

Postoperative Hungry Bone Syndrome Following VATS Resection of Ectopic Mediastinal Parathyroid Adenoma: A Case Report

Dr Khaldoon Abo Dakka¹, Dr Anil Kumar Pura Narayanaswamy², Dr Syed Zafar³, Dr Omar Fawzi Lootah⁴

¹ Specialist Thoracic Surgeon, Prime Hospital, Dubai, UAE

² Consultant Endocrinologist, Prime Hospital, Dubai, UAE

³ Medical Intern, Prime Hospital, Dubai, UAE

⁴ Medical Student, Year 5, University of Sharjah, Sharjah, UAE

Abstract:

Hungry bone syndrome (HBS) is characterized by persistent hypocalcemia and hypophosphatemia as a result of extensive remineralization following parathyroidectomy. It is a known but uncommon complication of parathyroid surgery, seen in 13–30% of cases. In this article, we report the case of a 36-year-old woman with increased thirst and polyuria as the first manifestation of primary hyperparathyroidism caused by an ectopic parathyroid adenoma located in the anterior mediastinum. After a single-port Thoracoscopic parathyroidectomy, the patient presented clinical signs of hypocalcemia consistent with hungry bone syndrome. This report highlights the diagnostic approach, surgical management, and postoperative care, emphasizing the importance of early recognition and intervention.

Key Words:

Hungry bone syndrome, ectopic parathyroid adenoma, primary hyperparathyroidism, VATS

Introduction:

Parathyroid adenoma remains the leading cause of primary hyperparathyroidism; however, in rare cases, these adenomas may be ectopically located in the mediastinum [1,2], as illustrated in this case. Surgical resection is the definitive management for such patients. While postoperative hypocalcemia is frequently observed following adenoma excision, the occurrence of hungry bone syndrome (HBS)—characterized by sustained hypocalcemia and hypophosphatemia due to marked skeletal remineralization—is relatively uncommon [3]. Here, we describe a 29-year-old female with a mediastinal ectopic parathyroid adenoma who developed HBS in the postoperative phase.

Case Report:

A 36-year-old lady was seen for her complaints of progressive worsening weakness, generalized body pain, increased thirst, and polyuria. Physical examination was unremarkable. She had gained 10 kg, unintentionally over two years without other systemic symptoms. Palpitations, sweating, headache, and diaphoresis were not present. The patient's and her family's past medical history was unremarkable. There was no personal or family history of endocrine disorders or multiple endocrine neoplasia (MEN).

Laboratory investigations revealed: Serum calcium: 12 mg/dL (normal: 8.2–10.6), Phosphorus: 2 mg/dL (normal: 2.5–4.5), PTH: 623 pg/mL, Magnesium: 2.3 mg/dL, Vitamin D: very low 6 ng/ml, Alkaline phosphatase: elevated, Other labs including urinalysis, renal and liver function were within normal limits.

Neck ultrasound of thyroid and parathyroid glands was within normal limits, while neck CT showed a well-defined ovoid avidly enhancing soft tissue density (56 HU) focus measuring 17 x 8 mm noted in the anterior mediastinum abutting the arch of aorta (Figure 1). A technetium-99m scan (Figure 2) and thoracic CT (Figure 3) suggested a focal metabolically active (approximately 0.7*1.0 cms in size) ectopic parathyroid adenoma seen in the left paraaortic region. The rest of the parathyroid glands were observed in the normal location.

Figure 1



Figure 2

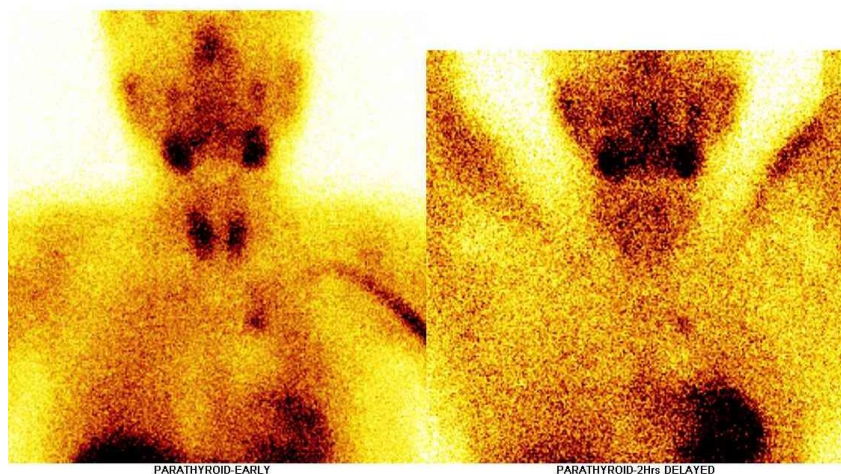
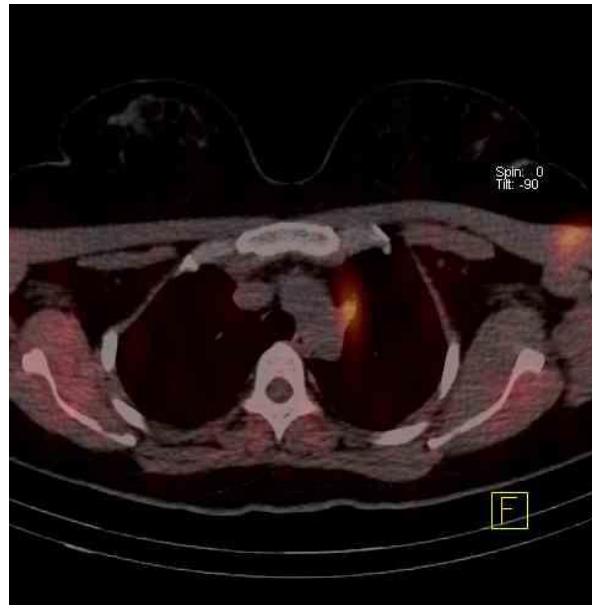
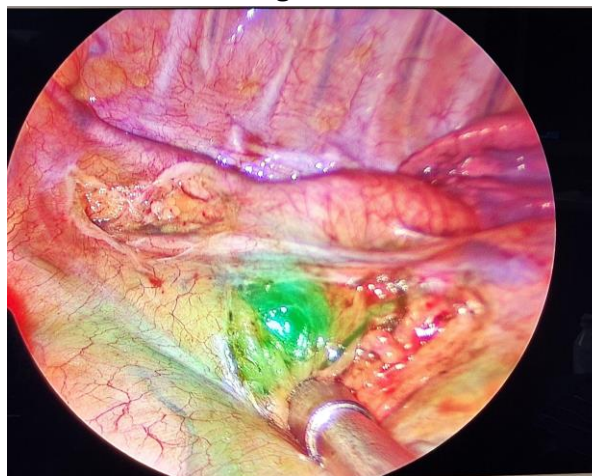


Figure 3



Treatment was initiated for hydration with saline infusion and correction of vitamin D deficiency. Three months later, after her serum calcium concentration decreased to 8.6 mg/dl, she underwent thoracoscopic surgery using single port - VATS, the mediastinal pleura was opened over the aortic arch between the phrenic nerve anteriorly and the vagus nerve posteriorly. 5 mg indocyanine green (ICG) was administered intravenously. A 1.5 cm solitary ectopic mediastinal parathyroid adenoma tumor was then localized and distinguished from the surrounding thymic and fatty tissues using a near-infrared indocyanine green (NIR-ICG) fluorescence thoracoscopic system (Figure 4). Complete dissection of the adenoma tumor, alongside the surrounding mediastinal fat tissue, was performed with preservation of the surrounding structures. The surgical duration was minimal, as clear identification of the ectopic parathyroid adenoma eliminated the necessity for a frozen section analysis. A significant reduction in serum parathyroid hormone (PTH) levels, confirmed by a sample taken 20 minutes post-excision (with more than a 50% drop), further validated the successful removal of the adenoma.

Figure 4.

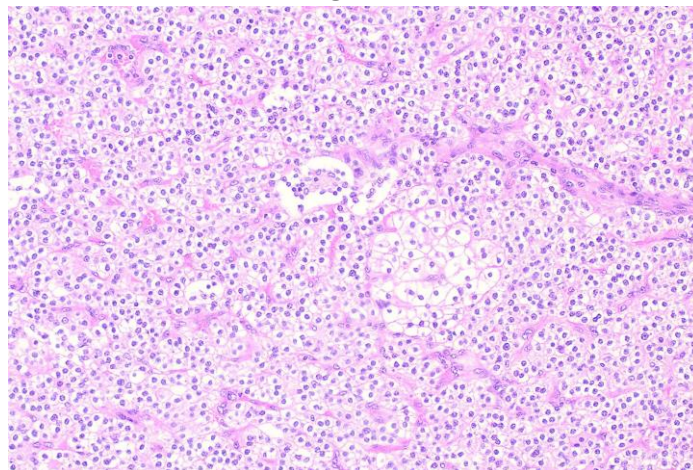


The lesion demonstrated a well-circumscribed, encapsulated ectopic parathyroid adenoma following surgical excision. It displayed a prominent green fluorescence due to the administration of indocyanine green (ICG) dye, enhancing intraoperative localization (Figure 5). Microscopically, the histology results revealed it to be a tumor composed of uniform, polygonal chief cells and clear cells, and the lesion was diagnosed as ectopic parathyroid adenoma (Figure 6).

Figure 5



Figure 6



The patient had an uneventful post-operative recovery and got discharged on day 2 on a high-dose oral calcium with vitamin D. Two days post discharge, the patient returned with paresthesia and muscle cramps. Clinical signs of hypocalcemia (Chvostek's and Trousseau's signs) were present. Labs showed: Serum calcium concentration decreased to 7.96 mg/dl; Alkaline Phosphate levels were elevated 246 IU/L, serum phosphorus 2.5 mg/dl, serum magnesium 1.47, PTH was normalized. She was diagnosed with HBS and treated with intravenous calcium and magnesium for six days. She was discharged on oral calcium

(400 mg TID) and calcitriol (1 µg BID). Calcium supplementation was tapered over follow-up as serum calcium levels normalized.

Discussion:

Primary hyperparathyroidism is most commonly attributed to a single parathyroid adenoma, which accounts for approximately 80% of cases. In about 15–20% of cases, hyperplasia affecting all four glands is observed, whereas parathyroid carcinoma is an exceedingly rare cause, responsible for less than 0.5% of cases [4]. Between 10–20% of parathyroid adenomas are ectopically located in the mediastinum. The embryological origin of the lower parathyroid glands from the third pharyngeal pouch, which also gives rise to the thymus, explains their frequent presence in the anterior superior mediastinum. These ectopic glands can also be found in other neck regions and throughout the mediastinum [5,6]. According to Richards et al., 5–10% of parathyroid glands may reside in the posterior mediastinum, with 20% found within the thymic tissue in the anterior mediastinum, 1% in the carotid sheath, and 5% embedded in the thyroid gland [7–9].

In our patient, both clinical presentation and laboratory results were indicative of primary hyperparathyroidism, leading to further evaluation with technetium-99m scintigraphy and thoracic CT. Parathyroid scintigraphy is a dependable diagnostic tool for detecting abnormal parathyroid glands in cases of primary hyperparathyroidism. It plays a crucial role in reducing surgical risks by facilitating precise localization of ectopic adenomas, decreasing operative time, and aiding in the identification of residual glands postoperatively [6,7]. In this case, imaging confirmed the presence of an ectopic parathyroid adenoma situated within the thymic tissue of the anterior mediastinum.

The majority of primary hyperparathyroidism cases are sporadic due to isolated adenomas. Nonetheless, inherited conditions like autosomal dominant hyperparathyroidism and syndromes such as MEN types I and IIA may also present with solitary adenomas. These syndromes typically become evident between the ages of 20 and 40 and are rarely diagnosed in childhood [10,11]. In our case, no familial predisposition or signs of MEN syndrome were identified upon thorough clinical evaluation and family history assessment.

One frequent postoperative complication of parathyroid surgery is hypocalcemia, which can occur due to several mechanisms: the surgical removal of parathyroid tissue, temporary ischemia of residual glands, or prolonged suppression of normal parathyroid function by long-standing hypercalcemia. Occurring in 13–30% of surgeries, significant skeletal remineralization results in sustained hypocalcemia and hypophosphatemia—characteristic of hungry bone syndrome (HBS) [12].

A transient increase in the number and intensity of lesions on bone scans observed after parathyroidectomy, known as the flare phenomenon, constitutes a healing response due to a rapid decrease in bone resorption as well as an increase in bone formation with a high uptake of calcium. Thus, it manifests radiographically as an improvement in bone mass [13].

Management requires aggressive and extended supplementation with calcium and vitamin D [12–14]. Factors increasing the risk for HBS include large adenoma size (exceeding 5 cm), elevated preoperative levels of parathyroid hormone, serum calcium, and ALP, the presence of osteitis fibrosa cystica, and older age [13,14]. Our patient had most of these risk factors.

Conclusion:

This case highlights the importance of recognizing atypical presentations of primary hyperparathyroidism and the potential for developing hungry bone syndrome following parathyroidectomy, particularly in cases involving ectopic mediastinal parathyroid adenomas. Early identification of risk factors and close postoperative monitoring are essential to prevent and manage this complication effectively.

A better understanding of the underlying mechanisms and predictive markers of HBS may improve patient outcomes and guide perioperative care.

References:

1. Loh KC, Duh QY, Shoback D, Gee L, Siperstein A, Clark OH (1998) Clinical profile of primary hyperparathyroidism in adolescents and young adults. *Clin Endocrinol (Oxf)* 48:435–443
2. Clark OH (1988) Mediastinal parathyroid tumors. *Arch Surg* 123:1096–1100
3. Kuzucu A, Soysal O, Savli H (2002) Giant mediastinal parathyroid adenoma presenting with a hyperparathyroid crisis and leading to postoperative hungry bone syndrome. *Eur J Surg* 168:747–749
4. Russell CF, Edis AJ, Scholz DA, Sheedy PF, van Heerden JA (1981) Mediastinal parathyroid tumors: experience with 38 tumors requiring mediastinotomy for removal. *Ann Surg* 193:805–809
5. Wynne AG, van Heerden J, Carney JA, Fitzpatrick LA (1992) Parathyroid carcinoma: clinical and pathologic features in 43 patients. *Med (Baltim)* 71:197–205
6. Foroulis CN, Rousogiannis S, Lioupis C, Koutarellos D, Kassi G, Lioupis A (2004) Ectopic paraesophageal mediastinal parathyroid adenoma, a rare cause of acute pancreatitis. *World J Surg Oncol* 2:41
7. Richards ML, Bondeson AG, Thomson NW (2000) Mediastinal parathyroid adenomas and carcinomas. In: Shields TWIII, Lo Cicero J, Ponn RB (eds) *General thoracic surgery*. Lippincott Williams & Wilkins, Philadelphia, pp 2383–2390
8. Bienvenu M, Amar L, Vignaux O, Fulla Y, Bonnichon P, Richard B, Bertagna X, Legmann P (2003) Diagnosis of ectopic mediastinal parathyroid adenomas: value of cardiac MRI. *J Radiol* 84:1969–1973
9. Akimoto T, Saito O, Muto S, Hasegawa T, Nokubi M, Numata A, Ando Y, Sohara Y, Saito K, Kusano E (2005) A case of thoracic hemorrhage due to ectopic parathyroid hyperplasia with chronic renal failure. *Am J Kidney Dis* 45:109–114
10. Levine MA, Zapalowski C, Kappy MS (2005) Disorders of calcium, phosphate, parathyroid hormone, and vitamin D. In: Kappy MS, Allen AB, Geffner ME (eds) *Principles and practice of pediatric endocrinology*. Thomas, Springfield, pp 695–814
11. Nishimura Y, Yamashita K, Yumita W, Yamazaki M, Katai M, Sakurai A, Hashizume K (2004) Multiple endocrine neoplasia type 1 with unusual concomitance of various neoplastic disorders. *Endocr J* 51:75–81
12. Kale N, Basaklar AC, Sonmez K, Uluoglu O, Demirsoy S (1992) Hungry bone syndrome in a child following parathyroid surgery. *J Pediatr Surg* 27:1502–1503
13. Bhattacharyya A, Buckler HM, New JP (2002) Hungry bone syndrome. *J R Coll Phys Edinb* 32:83–86
14. Brasier AR, Nussbaum SR (1988) Hungry bone syndrome: clinical and biochemical predictors of its occurrence after parathyroid surgery. *Am J Med* 84:654–660