Yemen Journal of Medicine 2024;3(2):172–174

Content available at: https://www.ipinnovative.com/open-access-journals

Yemen Journal of Medicine

Journal homepage: https://www.yjomonline.com/

Letter to Editor

Hungry Bone Syndrome following parathyroidectomy: Is it a preventable event?

Saifat Ullah khan[1]*, Muftah Othman[1], Memon Noor Ilahi[1], Zishan Nasir[1], Syed Hidayat Ali[1]

[1]*Nephology Unit, Hazm Mebaireek Hospital, Qatar*

ARTICLE INFO

Article history:
Received 29-07-2024
Accepted 30-08-2024
Available online 06-09-2024

The parathyroid gland is an important regulator of calcium metabolism and any lesion of this gland such as (parathyroid adenoma, parathyroid hyperplasia, and parathyroid carcinoma) can lead to primary hyperparathyroidism,[1] casing a high secretion of parathyroid hormone which causes calcium, phosphorous, and bone metabolism disorders that could be symptomatic or asymptomatic[2] Parathyroid crisis is rare, accounting for 1-2 percent of patients with primary hyperparathyroidism (PHPT).[3] Parathyroid crisis is associated with severe hypercalcemia with serum calcium concentration usually above 15 mg/dL (3.8 mmol/L) and symptoms of hypercalcemia including anorexia, nausea, constipation, polydipsia, and particularly central nervous system dysfunction. Definitive therapy for parathyroid crisis is parathyroidectomy, however, around 14.8% of patients undergoing parathyroidectomy for PHPT postoperatively develop a condition known as the hungry bone syndrome (HBS).[4] We herein report a case of a young man with a parathyroid crisis, who developed HBS after parathyroidectomy.

On September 19, 2022, a 23-year-old male patient with no known medical history was brought to the emergency department of Hazm Mebaireek General Hospital in Doha, Qatar, by emergency medical services (EMS), because of a balance-related fall caused by dizziness. The patient reported that when he tried to get out of bed, he was unable to balance himself and fell. He was unable to get up from a fall or to walk due to severe left groin pain. The patient also reported a loss of appetite and weight loss of >10 kilograms, and recurrent constipation for the last month. He was admitted to a hospital in his home country, but the cause was unclear (No documents).

Physical examination upon admission revealed a very cachexic patient with stable vital signs. His weight was 41 kg (height 171cm). His left thigh was tender upon palpation and he was unable to move or put weight on his left leg. The rest of his examination was unremarkable. An urgent hip X-ray revealed a left femur neck fracture. Blood investigation on admission showed high serum creatinine of 150 mmol/l with very high serum calcium 20 mg/dl (5.0 mmol/l), parathyroid hormone level 3500 pg/ml, and alkaline phosphatase >4000 U/l. The electrocardiogram showed no hypercalcemic changes.

The patient was admitted to the orthopedic ward. He received immediate treatment for high calcium with intravenous calcitonin 4 units/kg twice daily and isotonic saline hydration. Serum calcium initially decreased to 14 mg/dl (3.5mmol/l), and the patient operated for left femur fixation. Thyroid ultrasound showed a hypoechoic mass,17 × 13 x10 mm of the right lobe of the thyroid. The parathyroid nuclear scan showed a 17 x 12 mm presumed parathyroid adenoma in the same location. The patient was referred for parathyroid adenoma resection

* *Corresponding author*.
E-mail address: khansaif301@gmail.com (S. U. khan).

https://doi.org/10.18231/j.yjom.2024.017

but unfortunately, he refused and was discharged against medical advice on calcimimetic (cinacalcet 90 mg once daily). Two weeks after discharge he was readmitted with complaints of recurrent vomiting generalized body weakness, loss of appetite, and cramps. Blood investigation showed serum calcium 18.84 mg/dl (4.7 mmol/l) and PTH 4000 pg/ml. He was treated once again with aggressive hydration and received one dose of denosumab 60 mg subcutaneously. This time he agreed for surgery and underwent parathyroidectomy. Immediately following the surgery, his PTH decreased from 4000 to 500 pg/ml. Within 6 hours of the surgery, his PTH was in the normal range at 87 pg /ml. Parathyroid adenoma was diagnosed by intraoperative frozen section pathological examination. On the next postoperative day, the patient developed hungry bone syndrome (HBS), manifested by numbness, paresthesia, and tetany with a calcium level of 7.6 mg/dL (1.9 mmol/L) and PTH of 40 pg/ml. In addition, ECG changes including QTc prolongation occurred. Treatment was initiated with intravenous calcium gluconate infusions, along with oral calcium and calcitriol supplementation. He recovered well and was discharged after a week with serum calcium 8.8 mg/dl (2.2 mmol/l) and PTH 15 pg/ml and on high doses of calcium carbonate and calcitriol.

Hyperparathyroid crisis, or "parathyroid crisis" is a rare manifestation of primary hyperparathyroidism, characterized by sudden onset of symptomatic, severe hypercalcemia > 15 mg/dL (3.8 mmol/L). It is more frequently encountered in persons over 40 years old than in younger patients.[4,5] In contrast, our patient is 23 years old, which coincide with few reports in the literature.[6,7] The management strategies for parathyroid crises have evolved over time with the advent of new hypocalcemic drugs. The initial and most important step in the management of hypercalcemia is to administer intravenous isotonic saline for extracellular volume expansion and promote hypercalciuria, with or without the usage of loop diuretics to prevent volume overload.[8] Definitive therapy for parathyroid crisis is parathyroidectomy, however, around 14.8% of patients undergoing parathyroidectomy for PHPT postoperatively develop a condition known as the hungry bone syndrome (HBS).[4] Moreover, the exact timing for parathyroidectomy is not well-established.[8]

Although HBS does not have a consensus definition, it is described as a prolonged hypocalcemia after parathyroidectomy for hyperparathyroidism, followed by normal or elevated PTH levels.[4] It usually develops after parathyroidectomy for either primary or secondary hyperparathyroidism. Severe hypocalcemia results from the sudden drop of PTH secretion in the setting of a pre-existing severe bone resorption due to prolonged high levels of PTH. Risk factors for developing HBS include volume of parathyroid removed, elevated preoperative blood urea nitrogen (BUN), and/or older age.[4,8,9] HBS typically develops within 18 hours of parathyroid or thyroid surgery and may remain decreased for up to 3 months.[9] Careful monitoring of serum calcium in the immediate post-operative period (two to four times on the day of operation) is the key to the diagnosis. In our case, the patient is young and he developed HBS 24 hours after parathyroidectomy.

Treatment of HBS consists of calcium, active forms of vitamin D, and magnesium to prevent hypocalcemia and facilitate bone remineralization.[10] However, the treatment of HBS must be individually tailored to each patient in terms of dose and frequency and may take a longer period of time. Unfortunately, there is insufficient data-based evidence to prevent HBS or minimize severe complications of hypocalcemia before parathyroidectomy.[4]

In conclusion, HBS is a recognized complication of parathyroidectomy; if not recognized and corrected in time, it can cause significant morbidity linked to the consequences of hypocalcemia. Hence, our case illustrates the need for careful perioperative management and highlights the importance of anticipating HBS before surgical intervention. Literature review indicated that no evidence for possible preoperative prevention options.

Consent

Written informed consent was obtained from the patient for publication of this case report.

Authors' Contribution

All authors contributed to the completion of this work. The final manuscript was read and approved by all authors.

Source of Funding

None.

Conflict of Interest

None.

References

1. Townsend CM, Beauchamp RD, Evers BM. Sabiston textbook of surgery. Philadelphia: Elsevier; 2012. p. 933.
2. Costanzo LS. BRS physiology. 4th ed. Philadelphia: Lippincott Williams Wilkins; 2011. p. 247–8.
3. Corlew DS, Bryda SL, Bradley E, Digirolamo M. Observations on the course of untreated primary hyperparathyroidism. *Surgery*. 1985;98(6):1064.
4. Jakubauskas M, Beiša V, Strupas K. Risk factors of developing the hungry bone syndrome after parathyroidectomy for primary hyperparathyroidism. *Acta Med Litu*. 2018;25(1):45–51.
5. Ameerudden S, He X. Management and surgical treatment of parathyroid crisis secondary to parathyroid tumors: report of four cases. *Int Med Case Rep J*. 2011;4:59–66.
6. Mitchell D, Rybak LP, Glatz FR. Hyperparathyroid crisis in a pediatric patient. *Int J Pediatr Otorhinol*. 2004;68:237–41.
7. Wong P, Carmeci C, Jeffrey RB. Parathyroid crisis in a 20 year olddan unusual cause of hypercalcaemic crisis. *Postgrad Med J*. 2001;77(909):468–70.

8. Adeniran U, Hussein JB, Soni I. Parathyroid Crisis: A Case of Elective Parathyroidectomy. *Cureus*. 2023;15:40251.
9. Hungry Bone Syndrome. Available from: https://my.clevelandclinic.org/health/diseases/24954-hungry-bone-syndrome.
10. Witteveen JE, Thiel SV, Romijn JA, Hamdy N. Hungry bone syndrome: still a challenge in the post-operative management of primary hyperparathyroidism: a systematic review of the literature. *Eur J Endocrinol*. 2013;168(3):45–53.

Author biography

Saifat Ullah khan, Consultant https://orcid.org/orcid.org. 0000-0001-7531-0837

Muftah Othman, Consultant

Memon Noor Ilahi, Consultant

Zishan Nasir, Senior Consultant

Syed Hidayat Ali, Specialist

Cite this article: khan SU, Othman M, Ilahi MN, Nasir Z, Ali SH. Hungry Bone Syndrome following parathyroidectomy: Is it a preventable event?. *Yemen J Med* 2024;3(2):172-174.