Revista Mexicana de
Cirugía Bucal y Maxilofacial

Vol. 21, No. 1
January-April 2025. pp. 38-43

doi: 10.35366/119809

Received: November 2024.
Accepted: April 2025.

Clinical case

Maxillary brown tumor due to tertiary hyperparathyroidism. Case report and review of the literature

Tumor pardo maxilar por hiperparatiroidismo terciario. Reporte de un caso y revisión de la literatura

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ABSTRACT

Introduction: brown tumor is a benign giant cell lesion associated with hyperparathyroidism that for diagnostic accuracy should include a rigorous evaluation to determine the correct treatment option and avoid errors in management that can translate into repetitive management of lesions that can lead to irreversible sequelae. **Objective:** to analyze the clinical, radiographic and histologic characteristics of brown tumors due to hyperparathyroidism and to identify the importance of laboratory studies for a correct diagnosis of a probably undetected disease and to define treatment strategies. **Clinical case:** we present a 54-year-old male case with tertiary hyperparathyroidism who presented maxillary brown tumor. Clinical manifestations, laboratory, radiographic and histological tests of this lesion are analyzed for its correct diagnosis and ideal treatment plan. **Conclusion:** it is of vital importance the role of laboratory studies for an established clinical/radiographic diagnosis of a probably undetected

RESUMEN

Introducción: *el tumor pardo es una lesión benigna de células gigantes asociada al hiperparatiroidismo que para su precisión diagnóstica se debe incluir una evaluación rigurosa que permita determinar la opción de tratamiento correcta y evitar errores en el manejo, los cuales pueden traducirse en manejos repetitivos de las lesiones que pueden conllevar secuelas irreversibles.* ***Objetivo:*** *analizar las características clínicas, radiográficas e histológicas del tumor pardo por hiperparatiroidismo e identificar la importancia de los estudios de laboratorio para un diagnóstico correcto ante una enfermedad probablemente no detectada, así como definir las estrategias de tratamiento.* ***Caso clínico:*** *se presenta un caso de paciente masculino de 54 años con hiperparatiroidismo terciario que presenta tumor pardo maxilar, se analizan sus manifestaciones clínicas, pruebas de laboratorio, radiográficas e histológicas de dicha lesión para su correcto diagnóstico y plan de tratamiento ideal.* ***Conclusión:*** *es de vital importancia el rol que tienen los estudios de laboratorio para un diagnóstico clínico/radiográfico establecido ante una enfermedad pro-*

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How to cite: Hernández ZDP, Cadena AJL, Rodríguez GCS, Patlán VEA. Maxillary brown tumor due to tertiary hyperparathyroidism. Case report and review of the literature. Rev Mex Cir Bucal Maxilofac. 2025; 21 (1): 38-43. https://dx.doi.org/10.35366/119809

disease, in order to differentiate it from other entities and define treatment strategies.

Keywords: brown tumor, hyperparathyroidism, giant cell tumor.

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Palabras clave: *tumor pardo, hiperparatiroidismo, tumor de células gigantes.*

INTRODUCTION

Brown tumor is a benign giant cell lesion associated with hyperparathyroidism, they are late osteolytic lesions of giant cells secondary to the effects of parathyroid hormone on bone tissue.[1,2]

Hyperparathyroidism is defined as a metabolic state in which there is an increase in the secretion of parathyroid hormone (PTH) and serum calcium, which leads to high levels of this hormone in the bloodstream,[3] being a rare disease with a low prevalence of 1%,[4-6] although it is difficult to establish the exact frequency and the real prevalence due to asymptomatic cases that are not diagnosed.[7-9]

Within hyperparathyroidism we can distinguish three clinical forms: Primary hyperparathyroidism (PHPT), in which there is an autonomous secretion of PTH, and in which there is a high level of calcium in the bloodstream. PTH higher than the requirement for calcemia, being an uncontrolled production as a result of a parathyroid adenoma (80 to 90%), a parathyroid hyperplasia (10 to 15%) or a parathyroid carcinoma (1%).

In secondary hyperparathyroidism (SHPT) the production mechanism is related to adaptive changes of the parathyroid glands to hypocalcemia produced by other diseases (chronic renal failure, intestinal malabsorption syndrome, vitamin D4 deficiency), so the origin of the disorder will be unrelated to the glands themselves since a hypersecretion of PTH is manifested in response to chronic hypocalcemia.

In tertiary hyperparathyroidism (THPT) it occurs when despite correcting the underlying stimulus, the parathyroid glands continue to secrete high levels of PTH, this results from a long-term SHPT or for a long time due to renal failure, since the parathyroid glands become autonomous which causes hypercalcemias that are very difficult to manage.[2,10-13]

Therefore, the main regulator of PTH secretion is the serum concentration of calcium ion; elevated serum calcium suppresses PTH secretion (not completely) and low serum calcium concentrations induce PTH release. Within the pathophysiology of hyperparathyroidism, we have that, PTH is normally produced by parathyroid glands in response to a decrease in serum calcium levels.[4]

Therefore, increased PTH levels, regardless of their cause, produce an increase in osteoclastic activity, thus promoting bone resorption and the appearance of cystic lesions. The direct effect of PTH on the skeleton is to stimulate calcium output, so there is greater activity in osteoclasts, thus predominating bone resorption causing local destruction with hemorrhagic areas and hemosiderin deposits inside; As a result of intraosseous bleeding and tissue degeneration, groups of hemosiderin-laden macrophages, giant cells and fibroblasts may develop in these lytic lesions.[14-19]

Hemorrhages and hemosiderin deposits give the lesions their brown characteristics and give them the name «Brown Tumors».[16,17]

Therefore, the role of laboratory studies is of vital importance for an established clinical/radiographic diagnosis of an undetected disease and to be able to differentiate it from other entities in order to define the indicated treatment strategies.

CLINICAL CASE

A 54-year-old male from the State of Michoacán, Mexico, presented for a dental consultation for the placement of dental implants. Relevant family history includes: a diabetic and hypertensive father, a diabetic mother, and a brother with a history of kidney transplant. His personal medical history includes diagnosis with Henoch-Schonlein purpura, 24 years with Chronic Kidney Disease Stage G5 (renal failure), a history of bilateral papillary renal carcinoma followed by bilateral nephrectomy in the same year, subsequent kidney transplant from a related living donor, hypertension diagnosed at age 26 under treatment, pneumonia due to SARS-CoV-2 (2020), and a history of alcoholism and smoking.

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The patient reports asthenia and adynamia, muscle pain, extreme exhaustion after performing daily activities, dizziness, constipation, and paresthesia in the upper limbs.

During this consultation, a diagnosis of granuloma in the maxilla was made, and a biopsy was performed, which provided a histopathological result of Giant Cell Granuloma.

The following orthopantomogram is shown, which reveals radiolucent areas in the upper left maxilla and mandibular body *(Figure 1)*.

Relevant laboratory results include: parathyroid hormone level of 87.40 pg/mL, serum calcium of 14.03 mg/dL, and alkaline phosphatase of 278 U/L.

Due to the presence of severe hypercalcemia, the patient was treated surgically with head and neck surgery, where a selective subtotal parathyroidectomy was performed, including resection of the right superior parathyroid (3.2 g), left superior (220 mg), right inferior (120 mg), and biopsy of the left inferior (18 mg). Diagnostic-therapeutic evaluation for hyperparathyroidism was conducted, and follow-up was initiated.

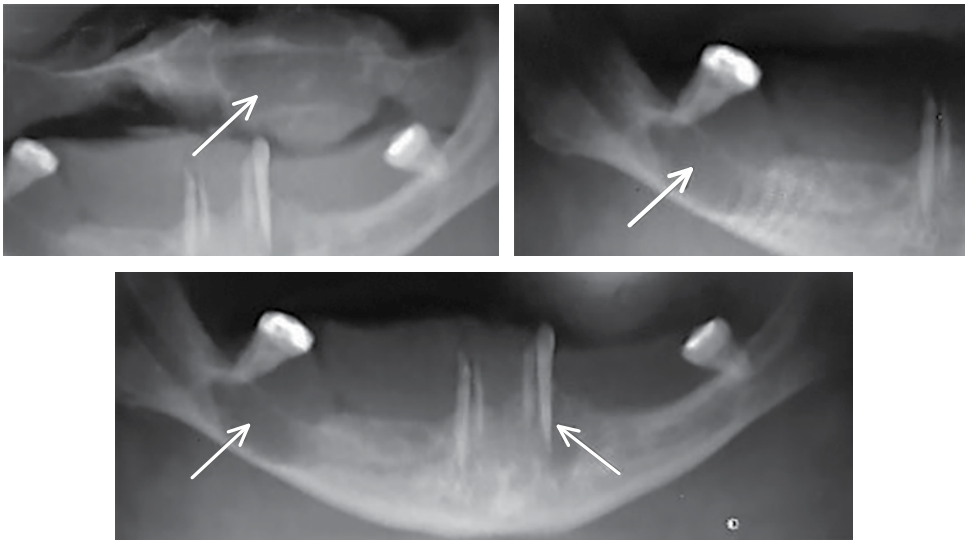

Figure 1:

Orthopantomogram showing lytic lesions in the maxilla and mandible in a 54-year-old male patient.

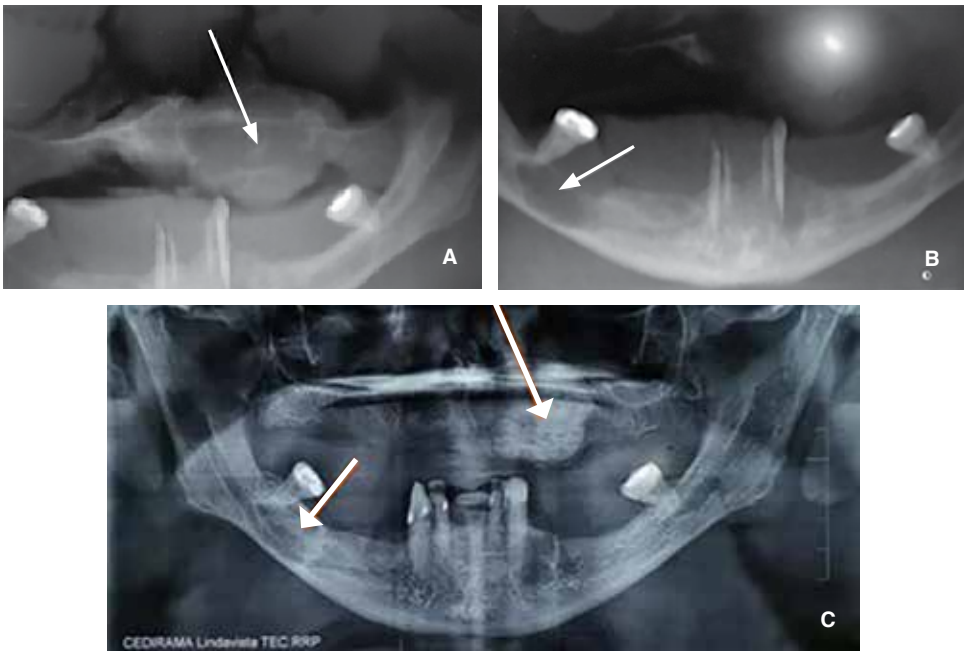

Figure 2:

A and **B)** Orthopantomogram without treatment for tertiary hyperparathyroidism. **C)** Orthopantomogram after surgical treatment for tertiary hyperparathyroidism.

Below is the radiograph of the patient showing the lytic lesions of the brown tumor due to tertiary hyperparathyroidism from Chronic Kidney Disease, which was managed by endocrinology *(Figure 2A-B)*.

Once under treatment for HPT, the patient presented with an updated panoramic radiograph *(Figure 2C)*, confirming partial involution of the lesions with an increase in bone density.

Subsequently, an evaluation for the treatment of sequelae is planned. Due to functional alterations, corrective maxillary osteoplasty is planned.

Facial tomography shows a hyperdense area in the posterior left maxillary region following the treatment for Tertiary Hyperparathyroidism, which causes severe deformity in the contour of the left maxillary alveolar process *(Figure 3)*.

Maxillary remodeling osteoplasty is planned for subsequent prosthetic rehabilitation *(Figure 4)*.

DISCUSSION

For diagnostic accuracy, a complete rigorous clinical evaluation, imaging studies, laboratory tests and biopsy of the lytic lesions must be included to reach the correct diagnosis and then correlate them.

There are 4 parathyroid glands located behind, at the ends of the thyroid lobes, which measure 8-9mm in length, 4-5mm in width and 4mm in thickness and have an approximate total weight of 125grs. While the patient presented this affected anatomy since only the selective parathyroidectomy of the upper left parathyroid (220 mg), lower right (120 mg) and biopsy of the lower left (18 mg) was performed.

Studies found that the incidence of brown tumors is higher in primary hyperparathyroidism[13] having that this patient presented was diagnosed with tertiary hyperparathyroidism.

The most frequent form in 80% of the cases is asymptomatic with a frequency in the facial bones is 2%, generally affects the mandible in 4.5% and very rarely the maxilla.[20,21] Although brown tumors have also been described in other areas of the facial mass including the orbital region.[22,23] Swelling, pain, functional alteration of the masticatory apparatus may occur due to dental mobility that can cause the injury.[24] Most patients are asymptomatic and the manifestations are bone and renal,[25] agreeing with the literature since it is a patient with chronic kidney disease and asymptomatic lesions, also with a presentation mostly of lytic lesions in the maxilla differing with the literature, although lesions in the mandible were also presented but in smaller size.

Histopathologically it is identical to the central giant cell granuloma, so it is important to mention that the only histopathological study is not enough to determine the diagnosis of brown tumor, since it is difficult to differentiate it from other giant cell lesions and the histological findings are not pathognomonic of the lesion, therefore remember that the hypersecretion of PTH is mainly characterized by:

1. Hypercalcemia
2. Hypophosphatemia
3. Abnormal bone metabolism

Hypercalcemia is the most common manifestation of hyperparathyroidism and is diagnosed when the serum calcium is above the normal range of 8.6 to 10 mg/dL, when the patient presents 14.03 mg/dL, a decreased serum phosphate level is also useful to confirm the diagnosis; the elevated PTH level with the patient presenting a level of 87.40 pg/mL, as well as the increased alkaline phosphatase level being a normal value of 44-147 IU/L thus presenting a value of 278 U/L (elevated) *(Table 1)*.

Therefore, hypercalcemia constitutes the key to the diagnosis of this disease, being the most important metabolic data.[15,26]

This circumstance determines the obligation of the study of the function of the parathyroid glands when lesions diagnosed histologically as giant cells are found and thus establish a treatment which will depend on the cause of the HPT, therefore, it begins with the stabilization of the HPT and it has been shown that after this the lesions begin to involute progressively[27] and when there is involution the time is variable, from six months to five years, the factor that most influences this involution is the age of the patient (with shorter times in the younger ones).[20,21,28]

CONCLUSION

With this bibliographic review and case report, all the clinical manifestations, laboratory, radiographic and histological tests of the so-called brown tumor due to hyperparathyroidism are analyzed.

It is essential to establish a definitive diagnosis to determine the correct treatment option and avoid management errors that can result in repetitive management of misdiagnosed giant cell lesions and

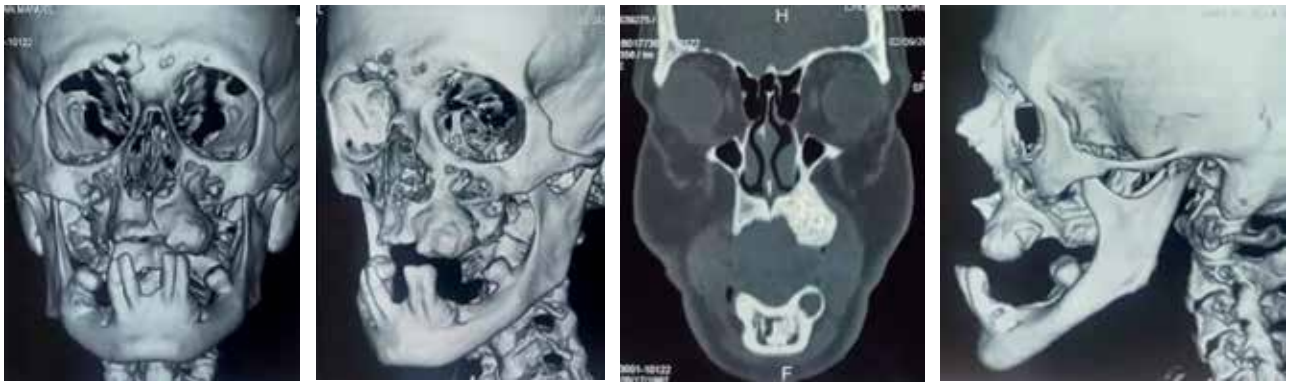

Figure 3: Tomography with hyperdense areas post-treatment for tertiary hyperparathyroidism.

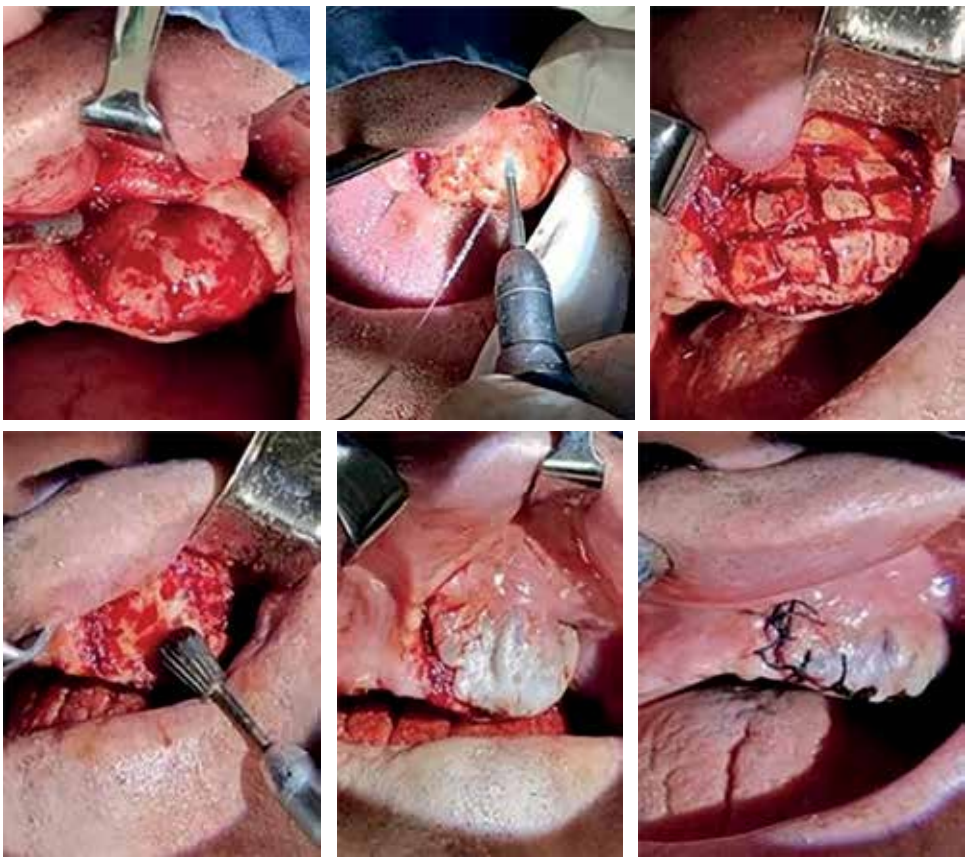

Figure 4:

Sequence of maxillary osteoplasty.

Table 1: Normal levels in Serum.[14]

Serum	Normal range
Calcium (mg/dL)	8.5-10.5
Phosphate (mg/dL)	2.3-4.7
Parathyroid hormone (pg/mL)	14.5-87.1
Vitamin D (ng/mL)	30.0-80.0

can lead to irreversible sequelae in misdiagnosed patients.

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Conflict of interests: no conflict of interest is declared.