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Primary Hyperparathyroidism Presenting With Pancreatic Pseudocyst: An Unusual Presentation

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IMPORTANCE The association between hyperparathyroidism and pancreatitis has been under debate for many years. The incidence of hyperparathyroidism related pancreatitis ranges between 1.5-7%. Here, we report a case of a parathyroid adenoma related pancreatitis in a 54-year-old lady, who had a month-long history of epigastric pain. The pancreatic pseudocyst rooted from hypercalcemia occurring secondary to excess PTH secretion by pituitary adenoma. The patient underwent surgery for both the adenoma and pseudocyst followed by normalization of serum calcium and PTH levels and resolution of the symptoms.

KEY WORDS Primary Hyperparathyroidism; Pancreatic Pseudocyst; Pancreatitis

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Case Report

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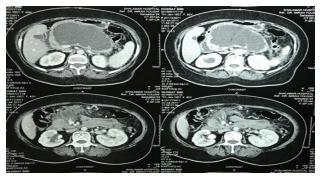
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rimary hyperparathyroidism (PHPT) is an endocrine disorder characterized by hypercalcemia and elevated levels of parathyroid hormone secreted by an overactive parathyroid gland.¹ A solitary pituitary adenoma accounts for PHPT in approximately 80% of the cases. Other causes include hyperplasia of multiple parathyroid glands and a rare parathyroid carcinoma.² Presentation of PHPT varies from being asymptomatic³ to the development of renal stones⁴, electrolyte imbalance, gastrointestinal disturbances and acute pancreatitis^{5,6}. Acute pancreatitis is inflammation of pancreas known to be caused by gallstones (30-60%), alcohol (15-30%), post Endoscopic Retrograde Cholangiopancreatography (ERCP) pancreatitis (5-10%), and hypertriglyceridemia (1.3-3.8%). It is very rare for PHPT to present as an acute pancreatitis since hypercalcemia itself is the cause of pancreatitis in 1.5-7% of the population7. Here, we report a rare case of pancreatic pseudocyst occurring secondary to parathyroid adenoma induced hypercalcemia which was subsequently confirmed on histopathology.

CASE PRESENTATION

A 54-year-old Pakistani lady had acute bout of acute pancreatitis and was conservatively treated. She again had epigastric pain a month later along with fullness in epigastrium. Her initial laboratory investigations showed normal LFTs and RFTs. Her serum calcium level was raised (12.8 mg/dl), serum sodium was low (130 mmol/L). Ultrasound abdomen and pelvis showed a large thickwalled cystic lesion with internal heterogenous contents

anterior to the pancreas, suggestive of pancreatic pseudocyst and a 21mm right renal pelvic partially obstructing calculus causing right sided hydronephrosis. Contrast enhanced CT scan was then performed confirming pancreatic pseudocyst showing a large unilocular thickwalled cyst (139×75×65 mm) anterior to pancreatic neck, body and tail with thick internal enhancing partial septations and surrounding changes of acute inflammation.



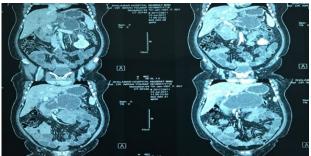


Fig 1a and b: CT scan film showing axial and coronal section.

Patient was diagnosed as a case of pancreatic pseudocyst. A thorough review of the case showed elevated calcium levels and the previous ultrasounds nullified the presence of gallstones. Her serum calcium, serum PTH and vitamin D levels were investigated and we found raised serum PTH level (541.9 pg/ml), hypercalcemia (11.8 mg/dl) and low levels of vitamin D (<7.50 nmol/l). On further evaluation, a well-defined lobulated hypo echoic lesion (15×6mm) inferior to right pole of lower lobe of thyroid gland was found on ultrasound neck representing parathyroid adenoma. Sestamibi scan further confirmed the findings. After proper hydration and resuscitative measures, patient underwent surgery of parathyroid adenoma which was found at the inferior pole. Right inferior parathyroidectomy was done and the specimen was sent for histopathology.





Fig 2 a & b: Gross parathyroid adenoma which was found at inferior pole (a) and recurrent laryngeal nerve is seen preserved following parathyroidectomy (b).

Post operatively patient's serum calcium and serum PTH levels dropped to 8.2 mg/dl and 2.5 pg/ml respectively. The histopathology report later confirmed a parathyroid adenoma of (2.5×1.0×0.5 cm) embedded in fibrofatty tissue. Patient's recovery period was smooth and she was discharged on 4th post-operative day with calcium supplements. Patient was readmitted for management of pancreatic pseudocyst. Imaging showed persistent

pancreatic pseudocyst ($14\times9.7\times8$ cm) for which open surgical cystogastrostomy was performed. Patient was discharged on 8^{th} post-operative day with full recovery and remained symptom free on follow up.

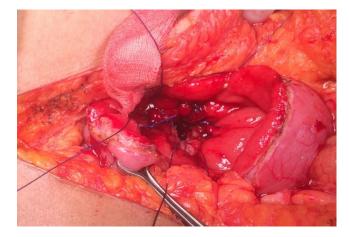


Fig 3: Open Cystogastrostomy was performed for pancreatic pseudocyst

DISCUSSION

Diagnosis of primary hyperparathyroidism (PHPT) is characterized by hypercalciuria, hypercalcemia and elevated PTH levels. The association between pancreatitis and PHPT has long been debated. There is no clear-cut pathophysiological basis for this presentation. However, it is thought that hypercalcemia induces acid lysosomal hydrolases to accelerate the conversion of trypsinogen to trypsin leading to autodigestion of pancreas.^{8 7 9} Others argue that precipitation of calcium in the pancreatic ductal system causes obstruction through calculi, inciting recurrent attacks of pancreatitis.¹⁰ It has been suggested that hypercalcemia poses 1.3 times increased risk of acquiring pancreatitis.11 Pancreatitis in PHPT occurs when calcium levels exceed beyond a threshold level as proved by certain studies demonstrating increased calcium levels in patients of PHPT with pancreatitis as compared to those without pancreatitis. 12 The incidence of PHPT presenting as pancreatitis was determined in an old Mayo clinical experience from 1950-1975, which turned out to be 1.5%.13 Jacob stated that out of 1284 patients admitted for pancreatic disease and 101 patients admitted for PHPT, 13 patients (1%) had concomitant PHPT and pancreatitis with 8 patients having no additional cause of pancreatitis other than hypercalcemia. He also proposed that PHPT may manifest as acute pancreatitis, recurrent pancreatitis without chronic pancreatitis, chronic pancreatitis with or without calcifications or PHPT complicated by pancreatitis in post-operative period.¹⁴ Cases have now been reported where pancreatitis was the only manifestation of PHPT¹⁵ ¹⁶ ⁷.The most effective treatment for PHPT is ultimately surgical removal of parathyroid gland, resulting in normalization of serum calcium levels and resolution of the associated symptoms. Our patient had no symptoms related to hypercalcemia other than epigastric pain and vomiting which turned out to be due to pancreatic pseudocyst formation as confirmed on CT abdomen. The serum calcium levels dropped to normal after parathyroidectomy but the symptoms vanished completely only after cystogastrostomy for the pancreatic pseudocyst was done.

CONCLUSION

Although, it is rare for PHPT to present as pancreatitis, we should never overlook its possibility. Pancreatitis is caused most commonly due to gall stones and alcohol but the workup must include the other less common causes including hypercalcemia. The disease has a good prognosis if diagnosed and managed promptly and effectively.

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