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Primary Hyperparathyroidism Revealed by a Brown Tumor of the Maxilla: A Case Report

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Abstract
Introduction: Brown tumors are osteolytic lesions that rarely reveal hyperparathyroidism. They usually occur at the terminal stage of primary or secondary hyperparathyroidism. We report the case of a patient with primary hyperparathyroidism revealed by a jaw's tumor, at the ENT and Maxillo-facial department of Mohamed VI University Hospital, Oujda, Morocco. Case report: A medical examination of A 48-year-old woman with a left nasal obstruction associated to an ipsilateral tumefaction progressively increasing in size. The CT scan showed an aggressive osteolytic process of the maxillary sinus. The diagnosis of the brown tumor was suspected on a biological assessment highlighting an hypercalcemia. Etiological research has revealed a parathyroid adenoma. The parathyroid hormone test: 322 pmol / L confirmed the diagnosis. The surgery consisted of a conservative lumpectomy with left parathyroidectomy. The anatomopathological result showed a benign giant cell tumor of the maxillary sinus. Conclusion: We recall through this observation, and view to the insidious side of brown tumors, two essential points: the difficulty of establishing the diagnosis of osteolytic processes of the maxillary sinus and the need to think about an hyperparathyroidism in front of a giant cell lesion.

Keywords: Maxillary Sinus, Brown tumor, Hyperparathyroidism

Introduction

The Brown tumor of the jaw is a lesion that affects patients with hyperparathyroidism. She appears as an expansive osteolytic lesion, mostly affecting the mandible, ribs, pelvis and femur. Maxillary involvement is exceptional. In 81% of cases, the tumor is due to an adenoma of the parathyroid gland. Sometimes, it is the result of a chronic renal failure. This tumor is usually painful with a slow growth and it can become aggressive and destructive (Ercihan Guney 2001).

In the maxillofacial region, this tumor is painful, difficult to palpate, can take a large volume and thus deform the bone, leading to a masticatory gene. This work describes a clinically and histologically diagnosed case of brown tumor that was located in the left maxillary sinus.

The aim of this work is to remind endocrinologists and maxillofacial surgeons that these tumors should be taken into account (Ercihan Guney 2001).

Case report

A 48-year-old patient, admitted for painful tumefaction of the left cheek, gradually progressing during the last 6 months. This tumefaction is associated to a nasal obstruction, without epistaxis or other ocular symptomatology. The clinical history does not include any personal or family history of endocrinopathy.

The clinical examination revealed facial asymmetry due to a large hard gingival-maxillary mass, filling the left upper vestibule and deforming the hard palate. The rest of the somatic examination did not reveal any abnormalities, including no adenopathy or bone pain. Nasofibroscopy revealed a huge mass filling the entire left nasal cavity without alteration of the pituitary mucosa.

Computed tomography showed osteolytic tissue mass, heterogeneous with numerous calcifications, centered on the left maxillary sinus measuring 50 / 40 mm, extending into the ipsilateral nasal cavity with involvement of the nasal septum. At the top, it lyses the floor of the orbit. At the bottom, it lyses the hard bony palate, which is prominent into the oral cavity. The anterior extension has passed the anterior bone wall of the maxillary sinus and extends to the soft parts [Fig. 1].

In front of these radiological data "osteolytic image", a biological assessment was carried out; the result indicates coexistence of hypercalcemia at 126 mg / L and hypophosphoremia at 21 mg / L. Alkaline phosphatase value was 217 U / L whereas the normal value is 30 - 100 U / L. The renal status (urea, creatinine) and protidemia were without abnormality.

Biologic exploration confirmed the diagnosis of primary hyperparathyroidism, initially expressed by a brown tumor of the maxilla, leading to an etiologic investigation. In addition, cervical sonography revealed a nodule strongly suggestive of a parathyroid adenoma in the left thyroid lobe [Fig. 2].

The bioassay of PTH indicated a high value of 322 pmol / L while the normal values have to be from 9 to 55 pmol / L. Thus, the removal of the parathyroid adenoma was performed. In parallel the anatomopathological report revealed a parathyroid adenoma.

The patient underwent a transfacial left medial maxillectomy [Fig. 3]. The removal of the tumor is performed in one piece followed by a histopathological study returned in favor of a benign tumor with giant cells of grade II. The post-operative blood check shows that calcemia and phosphoremia have returned to normal state. Five months later the Parathyroid hormone (PTH) assay shows normal value of 33 pmol/L.

Discussion

Currently, hyperparathyroidism is present in 75 to 80% of cases during asymptomatic hypercalcemia. It can also be revealed by renal failure in the terminal stage or by cardiovascular disorders. Bone manifestations of hyperparathyroidism: bone cysts, osteoporosis, sub-periosteal résorption and brown tumors are the late expression of the disease. These signs are few and occur in 5 to 15% of cases. In addition, it is exceptional that a brown tumor represents the first and only clinical sign of hyperparathyroidism. The reported incidence of these hyperparathyroidisms is 1.5 to 1.7% in secondary hyperparathyroidism and 3% in primary hyperparathyroidism (Meredith N 2002).

Most hyperparathyroidism cases are secondary to a primary hyperparathyroidism. This results from 80% of cases of parathyroidian adenoma, and more rarely of hyperplasia (15 %).

Primary hyperparathyroidism frequently affects patients over the age of 50, especially postmenopausal women, with a predilection for the female sex in benign hyperparathyroidism. Brown tumors can affect the entire skeleton. The most frequent locations are pelvis, flank, femur, mandible and hands. The maxillary localization is extremely rare.

Clinically, the severity of the symptoms depends on the size of the process and its location. Indeed, the brown tumour has no distinctive clinical features of other maxillary processes. It is generally in form of a jugal, palatal and/or gingival bone tumefaction with distortion and asymmetry of the face, pain and mobility, and even a fall of the teeth. The diagnosis is made fortuitously after a routine radiological examination (Meredith N 2002).

Radiologically, it is manifested by monogeodic or multilocular bone lysis with unspecified limits. This lysis is often responsible for a cortical rupture and can, therefore, suggest a malignant aspect. The CT scan showed a mass of tissue nature, taking the product of contrast and a perfect respect of the soft parts. The maxillary sinus is often filled with an intra-sinusal mass appearance. Standard radiographs of the skeleton let searching for other locations and looking for a renal lithiasis. Furthermore, other radiological manifestations of hyperparathyroidism include sub-periosteal resorption that are usually localized at level of the phalanges (C. Heimburger 2013).

It's known that cervical ultrasound and computed tomography are necessary to detect parathyroid lesions caused by hyperparathyroidism. CT after technetium injection (Tc - 99 m) is the best way to detect lesions in the parathyroid glands or ectopic tissue before surgery. In a 20-year retrospective study of 32 giant bone cell lesions, seven were located in the region of the head and neck. Four out of seven were giant cell granulomas, the remaining three were true giant cell tumors, which illustrates the exceptional character of the brown tumour. Thus if based only on histology data, the distinction between the Brown tumor and other giant cell lesions is not evident. Indeed, brown tumors are secondary to non-pathognomonic histological changes that can also be seen in a giant cell granuloma, an anevrysmal cyst or in fibrous dysplasia. The clinical history and the results of the phosphocalcic assessment are, therefore, essential, in particular the increase of the parathyroid hormone, to make the diagnosis (C. Heimburger 2013).

It is important to note that brown tumors are non-neoplastic lesions, without malignant potential, in comparison with giant cell tumors that are susceptible to malignant transformation with eventual lung metastases. The reparative granuloma is a quite different lesion from the brown tumor; it is a kind of tumor that affects young population. Mechanism of pathogenesis remains unknown, however, some authors define trauma as a triggering factor (Mohammed Farouk 2017).

It is important and logical to consider that the treatment of hyperparathyroidism should be the first step in the management of these patients. It is generally accepted that parathyroidectomy is the treatment of choice for primary hyperparathyroidism, but the management of bone lesions is not codified. Indeed, the evolution of Brown tumors after the parathyroidectomy is variable depending on their composition. Most of the authors believe that the spontaneous regression of these lesions is possible after the correction of hyperparathyroidism, without lytic bone lesions. According to some authors, the complete disappearance of the lesion was found six months after the treatment of hyperparathyroidism. Other authors reported that spontaneous bone regeneration might take several years to restore normal facial morphology (Mohammed Farouk 2017).

When the destructive lesions affect the function of an organ, the tissue lesions produced cannot be repaired despite obtaining a normal rate of calcemia. In these situations, when the lesions persist after treatment of hyperparathyroidism or continue to develop despite hormonal control, there are various recommendations. In this regard, Yamazak proposes the enucleation and the curettage of the tumor, however Cicconetti, recommends surgical excision of the tumor to stop bone destruction, followed by a second operation, parathyroidectomy, to suppress secretion of parathyroid hormone (A. Henry 2018).

Conclusions

A brown tumor rarely reveals a state of hyperparathyroidism. The main problem is to eliminate other osteolytic lesions. The presence of giant cells must evoke the diagnosis of a brown tumor and a hormonal assessment is

needed to look for hyperparathyroidism. The treatment is based on parathyroid adenoma surgery to avoid operating the tumor, which usually regresses after a parathyroidectomy (F. Antin 2018).

Figure 1: Computed tomography scan of maxilla. Soft tissue mass involving the left maxilla

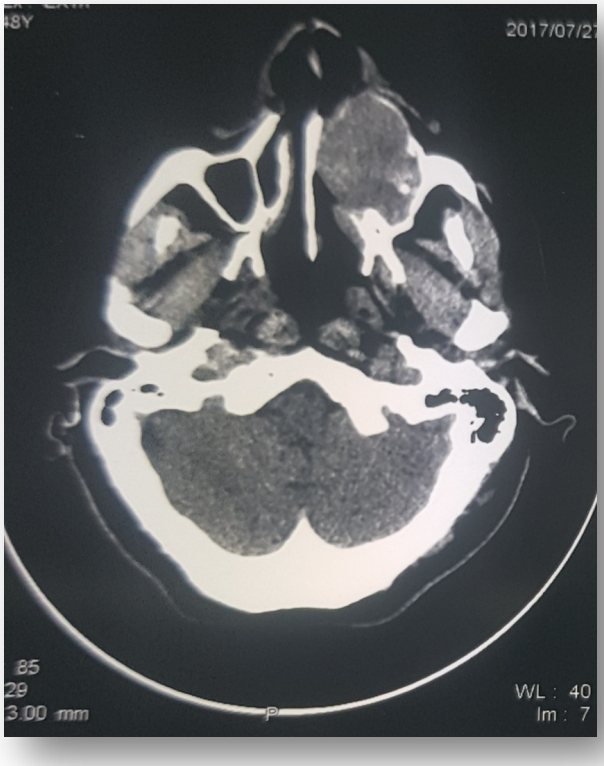

Figure 2: Cervical sonography showing a parathyroid adenoma

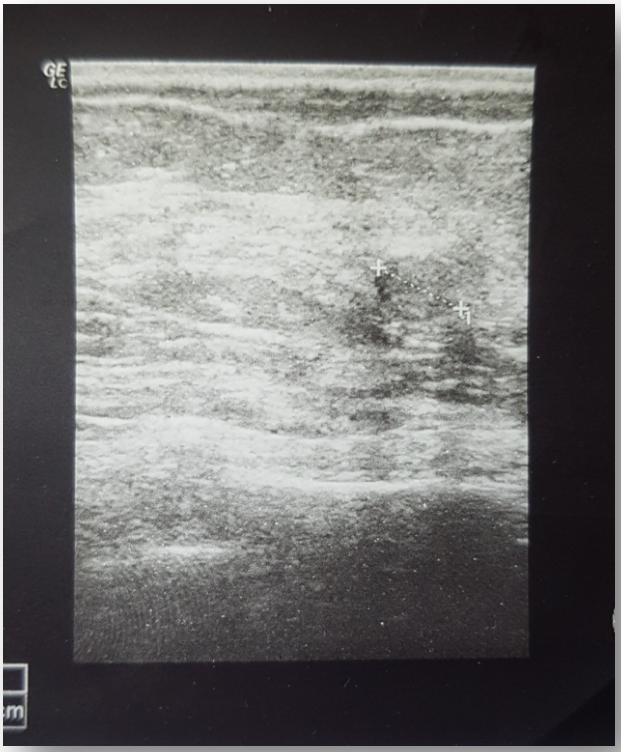

Figure 3: Transfacial left medial maxillectomy.

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