

## Recurrent pancreatitis due to parathyroid adenoma in pregnancy

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### ABSTRACT

Hyperparathyroidism complicating pancreatitis is relatively rare in pregnancy, but it is associated with significant maternal and fetal morbidity. This case report describes a pregnant lady with recurrent pancreatitis. Etiologic workup for recurrent pancreatitis revealed hypercalcemia secondary to hyperparathyroidism. The management of hyperparathyroidism includes treatment of hypercalcemia and definitive management of hyperparathyroidism which is the surgical excision of the abnormal gland. However, the timing of the surgery in a pregnant woman needs to be individualized. A multidisciplinary team approach is mandatory for better outcomes.

**Key words:** *Hypercalcemia, Pancreatitis, Parathyroid adenoma, Pregnancy*

**D**iagnosis of primary hyperparathyroidism (PHPT) in pregnancy and its management can be challenging as symptoms may be non-specific, and evaluation may involve procedures with radiation and surgery that are associated with risks during pregnancy. Acute pancreatitis complicating pregnancy is rare (incidence varies – 1 in 1000–1 in 10 000 births) and PHPT as the cause of pancreatitis in pregnancy is even rarer; gall stone pancreatitis (70%) and hypertriglyceridemia being the most common causes of acute pancreatitis in pregnancy [1]. Less than 200 cases of PHPT in pregnancy have been reported in the English literature, and its prevalence in the general population is reported to be around 0.15% [2]. Women are 3 times more affected than men. About 25% of these women are diagnosed in childbearing age. Maternal and fetal complications in pregnancy with hyperparathyroidism are as high as 67% and 80%, respectively [2].

We present the case of a pregnant lady with recurrent pancreatitis and hypercalcemia due to parathyroid adenoma. We wish to report this case, as this is a rare condition complicating pregnancy with recurrent pancreatitis due to parathyroid adenoma-induced hypercalcemia and discuss management dilemmas as there are no clear guidelines regarding the management of hyperparathyroidism and hypercalcemia in pregnancy.

### CASE REPORT

A 27-year-old second gravida with the previous miscarriage was admitted at 26 weeks of gestation (GA) with 1-day history of severe, continuous, and throbbing pain in the left upper abdomen radiating to the back. Her pain was better on sitting up and

was aggravated by food intake and on lying down. She had no history of fever, rash, alcohol consumption, or drug intake. She had no history of gall stones, and there was no family history of pancreatic or pituitary tumors suggestive of multiple endocrine neoplasia (MEN).

On clinical examination, she had tachycardia with a pulse rate of 110/min, blood pressure was 100/60 mm, and her per abdomen examination revealed mild tenderness in the epigastrium. Other systems examination was unremarkable.

Laboratory evaluation revealed elevated serum amylase (369.5 U/L, normal 20–96) and serum lipase (917.4 U/L, normal 0–160). Blood sugars and serum triglycerides were normal. Ultrasound abdomen showed mild bulky pancreas and gallbladder sludge. On the basis of these findings, a diagnosis of acute pancreatitis was made (sequential organ failure assessment score of 0 and Acute Physiology and Chronic Health Evaluation II score of 2 suggests that it is not severe pancreatitis). Her serum calcium was elevated (13.5 mg/dL, normal 8.7–10.2, corrected value for albumin of 3 mg/dL being 14.3), and inorganic phosphorus of 2.9 mg/dL. She was kept nil by mouth for 1 day until pain improved was given intravenous (IV) fluids and analgesia. The fetal well-being scan was normal (growth appropriate for GA age and normal liquor).

She was readmitted at 28+3 weeks with pain abdomen and vomiting. Her serum amylase (248 U/L), serum lipase (366 U/L), and serum calcium (13mg/dL) were abnormal. In view of the persistent hypercalcemia despite fluid therapy, her serum parathyroid hormone (PTH) was measured and was found to be elevated (111 pg/ml). Her Vitamin D level was low (12 ng/ml, normal being 30–100). She was started on oral

Vitamin D 2000 units/day. Ultrasonogram of the neck revealed a lobulated hypoechoic lesion posteroinferior to the right lobe of thyroid suggestive of parathyroid adenoma. A fetal scan revealed an appropriately grown fetus with polyhydramnios (amniotic fluid index [AFI] 23). She was managed with hydration, analgesics, and antacids. Her pain subsided in 1 day.

The multidisciplinary team's decision was to manage her conservatively with a plan for surgical excision of the parathyroid adenoma in the postpartum period. She was advised to consume at least 4 l of fluids per day. She was readmitted at 30+1 weeks with pain abdomen and irritable uterus. The ultrasound abdomen was no different from the previous one. A fetal well-being scan showed average fetal growth and worsening polyhydramnios (AFI 31). Steroids were administered for fetal lung maturity.

Surgical removal was planned in consultation with the surgical oncologist. In view of presumed fetal compromise, she was taken up for an emergency cesarean section at 30+4 weeks (under regional anesthesia), and a 1400 g female baby was delivered (APGAR 7/7/8). The baby was admitted to neonatal intensive care unit (NICU) with respiratory distress and hypocalcemia which were corrected. The baby was discharged after 15 days of NICU stay.

The patient underwent a Tc99 sestamibi parathyroid scan on 1<sup>st</sup> post-operative day which confirmed the diagnosis of the right inferior parathyroid adenoma. The right parathyroidectomy was performed on the seventh post-lower segment cesarean section day under general anesthesia. Postoperatively, the patient developed hypocalcemia with tingling and numbness, which was corrected with IV calcium chloride. The wound healed well, and she was discharged on Vitamin D3 and calcium supplements.

## DISCUSSION

Our patient had recurrent pancreatitis secondary to hypercalcemia. Further, the evaluation of hypercalcemia leads to a diagnosis of hyperparathyroidism due to parathyroid adenoma. Causes of hypercalcemia apart from hyperparathyroidism are sarcoidosis and other granulomatous diseases, Vitamin D toxicity, multiple myeloma, paraneoplastic syndrome, immobilization, etc., where the PTH levels are usually suppressed. Young patients with PHPT should be evaluated for MEN syndrome. The presence of pheochromocytoma, pituitary tumor, or thyroid cancer besides PHPT or family history of MEN syndrome should prompt us to evaluate for the presence of MEN syndrome [3].

Familial hypercalciuric hypocalcemia is also characterized by mild hypercalcemia and normal to mildly elevated PTH levels. Urinary quantification of urinary calcium was not done in our patient. She had concurrent Vitamin D deficiency which can cause hypocalciuria despite hyperparathyroidism which is the distinguishing feature between familial hypercalciuric hypercalcemia and PHPT. The presence of a parathyroid mass on ultrasound examination and the presence of severe hypercalcemia with recurrent pancreatitis were against the diagnosis of Familial hypercalciuric hypercalcemia [4].

Vitamin D deficiency is associated with compensatory elevation of PTH levels but has normal to low serum calcium levels. Our patient had Vitamin D deficiency, but she had hypercalcemia, high PTH levels, and parathyroid adenoma on ultrasound examination later confirmed by a Tc99 sestamibi parathyroid scan, hence the diagnosis of PHPT.

There is some published literature which states that primary hyperthyroidism tends to be overt in pregnant patients (four of every five patients) as compared to the non-pregnant women with this disorder, most frequent being nephrolithiasis [5,6]. The frequency of acute pancreatitis is higher in pregnant women with PHPT (7–13%) than non-pregnant patients with PHPT (1–2%) [5]. In a case report published by Krysiak *et al.*, acute pancreatitis was present in three of four pregnancies complicated by PHPT, while never occurred before and between pregnancies [5]. This case report supports the statistical data that pregnancy makes PHPT patients, particularly prone to the development of acute pancreatitis. Yang *et al.* reported a pregnant lady with severe necrotizing pancreatitis in the third trimester due to hypercalcemia caused by parathyroid adenoma [7]. She had parathyroid adenoma excision in the immediate postpartum period.

Tc99 sestamibi scan for localizing the adenoma during the antenatal period carries the risk of radiation exposure to the fetus and hence contraindicated in pregnancy. However, if the maternal condition warrants surgery in the antenatal period, the risk versus benefit of the radionuclide scan needs to be discussed with the patient. Dimarco *et al.* suggest avoiding ionizing radiation for localization of the tumor before surgery as the surgical intervention in a multidisciplinary set-up with high volume endocrine surgeries has excellent outcomes [8].

Management of hypercalcemia and hyperparathyroidism in pregnancy includes hydration, diuretics, bisphosphonates, and calcimimetics. Steroids are also used in sarcoidosis and lymphoma [9]. Definitive and curative management of parathyroid adenoma are surgical excision [2]. There are no clear guidelines regarding the timing of delivery as it is a rare disease complicating pregnancy. In patients with asymptomatic and non-severe hypercalcemia (serum calcium <14 mg/dL), surgical management of the parathyroid adenoma can be postponed until delivery.

In patients with symptomatic or severe hypercalcemia, surgical excision of the culprit tumor must be considered in the second trimester. During the third trimester, a decision regarding surgery needs to be individualized depending on the clinical status of the patient and GA age, where a fetus can be salvaged, which may vary from country to country. In the second trimester, surgical excision is preferred as the miscarriage rate is less, and in the third trimester, delivery is preferred as a sestamibi scan can be done for localization after the delivery. There have been case reports of pregnant women with hyperparathyroidism who had successful parathyroid surgeries in the third trimester without major complications [10].

A decision may be taken to deliver and then manage hyperparathyroidism when appropriate. In our patient, recurrent

pancreatitis and hyperparathyroidism both contributed to preterm labor, making delivery imminent at 30 weeks GA. Although the fetal morbidity is reported at 80%, hypocalcemia being the most common complication which was noted in our case too, the final outcome was good. Hypocalcemia after surgery is usually transient and easily correctable, as in our case, but there have been case reports of persistent hypocalcemia post-surgery [11].

## CONCLUSION

Investigation of the etiology of acute pancreatitis should include serum calcium levels. The presence of hypercalcemia as opposed to expected hypocalcemia should raise suspicion of hyperparathyroidism and needs evaluation accordingly, as the risk of preterm delivery and other complications is increased, and the management differs with this diagnosis. Recognition and appropriate treatment improve both maternal and fetal outcomes.

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