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Brief Report

BRIEF REPORT

Parathyroid carcinoma-related severe acute pancreatitis during pregnancy: a case report and literature review

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Introduction

Primary hyperparathyroidism (PHPT) is caused by hyperfunctioning parathyroid adenoma, hyperplasia, or carcinoma. Parathyroid carcinoma is a rare disease affecting <1% of PHPT patients and is associated with complications such as acute pancreatitis (AP), which can be complicated by pregnancy [1]. Currently, pregnant patients with AP or PHPT can only be treated empirically as there is no consensus approach. Here, we report a rare case of parathyroid carcinoma with adenoma-related AP during pregnancy. The detailed diagnosis and treatment process are described, and clinical features of similar cases are summarized. We hope to improve the diagnosis and treatment of PHPT-related pancreatitis during pregnancy.

Case report

A 27-year-old Chinese primigravida at 12 weeks of gestation was admitted to our hospital, unconscious. She had begun vomiting excessively 8 days previously. At emergency presentation, her blood pressure was high but fluctuated slightly (160–180/110–120 mmHg). Her leukocyte count was 13.2×10^9/L. A comprehensive metabolic panel revealed alanine transaminase 12 U/L, total bilirubin 6.5 μmol/L, serum creatinine 68.1 μmol/L, amylase 79.6 U/L, potassium 2.88 mmol/L, calcium 3.33 mmol/L, and triglyceride 1.72 mmol/L. Potassium and an antiemetic drug were given, after which vomiting was relieved. However, 12 h before presentation at our hospital, she had severe epigastric pain and lost consciousness. The patient's past history was notable for Stage 2 hypertension diagnosed 2 months before pregnancy without any treatment. She denied a history of alcohol consumption or medications.

On presentation, physical assessment showed a pulse rate of 150 beats/min, blood pressure of 81/67 mmHg, and Glasgow coma scale of E3V2M4. Palpation of the thyroid showed a solid immobile nodule. Abdominal physical examination showed absence of bowel sounds, abdominal distention, tenderness over the epigastrium, and enlarged uterus.

The white blood cell count was elevated (29.51×10^9/L) with a predominance of neutrophils (92.4%). A comprehensive metabolic panel revealed serum creatinine 214 μmol/L, calcium 4.07 mmol/L, amylase 1,113 U/L, and lipase 8,735 U/L. The computed tomography (CT) scan showed enlarged pancreas, peripancreatic fluid collection, a low-density mass along the right lobe of the thyroid gland, and bilateral renal calculi. Ultrasound examination did not find any gallstones or dilatation of the bile duct.

After the initial assessment, the patient was transferred to the intensive care unit. Considering the impact of the following

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examinations and treatments, her family decided to terminate the pregnancy. Uterine aspiration was performed on Day 4.

Treatment of severe AP was started immediately. Supportive care included fluid resuscitation, gastrointestinal decompression, and parenteral nutrition. On Day 7, the patient developed a continuous fever. CT images showed newly formed acute peripancreatic fluid collection (Figure 1A). Considering the possibility of abdominal infection, antibiotics were given and CT-guided percutaneous catheter drainage was performed.

Treatment to reduce calcium using salmon calcitonin and zoledronic acid was also initiated immediately, and continuous renal replacement therapy was applied. The patient regained consciousness on Day 2. One week later, serum calcium dropped to 1.96 mmol/L and creatinine to 101 μmol/L. The parathyroid hormone level was 1914.2 pg/mL, which suggested PHPT and parathyroid crisis. Ultrasound examination revealed a 1.2-cm hypoechoic nodule at the left lobe and a 3.4-cm hypoechoic nodule behind the right lobe (Figure 1B). Technetium-99m-sestamibi imaging showed increased uptake in the posterior of the right thyroid lobe. No pituitary, thyroid, adrenal, or enteropancreatic tumors were found. After preoperative localization, parathyroid exploration showed the right nodule conglutinated with the thyroid and esophagus. En bloc resection of the right thyroid, isthmus, and central compartment of cervical lymph nodes, as well as surgical removal of the left nodule were performed. Histopathology revealed the left nodule to be parathyroid adenoma and the right nodule to be parathyroid carcinoma (stage pT1N0M0, AJCC UICC 8th edition). Immunohistochemical staining of the carcinoma was positive for vascular endothelial growth factor (VEGF) and parathyroid hormone, negative for CD56, Galectin-3, and mesothelial cell, with a Ki-67 index of 7% (Figure 1C–E). The patient's PTH level dropped to 27.7 pg/mL and calcium level to 1.78 mmol/L immediately. Calcium carbonate tablets and calcitriol were given.

The patient was discharged on Day 45. Peripancreatic fluid was well absorbed (Figure 1F). CT images revealed a newly formed pancreatic pseudocyst and decreased peripancreatic fluid collection 3 months later (Figure 1G). The exudate was fully absorbed after 6 months (Figure 1H).

Discussion

Here we report a case of PHPT caused by parathyroid carcinoma and adenoma, which led to severe AP during pregnancy. For a better understanding of PHPT-related AP in pregnancy, we listed and preliminarily summarized the similarities and differences between all the 26 cases reported so far (Supplementary Table 1) [2–9]. PHPT-related AP prefers to attack in the third (50%) or second (34.6%) trimester. The majority exhibit mild AP, with common manifestations such as abdominal pain and nausea. The optimal treatment of parathyroid mass during the gestational period is parathyroidectomy, since there is a high risk of failure with pharmaceutical management. Two cases were successfully treated with ethanol ablation, which may be a promising alternative approach [8, 9]. Adenomas are the predominant parathyroid tumor type (24 in 26 cases). The other two cases were carcinomas (including the present one). No hyperplasia were reported. During pregnancy, the severity and fetal death rate are lower in patients with PHPT-related AP than in those with other types of AP [10]. We wish to emphasize two main laboratory findings. First, unlike most asymptomatic PHPT patients, with an average calcium level of <2.75 mmol/L, pregnant women with PHPT-related AP have highly increased serum calcium levels, especially in case of organ failures [1]. The line that indicates severe symptomatic AP could be drawn above the normally defined severe hypercalcemia (>3.5 mmol/L). Second, the serum PTH concentration seems to be correlated with the severity of AP and the pathological type of parathyroid mass. If the PTH concentration increases dramatically (10-fold higher than the upper limit of reference value), parathyroid carcinoma should be considered. However, since cases are limited, more studies are needed to prove the relationship between clinical findings and laboratory indices. In conclusion, we present a case with PHPT-related pancreatitis during pregnancy in detail, summarize the features of similar cases, and briefly discuss the

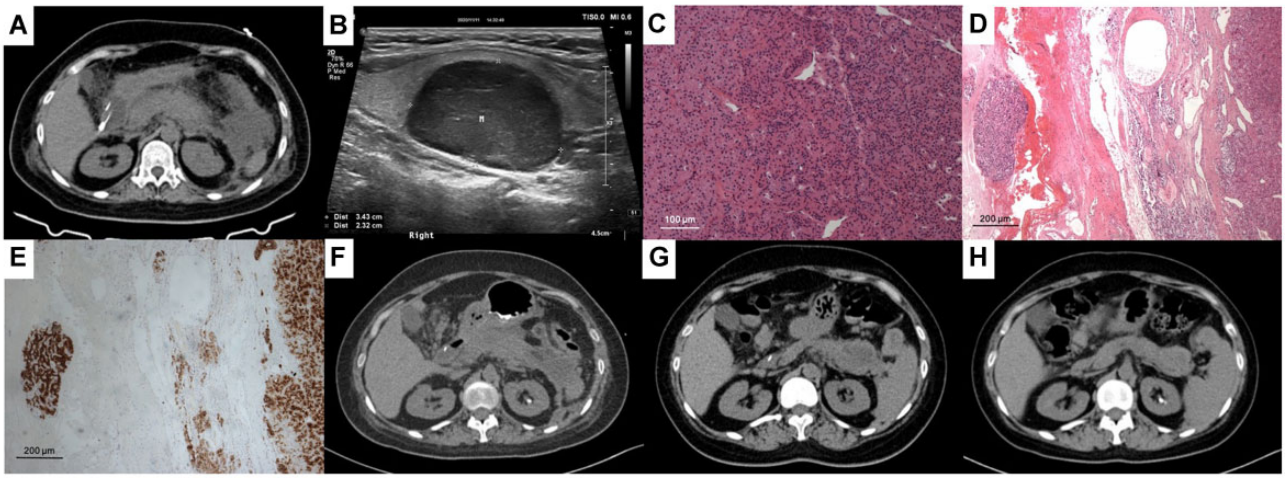

Figure 1. Imaging and pathological findings of the patient. (A) On Day 7 following admission, CT showed diffuse pancreatic swelling and acute peripancreatic fluid collection. (B) A hypoechoic nodule with a size of 3.4 × 2.9 × 2.3 cm protruding into the dorsal side of the right thyroid lobe. (C)–(E) Pathological findings of parathyroid carcinoma. (C) Hematoxylin-eosin staining (×10): tumor cells within the parathyroid gland. (D) and (E) Hematoxylin-eosin staining and immunohistochemical staining of PTH (×4): tumor localized to the parathyroid gland with extension limited to soft tissue. (F)–(H) Abdominal computed tomography (CT) axial images of the patient's pancreas during the recovery period. (F) On Day 44 following admission, the percutaneous catheter drainage volume was 0 mL and the pancreatic condition was indistinguishable from inflammatory changes. (G) Three months after discharge, a pancreatic pseudocyst formed in the tail of the pancreas. (H) Six months after discharge, the exudate was almost fully absorbed.

clinical findings for diagnosis and evaluation. We hope the details of management in our case may improve the understanding in such conditions.

Supplementary Data

Supplementary data is available at *Gastroenterology Report* online.

Authors' Contributions

Q.W. and Y.X.Z. conceived and designed the report. L.Z. and X.W. collected the data. Y.X.Z. reviewed the literature and wrote the manuscript. Q.W. and A.M.Y. revised and edited the manuscript. All authors read and approved the final version for submission.

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Conflict of Interest

The authors declare that they have no conflict of interest.

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