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Case Report

Peripheral giant cells granuloma as a rare early manifestation of primary hyperparathyroidism

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ABSTRACT

Primary hyperparathyroidism occurs due to parathyroid adenoma, which as initial presentation in most cases includes recurrent nephrolithiasis (10-25%). Giant cell tumors (GDD) also called osteoclastomas or brown tumors affect the second decade of life and are currently a rare manifestation of primary hyperthyroidism. The incidence of lesion appearance in the maxillary bones is 4.5%. We presented the case of a 36 year old female patient with history of hypertension, who developed an increase in volume in the right maxillary region of 4×3 cm, with incapability of complete occlusion of dental arch, solid dysphagia, biopsy was performed with peripheral giant cells granuloma as a result, PTH serum levels were requested, with result of 1175 pg/ml and serum calcium of 13.24 mg/dl. Parathyroid gammagram was performed with hyperfunctioning parathyroid tissue. Patient underwent a selective parathyroidectomy. She had an adequate postoperative evolution and was discharged without complications. The patient had adequate follow up by head and neck surgery in external consultation, serum calcium 7.66 mg/dl, decrease of volume in right maxillary region to 3×3 cm; pathology report with parathyroid adenoma. Surgical treatment of brown tumor is still pending by the maxillofacial surgery department.

Keywords: Giant cell tumors, Osteoclastomas, Brown tumors, Primary hyperparathyroidism, Parathyroid adenoma

INTRODUCTION

Hyperparathyroidism could be described as an endocrine disorder resulting from increased secretion of parathyroid hormone and is characterized by hypercalcemia due to increased mobilization of calcium from bone to circulation.[1] Primary hyperparathyroidism occurs due to parathyroid adenoma, which as initial presentation in most cases includes recurrent nephrolithiasis (10-25%), neuropsychiatric disorders, and peptic ulcer. GDD also called osteoclastomas or brown tumors when they are within the endocrinological sphere, are one of the least frequent, most controversial and least predictable tumors in their behavior. They are produced as a consequence of excessive osteoclastic activity, as occurs in the case of hyperparathyroidism.[2] They can be located in any bone, preferably affecting the ribs, the clavicle and the pelvis. The incidence of lesion appearance in the maxillary bones is 4.5%, being more frequent in the mandible than in the upper maxilla.[3]

CASE REPORT

A 36 year old female with a history of hypertension, consulted due to increase in volume in right maxillary region with incapability of complete occlusion of dental arch, solid dysphagia progressing afterward to liquids dysphagia,10kg weight loss in 3 months and repeated episodes of oral bleeding. A biopsy of the right retromolar area was performed by maxillofacial surgery

reporting peripheral giant cells granuloma. A head and neck tomography with contrast was performed reporting heterogeneous lytic lesions in the upper maxilla and right area of the jaw. Due to these findings and the association of giant cells granuloma, better known as brown tumor, with hyperthyroidism; PTH levels were requested, with a result of 1175 pg/ml and serum calcium 13.24 mg/dl.

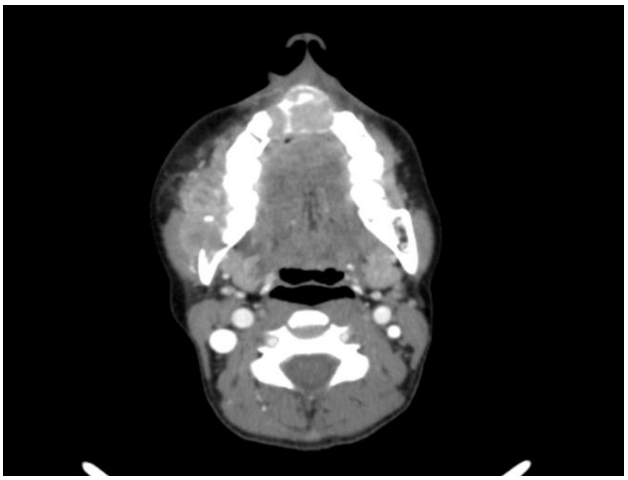

Figure 1: Preoperative abdominal CT, showing a lesion located at the central level in the upper maxilla that extends towards the hard palate 16×17×22 mm.

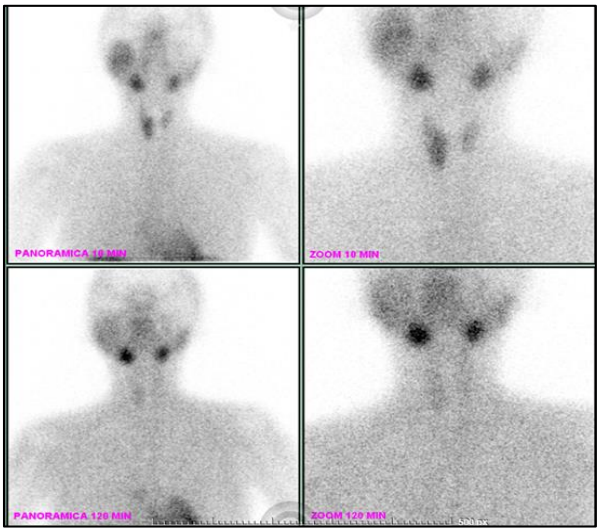

Figure 2: Parathyroid adenoma located in right paraesophageal area, adjacent to ipsilateral posterior thyroid border.

The patient was referred to endocrinology and a parathyroid gammagram was performed with hyperfunctioning parathyroid tissue, concluding parathyroid adenoma located in the right paraesophageal area, adjacent to ipsilateral posterior thyroid border. Maxillofacial surgery was consulted, and considered the patient candidate to resection of right maxillary tumor; however due to measurements of the tumor and finding of parathyroid adenoma, it was suggested as first approach right parathyroidectomy. On 20 September 2022, right parathyroidectomy was performed by head and neck surgery with the following findings: right parathyroid of approximately 4×2 cm with normal macroscopic characteristics; ipsilateral recurrent laryngeal nerve without damage. Subsequently, she had an adequate postoperative evolution and was discharged without complications. The patient had adequate follow up by head and neck surgery in external consultation, serum calcium 7.66 mg/dl, with no spontaneous oral bleeding and volume decrease in the right maxillary region, pathology report with parathyroid adenoma. Treatment by maxillofacial surgery is still pending, further volume decrease is expected to perform surgery.

DISCUSSION

Brown tumor, also called osteoclastoma, is one of the most characteristic bone manifestations of hyperparathyroidism that appears in the context of advanced disease. Although, it is more frequent as the first manifestation of primary hyperparathyroidism. It affects the second decade of life and is currently a rare manifestation due to early diagnosis by incorporating serum calcium into routine laboratory tests in daily clinical practice.[3] This was such the case presented before, as the patients was a young woman in her third decade and did not have advanced hyperparathyroidism, but a osteolytic lesion and after finding it was a giant cells granuloma the possibility of primary hyperparathyroidism was considered.[3]

Clinical symptoms caused by brown tumors depend on their location and size; they range from small asymptomatic lesion, discovered accidentally by radiological examination, to a large locally destructive lesion resulting in a variety of symptoms that are mostly related to facial deformation and disfiguration, such as difficulty in chewing, talking, and breathing.[1] This coincided with our patient who lost weight due to facial deformation that led to difficulty in chewing.

Brown tumor is defined as a focal and lytic bone lesion, non-neoplastic, secondary to a bone metabolic disorder. It is a localized form of cystic fibrous osteitis that is reached after bone marrow resorption that is progressively replaced by fibrous tissue with cystic changes. Due to the rapid bone turnover due to the action of PTH, areas of local hemorrhage, reparative granulation tissue and vascular fibrous proliferation are created that replace healthy bone.[3]

The radiological features of brown tumors in the reported cases mainly consisted of single or multiple well-defined osteolytic lesions, single or multi-lobular, usually with bone expansion and can demonstrate bony destruction and be associated with pathological fractures. There is also variability in its radiological characteristics, which can include an ill-defined lesion, mixed lytic/sclerotic lesion or margin, with adjacent soft tissue involvement.

In these situations, a malignant lesion should be considered in the differential diagnosis and histologic confirmation is recommended.[4]

Brown tumors do not show pathognomonic histological changes. Histological reports of injured tissue describe connective tissue rich in notched oval cells in which giant cells, hemorrhagic foci and hemosiderin can be observed.[5]

Treatment depends on the evolution of the patient, Silverman et al reported that the excision of a brown tumor was not necessary when HPT resolved, whereas Steinbach et al reported that brown tumors could be treated by curettage or local radiotherapy.[6,7] However, even though bone lesions tend to regress. After parathyroid surgery, the majority of the authors had reported a resection of any remaining brown tumors or when bone healing was compromised (symptomatic or disfiguring), but only after HPT was controlled.[8] In our case, maxillofacial surgery decided first to wait for the primary hyperparathyroidism to be resolved.

CONCLUSION

Brown tumors in upper maxilla is a rare entity, with only the incidence of 4.5% and is often related to primary hyperthyroidism. Early diagnosis has been possible before further osteolytic damage can occur due to PTH and calcium serum levels and CT scan for characterization of the lesion to take biopsy to establish diagnosis. Treatment priority is removal of the adenoma, whether the brown tumor requires surgical intervention, depends on the tumor evolution of the patient in the postoperative.

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