CASE REPORT

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Gastric Outlet Obstruction Following Recurrent Pancreatitis Uncovers a Giant Parathyroid Adenoma: A Case Report

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Abstract

A 35-year-old female presented with abdominal pain, fever, projectile vomiting, and a diffuse tender epigastric mass. She was diagnosed to have acute persistent pancreatitis with a pancreatic pseudocyst. Elevated serum calcium levels provided an etiologic link between hypercalcemia and pancreatitis. On examination, a nodule was found in the left side of her neck which was later diagnosed as a giant left inferior parathyroid adenoma. This report highlights the critical analysis of history, examination, and investigations to reach an ultimate diagnosis. Pseudocyst drainage and parathyroidectomy resolved her symptoms.

Key words: giant parathyroid adenoma, pancreatitis, gastric outlet obstruction, primary hyperparathyroidism

INTRODUCTION

Acute pancreatitis, an inflammation of the pancreas, can result in life-long morbidity and even mortality if not treated appropriately. Pancreatitis most commonly results from gallstone disease, alcohol intake, and following procedures like endoscopic retrograde cholangiopancreatography.[1] Primary hyperparathyroidism (PHPT) mediated hypercalcemia is comparatively a less common cause of pancreatitis (1.5-13%).[2]

Although PHPT is a common endocrine disorder, in Western literature, approximately 75-80% of patients are asymptomatic and are detected incidentally by routine calcium screening.[3] However, in our setup, due to lack of awareness and routine screening, patients are diagnosed in the symptomatic stage with the most common presenting features being bone pain and metabolic myopathy.[4] Seven percent of PHPT cases develop pancreatitis.[3]

However, PHPT manifesting as gastric outlet obstruction is rarely described in literature with no reports from India. Parathyroid adenomas are usually small (< 2 cm) and weigh less than 1 gram. However, in rare instances, adenomas may grow large and weigh more than 95th percentile or 3.5 grams. Such adenomas are labelled as "giant" adenomas.[5-7]

In this case report, we describe a young woman presenting with abdominal pain and non-bilious projectile vomiting. The features were suggestive of gastric outlet obstruction due to recurrent pancreatitis and pancreatic pseudocyst. An incidental finding of hypercalcemia was the only clue to an underlying PHPT leading to a whole gamut of signs and symptoms.

CASE

A 35-year-old female presented to the emergency department complaining of intermittent abdominal pain over 3 months which became severe over the past two days. It was associated with fever and persistent postprandial non-bilious vomiting for seven days. She had a history of intermittent generalized body ache and epigastric pain which subsided after pain relievers. She also sustained blunt trauma to the abdomen after a fall, following which she had severe pain in the epigastric region.

She consulted in the Gastroenterology department where she was diagnosed to have acute pancreatitis based on blood investigations showing raised serum amylase (112 U/L; normal range: 28 - 100 U/L) and lipase levels (138 U/L; normal range: 13 - 60 U/L). A contrast-enhanced computed tomography (CT) of the abdomen showed a bulky pancreas

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with peripancreatic fluid collection in the head, body and tail.

During this episode, her vital signs were stable. There was no pallor, icterus, cyanosis, or edema. Examination of her abdomen revealed a diffuse, firm, and tender epigastric mass which did not move with respiration. Given the present findings and clinical background, recurrent pancreatitis with pancreatic pseudocyst was considered.

Investigations

Ultrasound (USG) of her abdomen showed necrotic areas in the head of the pancreas with intra- and peripancreatic fluid collection. Contrast-enhanced CT (CECT) abdomen showed peripherally enhancing collection in the pancreatic bed replacing the entire pancreas with extension into the lesser sac and gastro-hepatic region with few air foci within suggestive of walled-off necrosis, likely a pseudocyst (Figure 1). These features were consistent with residual pancreatitis with a pseudocyst compressing the stomach leading to gastric outlet obstruction.

Hematologic parameters were normal except for leukocytosis (total count – 18000/mm^3). Blood chemistry showed normal serum amylase (38 U/L), lipase (25 U/L), and renal parameters. However, a remarkable finding was hypercalcemia (blood ionized calcium, iCa^{2+}- 1.9 mmol/L; normal range: 1.1 – 1.4 mmoL/L). Further evaluation of the cause of this hypercalcemia revealed raised total serum calcium (12.3 mg/dL; normal range: 8.6 – 10 mg/dL), and markedly raised parathormone (PTH) levels (PTH - 759 pg/mL; normal range: 15 – 65 pg/mL) with mild hypophosphatemia (serum phosphorus - 2.1 mg/dL; normal range: 2.5 – 4.5 mg/dL).

After noting the hypercalcemia, her neck was re-examined and a firm palpable nodule was discovered in the lower left side. Neck USG revealed a 2.3 x 1.1 x 1.8 cm hypoechoic lesion inferior to the left lobe of the thyroid. Further 4-dimensional CECT revealed a 1.5 x 1.5 cm lesion, posterolateral to the left lobe of the thyroid, with arterial enhancement and washout, suggesting a left inferior parathyroid adenoma. These findings were concordant with the results of the ^{18}F-Fluorocholine PET/CT scan of the patient (Figure 2).

Differential diagnosis

The possible causes of PHPT were parathyroid adenoma, parathyroid cyst, cystic adenoma and parathyroid carcinoma. USG, 4-dimensional CT, and ^{18}F-Fluorocholine PET/CT imaging studies ruled out the possibility of a thyroid nodule or ectopic parathyroid.

Treatment

The patient was finally diagnosed to have a left inferior parathyroid adenoma and hypercalcemia-induced recurrent pancreatitis with a pancreatic pseudocyst manifesting as gastric outlet obstruction. She was stabilized, a Freka's nasojejunal tube was inserted for feeding and a CT-guided abdominal external drain was placed to decompress the pseudocyst. The patient was prepared for focused parathyroidectomy in an elective setting with preoperative endocrine and anesthesia consults.

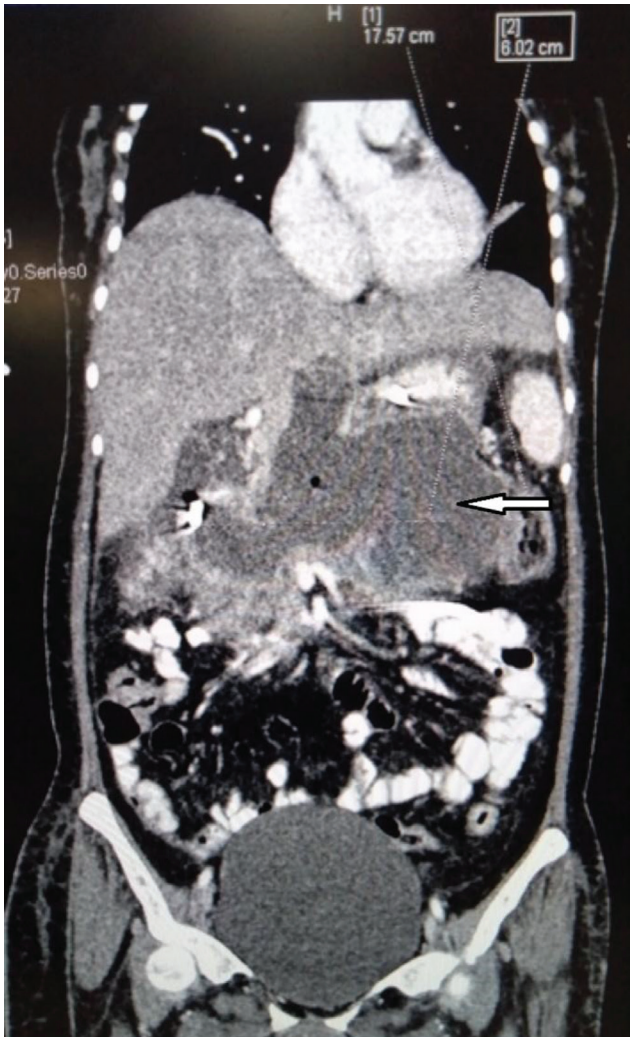

Figure 1. Contrast-enhanced CT scan of the abdomen showing pseudocyst of the pancreas (arrow) compressing the stomach.

Intraoperatively, a parathyroid nodule was identified at the inferior pole of the left lobe of the thyroid gland and was excised (Figure 3). Gross examination of the surgically excised parathyroid specimen revealed a 2.8 cm x 3 cm globular and well-circumscribed tumor weighing 4.8 grams (Figure 4). Histological examination of the specimen was compatible with a benign adenoma (Figure 5).

Intact parathormone (iPTH) levels declined drastically in the immediate postoperative period (from 759 pg/mL to 67.25 pg/mL). Serial monitoring of serum calcium revealed hypocalcemia on the second postoperative day (serum total calcium - 8.0 mg/dL) and third postoperative day (serum total calcium - 7.8 mg/dL) accompanied by tingling and

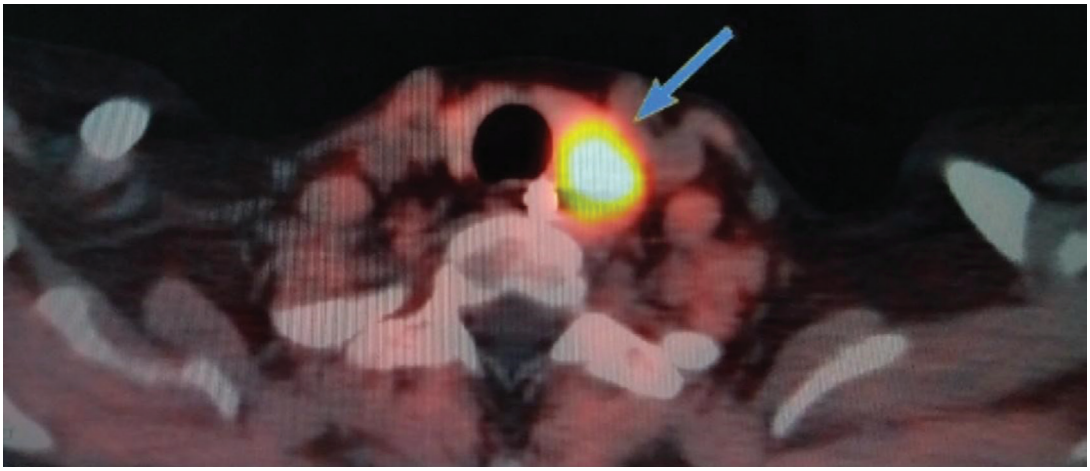

Figure 2. ^{18}F-Fluorocholine PET/CT scan showing transaxial image of intense tracer uptake behind left lobe of thyroid suggesting parathyroid hyperactivity (arrow).

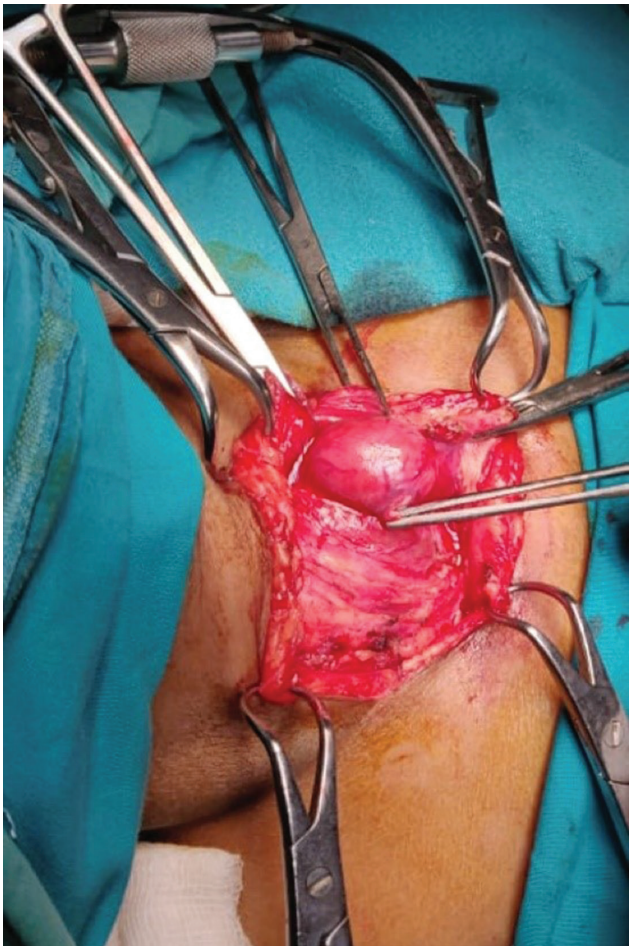

Figure 3. Intraoperative findings of focused parathyroidectomy showing enlarged left parathyroid.

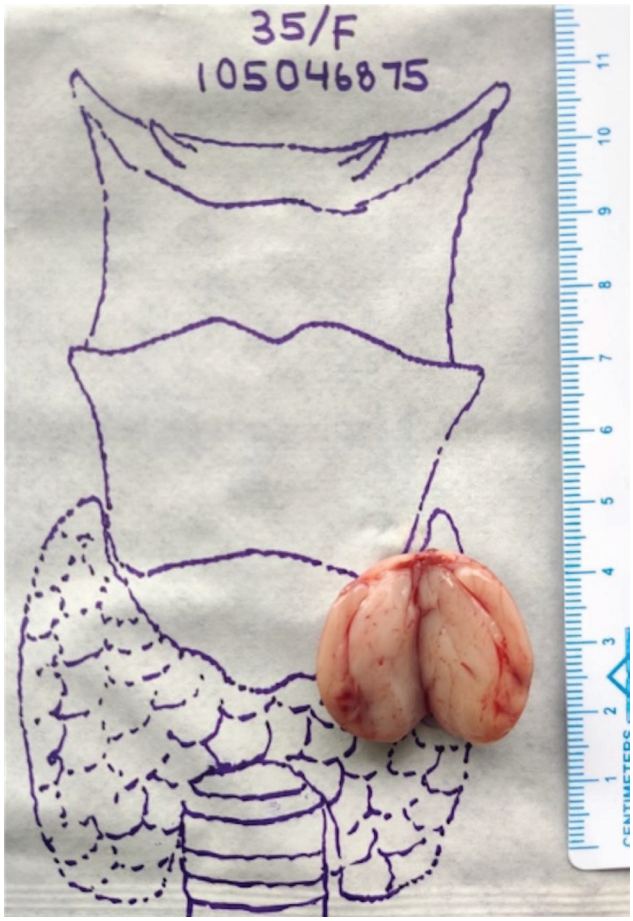

Figure 4. Schematic diagram showing the size and location of the parathyroid specimen with a cut section depicting gross features of adenoma.

numbness of the perioral region and extremities. These features were suggestive of hungry bone syndrome and managed with intravenous and oral calcium with vitamin D supplements.

The patient improved over 2 weeks with the resolution of abdominal pain. Postoperative abdominal USG revealed resolving pancreatitis. The nasojejunal tube was removed on the eighth postoperative day. The abdominal drain was removed on the tenth postoperative day. Oral feeding was gradually increased and was tolerated. She was discharged on the twelfth postoperative day. At 8 months follow-up, she was doing well and her serum calcium and parathormone levels were normal.

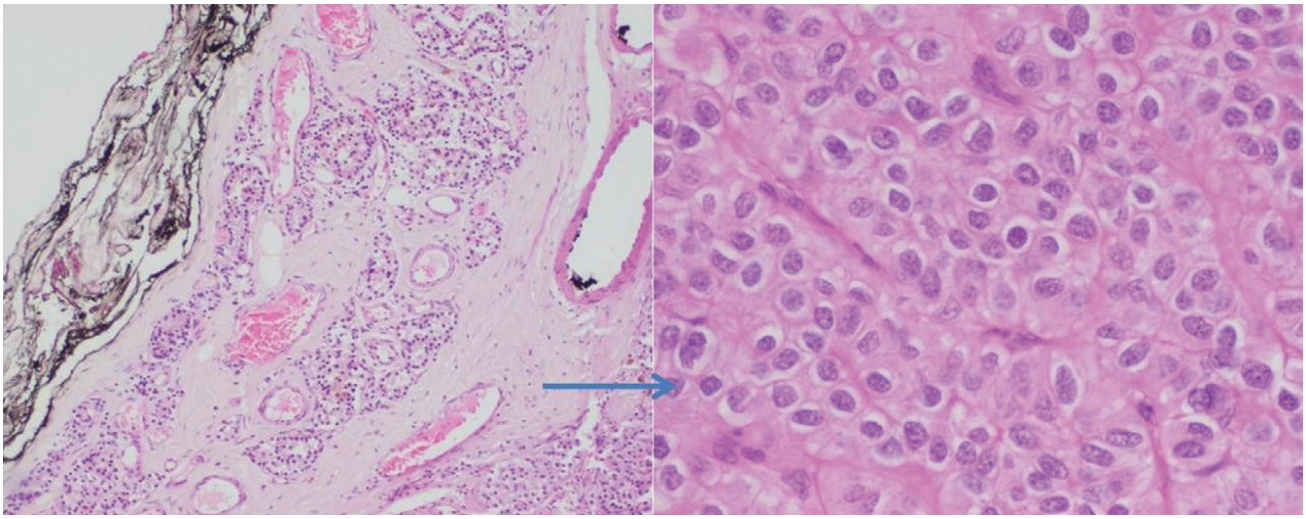

Figure 5. Histological examination revealed normal parathyroid tissue at the periphery of the cellular parathyroid tumor (arrow) (H&E, 100x). The inked resected surface can be seen on the upper left margin (H&E, 400x). A cellular parathyroid tumor composed of monomorphic population of chief cells. The tumor cells are arranged as nests separated by thin fibrovascular septae, showing minimal nuclear atypia and lacking mitotic activity (right).

DISCUSSION

Primary hyperparathyroidism (PHPT) is one of the common endocrine disorders characterized by increased secretion of parathormone (PTH). It predominately affects women from the fifth to seventh decades of life.[8] Most frequent causes of PHPT include solitary parathyroid adenoma (85–90%), followed by parathyroid hyperplasia (10-15%), multiple adenomas (5%), and, rarely, parathyroid cancer (<1-5%).[9] PHPT is most commonly diagnosed in the asymptomatic stage by routine biochemical screening.[3] The classical features associated with symptomatic disease include fatigue, myalgias, bone pain, constipation, nephrolithiasis and bone demineralization.[10]

The size of parathyroid adenoma in cases of PHPT is variable and a significant correlation has been described between gland weight, calcium and PTH levels.[7] Patients with giant adenomas are more likely to have asymptomatic disease and solitary gland involvement despite higher mean preoperative calcium and PTH levels. However, symptoms of postoperative hypocalcemia are more frequent compared to non-giant adenomas.[7] Although our patient had a giant adenoma, the symptoms related to hypercalcemia were remarkable with the rare manifestation of gastric outlet obstruction.

The association of pancreatitis with PHPT is between 1.5 – 15.0% in various studies.[9,11-13] with acute, recurrent, and chronic forms of pancreatitis reported.[2] Factors precipitating pancreatitis in cases with PHPT include hypercalcemia-mediated pancreatic duct stone formation and activation of intrapancreatic trypsinogen to trypsin, both of which lead to parenchymal injury.[2] Genetic predisposition may explain the development of pancreatitis only in a subset of patients with PHPT.[11,14] The prevalence of pancreatitis in PHPT ranges from 6.8 to 15 percent in the Indian population compared to 3.2 to 8.1 percent in the Western population.[11] The incidences of PHPT-associated acute and chronic pancreatitis are almost similar in the Indian population, but acute pancreatitis predominates in the Western population.[12]

Among patients with acute pancreatitis, 7% develop pancreatic pseudocysts[15], while among those with chronic pancreatitis, 30-40% develop pancreatic pseudocysts.[16] Approximately 50% of these may remain asymptomatic or regress spontaneously; the rest may develop acute or chronic complications in the form of infection, bleeding, rupture, bile duct dilatation, gastric outlet obstruction or thrombosis of the portal or splenic vein.[17]

Gastric outlet obstruction due to pressure effect has been reported in 8%[18] of acute pancreatitis (due to any cause) related pseudocysts and 15%[17] of chronic pancreatitis (due to any cause) related pseudocysts. There is no consensus on whether surgical drainage is required for acute pancreatitis-induced pseudocyst without infection.[18] However, persistent vomiting from gastric outlet obstruction due to pseudocyst predisposes to malnutrition.[19] The risk of pseudocyst infection increases in the malnourished patient ultimately worsening the prognosis.[18] Therefore, most clinicians agree that surgical drainage is an effective treatment for pancreatic pseudocyst with features of gastric outlet obstruction.[18,20]

Symptoms of bone pain, recurrent nephrolithiasis, neuromuscular weakness and psychiatric disorders must raise the suspicion of parathyroid disease and prompt further evaluation. Elevated levels of serum calcium and PTH confirm the diagnosis.[7]

Localization of parathyroid adenoma is done by imaging studies like cervical USG, sestamibi scintigraphy, and

magnetic resonance imaging (MRI).[6,8] A Tc99m Sestamibi scan of the parathyroid glands has a sensitivity of 92% while neck USG has a sensitivity of 80% for the preoperative localization of parathyroid adenomas.[21]

Newer modalities like PET-CT imaging with ^{18}F-fluorocholine (^{18}F-FCH) tracer carry advantages in terms of detection of smaller adenomas and reduced scanning time with a high sensitivity of 92% and specificity of 100%.[22] Moreover, PET-CT ^{18}F-FCH has an advantage in identifying smaller lesions very close to the thyroid, low gland mass, and multiple gland involvement.[23]

In this case, the diagnosis was made based on the elevated serum calcium and PTH levels. Initial screening of the neck was conducted using USG, while localization of the lesion was done by 4D-CT. We also performed a PET-CT ^{18}F-FCH to rule out ectopic parathyroid tissue.

Surgical excision of the parathyroid adenoma is the preferred treatment. Documenting intraoperative or immediate postoperative decline in iPTH levels is important to confirm successful removal of the adenoma. Close monitoring of serum calcium levels in the postoperative period is crucial for the diagnosis and management of life-threatening hypocalcemia.[24] Long-term follow-up is equally important as the postoperative course can be complicated by episodes of acute or chronic pancreatitis. However, various studies have shown significant improvement in abdominal symptoms and a low recurrence of pancreatitis when normocalcemia is achieved following removal of the parathyroid adenoma.[25,26] Our patient's symptoms also improved without any recurrence of pancreatitis over 8 months of follow-up.

CONCLUSION

The present case report emphasizes the importance of thorough history taking and clinical examination to reach a final diagnosis. It is equally important to analyze laboratory investigations and their correlation with clinical signs and symptoms because PHPT may have myriad manifestations, and pancreatitis in a young, non-alcoholic patient warrants evaluation of endocrinologic causes.

Ethical Consideration
Patient consent was obtained before the submission of the manuscript.

Statement of Authorship
All the authors certified fulfillment of ICMJE authorship criteria.

Author Disclosure
The authors declared no conflict of interest.

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