Case Report

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Parathyroid Cyst Presenting as Acute Pancreatitis: Report of a Case

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We report the first case of hypercalcemia-induced acute pancreatitis caused by a functioning parathyroid cyst in a 67-year-old man. Laboratory investigation revealed increased serum amylase and lipase, increased serum ionized calcium and parathyroid hormone (PTH) levels, and decreased serum phosphate, indicating pancreatitis and primary hyperparathyroidism (PHPT). Abdominal computed tomography (CT) revealed mild swelling of the pancreatic head with peri-pancreatic fat infiltration and fluid collection around the pancreatic tail. Ultrasonography and CT of the neck showed a cystic lesion at the inferior portion of the left thyroid gland, suggesting a parathyroid cyst. There was no evidence of parathyroid adenoma by 99mTc sestamibi scintigraphy. PHPT caused by a functioning parathyroid cyst was suspected. The patient underwent surgical resection of the functioning parathyroid cyst owing to his prolonged hypercalcemia. At 3 weeks after the operation, his serum levels of PTH, total calcium, ionized calcium, inorganic phosphate, amylase, and lipase were normalized. At the follow-up examinations, he has remained asymptomatic.

Key Words: *Pancreatitis; Hyperparathyroidism, Primary; Cysts*

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INTRODUCTION

Pancreatitis is pathologically defined as the histologic presence of inflammation within the parenchyma of the pancreas.[1] In adult patients, biliary tract diseases and heavy alcohol consumption account for up to 80% of cases of pancreatitis. The metabolic causes of acute pancreatitis include diabetic ketoacidosis, hypertriglyceridemia, and hypercalcemia with or without hyperparathyroidism.[2]

Acute pancreatitis secondary to hypercalcemia is an uncommon presentation of primary hyperparathyroidism (PHPT) and its prevalence is estimated to be between 1.5% and 7%.[3] PHPT is most commonly caused by parathyroid adenoma and rarely by parathyroid carcinoma or parathyroid cyst.[4] Parathyroid cyst is a rare condition arising in the neck and anterior mediastinum. Parathyroid cysts include two categories: functioning and nonfunctioning, the former being associated with clinical hyperparathyroidism. The majority of patients, however, have a nonfunctioning cyst.[5] To date, acute pancreatitis caused by a functioning parathyroid cyst has not been reported. Herein, we report the first such case of hypercalcemia-induced acute pancreatitis caused by a functioning parathyroid cyst.

CASE REPORT

A 67-year-old man was admitted to Chonnam National University Hospital with sudden onset of epigastric pain. The pain did not radiate along his back but was relieved to some extent by his leaning forward. He had a history of diabetes mellitus, essential hypertension, and an old cerebrovascular accident. He had no previous history of peptic ulcer diseases, cholecystitis with gallstones, any alcohol ingestion, or abdominal surgery. On admission, he was afebrile, his blood pressure and pulse were normal, and he appeared well-nourished. Scleral icterus was not present. His abdomen was mildly distended, with tenderness in the epigastrium. No abdominal masses were seen; the liver, gallbladder, and spleen were not palpable; and bowel sounds were slightly hyperactive. Laboratory indexes were as follows: white blood cell count, 9,000/mm^3; hemoglobin, 15.7 g/dl; platelet count, 135,000/mm^3; serum albumin, 4.4 g/dl; aspartate aminotransferase, 29 U/L; alanine aminotransferase, 29 U/L; alkaline phosphatase, 84 U/L; and

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γ-glutamyl transpeptidase, 148 U/L. Serum amylase and lipase were 293 U/L and 660 U/L, respectively. C-reactive protein was 2.6 mg/dl. The results of the renal function study were within normal limits. Serum triglyceride was 93 mg/dl and serum total calcium, ionized calcium, and inorganic phosphate were 10.3 mg/dl, 2.8 mEq/L, and 1.9 mg/dl, respectively. Abdominal computed tomography (CT) revealed mild swelling of the pancreatic head with peri-pancreatic fat infiltration (Fig. 1A) and fluid collection around the pancreatic tail (Fig. 1B). The follow-up serum ionized calcium level was still elevated at 3.0 mEq/L. The serum level of parathyroid hormone (PTH) was 113 pg/ml. The results indicated hypercalcemia caused by hyperparathyroidism.

Ultrasonography and CT of the neck showed an approximately 5-cm cystic lesion at the inferior portion of the left thyroid gland, which suggested a parathyroid cyst (Fig. 2). The fine needle aspiration biopsy specimen suggested a cystic lesion. 99mTc sestamibi scintigraphy showed no evidence of parathyroid adenoma. The patient had prolonged hypercalcemia despite hydration and diuretics. He underwent surgical resection, and the histological examination with PTH stain was interpreted as a parathyroid cyst with focal parathyroid hyperplasia (Fig. 3).

At 3 weeks after the operation, his serum levels of PTH, total calcium, ionized calcium, inorganic phosphate, amylase, and lipase were normalized. Since the operation, he has had no recurrent episodes of acute pancreatitis.

DISCUSSION

Many conditions predispose to acute pancreatitis to varying degrees. These conditions include gallstones, alcohol, hypertriglyceridemia, and hyperglycemia.[2] Any cause of hypercalcemia, including hyperparathyroidism, metastatic bone disease, total parenteral nutrition, sarcoidosis, vitamin D toxicity, and infusions of calcium, can lead to acute pancreatitis.[6] Gallstones and alcohol are common causes of acute pancreatitis. In this case, the patient had no previous history of cholecystitis with gallstones or any alcohol ingestion. Furthermore, his aspartate aminotransferase, alanine aminotransferase, and bilirubin were within normal limits. Thus, we excluded alcohol and gallstone as a cause of the pancreatitis.

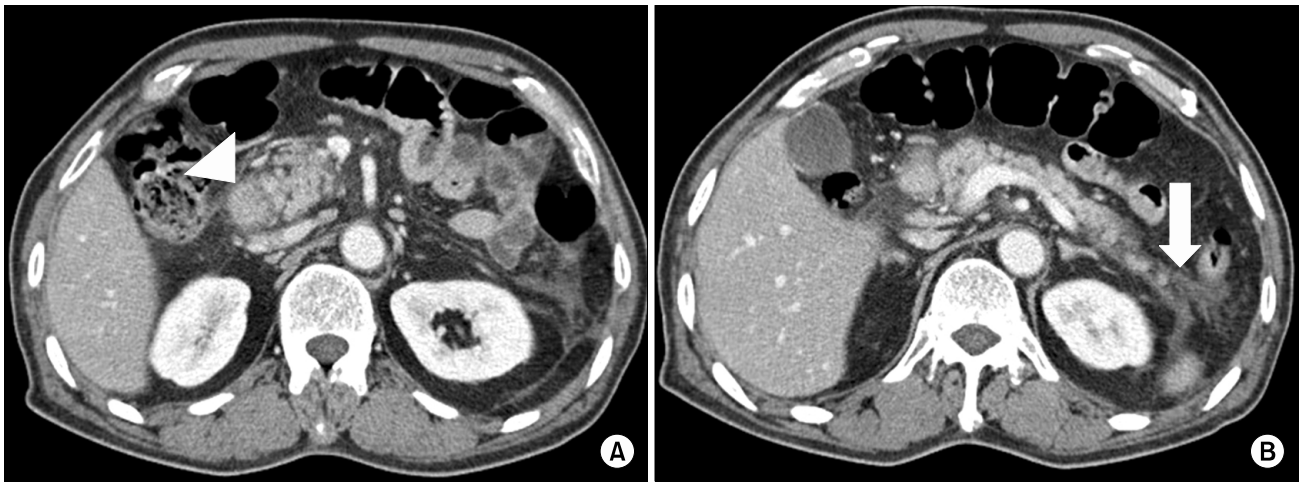

FIG. 1. Abdominal computed tomography (A, B) showed mild swelling of the pancreatic head with peri-pancreatic fat infiltration (arrow head) and fluid collection around the pancreatic tail (arrow).

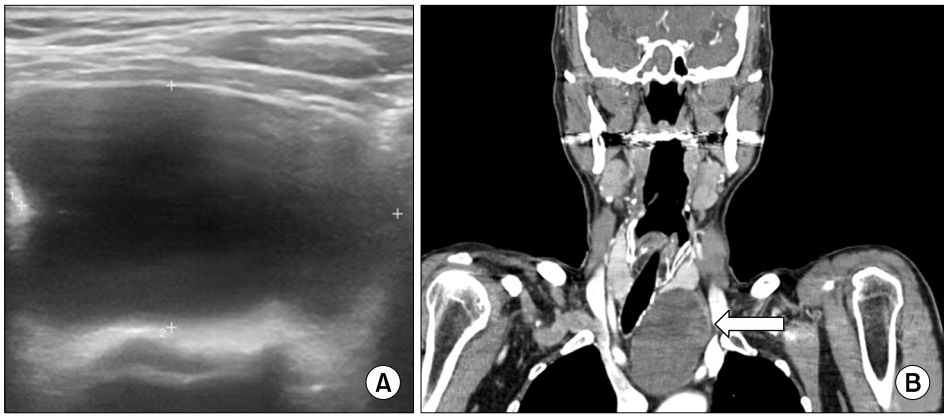

FIG. 2. Neck ultrasonography revealed a left infrathyroidal cystic lesion (A). Neck computed tomography revealed an approximately 5.4-cm sized low attenuated lesion (arrow) in the left the infrathyroidal area extending to the mediastinum (B).

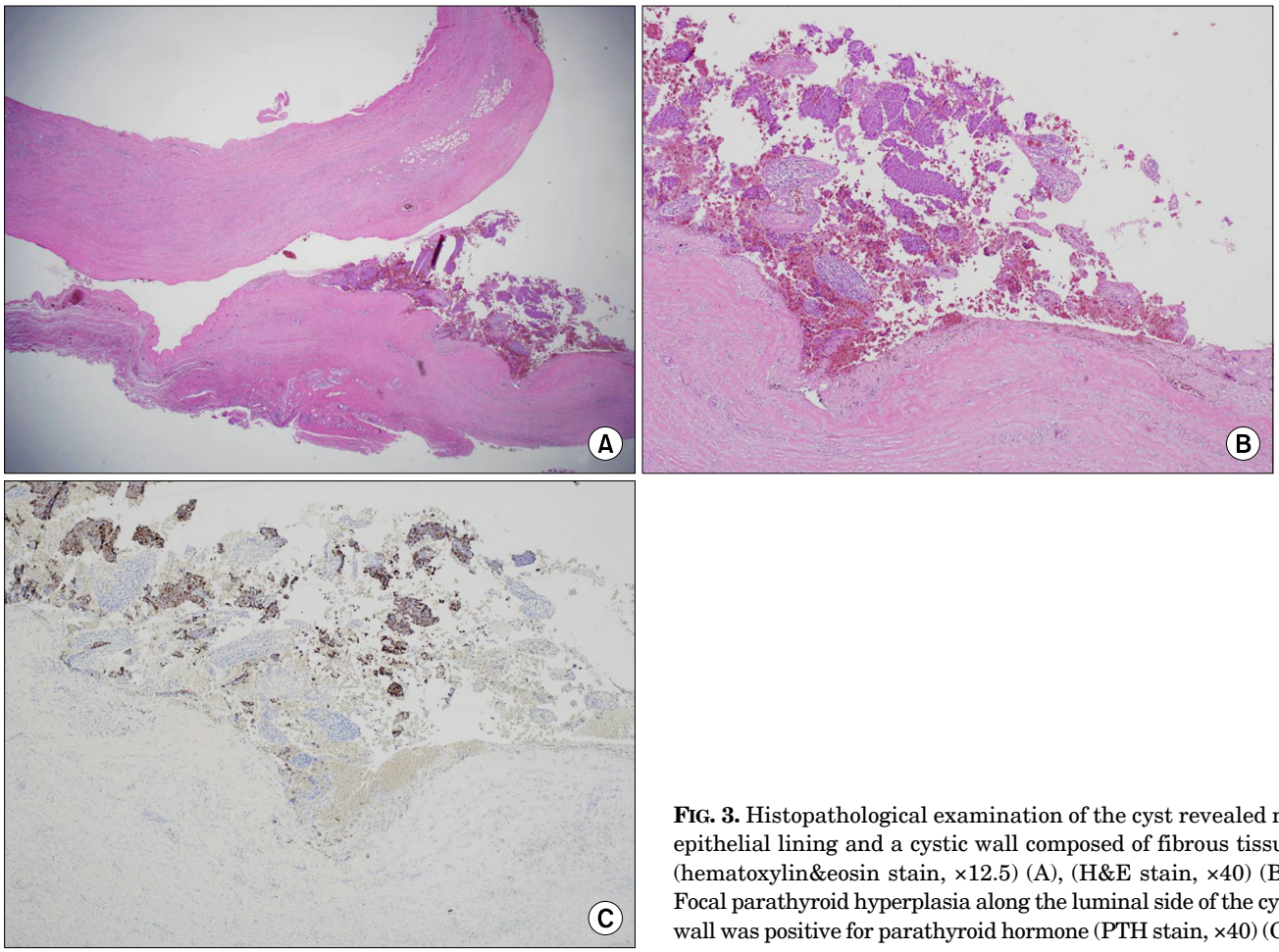

FIG. 3. Histopathological examination of the cyst revealed no epithelial lining and a cystic wall composed of fibrous tissue (hematoxylin&eosin stain, ×12.5) (A), (H&E stain, ×40) (B). Focal parathyroid hyperplasia along the luminal side of the cyst wall was positive for parathyroid hormone (PTH stain, ×40) (C).

The mechanisms of hypercalcemia-induced acute pancreatitis include deposition of calcium in the pancreatic duct and calcium activation of trypsinogen within the pancreatic parenchyma, with disruption of acinar cell ultrastructure and apoptosis of the acinar cell. However, PHPT causes less than 1% of all cases of acute pancreatitis, and the incidence of acute pancreatitis in hyperparathyroidism varies from 1.5% to 7%.[3,7]

PHPT represents a nonphysiological overproduction of PTH. The most common cause of PHPT is a single adenoma of the parathyroid gland. Rare causes include parathyroid hyperplasia, carcinoma, multiple endocrine neoplasia (MEN) type 1 and 2A, and parathyroid cysts.[2] Hypercalcemia-induced acute pancreatitis as one of the clinical manifestations of PHPT is very rare and is principally caused by parathyroid adenoma. Until now, however, there have been no reports of hypercalcemia-induced acute pancreatitis caused by a functioning parathyroid cyst.

Parathyroid cysts account for approximately 0.5% to 1% of all parathyroid pathologies.[8] Cystic degeneration of the parathyroid glands is not common, and a functioning parathyroid cyst with elevation of serum calcium and PTH levels occurs in only about 1% of all cases of hyperparathyroidism.[5] Most parathyroid cysts are nonfunctioning.

The pathophysiology of the evolution of parathyroid cysts is insufficiently understood. Rosenberg et al.[9] proposed two different settings. One is a true cyst containing an epithelial lining that will more frequently cause a nonfunctioning parathyroid cyst. The other is cystic degeneration of a parathyroid hyperplasia or adenoma. The latter mechanism may lead to a functioning parathyroid cyst. Similarly, the functioning parathyroid cyst in our case may have been derived from cystic degeneration of a parathyroid adenoma or hyperplasia.

The criteria for a functioning parathyroid cyst include preoperative biochemical and clinical evidence of hyperparathyroidism, identification of normal remaining parathyroid glands during surgery, histologic evidence of parathyroid tissue within the cyst wall, and correction of hypercalcemia after the operation.[10] In our case, laboratory investigation showed increased serum ionized calcium and PTH levels and decreased serum phosphate, thus indicating PHPT. Histologic examination of the surgical specimen confirmed a parathyroid cyst, and after the operation, serum levels of PTH, total calcium, ionized calcium, and inorganic phosphate were normalized. These findings indicate that our case had the features of a functioning parathyroid cyst as described above.

In summary, this is the first reported case of hypercalcemia-induced acute pancreatitis caused by a functioning parathyroid cyst. Second, a functioning parathyroid cyst, despite its rarity, should be considered in the differential diagnosis of the causes of acute pancreatitis. Third, accurate histopathologic diagnosis and surgical resection offer a chance for cure.

CONFLICT OF INTEREST STATEMENT

None declared.

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