

CASE REPORT

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Recurrent Pancreatic Pseudocyst Secondary to Hypercalcaemia with Underlying Parathyroid Adenoma

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ABSTRACT We report a case of recurrent pancreatic pseudocyst secondary to primary hyperparathyroidism (PHPT). The patient presented with a large left sided pancreatic pseudocyst (PP) complicated with pancreatic abscess requiring long term antibiotic therapy and image-guided drainage. The patient had persistent hypercalcaemia which after several investigations led us to the diagnosis of PHPT. We performed an elective parathyroidectomy after the expectant management for the pancreatitis complicated with pancreatic pseudocyst. Although hypercalcaemia is the 4th common cause of pancreatitis, its exact association between those primary hyperparathyroidism and pancreatitis is unclear.

Keywords: Hypercalcaemia; parathyroid adenoma; pancreatic pseudocyst; primary hyperparathyroidism

Pancreatitis is usually caused by gallstone disease and alcohol consumption. Although hypercalcaemia is a known cause for pancreatitis, the cases of primary hyperparathyroidism (PHPT) leading to formation of pancreatic pseudocyst (PP) are rare. We describe a case of recurrent PP which is caused by chronic pancreatitis secondary to PHPT and resolved after parathyroidectomy. Less than 1% of patients with pancreatitis have hyperparathyroidism. The appropriate therapy for patients with pancreatitis associated with hyperparathyroidism remains unclear especially when it is complicated with pancreatic pseudocyst.

CASE REPORT

This is a 46 y-o gentlemen with history of laparoscopic cholecystectomy in 2017. Presented with sudden onset of left hypochondrium (LHC) pain approximately 5 months post operation. The pain was radiating to the back, dull aching and usually appeared post meals. His abdomen was tender over the LHC without any signs of peritonism. Initial serum

amylase level was raised. Computer tomography (CT) was performed and the patient had multiloculated fluid in the peripancreatic region of the tail and distal portion of the body of the pancreas extending to the lesser sac, anteromedial to the spleen and in the left flank extending caudally to the level of left iliac crest, size 16.9 cmx14.6 cmx11 cm. Due to the presence of debris layering at the dependent aspect of the loculated fluid in the left flank, he underwent laparotomy and PP removal.

He presented again 1 month after the operation for persistent abdominal pain and was treated for adhesion colic. CT scan showed residual chronic PP at the tail of the pancreas (it measures approximately 8.0 cm in widest diameter and 20 cm craniocaudally) with a large solidification at the left retro-peritoneal area. We performed an ultrasound (USG)-guided percutaneous drainage of the collection. The drainage contained pus in which cultures grew *E.coli* (extended-spectrum beta-lactamases), hence required at least 6 weeks of antibiotics.

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We noticed that the serum calcium level was 3.03 mmol/L (normal: 2.15-2.55 mmol/L), alkaline phosphatase normal level was normal and intact parathyroid hormone (iPTH) 16 pmol/L (normal: 1.6-6.9 pmol/L). The USG showed right parathyroid mass. Correlating with blood investigations, provisional diagnosis was primary hyperparathyroidism. Tc-99m SESTAMIBI scan showed functioning right parathyroid adenoma in the inferior pole of the right thyroid gland. We performed right inferior parathyroidectomy and the intra-operative iPTH showed reduction to normal levels in 15 mins. The cause of the recurrent abdominal pain with pancreatic pseudocyst seems to be a sequela of the parathyroid adenoma. Currently, the patient is asymptomatic and recent USG shows no evidence of pancreatic collection (Figure 1, Figure 2, Figure 3, Figure 4). We have obtained informed consent from the patients for the details mentioned in this case report.

DISCUSSION

In patients with acute pancreatitis, the prevalence for pancreatic pseudocyst is approximately 6%-18%.¹ The prevalence of acute pancreatitis in PHPT has been estimated to be between 1.5% and 13%.² The diagnosis of pancreatitis is usually achieved with history, clinical examination and biochemical investigations such as serum amylase or lipase levels. Imaging studies such as USG and CT scans are often the method of choice for confirming diagnosis and monitor its complications. CT has 82% to 100% sensitivity and 98% specificity for diagnosing pancreatitis.³

Several studies have concluded that patients with acute PP with a diameter less than 4 cm regress spontaneously and requires no treatment, provided they are asymptomatic. The rate of spontaneous regression

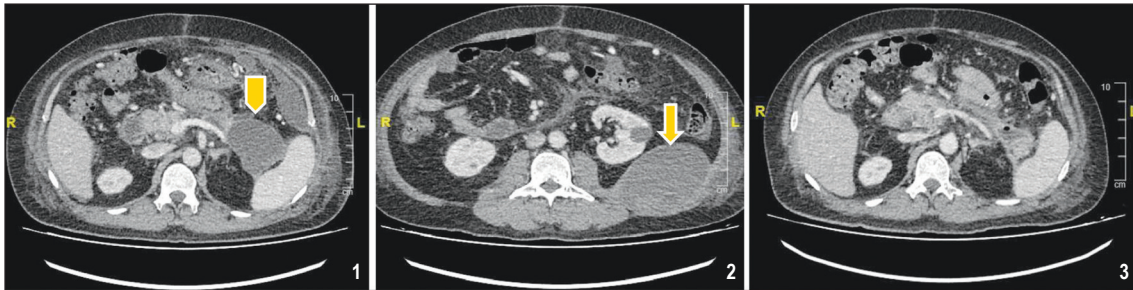


FIGURE 1, 2, 3: Figure 1 (left): Pancreatic collection at the tail. Figure 2 (right): Collection extending craniocaudally and confines within the anterior pararenal space and tracking posteroinferiorly, splitting the posterior perirenal fascia medially. Figure 3 (below): CT taken 2 weeks after the USG guided drainage showed reduction in collection and currently measures 5.0 cm (previously 8 cm) in widest diameter and 11.6 cm craniocaudally (previously 20 cm).

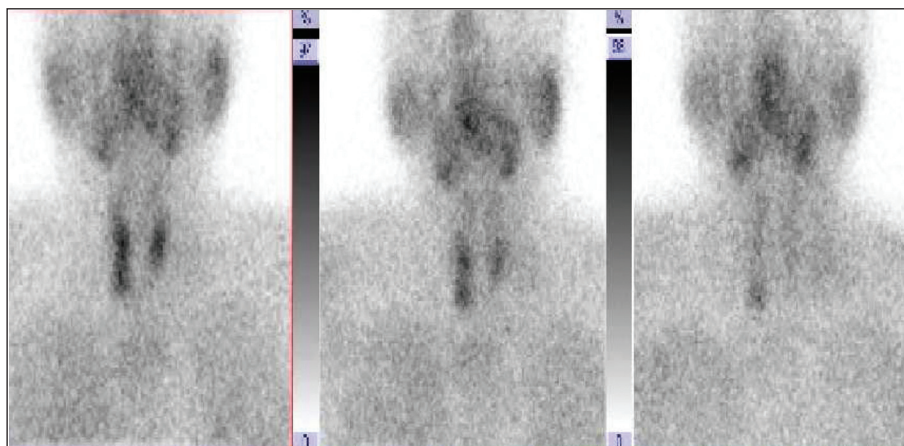


FIGURE 4: SESTAMIBI Tc-99m scan identifying the active parathyroid gland (arrow : Right inferior parathyroid gland).

in patients with chronic pancreatic pseudocyst is under 10%.¹ Factors which prevent spontaneous resolution include size more than 5cm, wall thickness more than 1cm, lack of communication with Wirsung's duct, enlargement of cyst on follow up evaluation and multiple cyst or underlying chronic pancreatitis.^{1,4} This patient has bouts of pain since 2017 and persisted after cholecystectomy. Hence, the possible cause would be chronic pancreatitis which requires expectant treatment.

The first successful surgical drainage of a PP was described by Bozeman in 1882. Recent developments in treatment of PP include endoscopic drainage, USG-guided drainage, laparoscopic and open drainage. This patient was given antibiotics for approximately 6 weeks, yet the collection persisted. After failure of antibiotics treatment, open surgical drainage was performed. However, the patient had recurrent PP complicated with pancreatic abscess which was attributed to his hypercalcaemic state due to PHPT. Without surgical treatment of parathyroid disease, pancreatitis and its sequelae such as PP is less likely to resolve spontaneously.⁴

Since the first illustrated symptoms of hyperparathyroidism in 1925, the presenting symptoms may vary broadly. The incidence of pancreatitis in patients with PHPT is approximately 7% to 10%.⁵ Although the exact association between pancreatitis and PHPT is unclear, it is suggested that an elective parathyroidectomy after the acute management of pancreatitis seems to prevent recurrent episodes of pancreatitis.^{6,7}

There are 2 mechanisms of hypercalcemia-induced pancreatitis. Hypercalcemia can lead to

de novo activation of trypsinogen to trypsin, resulting in autodigestion of the pancreas and subsequent pancreatitis. Another explanation is that hypercalcemia leads to the formation of pancreatic calculi, ductal obstruction, and subsequent attacks of acute or chronic pancreatitis. Also, genetic risk factors may predispose patients with PHPT to pancreatitis. The calcium level is probably of major importance in the development of pancreatitis. The mean calcium values among patients with PHPT and pancreatic disease have been reported to be significantly higher than those in patients with PHPT without pancreatic involvement.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Harinthiran Vijeyan, Ikhwan Sani Mohamad, Maya Mazuwin Yahya, Leow Voon Meng; **Control/Supervision:** Ikhwan Sani Mohamad, Maya Mazuwin Yahya, Leow Voon Meng; **Writing the Article:** Harinthiran Vijeyan; **References and Fundings:** Ikhwan Sani Mohamad, Maya Mazuwin Yahya, Leow Voon Meng.

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