A secondary hyperparathyroidism revealed by a mandibular radiolucency: case report

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Introduction:

Brown tumors of the jaw, also known as fibrocystic osteitis, are non-neoplastic osteolytic lesions characterized by proliferation of multinucleated giant cells in the maxilla and/or mandible, resulting from abnormal bone metabolism related to with hyperparathyroidism (HPT).

HPT is a frequent endocrine disorder (third most common of endocrine pathologies) which is characterized by hypersecretion of parathormone (PTH). [1,2] A distinction is made between primary HPT due to a disorder of the parathyroid glands and secondary HPT due to non-parathyroid disorders inducing chronic hypersecretion of PTH. Tertiary HPT is due to an autonomous secretion of parathyroid hormone in patients with long-standing secondary HPT.

Brown tumors are the terminal stage of bone remodeling occurring during primary, secondary or tertiary hyperparathyroidism and have specific clinical and histological features allowing the diagnosis establishment and its management. Early diagnosis and treatment of hyperparathyroidism has made the clinical appearance of associated bone and especially jaw lesions rare.

Case report:

Case History / examination:

A 28-year-old patient with chronic renal failure and undergoing dialysis 3 sessions per week was referred by the nephrology department to the dental medicine department for the oral cavity checkup before a kidney transplant. Intra-oral examination revealed insufficient hygiene without any tooth mobility. The panoramic X ray showed mandibular well limited radiolucency in the left and right molar area. (Figure 1)

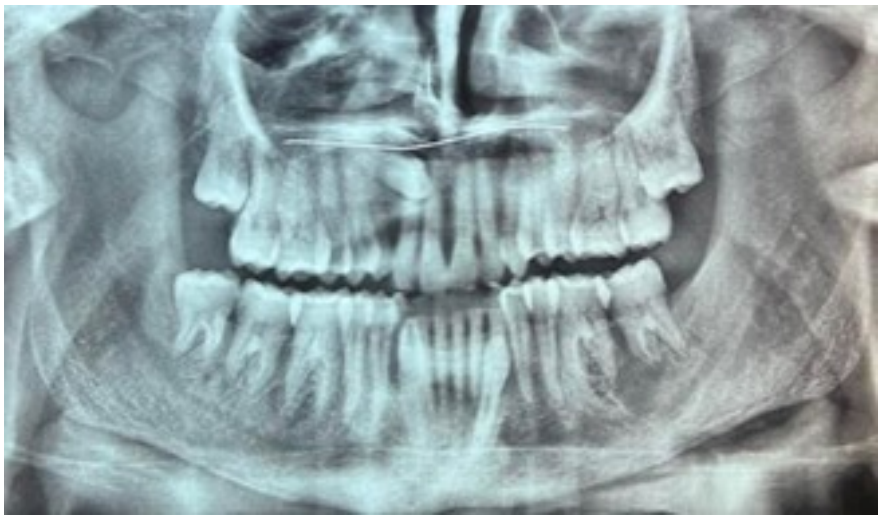

Figure 1: Panoramic radiograph revealed well-defined radiolucent lesions in the molar regions of both the left and right mandible.

Methods:

At this stage two differential diagnosis have been proposed:

- Periapical or latero-radicular cysts of inflammatory origin have been ruled out since both teeth were free from decay with a positive vitality test.

- Brown tumor, which might be secondary to renal failure, like in this case

Biological examination: showed hyperparathyroidism (PTH = 3919.0 pg/ml while the normal value is between 15 and 65 pg/ml) (Calcium = 2.38 mmol/l Normokalaemia) (Phosphorus = 1.70 mmom/l) and hyperphosphatemia. These values were in favor of hyperparathyroidism secondary to chronic renal failure.The patient was referred to an endocrinologist for more explorations. A thyroid gland ultrasound revealed the presence of parathyroid adenoma.

Conclusions and Results:

Since the treatment of brown tumors begins with the resolution of hyperparathyroidism, the patient was therefore referred for the surgical removal of the parathyroid adenoma. A panoramic X-ray 1 year after the intervention shows a reossification of mandibular radiolucency without resorting to surgery (Figure 2 and 3).

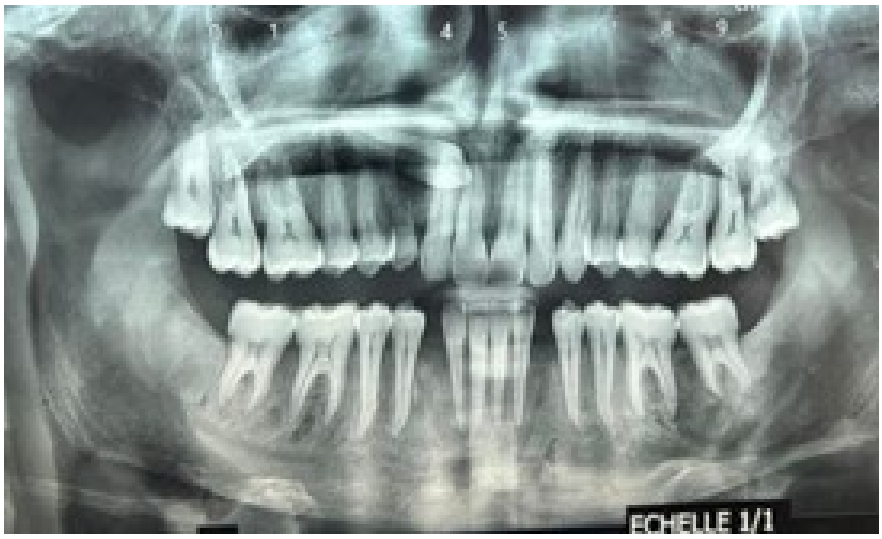

Figure 2: A panoramic radiograph obtained one-year post-intervention demonstrated evidence of reossification within the previously observed mandibular radiolucency, suggesting successful bone healing without the need for surgical intervention.

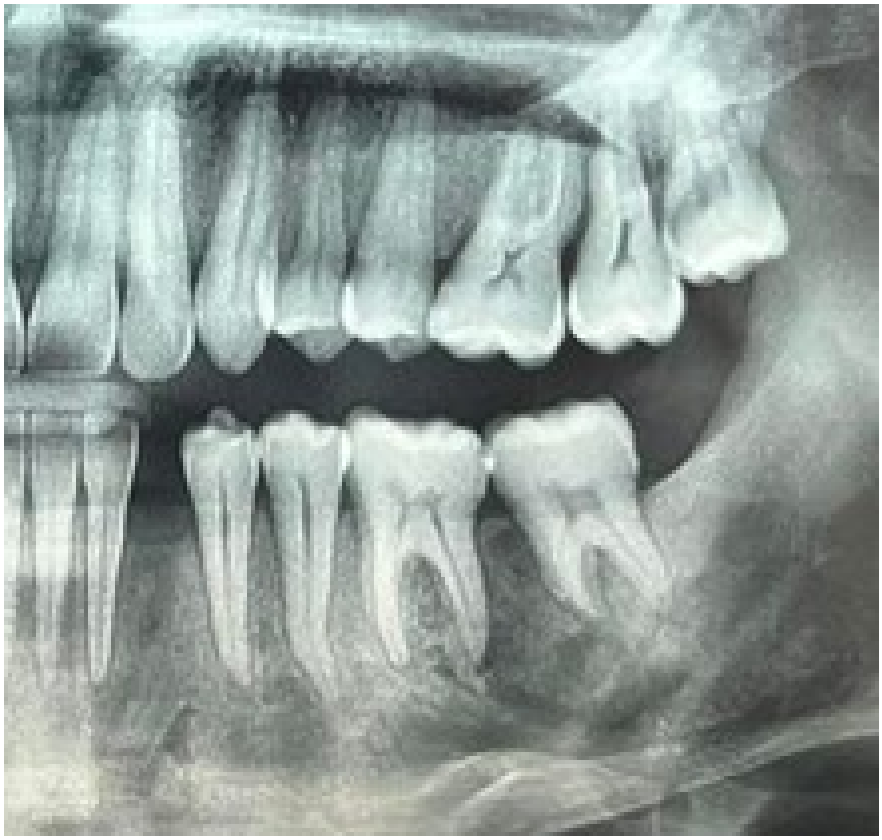

Figure 3: A panoramic radiograph obtained one year after the intervention demonstrated a significant reduction in the size and radiolucency of the previously observed mandibular lesion, suggesting successful treatment.

Discussion :

As found in the literature, brown tumors target a rather young population, with a highest incidence between 30 and 60 years old with female predilection, ration 2/3.[3,4] The predominant clinical manifestation is facial swelling which can be painful or painless. [5] When the mandibular lesion is not yet externalized, the discovery is then fortuitous as in our case. Brown tumors most often have many locations, particularly in the long bones such as the femur, tibia, clavicle, ribs and appear more rarely at the maxillofacial area.[6] When they develop in the facial region, the most affected bone is the mandibular bone, as in the case reported.[5,7] Bone demineralization is caused by hypersecretion of PTH, PTH stimulates osteolysis which allows the release of calcium into the blood. Primary hyperparathyroidism is caused by gland adenoma in 80% of cases. Secondary hyperparathyroidism is related to chronic renal failure or vitamin D deficiency. Tertiary hyperparathyroidism is caused by a long standing non treated secondary hyperparathyroidism leading to parathyroid hyperplasia. Diagnosis of maxillary bone lesions caused by hyperparathyroidism is based on clinical, radiological, biological and anatomopathological examinations.[6,8] Panoramic X-ray is a two-dimensional dental X-ray examination that shows maxilla mandibular structures and lesions. It can show fortuitously bone demineralization or well-defined bone radiolucency. Cervical ultrasound when hyperparathyroidism is suspected, can show adenoma or hyperplasia of the gland. Scintigraphy on MIBI is indicated in case of ectopic and posterior adenoma. Biological examination usually shows hyperparathyroidism associated to:

- Hypercalcemia and hypophosphatemia for Primary HPT

- Hypocalcemia and hyperphosphatemia for Secondary HPT

- increased alkaline phosphatase in both cases

The case reported here presented hyperphosphatemia (1.70 mmol/l) which is in favor of secondary hyperparathyroidism. The pathological examination with biopsy of the tumor was not carried out because there was no indication for surgical intervention for this patient. Regarding dental condition in the injured area, dental displacements and / or dental mobility due to the loss of the lamina dura, as well as bone lysis in the periapical region can be observed, in our patient the lesion was discovered at an early stage with absence of tooth mobility. The evolution and resolution of the brown tumor will be more or less rapid depending on the importance of the bone demineralization as well as the tumor localization. Either a resolution without sequelae (disappearance of mobility) or an early loss of teeth will be observed.[4,5] A loss of teeth vitality related to the lesion and/or canals calcification are also observed, which can lead to a clinical misdiagnosis and erroneous management consisting of antibiotics prescription or endodontic treatment. [2]The characteristics of the brown tumor are as follows:

- Macroscopic appearance: hard nodular type lesion, firm on palpation, with or without ulcerated oral mucosa, gray/brown in color, and haemorrhagic.[9,10]

- Microscopic appearance: it is a fibrous tissue composed of fibroblasts, multinucleated giant cells,surrounded by a stroma composed of numerous blood vessels with clusters of macrophages filled with deposits of hemosiderin and hemorrhagic areas.[9,10]

- Radiological aspect: radiolucent osteolytic lesion, uni or multilocular with defined contours, without invasion of the adjacent structures bone remodeling zones can be observed associated to significant osteoclastic activity.[9,10]

Loss of cortical bone and lamina dura at the level of the affected teeth may occur in more locally aggressive forms.[1,6] Brown tumors treatment begins with the resolution of the hyperparathyroidism to hope for the tumor's resorption and, if necessary, perform a secondary mass excision.[3,6,8,9] Total or subtotal parathyroidectomy is indicated especially in cases of hyperplasia or parathyroid adenoma depending on the number of glands affected, as is the case with our patient, the bone lesion spontaneously regressed after normalization of the constants, a control X-ray panoramic after one year is recommanded to objectify the regression of the lesion

In cases of secondary hyperparathyroidism, its resolution requires renal therapy (graft, renal transplantation, hemodialysis) and/or calcium and vitamin D supplementation.[7]

Conclusion:

Although rare, brown bone tumors should always be considered in cases of lytic bone lesions to avoid overlooking hyperparathyroidism.

Treatment of hyperparathyroidism brown tumors requires normalization of serum PTH, calcium, and phosphorus levels prior to any surgical procedure. Resolution of the lesion is expected after gland function has stabilized. In some patients, the brown tumor continues to grow even after hyperparathyroidism has been controlled; in these cases, resection is mandatory. The brown tumor is considered a reparative granuloma rather than a true neoplasm and has no potential for malignant transformation.

Key clinical message: This case report highlights the importance of considering secondary hyperparathyroidism as a potential cause of jawbone lesions. Early recognition and treatment of the underlying hyperparathyroidism can lead to resolution of the bone lesions and improve overall patient outcomes.

References:

1. Bilezikian JP, Khan AA, Silverberg SJ, et al. Evaluation and Management of Primary Hyperparathyroidism: Summary Statement and Guidelines from the Fifth International Workshop. *J Bone Miner Res* . 2022;37(11):2293-2314. doi:10.1002/jbmr.46772. Bilezikian JP. Primary Hyperparathyroidism. *J Clin Endocrinol Metab* . 2018;103(11):3993-4004. doi:10.1210/jc.2018-012253. Shavlokhova V, Goeppert B, Gaida

MM, et al. Mandibular Brown Tumor as a Result of Secondary Hyperparathyroidism: A Case Report with 5 Years Follow-Up and Review of the Literature. *Int J Environ Res Public Health* . 2021;18(14):7370. doi:10.3390/ijerph181473704. Manjunatha BS, Purohit S, Harsh A, Vangala N. A complex case of brown tumors as initial manifestation of primary hyperparathyroidism in a young female. *J Oral Maxillofac Pathol JOMFP* . 2019;23(3):477. doi:10.4103/jomfp.JOMFP_166_195. Heimburger C, Andres E, Rust E, et al. Imagerie morphologique et fonctionnelle des tumeurs brunes. À propos d'un cas de localisation maxillo-mandibulaire. *Rev Médecine Interne* . 2013;34(6):377-381. doi:10.1016/j.revmed.2013.02.0076. Hong WS, Sung MS, Chun KA, et al. Emphasis on the MR imaging findings of brown tumor: a report of five cases. *Skeletal Radiol* . 2011;40(2):205-213. doi:10.1007/s00256-010-0979-07. Brabyn P, Capote A, Belloti M, Zylberberg I. Hyperparathyroidism Diagnosed Due to Brown Tumors of the Jaw: A Case Report and Literature Review. *J Oral Maxillofac Surg Off J Am Assoc Oral Maxillofac Surg* . 2017;75(10):2162-2169. doi:10.1016/j.joms.2017.03.0138. Palla B, Burian E, Fliefel R, Otto S. Systematic review of oral manifestations related to hyperparathyroidism. *Clin Oral Investig* . 2018;22(1):1-27. doi:10.1007/s00784-017-2124-09. Xie C, Tsakok M, Taylor N, Partington K. Imaging of brown tumours: a pictorial review. *Insights Imaging* . 2019;10(1):75. doi:10.1186/s13244-019-0757-z10. Adnane A, Merzem A, Harmak M, et al. Imagerie des tumeurs brunes maxillo-mandibulaires multiples: à propos d´un cas. *Pan Afr Med J* . 2021;38(4). doi:10.11604/pamj.2021.38.4.27141

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