Maxillary brown tumor as a first sign of parathyroid adenoma : A case report

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ABSTRACT

Introduction: Brown tumor is a rare late-stage skeletal change caused by long-term stimulation of excess parathyroid hormone. It is not neoplastic, but a reparative cellular process. Solitary maxillary brown tumor as initial presentation of primary hyperparathyroidism is rare.

Case presentation: We present the first case of solitary maxillary brown tumor in a 67-year-old woman with initial presentation of a tumor filling the left maxilla. Underlying long-standing primary hyperparathyroidism caused by a large parathyroid adenoma was finally diagnosed.

Conclusion: Biopsy of the suspicious bone tumor and blood tests for calcium and parathyroid hormone level are crucial and essential to reach the correct diagnosis.

Key words: Brown Tumor, Hyperparathyroidism, Giant Cell Tumor, Maxilla

INTRODUCTION

Brown tumor is an unusual bone change created by primary hyperparathyroidism affected less than 2% of primary hyperparathyroidism patients [1, 2]. Brown tumors have no neoplastic behavior but are a reparative cellular response. The term Brown tumor is derived from the characteristic macroscopic appearance of brownish material within the cystic lesion [2]. Brown tumors often develop at multiples sites including clavicle, ribs, tibia, femur, and pelvic bones [3]. Maxillofacial brown tumors are rare; when they occur in this region, they usually involve the mandible, whereas the maxilla is only rarely affected [3-5].

We present the case of an elderly woman with a maxillary swelling that was found to be the first clinical manifestation of primary hyperparathyroidism.

CASE REPORT

A 67-year-old Tunisian woman was referred to our Department, complaining of facial pain and deformity began 4 months ago. There was no family history of peptic ulcer, parathyroid disease, or any other endocrinopathy. There was no personal history of trauma to the facial bones, nasal obstruction, dental problem, epistaxis, visual change, urolithiasis, drugs consumption, exposure to ionizing radiation or industrial toxins, or use of tobacco or alcohol.

On physical examination, the patient appeared well. A facial asymmetry was found due to swelling of left maxilla. The lesion had 4 cm in major axis, fixed, painless and covered with healthy skin.

Her head and neck were normal; no cervical masses were palpated. Rigid nasal endoscopy, with a 0° endoscope, was normal: no medialization of the left lateral nasal wall and no signs of mucosal alteration.

CT scan of the maxillofacial region revealed an osteolytic, bone-expanding lesion depending of frontal process of the left maxillary sinus (20 × 18 × 16 mm) (Fig 1).

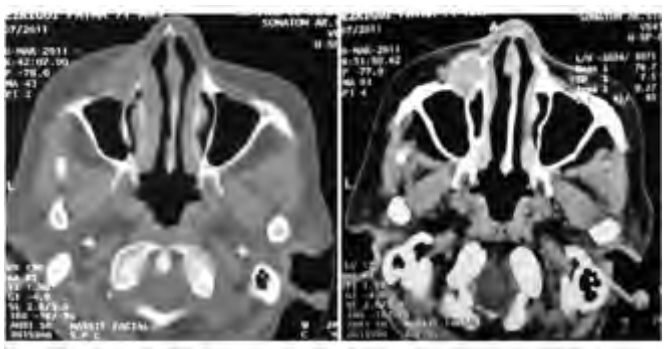

Figure 1 : Axial computed tomography of the paranasal sinuses without and post contrast showing a round osteolytic lesion lesion depending of frontal process of the left maxillary sinus (red arrow).

Under general anesthesia, an excision biopsy was performed using the Caldwell-Luc approach (fig 2).

Figure 2 : Per operative view of the lesion.

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The histological section ware compatible with a giant cell lesion, a brown tumor among the possible causes. Blood tests after histology demonstrated elevated calcium (2,6 mmol/L) and parathyroid hormone (PTH) concentrations (645 pg/mL). According to laboratory tests, ultraso¬nography was con¬sidered.
In ultrasonography, a 11 × 14 × 27 mm mixed hypo echo mass lesion was seen in posterior aspect of left thyroid lobe compatible with probable vascular parathy¬roid mass.
After diagnosis of parathyroid mass, surgical exci¬sion was performed. Pathological and surgical findings were compatible and consistent with parathyroid hyperplasia.
Postoperatively the patient became hypocalcemic and we began oral calcium replacement therapy, the parathyroid hormone level was normal. The patient was free of symptoms and living well after two years of follow-up.
Diagnosis of brown tumor due to primary hyperpara thyroidism related to parathyroid hyperplasia was achieved according to clinical and pathological findings.

DISCUSSION

Brown tumor is caused by hyperparathyroidism, which may be primary, secondary or tertiary. The major determinant may be the chronicity of excess secretion of and stimulation by PTH, rather than the PTH level per se [6]. The combination of vitamin D deficiency and primary hyperparathyroidism predisposes patients to develop the classic skeletal changes of primary hyperparathyroidism [4].
The incidence of bone lesions in patients with hyperparathyroidism has fallen from 80% to a current 15%, a reduction that is attributed to better hypocalcemia monitoring in asymptomatic patients, and to the wider use of biochemical analyses [7].
Primary hyperparathyroidism affects women more than men by almost 3/1. It can occur at any age, but the majority of cases are postmenopausal women. Resendiz-Colosia et al. have reported a series of 22 cases of maxillofacial brown tumor, showing that 91% of them were women [4].
The symptoms caused by these lesions depend on their size and location. In the maxilla, they can cause pain or deformity, as in our patient. In other cases, the lesions were asymptomatic and the diagnosis occurred accidentally as a result of a radiological examination [7].
Brown tumors are consequence of an imbalance between osteoclast and osteoblast cells activity, which can lead to bone resorption and its substitution by fibrous tissue [8].
So brown tumor is just a reaction rather than a true tumor, but it may be recognized as a primary bone tumor and be excised, as in our case.
Although surgical resection may not be necessary, biopsy of the tumor is a crucial and essential step to exclude malignancy and other bone disease, and it helps in avoiding unnecessary extensive operation [1, 2]. The differential diagnosis should include giant-cell tumor of the bone, giant-cell reparative granuloma, aneurismal bone cyst, and osteosarcoma [9].
Pathology cannot distinguish giant cell tumor of the bone from the brown tumor of hyperparathyroidism [10], however, they can be easily differentiated on the basis of laboratory findings.
Blood calcium and PTH levels should both be checked, because a primary hyperparathyroid patient may have a normal blood calcium level due to many factors, such as vitamin D deficiency [2, 6].
There is agreement as to the treatment of choice for primary hyperparathyroidism being parathyroidectomy; however, opinions are divided as to the treatment of bone lesions.
Most of the bone lesions will regress with time after parathyroidectomy, thus, surgical removal of the brown tumor may not be necessary [6] . In the case of large destructive cysts, the amount of tissue damaged may be so great that there are few possibilities of remodeling once normocalcemia has been achieved [7]. In these situations, or in cases of special anatomical sites especially the skull base, the excision of the brown tumor should be performed to avoid facial deformation or damage to vital structures [6]. Some authors recommend initial treatment with systemic corticosteroids in order to reduce the tumor size [5].

CONCLUSION

The incidence of hyperparathyroidism with advanced bone lesions is rare. Due to recent improvements in analytical techniques, the diagnosis usually occurs when the disease is in an asymptomatic phase. The treatment of choice for bone lesions is a parathyroidectomy; however, in the case of larger lesions, or those that persistently grow in spite of treatment, or those lesions causing incapacity, curettage and associated enucleation should be conducted.

Conflicts of interest: Authors have declared that no competing interests exist.

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