreatitis could not be done due to financial constraints. Thus a diagnosis of Calcific Ideopathic Chronic Pancreatitis (CICP) with pancreatico pleural fistula was reached. The Patient obviously needed ERCP, stone removal and stenting.

We attempted ERCP after about 20 days of admission. At ERCP, selective Pancreatic Duct cannulation showed leak from the head region, thus explaining the PPF. MPD was dilated with multiple filling defects in the head and body region. Wide pancreatic duct sphincterotomy was done. Repeated balloon sweepings brought out multiple soft pancreatic calculi. Total duct clearance was achieved. A 5Fr, 7cm straight pancreatic stent was deployed. After the stenting, ICD output dropped drastically. Lung expanded and ICD was removed by 7th post procedure day (Figure 3).

Inj. Octreotide was tapered and omitted. Patient was gradually put on full oral diet and her serum amylase and lipase came back to normal. Patient was discharged on pancreatic enzyme supplements. At one year follow up, MRCP showed no leak or stones or stricture. The stent was not seen. The patient was totally asymptomatic, was eating well and had put on weight.

3. Discussion

As per International Study Group of Paediatric Pancreatitis (INSPIRE), incidence of pancreatitis in children is 3.6 to 13.2 per 100,000 population[1]. PPF is seen in 0.4–7% of patients of Chronic pancreatitis[5].

Alcoholism is the commonest cause of Chronic Pancreatitis in adults, 60% patients being males, whereas ductal obstruction is the primary cause in children[2]. Trauma contributes 0.5% of cases. A Pseudocyst is found in 43–79% of cases.

The hallmark of PPF is the massive and recurrent Pleural Effusion (PE) which is rich in Amylase, usually more than 1000 IU[2]. This can be easily distinguished from lesser degrees of PE associated with Acute Pancreatitis.

In PPF, pancreatic fluid leaks into pleural cavity through posterior disruption of MPD, finding its way to pleural cavity through natural diaphragmatic hiatus. Alternatively, a Pseudocyst can also rupture into mediastinum giving rise to PE. PE due to PPF is on Left side in 76% cases, on Right side in 19% cases and Bilateral in 14% cases[2].

Differential Diagnosis of Amylase rich PE in children are Acute or Chronic Pancreatitis, Pancreatic Trauma, Iatrogenic Rupture of Pancreatic duct[6]. MRCP is the 'gold standard' in PPF. It is Non-invasive and delineates the pancreatic ductal anatomy accurately. It also depicts parenchymal pathology, presence of pseudocyst and peri-pancreatic collection if any[4].

The sensitivity of MRCP is estimated at 80%. False negative results may be caused by multiple pseodocysts or ascites which reduce the sensitivity[2]. J Zhang *et al.*, reported that only in 53% of PPF cases, actual anatomy of the fistulous tract can be demonstrated on various

imaging techniques[6]. A Therapeutic Plan on the basis of Ductal Anatomy on MRCP was proposed by Wronski *et al.*[4]. A normal or mildly dilated MPD responds well to chest drainage with Inj.Octreotide; while Endoscopic Stent Placement benefits patients with ductal disruption located in head or body of pancreas.

All patients with suspected PPF should undergo ERCP as it is diagnostic and therapeutic as well. ERCP leads to diagnosis in 80% cases and demonstrates fistulous tract in 59–74% cases[5]. Pancreatic duct stones can be extracted and stenting bridges the site of ductal disruption thus reducing the fistula output. Failure of the above requires surgical intervention. Primary Surgical Management may be tried in patients with Complete duct obstruction, Ductal obstruction proximal to fistula site and Ductal disruption in the tail of pancreas[3].

Lateral Pancreaticojejunostomy or Puestow's Procedure is the procedure of choice when the MPD is dilated. Cyst Enterostomy with Roux en Y drainage may be sufficient when the MPD is non dilated. Partial Pancreatic Resection is recommended when the leak is in the tail region due ductal disruption. Success rate of ERCP is reported to be 96% as compared to 95% with Surgical Procedures[3].

4. Conclusion

Early recognition of PPF and early intervention either endoscopic or surgical, prevents lot of morbidity, whether it is effective in preventing long term complications like Endocrine and Exocrine Pancreatic insufficiency remains to be seen.

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