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Radiological Appearance of Brown Tumors of the Maxilla Indicative of Primary Hyperparathyroidism: A Case Report

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Abstract **Case Report**

Brown tumors are a rare complication of hyperparathyroidism resulting from abnormalities in bone metabolism. Advances in biological exploration techniques enable early diagnosis before the onset of bone manifestations. While mandibular involvement is the most common in the maxillofacial region, maxillary involvement is extremely rare. We report a case of a patient with primary hyperparathyroidism revealed by a maxillary tumor. The pathological result concluded a benign giant cell tumor of the maxilla and fibrous dysplasia. The diagnosis of a brown tumor was considered based on the association of radiological data (osteolytic image and parathyroid adenoma), histological findings (giant cell tumor), and biological markers (hypercalcemia, hypophosphatemia, and elevated Parathyroid Hormone (PTH). This case highlights the difficulty in establishing a correct diagnosis in patients with an osteolytic maxillary process and the need to investigate hyperparathyroidism in the presence of a giant cell lesion.

Keywords: Brown tumors, bone metabolism, hyperparathyroidism, Parathyroid Hormone (PTH).

INTRODUCTION

Brown tumors, also known as von Recklinghausen's fibrous osteitis, are non-neoplastic lesions resulting from a bone metabolism anomaly in the context of primary or secondary hyperparathyroidism. They are a direct result of parathyroid hormone action on the bone matrix. They are secondary, in most cases, to primary hyperparathyroidism, which in over 80% of cases is caused by a parathyroid adenoma [1]. They are reported in 4.5% of patients with primary hyperparathyroidism and 1.5 to 1.7% of those with secondary hyperparathyroidism [2].

In 1934, Albright first described them in the facial skeleton [3]. Brown tumors can affect the entire skeleton, with maxillomandibular involvement being unusual [4]. They typically present as a progressive, painful buccal swelling. They can be locally aggressive without metastatic potential [5]. Histologically, they are classified as giant cell lesions of the maxillae [5]. Thus, a definitive diagnosis requires systemic investigations: measuring calcium and Parathyroid Hormone **(**PTH) levels to differentiate between these lesions.

The aim of our study is to describe the circumstances of discovery, and the radiological, clinical, and biological characteristics of brown tumors.

PATIENTS AND METHODS

: We present a patient followed in the department for brown tumors in the context of primary hyperparathyroidism.

OBSERVATIONS

Ms. C. B., a 26-year-old patient with a history of undocumented psychiatric follow-up, had undergone maxillofacial surgery in 2015 for a mandibular ameloblastoma. This surgery included a hemimandibulectomy with reconstruction using a maxillary plate and a bone graft (histology from that time is undocumented). She presented for consultation in the trauma department for a pathological fracture of the right trochanteric region and the left distal femur. The patient complained of a maxillary swelling, which was mildly painful and caused chewing difficulties, evolving since 2018. She had previously undergone CT scans and pathological examinations.

The initial histological examination in 2018 concluded a giant cell tumor with no signs of

malignancy. The second examination in 2019 suggested fibrous dysplasia without evident malignancy or signs of a giant cell tumor. The latest examination of giant cell lesions initially suggested a giant cell reparative granuloma, considering the patient's surgical history. However, a close correlation with clinical, radiological, and evolutionary data was necessary.

CT imaging revealed a mixed bone lesion of the maxillary bone with alveolar and endosinusoidal extension on the same side, associated with multiple lytic bone lesions in the frontal-parietal regions, both clavicles, and the right humeral epiphysis, with cortical thinning and rupture in some areas (Fig. 1 and 2).

Given these findings, several diagnostic hypotheses were considered. Based on the association of radiological data (osteolytic image), histological findings (giant cell tumor), and biological markers (hypercalcemia, hypophosphatemia, and elevated Parathyroid Hormone (PTH)), the diagnosis of brown tumors in the context of primary hyperparathyroidism was established. Cervical ultrasound revealed a hypervascularized heterogeneous hypoechoic nodule behind the right thyroid gland, suggestive of a parathyroid adenoma (Fig. 3). Parathyroid scintigraphy was performed and returned positive.

The Parathyroid Hormone **(**PTH) level was very high at 1825 pmol/L (normal range: 9-55).

After one month from the diagnosis, the parathyroid adenoma was surgically removed. Post-operative calcium-phosphorus balance and Parathyroid Hormone **(**PTH) levels returned to normal.

The pathological report confirmed a parathyroid adenoma.

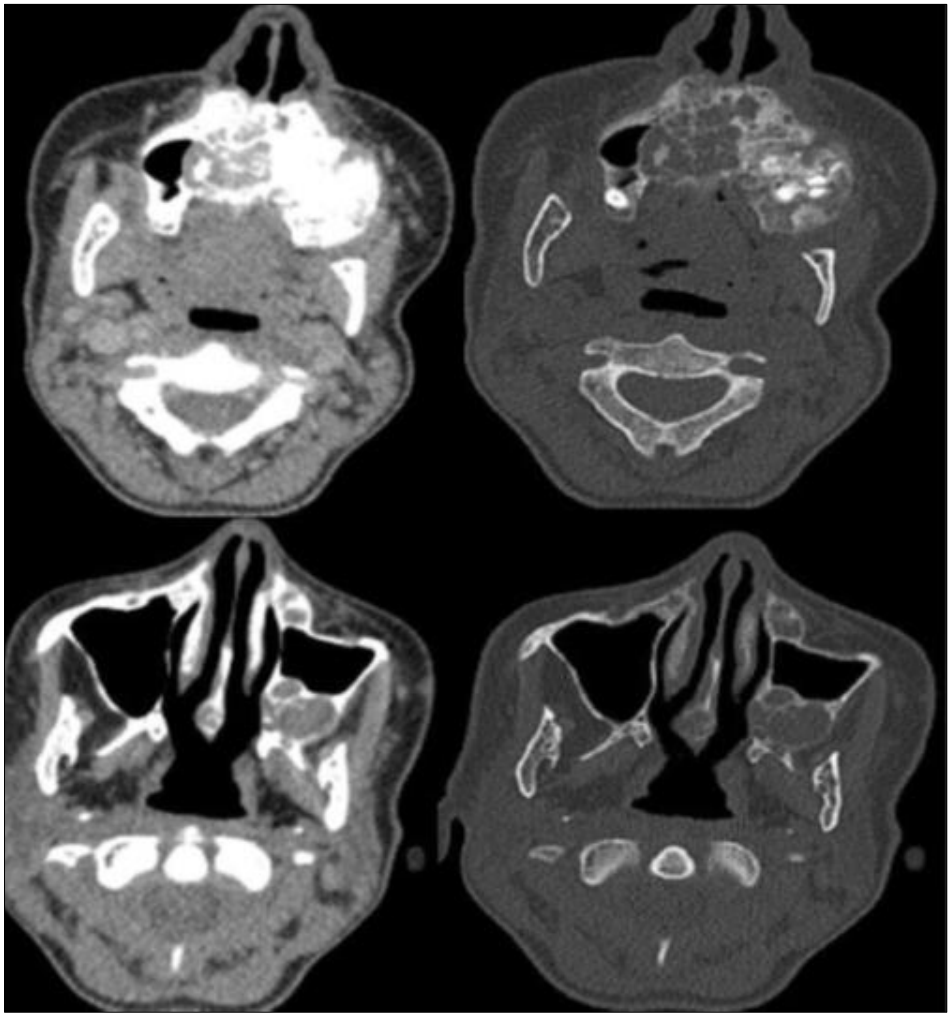

Fig. 1: Axial CT scan with soft tissue and bone window settings

Lytic and sclerotic bone lesion with a glassy matrix affecting the left maxillary bone, involving the alveolar and palatine processes, and extending into the hard palate with endo-sinus extension.

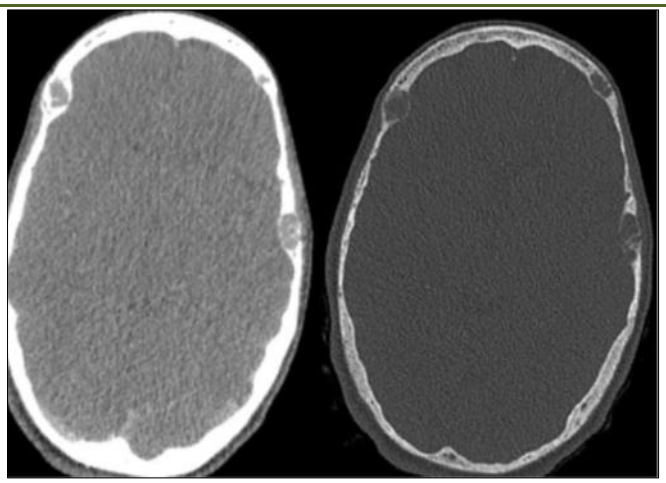

Fig. 2: Axial CT scan with soft tissue and bone window settings

Lytic lesion of the frontal and parietal vault with fine septation, and areas of thinning and rupture of both the inner and outer tables.

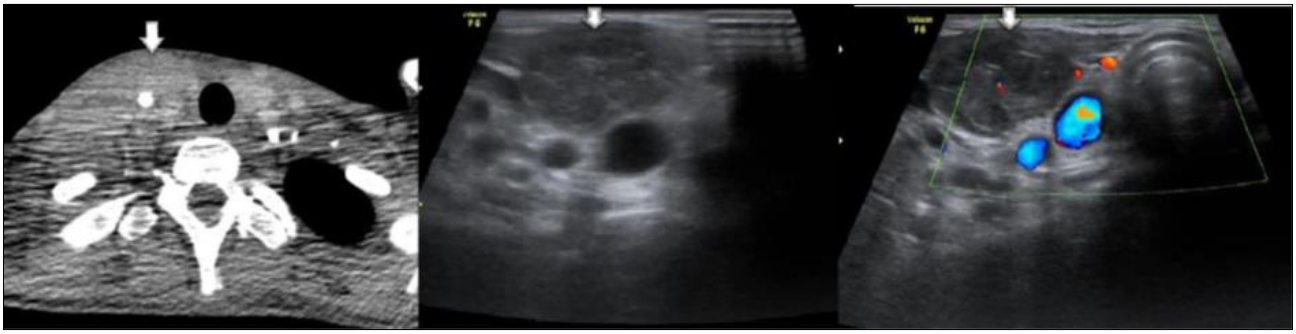

Fig. 3: Axial CT scan with soft tissue window settings and cervical ultrasound with and without color Doppler

Heterogeneous hypoechoic nodular formation in the right retrothyroid region with central and peripheral vascularization, consistent with a parathyroid adenoma.

DISCUSSION

Brown tumors are present in 4.5% of cases of hyperparathyroidism and primarily affect the ribs, pelvis, femur, and mandible, with a predominance in young women. Maxillary involvement is rare, occurring in about 4.5% of cases [6]. Primary hyperparathyroidism results from excessive production of parathyroid hormone (PTH), most often due to a single adenoma, as seen in our patient.

Currently, primary hyperparathyroidism is often discovered incidentally in 75 to 80% of cases during blood tests showing asymptomatic hypercalcemia [7]. It can also be revealed by kidney stones or cardiovascular issues [7]. Bone cysts, osteoporosis, and brown tumors are late manifestations of the disease and are rare [8,9]. Primary hyperparathyroidism is caused by a parathyroid adenoma in over 80% of cases, and less commonly by hyperplasia (15%) [10, 11]. There is a female predominance in benign hyperparathyroidism [12]. Maxillary brown tumors are extremely rare [16]. Brown tumors can present similarly to other tumors and pseudotumors of the maxilla.

Radiologically, a brown tumor manifests as nonspecific osteolysis that can appear as either a unilocular or multilocular bone lysis. CT imaging shows a mixed osteolytic mass that enhances with contrast but does not invade soft tissues or cause any periosteal reaction [13].

Cervical ultrasound is used to detect parathyroid lesions responsible for hyperparathyroidism.

Brown tumors manifest with histological transformations that are not pathognomonic and can be observed in true giant cell tumors, reparative granulomas, aneurysmal bone cysts, and fibrous dysplasia [14-6].

The treatment of hyperparathyroidism should be the primary step in managing these patients, although opinions differ on the management of secondary bone lesions. The evolution of brown tumors after parathyroidectomy is variable. In cases where the lesions are extensive or significantly destructive, affecting organ function, the resulting tissue damage may not be repaired

despite achieving normocalcemia. When lesions persist after treatment for hyperparathyroidism, Yamazaki [2], recommends curettage and enucleation of the tumor.

CONCLUSION

Primary or secondary hyperparathyroidism can sometimes present solely with osteolytic lesions of the facial bones. Therefore, a giant cell lesion of the maxilla should systematically prompt a search for hyperparathyroidism through a calcium-phosphorus balance test and parathyroid hormone (PTH) measurement. Diagnosing primary hyperparathyroidism helps avoid unnecessary surgery on brown tumors of the maxilla, which should regress following the excision of the parathyroid lesion [14]. The case presented highlights the difficulty in establishing an accurate diagnosis in patients with an osteolytic maxillary process appearing histologically as a giant cell lesion, which is crucial for proposing appropriate treatment.

Conflicts of Interest: The authors declare no conflicts of interest.

Contributions of the Authors: All authors contributed to the conduct of this work. They have read and approved the final version of the manuscript.

REFERENCE

1. Khochtali, H., Ach, K., Jlidi, R., Bouhaouala, H., Ben Gaid Hassine, N., & Essakly, M. K. (1991). Tumeur brune bilatérale des maxillaires. A propos d'un cas. *Revue de stomatologie et de chirurgie maxillo-faciale*, *92*(2), 116-119.
2. Keyser, J. S., & Postma, G. N. (1996). Brown tumor of the mandible. *American journal of otolaryngology*, *17*(6), 407-410.
3. Smith, G. A., & Ward, P. H. (1978). Giant-cell lesions of the facial skeleton. *Archives of Otolaryngology*, *104*(4), 186-190.
4. Cicconetti, A., Matteini, C., & Piro, F. R. (1999). Differential diagnosis in a case of brown tumor caused by primary hyperparathyroidism. *Minerva stomatologica*, *48*(11), 553-558.
5. Schweitzer, V. G., Thompson, N. W., & McClatchey, K. D. (1986). Sphenoid sinus brown tumor, hypercalcemia, and blindness: An unusual presentation of primary hyperparathyroidism. *Head Neck Surg*, *8*, 379-381.
6. Aoune, S., Khochtali, H., Dahadouh, C., Turki, A., Mokni, M., & Bakir. A. (2000). Lésion à cellules géantes des maxillaires révélatrices d'hyperparathyroïdie primaire. *Rev Stomatol Chir Maxillofac*, *101*, 86-89. Guney, E., Yigitbasi, O., Bayram, F., Oser, V., & Canoz, O. (2001). Brown tumor of the maxilla associated with primary hyperparathyroidism. *Auris Nasus Larynx*, *28*, 369–72.
7. Rodgers, S. E., Lew, J. I., & Solórzano, C. C. (2008). Primary hyperparathyroidism. *Current Opinion in Oncology*, *20*(1), 52-58.
8. Horowitz, M., Wishart, J. M., Need, A. G., Morris, H. A., & Nordin, B. C. (1994). Primary hyperparathyroidism. *Clinics in geriatric medicine*, *10*(4), 757-775.
9. Marx, R. E., & Stern, D. (2000). Oral and maxillofacial pathology. A rationale for diagnosis and Treatment. 2002. Chicago, IL. Quintessence Publishing. Secondary hyperparathyroidism. *Report of a case, Joint Bone Spine*, *67*, 230–233.
10. Masson, E. A., MacFarlane, I. A., Bodmer, C. W., & Vaughan, E. D. (1993). Parathyroid carcinoma presenting with a brown tumour of the mandible in a young man. *British Journal of Oral and Maxillofacial Surgery*, *31*(2), 117-119.
11. Owens, B. B. (2009). A review of primary hyperparathyroidism. *Journal of Infusion Nursing*, *32*(2), 87-92.
12. Mundy, G., Cove, D., & Fisken, R. (1980). Primary hyperparathyroidism: changes in the pattern of clinical presentation. *The Lancet*, *315*(8182), 1317-1320.
13. Oral and Maxillofacial Surgery Clinics of North America. Philadelphia: Saunders; 1997. Fibro-osseous diseases: considerations in diagnosis and management
14. Presentation of primary hyperparathyroidism: (2008). Severe hypercalcemia and multiple brown tumors. *Revue du Rhumatisme*, *75*(3), 289–291.