

Mediastinal ectopic parathyroid adenoma. A case report

Adenoma paratiroideo ectópico mediastinal. A propósito de un caso

***Marco Antonio Jiménez-Rodríguez**¹

****Jorge Aurelio Gutiérrez-González**¹

*****Marco Antonio Treviño-Lozano**¹

#**Luis José Bellorín-Figueroa**¹

##**Gerardo Enrique Muñoz-Maldonado**¹

¹Universidad Autónoma de Nuevo León, Servicio de Cirugía General del Hospital Universitario "Dr. José Eleuterio González", Monterrey, Nuevo León, México.

ABSTRACT

Primary hyperparathyroidism is caused by increased secretion of parathyroid hormone and was first described in 1930. Eighty percent are caused by a benign parathyroid adenoma, about 16% may present as ectopic. A 39-year-old female with multiple urinary tract infections, urolithiasis, hypercalcemia and increased parathyroid hormone. Gamma-gram showed ectopic parathyroid adenoma in superior mediastinum, by transverse cervicotomy, median sternotomy and gamma probe, the lesion was resected without eventualities.

Keywords: parathyroidectomy, hyperparathyroidism, ectopic parathyroid, mediastinal parathyroid.

RESUMEN

El hiperparatiroidismo primario es causado por el aumento de la secreción de paratohormona, y se describió por primera vez en 1930. 80% de los casos son causados por un adenoma paratiroideo benigno y hasta el 16% pueden presentarse como ectópico. Femenina de 39 años, con múltiples infecciones urinarias, urolitiasis, hipercalcemia y paratohormona aumentado. Gammagrama demostró adenoma paratiroideo ectópico en mediastino superior, mediante cervicotomía transversa, esternotomía media y gamma sonda, se resecó lesión sin eventualidades.

Palabras clave: paratiroidectomía, hiperparatiroidismo, paratiroides ectópica, paratiroides mediastinal.

INTRODUCTION

Primary hyperparathyroidism (PHPT) is due to an increased secretion of parathyroid hormone (PTH). It is the most common cause of hypercalcemia, above all, during postmenopause. It was described for the first time back in 1930⁽¹⁾.

Both its rate and prevalence have increased over the past decades thanks to the greater detection of mild hypercalcemia with the use of more biochemical tests with serum calcium that has increased the number of diagnoses of asymptomatic cases of PHPT. Its significance lies in its progression: up to a fourth of all patients ends up developing symptoms within the next 5 years^(1,2).

A total of 80% of all PHPTs are benign parathyroid adenomas, 15% to 20% are parathyroid hyperplasias, and less than 1% parathyroid carcinomas⁽¹⁾.

Ectopic parathyroid glands (EPG), described in up to 16% of the cases, are due to the anomalous migration of these during development. They can be found in tissues that share their embryologic origin⁽³⁾. Imaging modalities are important for the diagnosis, location, and surgical planning of this entity. Parathyroidectomy (PT) is the ultimate treatment here^(3,4).

This is the case report of a female patient with PHPT due to ectopic parathyroid adenoma.

* Fifth-year Surgical Resident.

** Third-year Surgical Resident.

*** Oncological surgeon, Professor at the General Surgery unit

Thoracic surgeon, Professor at the General Surgery unit

Professor and Head of General Surgery Unit

Corresponding author: Dr. Gerardo Enrique Muñoz Maldonado

E-mail address: cevam99@gmail.com - Address: Avenida Francisco I. Madero y Gonzalitos S/N, Colonia Mitras Centro, C.P. 64460 Monterrey, Nuevo León, Mexico.

Date of reception: 28/06/2022 - Date of approval: 19/09/2022

 This is an open access article published under Creative Commons License.



Figure 1. Computed tomography scan. Panel A: Sagittal view showing ectopic parathyroid adenoma 2 cm away from the sternal notch. Panel B: Axial view showing an oval-shaped nodular image with demarcated borders.

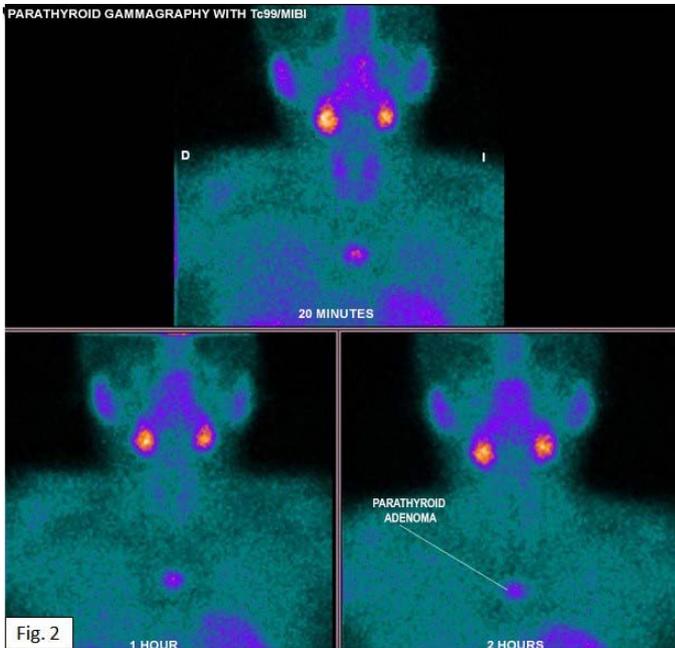


Figure 2. Images acquired through parathyroid gammagraphy with ^{99m}Tc -Sestamib showing the ectopic parathyroid adenoma at the superior mediastinum central region.

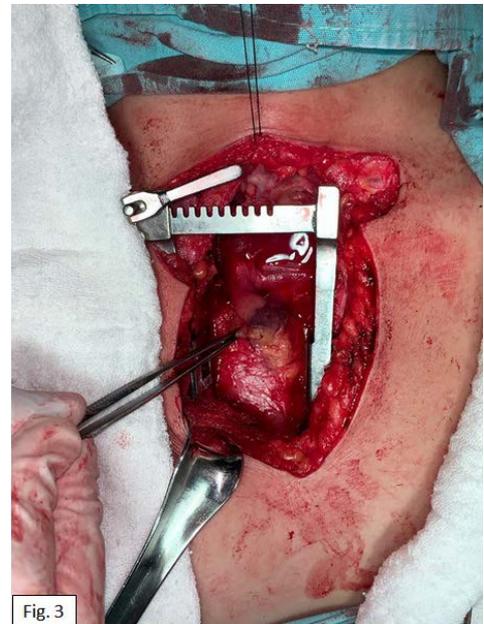


Figure 3. Intraoperative image of ectopic parathyroid adenoma at superior mediastinal level.



Figure 4. Resection of ectopic parathyroid adenoma measuring 2 cm x 1.6 cm x 0.8 cm and weighting 1.7 g.

CLINICAL SIGNS

This is the case of a 39-year-old woman with multiple episodes of urinary tract infections, renal colics, and nephrolithiasis. She presents with weakness, chronic fatigue, and hypercalcemia (10.9 mg/dL). PTH is analyzed and since elevated levels are found (138.3 pg/mL) PHPT is finally diagnosed.

Management of the patient included a computed tomography scan (see Figure 1) and a gammagraphy with Tc99-sestamibi (Figure 2) that revealed the presence of ectopic parathyroid adenoma of superior mediastinum location. Transverse cervicotomy access was used and medium sternotomy with pneumatic saw was performed under balanced general anesthesia. The radio-and-gamma probe guided minimally invasive parathyroidectomy performed revealed the presence of parathyroid adenoma at mediastinum level (Figure 3) that was resected uneventfully (Figure 4). No events were reported during the post-operative period resulting in lower levels of calcium and PTH.

DISCUSSION

PHPT is more common in women on a 4:1 ratio compared to men. The highest incidence rate has been reported within the first few years after menopause, which is consistent with estrogen loss. Up to 20% of the patients have a past medical history of renal colics⁽¹⁾. Our case is consistent with the cases reported in the medical literature available today.

There are 3 different presentations of PHPT: symptomatic or classic (elevated calcium and PTH levels), asymptomatic or normohormonal (elevated calcium and normal PTH levels) and normocalcemic (normal calcium and elevated PTH levels). The latter can be a precursor of traditional PHPT^(1,2). Our patient had classic signs.

The risk factors reported are low calcium intake, low physical activity, large abdominal circumference, obesity, hypertension, neck radiation or nuclear exposure and prolonged lithium therapy⁽¹⁾.

Up to 16% of the patients show EPG. Three different studies concluded that the most common regions are the thymus, the retro/paraesophageal space, and thyroid, which represent 56% to 89% combined. Between 6% and 26% have been described in the mediastinum^(5,6).

Signs and symptoms are due to hypercalcemia, among them, fatigue, weakness, polyuria, polydipsia, constipation, anorexia, vomiting, dehydration, arrhythmias, anxiety, altered mental state, and mood swings. Our patient also complained of fatigue, and chronic weakness⁽¹⁾. Frailty-induced fractures, skeletal deformities, and bone pain are also possible. Also, presentation through hypercalciuria, nephrolithiasis, nephrocalcinosis, and reduced renal function⁽¹⁾, as it happened in our case and its pre-

sentation of classical phenotype.

Diagnosis is achieved after detecting elevated hypercalcemia. Also, elevated PTH levels with ≥ 20 pg/mL in the context of hypercalcemia is consistent with the diagnosis^(1,7).

Preoperative imaging modalities facilitate surgery by accurately finding the adenoma like ultrasound, gammagraphy with 99m-technecium-sestamibi, computed tomography scan, magnetic resonance imaging, and PET⁽⁸⁾. Gammagraphy has higher sensitivity (89%) and ultrasound has less sensitivity (59% to 76%) although the latter is more widely available. Gammagraphy is considered the gold standard^(3,8).

Surgery is clearly indicated in patients with symptoms due to hypercalcemia or target organ failure (frailty-induced fractures or colics)^(1,7). Definitive treatment of choice is parathyroidectomy⁽⁸⁾. Cervical approach is often enough. However, 2% of the cases of mediastinal location require transsternal or transthoracic approach⁽³⁾.

The rates of healing for an experienced surgeon exceed 95% with a very low rate of complications (< 1% to 3%) like hemorrhages, hypocalcemia, and recurring laryngeal nerve injuries^(1,2). Once parathyroidectomy has been performed, biochemical markers often go back to normal, and bone density goes up, thus reducing the risk of fracture and nephrolithiasis⁽⁸⁾.

Authors' contributions: MAJR and JAGG were involved in surgical treatment, bibliographic search, and drafting of the manuscript. MATL and LJBF were involved in surgical treatment, counselling while drafting the manuscript, and proofreading tasks. GEMM was involved in the process of drafting a nd proofreading the manuscript. The authors declared no conflicts of interest whatsoever.

Conflicts of interest: none reported.

Financing: the authors declare that they have not received any type of funding for the realization of this article.

REFERENCES

1. Silva BC, Cusano NE, Bilezikian JP. Primary hyperparathyroidism. *Best Pract Res Clin Endocrinol Metab.* 2018 Oct 1;32(5):593–607.
2. Zhu CY, Sturgeon C, Yeh MW. Diagnosis and Management of Primary Hyperparathyroidism. *JAMA [Internet].* 2020 Mar 24 [cited 2022 May 17];323(12):1186–7. Available from: <https://pubmed.ncbi.nlm.nih.gov/32031566/>
3. Boccalatte LA, Gómez NL, Olivera López S, Abuawad CY. Hiperparatiroidismo ectópico: Detección de la localización mediastinal. *Med (B Aires) [Internet].* 2020 May 20 [cited 2022 May 20];80(1):39–47. Available from: http://www.scielo.org.ar/scielo.php?script=sci_arttext&id=S0025-76802020000100006
4. Udelsman R, Lin Z, Donovan P. The superiority of minimally invasive parathyroidectomy based on 1650 consecutive patients with primary hyperparathyroidism. *Ann Surg [Internet].* 2011 Mar [cited 2022 May 17];253(3):585–91. Available from: <https://pubmed.ncbi.nlm.nih.gov/21183844/>
5. Vaidya A, Curhan GC, Paik JM, Wang M, Taylor EN. Physical activity and the risk of primary hyperparathyroidism. *J Clin Endocrinol Metab [Internet].* 2016 Apr 1 [cited 2022 May 17];101(4):1590–7. Available from: [pmc/articles/PMC4880164/](https://pubmed.ncbi.nlm.nih.gov/264880164/)
6. Roy M, Mazeh H, Chen H, Sippel RS. Incidence and localization of ectopic parathyroid adenomas in previously unexplored patients. *World J Surg [Internet].* 2013 Jan 12 [cited 2022 Mar 23];37(1):102–6. Available from: <https://link.springer.com/article/10.1007/s00268-012-1773-z>
7. Shen W, Düren M, Morita E, Higgins C, Duh QY, Siperstein AE, et al. Reoperation for persistent or recurrent primary hyperparathyroidism. *Arch Surg [Internet].* 1996 [cited 2022 May 17];131(8):861–9. Available from: <https://pubmed.ncbi.nlm.nih.gov/8712911/>
8. Khan AA, Hanley DA, Rizzoli R, Bollerslev J, Young JEM, Rejnmark L, et al. Primary hyperparathyroidism: review and recommendations on evaluation, diagnosis, and management. A Canadian and international consensus. *Osteoporos Int [Internet].* 2017 Jan 1 [cited 2022 May 17];28(1):1. Available from: [pmc/articles/PMC5206263/](https://pubmed.ncbi.nlm.nih.gov/2806263/)