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Simultaneous Maxillary and Mandibular Brown Tumors as the First Presentation of Secondary Hyperparathyroidism

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Abstract Brown tumor represents a serious and late bony complications of advanced hyperparathyroidism with a frequency of 1.5–1.75 percent in secondary hyperparathyroidism and 3–4 percent in primary hyperparathyroidism. [1,2] Here we describe an extremely rare case of a 19-year-old male patient with a brown tumor localised in the maxilla and mandibular due to secondary hyperparathyroidism compromising mastication, phonation and social ease of the patient. An early diagnosis and a timely parathyroidectomy in a medically resistant hyperparathyroidism are an optimal option to control the growth of the bony lesion

Keywords: *secondary hyperparathyroidism; renal osteodystrophy; maxillary and mandibular brown tumors*

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1. Introduction

Secondary hyperparathyroidism is a frequent complication of chronic renal failure as a consequence of renal osteodystrophy. Brown tumor represents a serious complication of advanced hyperparathyroidism with a frequency of 1.5–1.75 percent in secondary hyperparathyroidism and 3–4 percent in primary hyperparathyroidism [1,2].

Histopathological examination of brown tumors can suggest the diagnosis but may not be sufficient to differentiate it conclusively from other lesions which can have similar microscopic and macroscopic features; final diagnosis can be defined only by evaluating the radiological findings with histopathological, laboratory, radiological and clinical data. This entity highlights the importance of early diagnosis of hyperparathyroidism with a thorough diagnostic work-up [1,2,3].

2. Case Report

A male patient aged 24 years complained of a painful tumor bilaterally on the jaw and in the mouth. It had started six months before; however, its volume increased rapidly. The patient was on hemodyalisis for the past four years due to end-stage renal disease. The physical examination showed a bilaterally enlarged jaw with limitation of mouth opening compromising mastication, phonation and social ease of the patient (Figure 1).

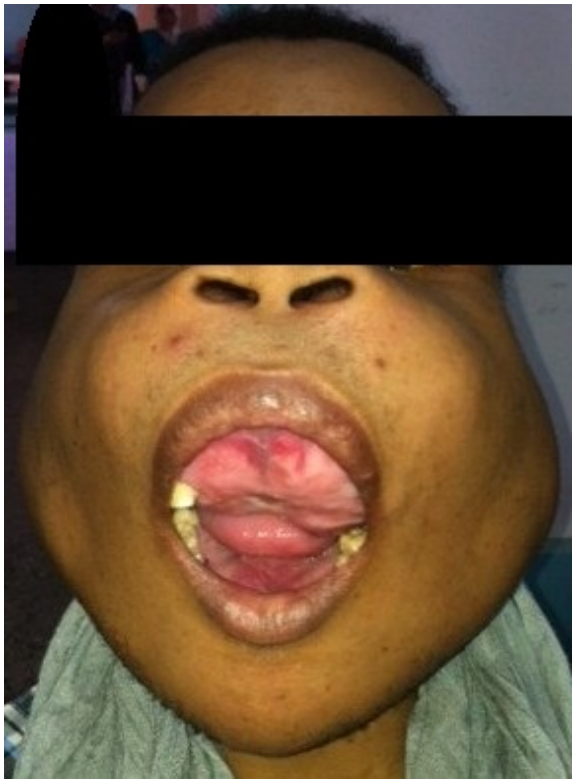

Figure 1. Clinical feature of facial lesion

Laboratory work up showed elevated creatinine, urea, alkaline phosphatase (2996 IU / l) and parathormone or PTH (4042 pg / ml pg/ml; normal range 12 to 72pg/ mL). hypocalcemia at 70 mg / l, and hyperphosphatemia to 66 mg / l.

A radiograph of the face showed a large lesion in the face. Computed tomography revealed a diffuse and heterogeneous multiloculated bony lesion, which enlarged the mandible and maxilla bilaterally (Figure 2).

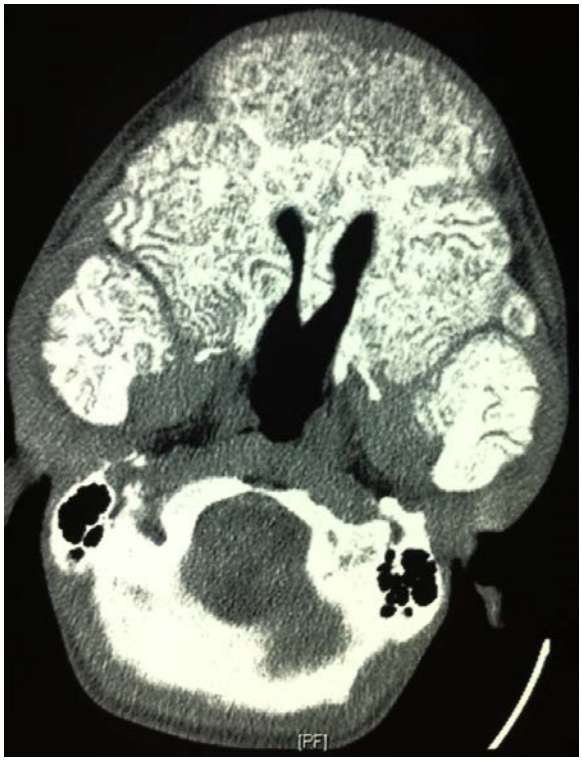

Figure 2. Computerized tomography image of lesion

The parathyroid glands were enlarged on the neck ultrasound. Based on clinical findings and patient's history, our initial diagnosis included brown tumor, central giant cell granuloma (CGCG) and fibrous dysplasia.

All of these lesions have almost similar age distribution, clinical features and radiographic manifestation. Most CGCGs are asymptomatic with gradual painless bony enlargement. An incisional biopsy was performed and specimens Specimens were analyzed histopathologically.

Microscopic evaluation showed numerous multinucleated giant cells in a hypercellular fibrous background with hemosiderin pigmentation. A histological diagnosis of brown tumor of hyperparathyroidism was rendered, given the history of renal failure and secondary HPT.

The patient underwent subtotal parathyroidectomy; pathology revealed the presence of a hyperplasia. On the twentieth postoperative day the tumor could be seen to have regressed significantly. At 2 months follow-up, the tumor remains stable, but did not regress any further compared to the first few weeks after surgery. Unfortunately, the evolution was marked by the onset of severe hypocalcemia at 40 mg / l complicated generalized convulsions. The patient died due to severe aspiration pneumonia.

3. Discussion

Hyperparathyroidism (HPT) is a disease in which there may be a complex of biochemical, anatomic and clinical abnormalities resulting from increased secretion of parathyroid hormone (PTH). It may occur in primary, secondary and tertiary form [1,2,4].

Secondary HPT develops in response to chronic low levels of calcium usually associated with chronic renal disease. Incidence of HPT in chronic renal failure differs from 18% after one year on dialysis to 92% after more than 2 years [4,5].

Bony complications of HPT have started to decline rapidly. With new and more objective diagnostic PTH radioimmunoassay techniques, along with the successful treatment of the disease, In some cases like the present one, HPT is diagnosed by the presence of osteolytic lesions called brown tumors [2,3].

The name ‘‘brown tumour’’ derives from the colour, which is caused by the vascularity, haemorrhage, and deposits of haemosiderin [2,5].

Brown tumour or osteoclastomas, is actually a giant cell lesion, caused by alterations in the trabecular bone pattern, demineralization, and replacement by loose connective tissue. They present as uni/multilocular radiolucencies, and/or mixed lesions with bone expansion, bone deformity, tooth mobility and loss of lamina dura [3,5].

Except in ESRD (end-stage renal disease), brown tumors are rarely reported in other conditions resulting to secondary hyperparathyroidism like rickets, anticonvulsant medications and malabsorption [4,5].

Its prevalence is about 0.1%, it can affect the base of the skull, orbits, paranasal sinuses, spinal column as well femur, tibia, humerus, clavicles and scapula [4,5]. However, it is relatively rare in the maxilla with a frequency of 4.5–11.8 percent. Contrary to our patient, simultaneous presence of maxillary and mandibular lesions is completely rare. And it is three times more common in women than in men [4,6].

The most important complications of this neoformation are related to its position and size and the possible effects on nearby structures. It can be a cause of an increased risk of fracture, of spinal cord compression or of facial disfiguration, compromising normal functions such as mastication, compression, phonation, and social ease of the patient. Other neurological complications include diplopia if the optical nerve is involved [1,4,7].

The differential diagnosis is based on the clinical and radiological findings and the presence of hyperparathyroidism, which is confirmed with biochemical tests including PTH level [2,4].

The radiographic manifestations are in many cases incidental findings and may be multiple. Demineralization and thinning of cortical boundaries often occur in the jaws in cortical boundaries such as the inferior border, mandibular canal, and the cortical outlines of the maxillary sinuses. The density of the jaws is decreased, resulting in a radiolucent appearance that contrasts with the density of the teeth. The teeth stand out in contrast to the radiolucent jaws. CT scans were especially valuable for the determination of the exact borders and size of the brown tumours [2,4,6].

Ultrasound and parathyroid scintiscans were also very useful in localizing the abnormalities in the skeletal bones and parathyroid glands. Further workup with osseous scintigraphy (using technetium 99m—methylene diphosphonate) may

be indicated to locate other areas of osseous metabolic disease as discussed by Prado et al [9].

Histologically Brown tumor of HPT is similar to giant cell lesion (GCL): multinucleated giant cells in a background of spindle cell proliferation containing a large amount of hemosiderin, GCLs are non-encapsulated and consist of fibrous connective tissue, with an important proliferation of fibroblasts and multinucleated giant cells containing variable numbers of nuclei such as giant cell granuloma, giant cell tumour, aneurysmatic cyst, cherubism, Paget's disease, odontologic bone tumour, and nonodontologic fibrous dysplasia [3,5,9].

The various types of HPT manifest different laboratory findings. Patients with secondary HPT, usually present with hypocalcaemia and hyperphosphataemia: Decreased glomerular filtration occurs with reduced nephron function, which results in decreased 1,25-dihydroxyvitamin D synthesis by the kidney leading to decreased calcium absorption by the gut. Consequently an increased level of serum phosphate is also seen. Phosphate is the driving force of bone mineralization, excess phosphate tends to cause serum calcium to be deposited in bone, leading to a decreased serum calcium level and structurally deficient bones. In response to low serum calcium, the parathyroid glands are stimulated to secrete PTH, which results in secondary HPT. The patient becomes hypocalcemic and hyperphosphatemic, opposite of primary HPT [2,4,8].

A final diagnosis can be defined only by evaluating the radiological findings with histopathological, laboratory, and clinical data. This phenomenon is considered as pathognomonic of hyperparathyroidism secondary to renal failure, especially in patients on long-term hemodialysis [2,4].

The therapy of choice is to control the hyperparathyroidism. Tumor regression or complete remission following parathyroidectomy has been well documented in primary and secondary hyperparathyroidism due to ESRD. Several authors consider this approach the only correct therapy [1,2,7].

The treatment of choice for the patient in the present case was subtotal parathyroidectomy, after which the tumor regressed within the first few weeks following surgery.

Triantafillidou et al [11] stated that the significant bone disease associated with secondary HPT may be prevented or reduced by medical treatment, such as calcium carbonate, vitamin D and aluminum hydroxide antacids for hyperphosphataemia.

In contemporary medical treatment alternatives, lanthanum carbonate, sevelamer and calcimimetics such as cinacalcet are more commonly preferred [2]. Systemic corticosteroids can be used to reduce the size of the lesion; sometimes intralesional corticosteroid injections also give satisfactory results [10].

Large lesions may resolve very slowly or may regress with resultant asymmetry on the face. Surgery in the form of excision of the brown tumor and recontouring of the bone should therefore be done in such cases [8]. In the case presented, parathyroidectomy was indicated. With the removal of the autonomous parathyroid glands, the PTH level would eventually return to normal, thus leading to a spontaneous decrease in the sizes of the intraoral lesions within a few weeks.

4. Conclusion

Brown tumors represent a serious complication of advanced hyperparathyroidism; the most important complications of this neoformation are related to its position and size and the possible effects on nearby structures. Therefore an early diagnosis and a timely parathyroidectomy in a medically resistant hyperparathyroidism are an optimal option to control the growth of the bony lesion and to avoid further weakening of bone structure and consequent increased risk of fractures, compression of contiguous structures, deformities, and functional alteration of other involved areas of the skeleton.

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