

## Primary Hyperparathyroidism Revealed by a Giant Cell Granuloma of the Maxilla About 1 Case

Najoua Belhaj<sup>1\*</sup>, Razika Bencheikh<sup>2</sup>, Hanaa Rahim<sup>1</sup>, Wah Sidelmoctar Abdellahi<sup>1</sup>, Mohammed Anass Benbouzid<sup>2</sup>, Nadia Cherradi<sup>3</sup>, Leila Essakalli<sup>2</sup>

<sup>1</sup>Resident Physician in Otorhinolaryngology, Department of Otorhinolaryngology, Head and Neck Surgery, Ibn Sina University Hospital, Faculty of Medicine, Mohammed V University, Rabat, Morocco

<sup>2</sup>Professor of Otorhinolaryngology, Department of Otorhinolaryngology, Head and Neck Surgery, Ibn Sina University Hospital, Faculty of Medicine, Mohammed V University, Rabat, Morocco

<sup>3</sup>Professor of Anatomopathology, Department of anatomopathology Ibn Sina University Hospital, Faculty of Medicine, Mohammed V University, Rabat, Morocco

DOI: [10.36348/sjm.2021.v06i05.001](https://doi.org/10.36348/sjm.2021.v06i05.001)

| Received: 22.03.2021 | Accepted: 30.04.2021 | Published: 07.05.2021

\*Corresponding Author: Belhaj Najoua

### Abstract

Giant Cell Granuloma is a rare bony lesion in the Head and Neck region. It mainly affects young adults between the age of 20 and 40. Occasionally, soft parts may be invaded, resulting from the direct effects of the parathyroid hormone. We report in this work the observation of a 36-year-old patient who presented for a maxillary tumor that revealed a parathyroid adenoma with associated hyperparathyroidism. The histopathological study of maxillary tumor's material confirms the diagnostic of a giant cell granuloma.

**Keywords:** Giant Cell Granuloma, Head and Neck region, young adults, hyperparathyroidism.

**Copyright © 2021 The Author(s):** This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

### INTRODUCTION

The giant cell tumor is a benign tumor that mainly affects the epiphyses of long bones. It mainly affects young adults between the age of 20 and 40. Its origin remains uncertain. Simple shots are usually characteristic enough to suggest diagnosis. It is a purely lytic and generally aggressive tumor that destroys the bone, even leading to a fracture. Occasionally, soft parts may be invaded [1], resulting from the direct effects of the parathyroid hormone (PTH) [2]. We report in this work the observation of a 36-year-old patient who presented for a maxillary tumor that revealed a parathyroid adenoma with associated hyperparathyroidism.

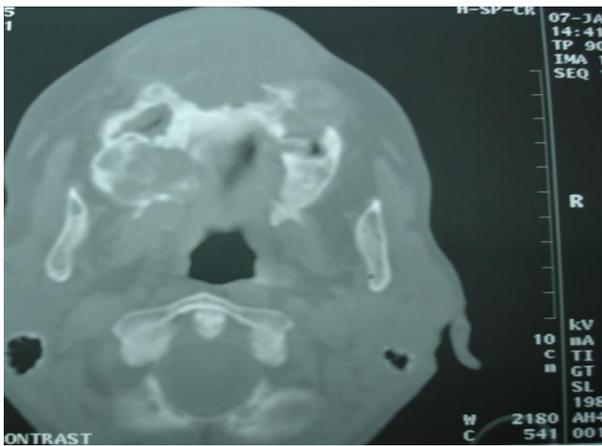
### CASE REPORT

This is a 36-year-old patient with no significant pathological history who consults for a tumor in the maxilla since 1 year. The patient reports discomfort when chewing and speaking, and a few episodes of oral bleeding. Clinical examination shows a tumor developed at the expense of the maxilla (Figure-1).

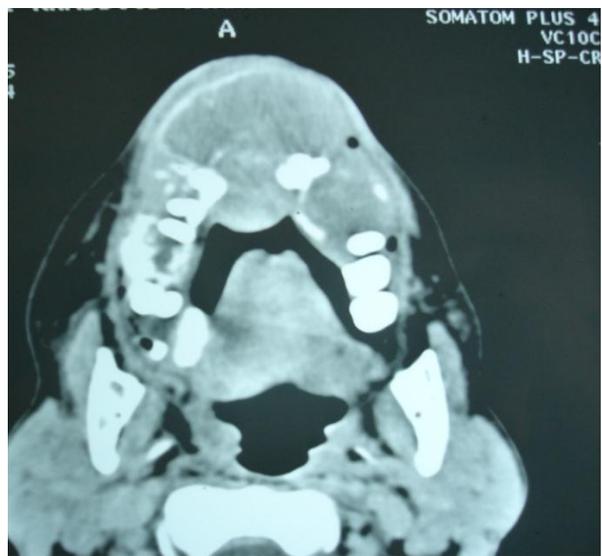
Palpation is not painful. The skin in the face is normal. There is no satellite adenopathy. Chest X-rays are normal. CT scans of the face show an osteolytic tissue tumor of the maxillary bone that combines boxes with thin calcified spans (Figure 2, 3 & 4).



**Fig-1: Photo of the patient showing the giant Cell granuloma of the maxillary**



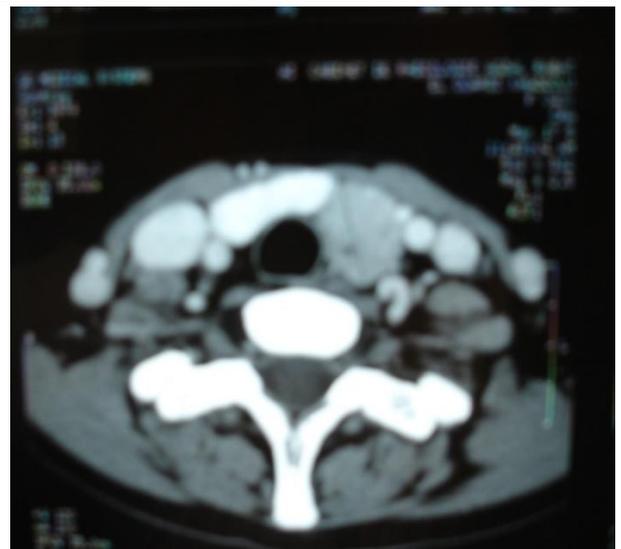
**Fig-2: Axial CT slice showing the lytic process of the maxilla**



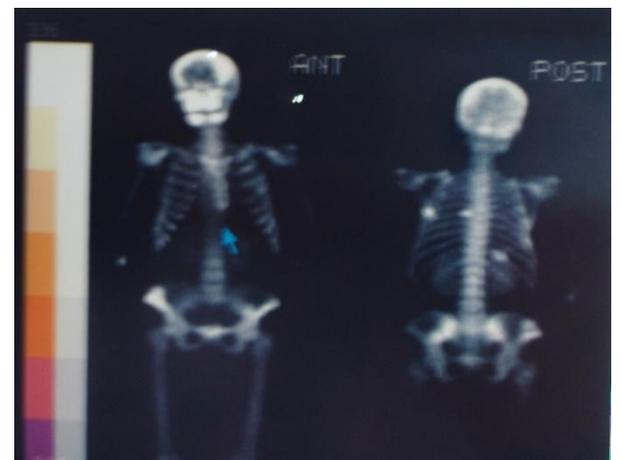
**Fig-3: Axial CT slice showing the lytic process of The maxilla**



**Fig-4: Coronal CT slice showing the lytic process**

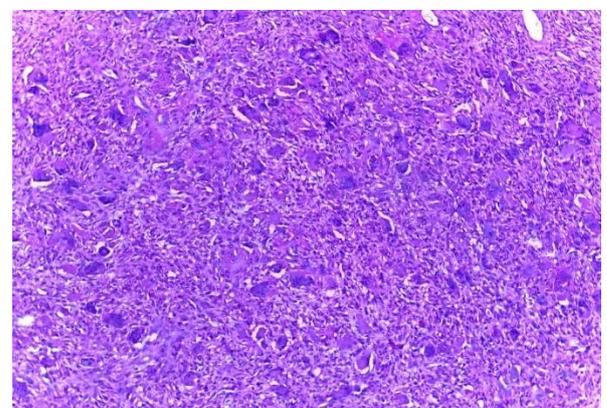


**Fig-5: Axial section of a cervical CT scan showing the parathyroid adenoma**

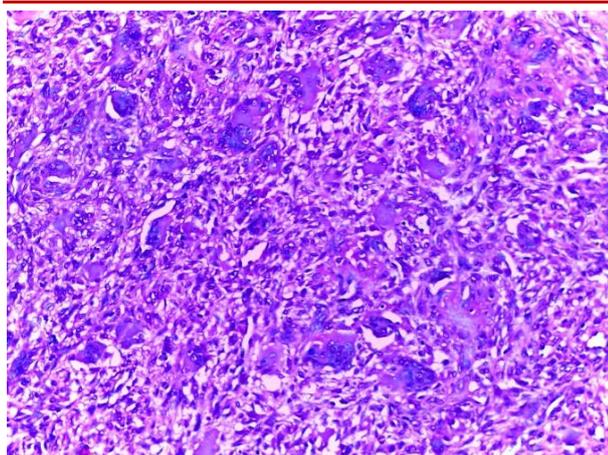


**Fig-6: Sesta MIBI scan showing the hyperfixation in the parathyroid gland**

This tumor represses, without invading the neighborhood structures such as sinus. We perform a tumor biopsy, the histopathological study of material confirms the diagnostic of a giant cell granuloma (Figure 7 & 8).



**Fig-7: H. E/E\*10 (HE: hematoxylin and eosin staining/Enlargement)**



**Fig-8: H.E/E\*20**

Figure 7 & 8: Hematoxylin and Eosin staining: Many multi-nucleated giant cells can be seen amidst a background of stromal cells.

The biological findings: The PTH dosage shows values at 500 pg/ml, for normal values between 15 and 65 pg/ml. Kidney function and calcemia are normal; X-rays of the skull, long bones and spine x-rays are normal. The cervical ultrasound objective a left basithyroid cervical nodule. The sesta-MIBI scan shows a hyperfixation outbreak in the left lower parathyroid gland. The balance was supplemented by a cervical ultrasound and a cervical scan showing a left parathyroid mass of 3 cm of large axis (Figure 5 & 6).

Surgical treatment was based on exeresis of the maxillary tumor, well encapsulated. Exeresis of the parathyroid nodule is made, at the same time operating by a cervical incision.

## DISCUSSION

Giant Cell Granuloma is a rare bony lesion in the Head and Neck region. It is a non-odontogenic tumor never seen in any other bone of the skeleton. It most commonly affects maxilla followed by the mandible. Although benign, it can locally be destructive. Surgery is the most accepted method of treating the condition. World Health Organization defines it as an intra-osseous lesion consisting of cellular fibrous tissue and contains many foci of hemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone. Frequently it is only a painless swelling, but growth in some cases is so rapid and the mass can also rarely erode through bone particularly of the alveolar ridge to produce a soft tissue swelling. It can occur at any age but presents most frequently in the 2nd and 3rd decades and involves the maxilla more than the mandible. It is twice as frequent in females. Despite the fact that the course of the disease is considered benign, there still exist some reports in literature where metastasis has been observed. Furthermore, malignant transformations to osteosarcoma or fibrosarcoma have been reported.

Histologically, it is indistinguishable from other giant cell lesions of the bone like cherubism and aneurysmal bone cyst. Giant cell granuloma forms a lobulated mass of proliferative vascular connective tissue packed with giant cells. These giant cells are seen lying in vascular stroma. These giant cells have a patchy distribution and signs of bleeding into the mass and deposits of hemosiderin are frequently seen [3-5].

Ultra-structurally the proliferating cells include spindle-shaped fibroblasts myofibroblasts and inflammatory mononuclear cells. Sparse strands of collagen fibers partly subdivide the lesion which may contain a few trabeculae of osteoid or bone. Surgery is the most accepted and traditional form of treatment. However, tissue removal ranges from simple curettage to bloc resection.

The main differential diagnosis is that of a giant cell tumor. Morphologically, there seem to be some nuances in the giant cell tumor: fewer fibroblasts, very large multinucleated giant cells, less osteoid tissue production. Differential diagnosis is made on histochemical and immunological research and markings. Other differential diagnoses are those of aneurysmal cyst and we have seen the community that these two entities may present or brown tumors of hyperparathyroidism [5, 6].

HPT is characterized by high levels of PTH in the blood, usually responsible for hypercalcemia [7]. The etiology of primary HPT is dominated by parathyroid adenoma (90% of cases) [8]. Primary HPT is most common in adults between the ages of 40 and 50. Female achievement predominates. Primary HPT is often detected by biological examination [9]. The clinical picture is not specific [7, 8].

It is dominated by urinary, digestive or neuropsychic symptoms. HPT causes long-term bone remodeling, of which so-called "brown" tumors represent a rare appearance. Only a dozen observations of primary HPT revealed by a maxillofacial tumor have been published [8-10].

These bone tumors are single or multifocal and are primarily of interest to the axial skeleton. In the facial area, mandibular damage is the most common. Maxillary involvement is extremely rare [10, 11]. Damage to the other bones of the face has also been reported [11].

Histologically, giant cell granulomas brown tumor poses a differential diagnostic problem with brown tumors, giant cell repairing granuloma, non-ossifying fibroid, aneurysmal cyst, cherubism, giant cell sarcoma [12-14].

For treatment, the maxillary locations of the GRCCG, non-surgical treatments have been proposed: intralesional injection of corticosteroids or general administration of calcitonin or interferon  $\alpha$  2a. Some observations, with varying successes, have been reported mainly in forms with the most aggressive behavior [14] always for maxillary locations. De Lange *et al.*, [16, 17] reported a randomized prospective study comparing in a group of 14 patients a general calcitonin treatment versus a placebo. The authors do not find a decisive difference with a follow-up of more than 1 year. Note that they describe some spontaneous regressions including in the control group, but no spontaneous complete disappearance.

Etiological treatment consists of surgical exeresis of adenoma to eliminate active hormone secretion by transverse cervicectomy with exploration of the four parathyroid glands, or by targeted mini-incision on a single gland. This technique requires locating the pathological gland by ultrasound and scan at sesta-MIBI. It requires an intraoperative dosage of PTH, which should decrease by more than 50% to 15 minutes. After treatment, the regression of bone lesions is more or less complete. In our patient, we opted for an exeresis of the tumor, given its large volume. The prognosis depends mainly on the etiology of the HPT. Surgical treatment of parathyroid adenoma is most often successful.

## CONCLUSION

In the face of vestibular swelling, without pathognomonic criteria, associated with a radiological aspect of lytic image and histological criteria for the presence of giant cells, if it is, moreover, a young female subject, the diagnosis of GRCCG is then possible. A physiological phospholcacic biological test as well as a dosage of PTH, before a high rate of the latter test must be supplemented by an ultrasound see a scan.

## REFERENCES

1. Bernard, F., Troude, L., Bouvier, C., & Roche, P. H. (2018). Le granulome réparateur à cellules géantes: un diagnostic différentiel exceptionnel pour une lésion lytique de l'os temporal. *Neurochirurgie*, 64(3), 240.
2. Qiu, Z. L., Wu, C. G., Zhu, R. S., Xue, Y. L., & Luo, Q. Y. (2013). Unusual case of solitary functioning bone metastasis from a "parathyroid adenoma": imagiologic diagnosis and treatment with percutaneous vertebroplasty—case report and literature review. *The Journal of Clinical Endocrinology & Metabolism*, 98(9), 3555-3561.
3. Garg, P., Jain, J., De, N., & Chatterjee, K. (2017). Un granulome géant central de cellules dans la partie postérieure du maxillaire—Un rapport de cas. *International Journal of Surgery Case Reports*, 30, 222-225. doi:10.1016/j.ijscr.2016.11.015

4. Usman, H. U., & Iram, M. (2007). Case Reportgiant Cell Granuloma of the Maxilla. *J Ayub Med Coll Abbottabad*. 19(3):93.
5. Enrique Palacios, MD, FACR, DABR, Department of Radiology/ Neuroradiology, Tulane University School of Medicine, New Orleans, LA 70112, USA. Email: drpalacios@aol.com.
6. Dagang, D. J. T., Gutierrez, J. B., Sandoval, M. A. S., & Lantion-Ang, F. L. (2016). Multiple brown tumours from parathyroid carcinoma. *Case Reports*, 2016, bcr2016215961.
7. Melton, L. J 3rd. (2002). The epidemiology of primary hyperparathyroidism in North America. *J Bone Miner Res*, 17(Suppl. 2):N12-7.
8. Pahlavan, P. S., & Severin, M. C. (2006). Parathyroid carcinoma: a rare case with mandibular brown tumor. *Wien Klin Wochenschr*; 118:175-9.
9. Chammakhi-Jemli, C., Saddoud, N., Zouaoui, W., Gamra, B., Mzabi, H., Sehili, S., ... & Daghfous, M. H. (2006). What is your diagnosis? Brown tumor of the right maxillary sinus. *Journal de radiologie*, 87(7-8), 977-979.
10. Naiman, J., Green, W. R., d'Heurle, D., Iliff, W. J., Benedict, G. W., Eggleston, J. C., & Dorfman, H. D. (1980). Brown tumor of the orbit associated with primary hyperparathyroidism. *American journal of ophthalmology*, 90(4), 565-571.
11. Aderdour, L., Maliki, O., Hassani, R., Elfakiri, M., & Bouchoua, F. (2009). Tumeur brune de la mandibule révélatrice d'une hyperparathyroïdie primaire à calcémie normale. *La Lettre d'oto-rhino-laryngologie et de chirurgie cervico-faciale*, (319), 20-22.
12. Tobón-Arroyave, S. I., Franco-González, L. M., Isaza-Guzmán, D. M., Floréz-Moreno, G. A., Bravo-Vásquez, T., Castañeda-Peláez, D. A., & Vieco-Durán, B. (2005). Immunohistochemical expression of RANK, GR $\alpha$  and CTR in central giant cell granuloma of the jaws. *Oral oncology*, 41(5), 480-488.
13. Abrams, B., & Shear, M. (1974). A histological comparison of the giant cells in the central giant cell granuloma of the jaws and the giant cell tumor of long bone. *Journal of Oral Pathology & Medicine*, 3(5), 217-223.
14. Chomette, G., Auriol, M., Labrousse, F., & Guilbert, F. (1989). Granulome réparateur central à cellules géantes des maxillaires: étude morphologique. *Revue de stomatologie et de chirurgie maxillo-faciale*, 90(2), 131-135.
15. Stimson, P. G., & McDaniel, R. K. (1989). Traumatic bone cyst, aneurysmal bone cyst, and central giant cell granuloma—pathogenetically related lesions?. *Journal of endodontics*, 15(4), 164-167.
16. De Lange, J., Van den Akker, H. P., Van Zanten, G. V., Engelshove, H. A., Van den Berg, H., & Klip, H. (2006). Calcitonin therapy in central giant cell granuloma of the jaw: a randomized double-blind placebo-controlled study. *International*

journal of oral and maxillofacial surgery, 35(9), 791-795.

17. EMC-2011. Granulome réparateur à cellules géantes. F.H. Dujardin, Professeur des Universités, chef de service (Franck.Dujardin@chu-rouen.fr). Service de chirurgie orthopédique, Clinique universitaire de chirurgie orthopédique et

traumatologique, 1, rue de Germont, 76031 Rouen cedex, France.

18. Marescaux, J., Miranda, E., Lantz, J. M., Dumény, E., & Pinget, M. (1989). Pièges diagnostiques et techniques de l'hyperparathyroïdisme primaire : réflexion à propos de 120 cervicotomies récentes. *Chirurgie*, 115:705-715.