Case Report

Don't pin it all on a stereotype

L U Chirag[1], Pramila Kalra[2], Arivazhagan[3], Abhinith Shashidhar[4], Gyani Jail Singh[4], Jitender Saini[5]

From [1]Senior Resident, [2]HOD and Professor, Department of Endocrinology, Ramaiah Medical College, [3]Associate Professor, [4]Assistant Professor, Department of Neurosurgery, [5]Assistant Orofessor, Department of Neuroradiology, NIMHANS, Bengaluru, Karnataka, India

ABSTRACT

Hypercalcemia is one of the most frequently encountered problems in endocrinology OPD. Although the evaluation may not always be straightforward in all scenarios. Common factors affecting calcium levels such as dehydration, improper sample collection, and vitamin D supplementation may mask a serious underlying disorder. Here, we discuss a case of an elderly female who had symptoms of myelopathy and hypercalcemia whose etiology was initially attributed to excessive supplementation of calcium, vitamin D, and age-related bone changes; had another serious underlying calcium metabolism disorder.

Keywords: Hypercalcemia, Parathyroid adenoma, Vitamin D, Hyperparathyroidism, Renal tubular acidosis

Acute hypercalcemia is one of the most frequently encountered problems in endocrinology practice [1]. Although the evaluation may not always be straightforward in all scenarios. The factors affecting calcium levels such as dehydration, improper sample collection, and Vitamin D supplementation may mask a serious underlying disorder [2]. Parathyroid adenoma is part of a spectrum of parathyroid proliferative disorders that includes parathyroid hyperplasia, parathyroid adenoma, and parathyroid carcinoma [3].

Here, we discuss a case of an elderly female aged about 65 years who had symptoms of myelopathy and presented with acute hypercalcemia whose etiology was initially attributed to excessive supplementation of calcium, Vitamin D, and age-related bone changes; had another serious underlying calcium metabolism disorder.

CASE REPORT

A 65-year-old lady presented with complaints of mid backache for 11 years, radiation to both lower limbs for 6 years with progressive weakness of both lower limbs, and difficulty in walking for 2 years. She was on medication for hypertension for 8 years (tab telmisartan 40 mg OD and tab amlodipine 5 mg OD). She was also on calcium and Vitamin D3 supplements.

The patient was moderately built and nourished. Her pulse rate was 78/min regular in rhythm, respiratory rate was 24/min, blood pressure was 128/78 mm Hg, afebrile, and oxygen saturation was 97% on room air. Higher mental functions were normal. Cranial nerve functions were normal. Her motor examination revealed normal bulk and tone. Power in the lower limbs was affected both distally and proximally. The sensory examination was normal. Deep tendon reflexes were normal and plantar response was normal. She needed support while walking.

Her blood reports revealed creatinine 0.5 mg/dL, corrected calcium 12.4 mg/dL, alkaline phosphatase 1193 U/L, sodium 138, and potassium 3.0. The rest of the reports were within the normal range. Serum protein electrophoresis was normal and revealed no M band. Urine was negative for Bence Jones Proteins (Table 1). Her calcium and Vitamin D supplements were stopped. She was treated with IV fluids and her calcium normalized.

Her magnetic resonance imaging (MRI) showed disc osteophyte complexes at C4-5 causing spinal stenosis but no cord compression MRI spine showed partial collapse at the D9 vertebra with convex posterior margin, mild cord compression and bilateral neural foramina compromise. A probable diagnosis of a metastatic lesion causing D9 vertebra destruction was considered, though the long history was unusual. She was evaluated with a fluorodeoxyglucose-positron emission tomography scan which showed hypermetabolism over the D9 vertebral body and no focal uptake seen in soft tissue.

To correct the instability caused by the collapsed D9 vertebrae and to obtain a tissue diagnosis, she underwent D9 laminectomy and biopsy of the lesion with D8-D10 fixation using transpedicular screws and rods. Intraoperatively, the collapsed D9 vertebra was

Access this article online	
Received - 27 December 2022 Initial Review - 12 January 2022 Accepted - 07 February 2022	Quick Response code
DOI: 10.32677/ijcr.v9i2.3807	

Correspondence to: Dr. Pramila Kalra, Department of Endocrinology, Ramaiah Medical College, MS Ramaiah Nagar, Mathikere, Bengaluru, Karnataka, India. E-mail: kalrapramila@gmail.com

Figure 1: Sestambi scan (a) early phase; (b) delayed phase showing uptake in the right inferior parathyroid gland

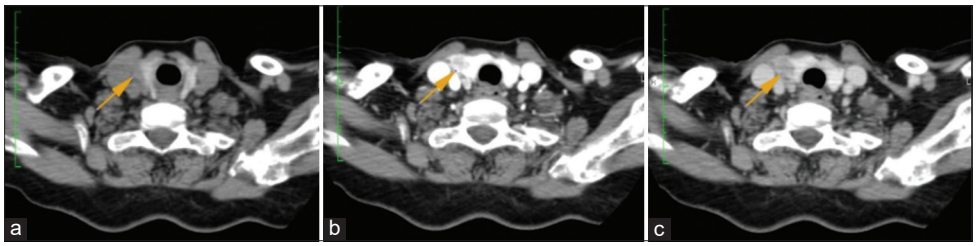

Figure 2: (a-c) 4D CT scan of neck showing lesion in the right inferior parathyroid gland in early intermediate and delayed phase

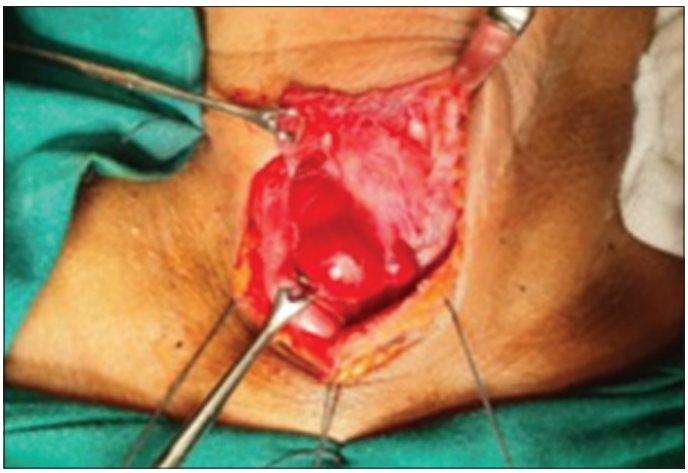

Figure 3: Intraoperative visualization of parathyroid mass

seen to be made of brittle and osteoporotic bone with no definite mass lesion. The tissue was sent for biopsy.

Postoperatively, the patient developed abdominal pain which was managed conservatively. Ultrasound whole abdomen showed non-obstructing renal calculi, bulky uterus and pancreas with a heterogeneous echo texture.

Her blood reports was repeated after 1 week which showed elevated calcium levels (13.45 mg/dL) with phosphorous 1.9 mg/dL, Vitamin D 14.42 ng/mL, and Lipase 219 U/L (Table 2). She was referred to endocrinology OPD for persistently elevated calcium levels. Her serum intraoperative parathyroid hormone (iPTH) level was 2598 pg/mL. She underwent a Tc-99 sestamibi (Fig. 1) scan which revealed an avid tissue to the lower pole of the right lobe of the thyroid gland – suggestive of right inferior parathyroid adenoma. 4D CT scan of the neck (Fig. 2) showed a 14 × 21 mm heterogeneously enhancing hypoattenuating lesion posterolateral to the right lobe of the thyroid gland predominantly enhancing in the arterial phase and showing washout in the delayed phase was noted. There was also diffuse sclerosis and thickening of the skull vault and ill-defined lytic lesions in the manubrium sternum, the lateral end of the clavicle.

She was posted for the right inferior parathyroidectomy (Fig. 3). Her pre-operative investigations were iPTH 2151 pg/mL, Vitamin D 3 27.27 ng/mL, corrected calcium

Table 1: Blood and urine analysis of the patient

Investigation	Patient value	Reference range
Hemoglobin (g/dL)	12.9	12–15
Total leucocyte count (cells/cumm^3)	7,800	4000–11,000
Platelet count (cells/cumm^3)	1,80,000	1,50,000–4,50,000
ESR (mm/h)	12	0–28
Serum creatinine (mg/dL)	0.8	0.5–1.1
Serum sodium (meq/L)	138	135–145
Serum potassium (meq/L)	3.0	3.5–5.1
SGOT (U/L)	18	8–48
SGPT (U/L)	22	7–55
HbA1c (%)	5.7	4–5.6
Fasting blood sugar (mg/dL)	102	70–102
Albumin (g/dL)	3.7	3.5–5.5
Alkaline phosphatase (IU/L)	1193	44–147
Lactate dehydrogenase (U/L)	121	120–280
Serum electrophoresis	No M Band	
Urine routine	Protein – Nil Sugar – Absent No Bence Jones proteins	

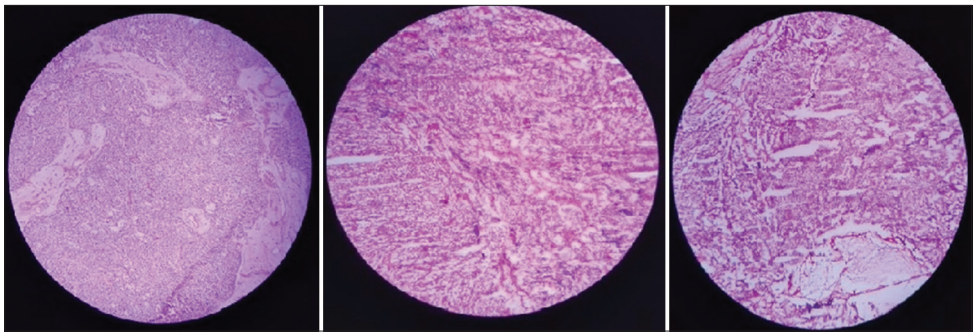

Figure 4: Histopathology of resected mass

Table 2: Key laboratory parameters preoperatively and postoperatively

Investigation	Pre-operative	Intraoperative	Post-operative
Corrected serum calcium	13.45 mg/dL 12.1 mg/dL		9.48 mg/dL
Serum phosphorous	2.8 mg/dL 2.1 mg/dL		3.4 mg/dL
iPTH	2598 pg/mL 2151 pg/mL	212 pg/mL	118 pg/mL
Vit D3	28.29 ng/dL		

12.9 mg/dL, phosphorous 2.1 mg/dL, and sodium 141 K 3.0. She was started on IV fluids, potassium supplementation and injection of calcitonin 50 IU 6th hourly to control calcium levels and optimize hydration. Her calcium level was controlled and was taken up for the procedure. Her intraoperative course was uneventful. The excised lesion showed a partly encapsulated lesion composed of sheets and trabeculae of polygonal cells exhibiting smooth contoured hyperchromatic nuclei and clear to acidophilic cytoplasm. Histopathology revealed parathyroid adenoma (Fig. 4). Her iPTH was 212.2 pg/mL.

Although the patient was asymptomatic on post-operative day 1, serial calcium monitoring revealed dropping calcium levels which were maintained on infusion at 0.5 mg/kg/h. Her infusion was stopped and she was shifted to oral calcium and active Vitamin D supplementation. Her calcium and phosphorous levels were monitored and remained stable on follow-up. She is currently on 2 g/day of elemental calcium and 0.5 mcg/day of active Vitamin D3.

DISCUSSION

Primary hyperparathyroidism (PHPT) is a common endocrine disorder of calcium metabolism characterized by hypercalcemia and elevated or inappropriately normal concentrations of PTH. It is due to a benign overgrowth of parathyroid tissue either as a single gland (80% of cases) or as a multiple gland disorder (15–20% of cases) [4]. The genetics of PHPT is usually monoclonal when a single gland is involved and polyclonal when the multiglandular disease is present [5].

Calcium and Vitamin D supplementation is thought to have general health and preventive benefits and is also recommended for the treatment of osteoporosis and Vitamin D deficiency [6]. However, it is becoming evident that liberal calcium intake in susceptible individuals may not be entirely benign, with consequences not unlike the classic milk-alkali syndrome of the past [7]. In a report by Asghar *et al*., a hypercalcemic crisis was precipitated by a Vitamin D supplementation and the underlying etiology was a cystic parathyroid adenoma mimicking carcinoma [8].

The index patient initially presented with chronic backache and was thought to have compressive myelopathy symptoms. Her hypercalcemia was attributed to calcium and Vitamin D supplements and was stopped. She was treated with intravenous hydration. When the patient was further evaluated post-surgery; her hypercalcemia persisted and led to an acute abdomen-like presentation.

PTH was found to be elevated and further investigation was undertaken to find out the etiology of the hypercalcemia. She also had low potassium levels which may be seen in long-standing hyperparathyroidism leading to distal renal tubular acidosis (RTA). In a case series by Muthukrishnan *et al*., they reported reversible distal RTA in their three patients which occurred postoperatively [9]. In concordance with their case series, our patient's potassium level normalized postoperatively.

Postoperatively, she developed low calcium levels which were managed with calcium infusion. The etiology of post-operative hypocalcemia is most commonly transient vascular insult, hungry bone syndrome, concomitant magnesium deficiency, and permanent hypoparathyroidism [10]. Considering our patient's high alkaline phosphate levels, it may indicate that hungry bone syndrome may have been the likely cause.

CONCLUSION

This case underlines the importance of detailed evaluation for causes of hypercalcemia in all age groups. Patients developing severe hypercalcemia after calcium and vitamin D in the background of osteoporosis may have other underlying causes and should be evaluated for the same. In long-standing hyperparathyroidism, there may be associated hypokalemia due to distal RTA. It is usually reversible after the correction of hyperparathyroidism.

Patient Perspective

I had backache for a long time and received calcium and Vitamin D. It never helped me as I was always feeling tired. I thought the

surgery would finally help relieve my pain but unfortunately, the pain increased. I am very thankful to the doctors who accurately diagnosed the root problem. I was educated in detail about the gland which I was not aware was a part of our body and it causes a lot of problems. After my second surgery, I started to feel more energetic. I understand that my disorder needs regular follow-up. I am happy that my case is helping doctors all over the world in helping to diagnose the condition more accurately. I do not have any objection to my case record being published.

REFERENCES

1. Bilezikian JP. Management of acute hypercalcemia. N Engl J Med 1992;326:1196-203.
2. Weiss-Guillet EM, Takala J, Jakob SM. Diagnosis and management of electrolyte emergencies. Best Pract Res Clin Endocrinol Metab 2003;17:623-51.
3. Wieneke JA, Smith A. Parathyroid adenoma. Head Neck Pathol 2008;2:305-8.
4. Bilezikian JP, Bandeira L, Khan A, Cusano NE. Hyperparathyroidism. Lancet 2018;391:168-78.
5. Bilezikian JP, Cusano NE, Khan AA, Liu JM, Marcocci C, Bandeira F. Primary hyperparathyroidism. Nat Rev Dis Primers 2016;2:16033.
6. Vieth R. Vitamin D toxicity, policy, and science. J Bone Miner Res 2007;22:V64-8.
7. Machado MC, Bruce-Mensah A, Whitmire M, Rizvi AA. Hypercalcemia associated with calcium supplement use: Prevalence and characteristics in hospitalized patients. J Clin Med 2015;4:414-24.
8. Asghar A, Ikram M, Islam N. A case report: Giant cystic parathyroid adenoma presenting with parathyroid crisis after Vitamin D replacement. BMC Endocr Disord 2012;12:14.
9. Muthukrishnan J, Kumar KH, Jha R, Jha S, Modi KD. Distal renal tubular acidosis due to primary hyperparathyroidism. Endocr Pract 2008;14:1133-6.
10. Mittendorf EA, Merlino JI, McHenry CR. Post-parathyroidectomy hypocalcemia: Incidence, risk factors, and management. Am Surg 2004;70:114-9; discussion 119-20.

Funding: Nil; Conflicts of interest: Nil.

How to cite this article: Chirag LU, Kalra P, Arivazhagan A, Shashidhar A, Singh GJ, Saini J. Don't pin it all on a stereotype. Indian J Case Reports. 2023; 9(2):34-37.