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A giant brown tumor of the face caused by secondary hyperparathyroidism in a young chronic hemodialysis patient: a case report

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ABSTRACT

Brown tumour represents a serious complication of hyperparathyroidism. Differential diagnosis, based on histological examination, is only presumptive and clinical, radiological and laboratory data are necessary for definitive diagnosis. Here we describe a case of a brown tumour localised in the maxilla due to secondary hyperparathyroidism in a young male with chronic renal failure. Hemodialysis and pharmacological treatment were unsuccessful in controlling secondary hyperparathyroidism making it necessary to proceed with a subtotal parathyroidectomy. The proper timing of the parathyroidectomy and its favourable effect on regression of the brown tumor made it possible to avoid a potentially disfiguring surgical removal of the brown tumor.

Keywords: brown tumor, hyperparathyroidism

INTRODUCTION

Secondary hyperparathyroidism is a common complication in chronic renal failure. It is characterized by excessive synthesis and secretion of parathyroid hormone, parathyroid hyperplasia and abnormal bone mineral metabolism [1, 2]. Brown tumors are classic manifestations of hyperparathyroidism. They usually occur during severe forms. These are benign lesions with osteolytic giant cells secondary to the action of parathyroid hormone on bone. These lesions are rare and occur in 1.5 to 1.7% of patients with secondary hyperparathyroidism [3, 4]. The most common sites are the ribs, clavicles, pelvic bone and the mandible. The maxillary brown tumor is rare [5]. Different techniques **for** bone imaging are necessary for the diagnosis and monitoring of these bone tumors, and should be combined with those for the etiologic diagnosis of hyperparathyroidism. The diagnosis of hyperparathyroidism avoids the surgery of maxillary brown tumors that usually regress after removal of the parathyroid lesion. We report an unusual large brown tumor of jawbones after secondary hyperparathyroidism in patient with end stage renal disease. The originality of this case

is the gigantic volume of the brown tumor, in addition to the unusual location, and the severe nature of hyperparathyroidism.

MATERIALS AND METHOD

Observation:

We report the case of Mr B.T who was born in 1988, in terminal chronic renal failure following an indefinite nephropathy who has been on periodic hemodialysis for 5 years with two sessions per week for 5 hours per session. In March 2013, the patient was seen for a tumefaction of the face with a rapidly increasing volume (Figure 1). He reported that the lesion appeared two months before. Clinical examination of the face objectified a tumefaction measuring approximately 8cm x 4cm x 3cm, painless, symmetrical, facial, hard, with limited mouth opening without involvement of the facial and trigeminal nerves. The intraoral examination showed no abnormality. The cervical examination has not objectified any mass. The generalbody examination was without abnormalities. A CT scan of the face objectified a bloated appearance and bone blown of the upper and lower jaws, which are dense with serpiginous resorption bays, that are regular and symmetrical, with a thickened pepper and salt appearance of the cranial vault, without anomalies of the soft parts, and without lysis or alveolar condensation (Figure 2).

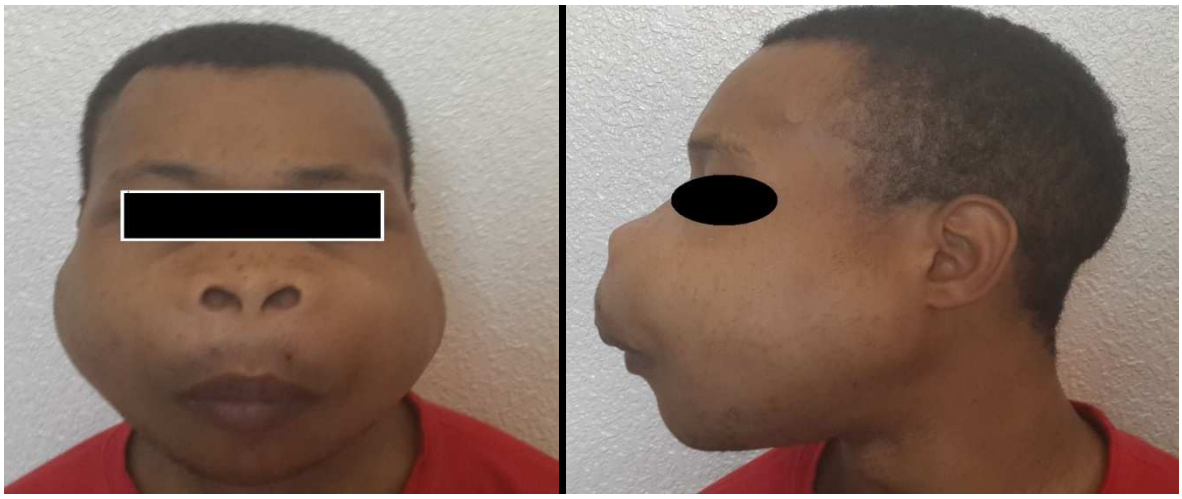
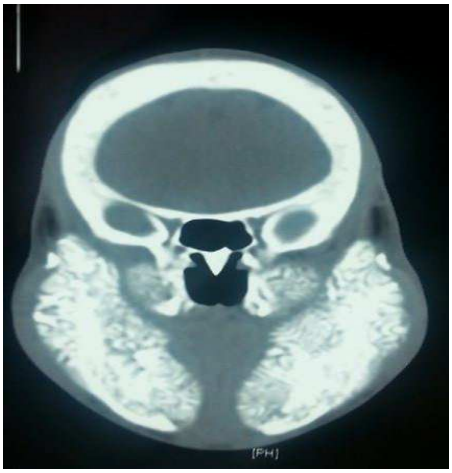
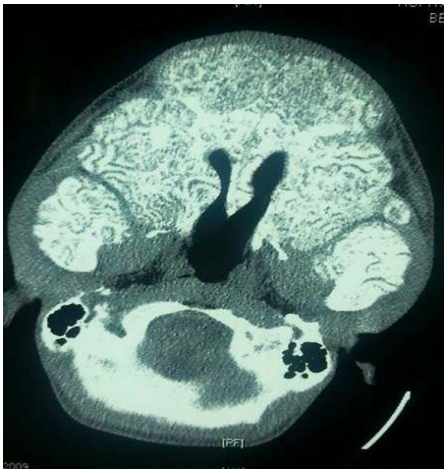
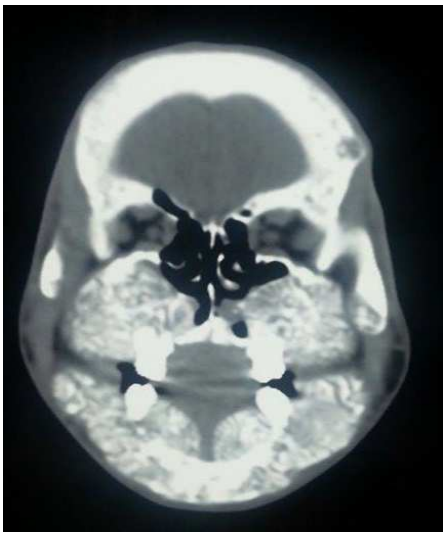
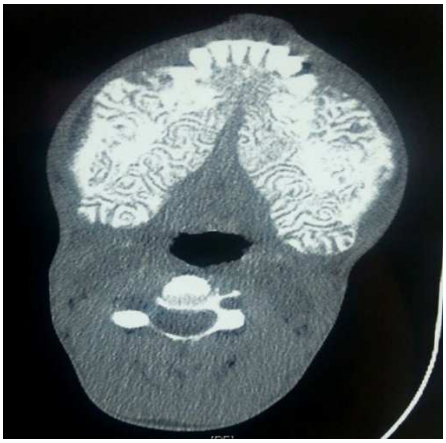


Figure1: large tumefaction of the face









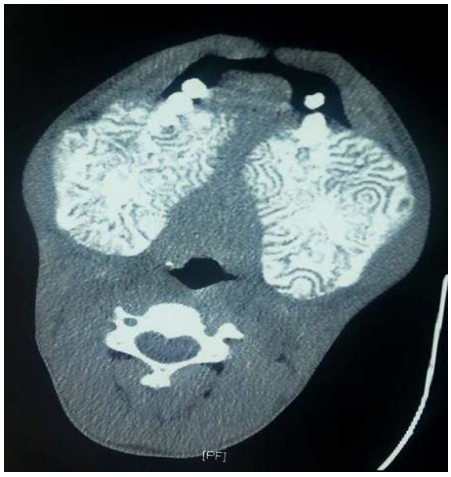


Figure 2: A CT scan of the face objectified a bloated appearance and bone blown of the upper and lower jaws.

One bone biopsy was performed, and the histologic study objectified fibroblastic cell proliferation, with inflammatory granulation tissue that is containing giant cells and a very abundant hemosiderin pigment. Laboratory tests showed a hyperparathyroidism with a parathormone rate of 4042 pg / ml, alkaline phosphatase to 2996 IU / l, hypocalcemia at 70 mg / l, hyperphosphataemia to 66 mg / l, and aplastic normocytic normochromic anemia of 8 g / dl. The rest of the biological assessment was without abnormalities, including rates of ferritin, vitamin D, and serum aluminum. Cervical ultrasound revealed retrothyroidien nodules, that are suggestive of bilateral parathyroid adenoma. Then, the diagnosis of secondary hyperparathyroidism complicated by a brown tumor of the face was made. Surgical treatment with subtotal parathyroidectomy of 7.8 was performed in this patient to curb the osteolytic process. In the immediate postoperative period, the biological tests showed profound hypocalcemia at 30 mg / l, hypophosphatemia at 15 mg / l, and hypoparathyroidism to 45pg / l. Then, the patient was placed under intravenous calcium supplementation, with a control serum calcium at 80 mg / l. A month after of the surgery, the patient was declared out of the hospital with a prescription of 4 g of calcium gluconate per day orally. Seven days after, the patient has stopped the calcium therapy and developed generalized seizures with impaired consciousness. Laboratory tests showed a profound hypocalcemia at 56 mg / l. A brain CT scan was normal. The treatment consisted of an antiepileptic drug (diazepam) with a correction of hypocalcemia with intravenous calcium gluconate. The evolution was marked by the decline of seizures with onset of dyspnea and fever. Laboratory tests showed a serum calcium control at 75 mg / l with a high C-reactive protein of 150mg / l. A chest radiograph performed objectified a white right lung. Then, the patient was put under bi-antibiotics for severe aspiration pneumonia, but no improvement was noticed, the patient passed away because of severe sepsis.

RESULTS AND DISCUSSION

Our case is a secondary hyperparathyroidism complicated by a large brown tumor of unusual location in a patient in end stage renal disease. In some cases like the present one, HPT is diagnosed by the presence of osteolytic lesions called brown tumors. In fact, brown tumors are one of the complex pathological expressions of osteitis fibrosa cystica. Osteitis fibrosa cystica is a late manifestation of severe hyperparathyroidism. Other findings of osteitis fibrosa cystica include generalized demineralization of bone, “salt and pepper” appearance of the skull, bone cysts, and

brown tumors [6]. It is well known that brown tumors, or osteoclastomas, are caused by localized, rapid, osteoclastic removal of bone secondary to the direct effects of PTH on the bone; it is actually a giant cell lesion and often appears as an expansile osteolytic lesion of the bone. Common involvements are long bones, ribs, clavicle, hand, skull, pelvic girdle, and mandible. While the mandible is the most frequently involved bone in the head and neck region, atypical involvement of the cranium in the area of the maxillary sinus, as presented in our case, is rare [7]. Clinically, maxillary brown tumors appear as bony tumefacation that is in some cases responsible of facial deformities, tooth falls or there mobility [8]. These tumors are mostly asymptomatic, but pain or fractures may occur. [9] Our patient had a large tumefaction of the face that was distorting, painless and uncomplicated dental affect or fracture. On CT scan, brown tumors appear as a lytic or sclerotic lesion without periosteal reaction. The lesion may be hyperdense or heterogeneous and well-circumscribed or expansile lucent lesion with a rim of calcification and remodeling of surrounding bone. After injection of contrast medium, brown tumors appear as heterogeneously enhancing masses [10]. There is no periosteal reaction or soft tissue invasion [10]. The appearance on CT is challenging and not always specific and mimics metastasis [10]. Histologically, brown tumors are nonencapsulated and are characterized by abundant stroma consisting of fibrous connective tissue, with important proliferation of fibroblasts and several multinucleated osteoclast-like giant cells containing variable numbers of nuclei. Calcified material and areas with extravasations of red blood cell and hemosiderin in histiocyte can be found [11]. The cystic spaces are filled up by clusters of giant cells, hemosiderin-laden macrophages, and plump fibroblasts. Cystic formation may develop as a result of bleeding and tissue degeneration. Giant cell masses or brown tumors may result from these changes and are usually seen as focal bone lesions. Giant cells are similar to the other giant cell lesions (true giant cell tumor, reparative giant cell granuloma, cherubism, and anevrysmal bone cyst). Therefore, the brown tumour is actually a kind of giant cell lesion and often appears as a multiple and expansive osteolytic lesion of the bone. Because it is difficult to distinguish histopathologically brown tumour from other giant cell lesions, a clinical diagnosis is made based on the association with HPT [12]. There is no single test that establishes the diagnosis of secondary HPT. However, the diagnosis is made based on the high level of PTH associated with low or normal serum calcium [13]. In the present case, the patient presented chronic progressive renal failure together with the high level of PTH and low serum calcium associated with a giant cell tumor. So, we retained a diagnosis of a brown tumor secondary to hyperparathyroidism. There is agreement as to the treatment of choice for hyperparathyroidism being parathyroidectomy; however, opinions are divided as to the treatment of bone lesions. It is possible to avoid surgical of a large brown tumour even though it compromised the physical appearance of the patient and caused dysfunction of the masticatory apparatus. The surgical resolution of hyperparathyroidism was enough to correct the calcium-phosphate-PTH imbalance and to result in regression of the jaw lesions A. Authors such as Scott et al [14] believe that bone lesions reappear spontaneously following removal of the diseased parathyroid gland; others such as Martinez-Gavidia et al [15] recommend initial treatment with systemic corticosteroids in order to reduce the tumor size, followed by surgical removal of the residual lesion. Surgical excision of the brown tumours is indicated for large and disfiguring lesions and in case that the affected bone is weakened [16] In our case, we opted for a parathyroidectomy followed by surgical removal of the residual lesion. Mais, le patient a présenté une hypocalcémie profonde compliquée de crises convulsives, et puis il est décédé dans un tableau de choc septique suite à une pneumopathie sévère d'inhalation. But the patient had a hypocalcemia complicated by seizures, and then he died in an array of septic shock following a severe aspiration pneumonia.

CONCLUSION

Due to recent improvements in analytical techniques, the diagnosis of hyperparathyroidism usually occurs when the disease is in an asymptomatic phase, and the incidence of patients with advanced bone lesions is rare. 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. This case should attract the attention of general practitioner dentists, oral and maxillofacial surgeons, endocrinologists, and especially radiologists whose consultation is essential in including this entity in the differential diagnosis to avoid unnecessary surgical removal. Accurate diagnosis enabled the proper treatment to be carried out, avoiding unnecessary harm to the patient.

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