Hypokalemic periodic paralysis: an unusual presentation of primary hyperparathyroidism

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ABSTRACT

Primary hyperparathyroidism is an endocrine condition characterized by hyper secretion of parathyroid hormone (PTH). It has a wide varied clinical presentation from mild nonspecific symptoms to classical disease. We herein report two cases of primary hyperparathyroidism who were presented as hypokalemic periodic paralysis. The aim of this case report is to highlight this unusual presentation of primary hyperparathyroidism. Patients presenting with hypokalemic periodic paralysis should be evaluated for the possibility of primary hyperparathyroidism. Surgeons should be aware of this unusual presentation of primary hyperparathyroidism thereby it can be detected earlier, and prompt treatment can be offered before the disease progresses.

Keywords: Hypokalemia, Intrathyroidal parathyroid, Primary hyperparathyroidism, Periodic paralysis, Oncocytic adenoma

INTRODUCTION

Primary hyperparathyroidism (PHPT) is due to increased PTH production from abnormal parathyroid glands. The normal feedback mechanism exerted by serum calcium is deranged. The underlying pathology in the majority of cases is due to an adenoma (80% to 90%) followed by hyperplasia of all four glands (10% to 15%). Parathyroid adenoma is a benign encapsulated neoplasm which usually affects a single gland, but double adenomas are reported in 2% to 5% of patients with PHPT. Adenoma can also be found within the thyroid gland and the reported rates of intra thyroidal adenoma are 1.4 to 6% in the literature.1 PHPT is more commonly seen in middle aged persons between 50 to 60 years of age and more common in females than in males, with a ratio of approximately 3:1. Nowadays, patients with PHPT are often diagnosed on routine health screening or during evaluation for an unrelated medical problem. But patients may have non-specific symptoms like weakness, fatigue, bone and joint pain, polydipsia, polyuria, nocturia, decreased appetite, nausea, heartburn, constipation, pruritus, depression, and memory loss. Electrolyte abnormalities like hypomagnesemia, hypernatremia and hypokalemia are also reported.2 Our cases have been diagnosed to have primary hyperparathyroidism on evaluation for hypokalemic periodic paralysis.

CASE REPORT

Case 1

A 48-year-old female presented with acute onset of weakness of both upper and lower limbs which on initial evaluation found to be due to profound hypokalemia (serum potassium 2.5 mEq/l). The patient was treated as hypokalemic periodic paralysis in the nephrology department. The patient had a history of multiple joint pain, generalized myalgia, and low backache. There was no history of vomiting or diarrhea, oliguria or uraemic
symptoms. Family history was not contributory. Past history of ankle fracture present following a trivial fall. Physical examination revealed a 3X2 cm swelling anterior aspect of the neck above the medial end of the left side clavicle.

On further evaluation of the reason for hypokalemia, she was found to have serum calcium 15.4 mg/dl (normal values 9-11mg/dl), ionized calcium 2.39 (normal values 1.15-1.35), phosphate 2.4 mg/dl (normal values 2.7-4.2) and alkaline phosphatase 826 IU/ml (normal values 40-129). Other electrolytes and renal parameters were within normal range. Serum parathormone was markedly elevated to 932 pg/ml (normal values 15-72 pg/ml). Serum 25-hydroxyvitamin D was in deficient range 10.85 (>30 ng/ml in normal). Bone mineral density assessed by dual energy x-ray absorptiometry revealed osteoporosis. Single intramuscular injection of 600,000 IU of cholecalcciferol (arachitol) was given preoperatively. With the biochemical diagnosis of primary hyperparathyroidism, further imaging was done to localize the lesion.

Ultrasonogram of the neck showed 2.5X1.6 cm well defined heteroechoic lesion in the left paratracheal region below left lobe of thyroid. Computerized tomography of the neck showed a well defined heterogenous nodular lesion of size 4X2.2X2.3 cm in the left side of neck abutting the posteroinferior aspect of the left thyroid gland with few areas of necrosis seen.

Findings were consistent with left parathyroid adenoma (Figure 1A). Tc-99m sestamibi scan showed an area of tracer retention close to left lower pole of thyroid gland strongly suggestive of left inferior parathyroid adenoma (Figure 1B).

With these findings of concordant imaging, the patient proceeded with four-gland exploration. Intraoperatively left inferior parathyroid gland measured 3X3 cm and all other parathyroid glands were normal (Figure 1C). Left inferior parathyroidectomy was performed (Figure 1D).

Intraoperative parathormone (IOPTH) done 20 minutes after removal of the inferior parathyroid showed a value of 165 pg/ml which was more than 50% fall from the pre-incision value, confirming the removal of the abnormal gland. 24 hrs after surgery, she developed symptomatic hypocalcemia with serum calcium 8.5 mg/dl and iPTH of 13 pg/ml.

She was managed with intravenous calcium infusion, oral cholecalciferol, and calcitriol with frequent monitoring of serum calcium levels. Her symptoms improved and she was discharged on the 5th postoperative day and kept under regular follow up. Histopathology confirmed the diagnosis of parathyroid adenoma. Serum calcium, parathormone, 25-hydroxy vitamin D and potassium did two weeks after surgery was 8.9 mg/dl, 21pg/ml, 32 ng/ml and 3.8 mg/dl respectively.

Case 2

A 30-year-old male presented with acute onset weakness of both lower limbs and on initial evaluation found to have a serum potassium level of 2.5 mEq/l. The patient was treated in the medical ward as hypokalemic paralysis and on further evaluation for the cause of hypokalemia, revealed high serum calcium (12 mg/dl) and markedly elevated parathormone levels (1513pg/ml). Physical examination revealed 4X3 cm swelling anterior aspect of the neck towards the right of midline which was hard with no palpable cervical lymph nodes. Ultrasonogram of the neck showed a hypoechoic lesion 2X3 cm in the right lobe of thyroid. Tc-99m sestamibi scan showed increased tracer uptake near the right lower pole of thyroid gland consistent with right inferior parathyroid (Figure 2A). With these findings, the patient was diagnosed with right inferior parathyroid adenoma and a four-gland exploration was performed. Intraoperatively, 5X3X4 cm nodule involving the right lobe of thyroid was noted and the right inferior parathyroid was not made out separately (Figure 2B). Left inferior parathyroid was found to be enlarged. Right and left superior parathyroids were normal. Hence with a suspicion of intra thyroidal parathyroid within the right solitary thyroid nodule, a right hemithyroidectomy was performed along with a portion of left inferior parathyroid (Figure 2C). Both superior parathyroids were left in situ as they are found to be normal and there was a fall in IOPTH levels (210 pg/ml) Postoperatively patient had hypocalcemia which...
was managed with intravenous and oral calcium along with active vitamin D supplements. Post operatively, serum calcium and PTH levels were 8.6 mg/dl and 34 pg/ml respectively. Serum potassium level normalizes to 3.9 mg/dl and patient was symptomatically improved. Histopathology was consistent with intrathyroidal oncocyctic variant of parathyroid adenoma (Figure 2D).

![Image](image.png)

**Figure 1:** A: Sestamibi scan showing increased tracer uptake in right inferior parathyroid region, B: Intra operative image showing solitary nodule in the right lobe of thyroid, C: Right hemithyroidectomy specimen with a portion of left inferior parathyroid, D: Histopathology showing oncocyctic variant of parathyroid adenoma with adjacent normal thyroid follicular cells.

**DISCUSSION**

Primary hyperparathyroidism is a common endocrine disorder, with an estimated incidence of 20-70 per 1,00,000 persons/year and a prevalence of approximately 0.7% in the western world. Primary hyperparathyroidism occurs most commonly due to a single parathyroid adenoma. Other less common causes are multiple adenomas or hyperplasia.

Formerly patients with primary hyperparathyroidism (PHPT) presented with the "classic" pentad of symptoms (i.e., kidney stones, painful bones, abdominal groans, psychic moans, and fatigue overtones). Nowadays these classical symptoms are hardly seen because of early detection following widespread use of routine calcium screening in patients with minimal or atypical symptoms. Nonspecific complaints such as fatigue, lethargy, and depression are now most commonly reported. But in India, bone and renal diseases are still the commonest presentations which may be due to lack of awareness of routine calcium screening or due to co-existing Vitamin D deficiency.2

PHPT is associated with various electrolyte abnormalities. Other than hypercalcemia and hypophosphatemia, electrolyte disturbances such as hypokalemia, hypomagnesemia, and hypernatremia are also occasionally seen. There are various proposed mechanisms for hypokalemia in PHPT. It was postulated that excess calcium delivering into the distal tubule of nephrons drives the excess sodium ions into the distal tubule which in turn gets absorbed in exchange for potassium ion leading to hypokalemia. Another mechanism leading to hypokalemia is excess PTH secretion causing metabolic alkalosis.

A study by Aldinger and Samaan in 1977 reported an association between hyperparathyroidism and hypokalaemia at 16.9%.2 Also, the degree of hypokalemia correlates with higher calcium levels. Hypokalemia as such can cause various complications depending on its severity. There are anecdotal reports of PHPT with hypokalemia presenting as seizures, central pontine myelinolysis, and PHPT complicating pregnancy.3-7 Our cases presented as hypokalemic periodic paralysis which is an atypical presentation of PHPT.

The only definitive treatment of primary hyperparathyroidism is surgery. Treatment of single adenoma is excision of abnormal gland. Successful parathyroidectomy not only results in resolution of classical symptoms but also of nonspecific manifestations like fatigue, bone, joint pain and fibromyalgia. Our patients had a resolution of hypokalemia and muscle weakness following surgery.

Both patients had hypocalcemia in the postoperative period which might be due to the stunning effect of other parathyroids because of persistent hypercalcemia or due to the hungry bone syndrome. Temporary hypocalcemia following single adenoma excision has been reported to be 0 to 30% in the literature.8 This is managed with oral calcium and vitamin D supplementations for a shorter period of time and gradually withdrawn.

Intra thyroidal parathyroid adenomas presenting as primary hyperparathyroidism is very rare with a prevalence of 1.4 to 6%.1 Our second case is an oncocyctic variant of intrathyroidal parathyroid adenoma which is extremely rare and very few cases are reported in the literature.9,10,11

There is one anecdotal report of intrathyroid parathyroid adenoma presenting with neuromuscular manifestation.12 But to our knowledge, there are no reports of an oncocyctic variant of intrathyroidal parathyroid adenoma presenting with hypokalemic periodic paralysis in the literature till date and this is the first case to be reported with such an unusual presentation.
CONCLUSION

Primary hyperparathyroidism should be considered as a differential diagnosis when a patient presents with hypokalemia and paralysis. Serum calcium estimation should be done in initial evaluation of patients with hypokalemic periodic paralysis. Parathyroidectomy can reverse hypokalemia and offers the best cure with minimal morbidity. Hemithyroidectomy should be the surgical procedure of choice whenever an intrathyroidal parathyroid adenoma is suspected.

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