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Brown Tumors of the Mandible Revealing Hyperparathyroidism: About Two Cases

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ABSTRACT

The diagnosis of osteolytic lesions of the jaws can be challenging.
Case Reports: Two cases of brown tumor of hyperparathyroidism were reported. A 76- year-old female patient presented with indolent swelling of her right lower jaw measuring approximately 5 cm /6 cm. The panoramic radiograph showed a well-defined osteolytic radiolucency involving the entire mandibular symphysis. Blood investigations revealed High level of parathyroid Hormone (PTH): 102pg/ml. The diagnosis of a brown tumor of hyperparathyroidism was suspected. A parathyroid technetium scintiscan revealed abnormally high uptake at the lower pole of the thyroid lobe interpreted as hyperplasia of right inferior parathyroid gland with possible brown tumor of the mandible. Second case: A 36- year-old female patient presented for the replacement of her missing teeth. Her medical history revealed chronic renal failure and a recent surgical excision of an Osteitis fibrosa cystica of her fifth left proximal phalange. Panoramic radiograph showed multiple well defined osteolytic lesions of the mandible. The diagnosis of a brown tumor of the mandible secondary to hyperparathyroidism was suspected. Laboratory investigations showed increased PTH level, serum hypocalcemia and hyperphosphatemia and vitamin D deficiency. The patient was referred to the department of endocrinology for further investigation and the correction of PTH level. At Six months follow up all the lesions disappeared on radiological control.
Discussion: Brown tumor of hyperparathyroidism is a metabolic disorder causing bone resorption that can affect the jaw bones. Clinical symptoms depend on the size and the location of the lesion. Radiographically, it appears as radiolucent unique or multiple well-defined intra-osseous radiolucency. Biological examination is the key to the diagnosis and it is marked by high level of parathyroid hormone (PTH).

Key Words: Jaw, Tumors, Osteitis Fibrosa Cystica, Hyperparathyroidism, Diagnosis

INTRODUCTION

Oral lesions are in several cases revealing of underlying systematic diseases which require multidisciplinary interventions to prevent further complications and to offer better treatment options. Osteolytic lesions of the jaw bones can be a diagnosis challenge especially when no evident clinical or radiological signs are present. Histopathological examination is not always determinant; furthermore, surgical approach should not be our first option [1, 2]. Brown tumor of the jaw is an osteolytic bone disease that can be due to primary, secondary or tertiary hyperparathyroidism. The diagnosis could only be confirmed after correlation of clinical, radiological and biological investigations. Bone lesions secondary to hyperparathyroidism often appear in advanced stage of the disease; however, in some cases these lesions can reveal the metabolic disorder [2]. The aim of this paper was to report two cases of brown

tumors of Hyperparathyroidism involving the mandibular bone. Diagnosis as well as management of this condition is discussed.

CASE REPORT

1st case

A 76- year-old female patient was referred to the department of oral medicine with indolent swelling on the right lower jaw. The patient had a controlled type I diabetes. No associated symptoms neither systemic manifestation was reported.

Extra oral examination revealed a facial asymmetry with a firm swelling on the right side of the mandible, with no inflammatory signs (Figure 1). Palpation revealed no clinically detectable cervical lymphadenopathy. Intra oral examination showed an edentulous mandible and a large swelling on the right alveolar process, measuring approximately 5 cm /6 cm. The swelling caused a deformity of the buccal and lingual right mandibular plates. It was firm and painless on palpation. The overlying mucosa was slightly erythematous (Figure 2). The panoramic radiograph showed a well-defined osteolytic radiolucency extended from the right mandibular body to the left side involving the entire mandibular symphysis (Figure 3). The patient was advised blood investigations that revealed PTH level of 102pg/ml (normal value 15-65 pg/ml).

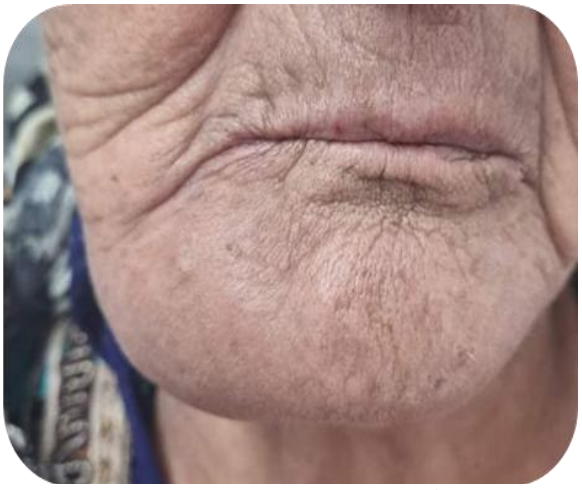

Figure1 :Extra-oral examination revealing a facial asymetry with a deformity of the right side of the mandible

The possible diagnosis of a brown tumor of hyperparathyroidism was suspected. A parathyroid technetium scintiscan ($^{99}Tc^{m}$ SESTAMIBI; Technetium-99 MIBI; methoxy-isobutyl-isonitrile) was indicated and revealed abnormally high uptake at the medium pole of the thyroid lobe interpreted as hyperplasia of the parathyroid gland with possible brown tumor of the mandible.

The patient was referred to the department of endocrinology for further investigations.

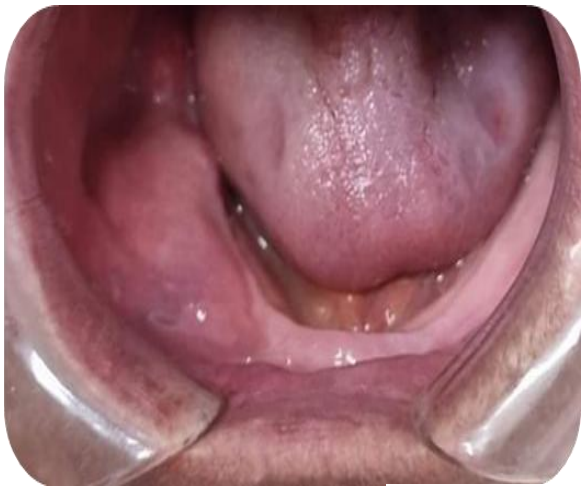

Figure2: intraoral examination revealing large swelling on the right alveolar process, measuring approximately 5 cm /6 cm causing the deformity of the buccal and lingual right mandibular plates.

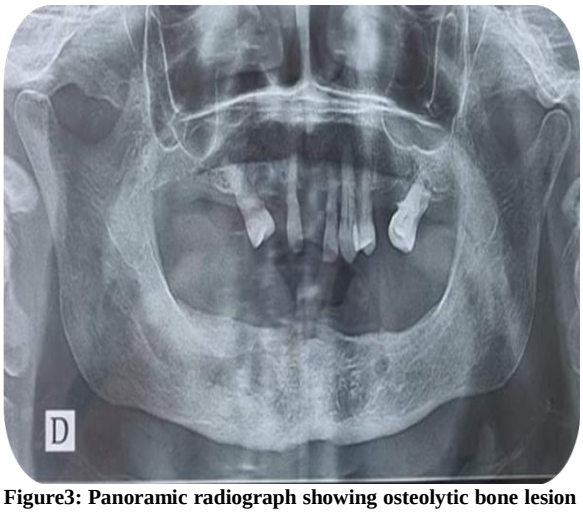

Figure3: Panoramic radiograph showing osteolytic bone lesion with heterogeneous pattern extended to the entire mandibular symphysis

2nd Case

A 36- year- old female patient presented at the department of oral medicine

for the replacement of her missing teeth. Her medical history involved chronic renal failure. The patient was recently operated for an Osteitis fibrosa cystica of her fifth proximal phalange of the left hand (Figure 4).

Extra oral examination didn't show any abnormality (Figure 5). Intra oral examination revealed poor oral hygiene (figure 6). Panoramic radiograph showed multiple well defined osteolytic lesions of the mandible. The lesions were located on the right mandibular premolar region without any contact with the adjacent teeth. A second lesion was appended to the roots of the left mandibular canine and of the first premolar (teeth 33 and 34), measuring 1 cm of diameter and mimicking an odontogenic cystic lesion. A third radiolucent image was detectable on the apical area of the left first mandibular molar (tooth 36) and a fourth well defined radiolucent image appeared on the left coronoid process (Figure 7).

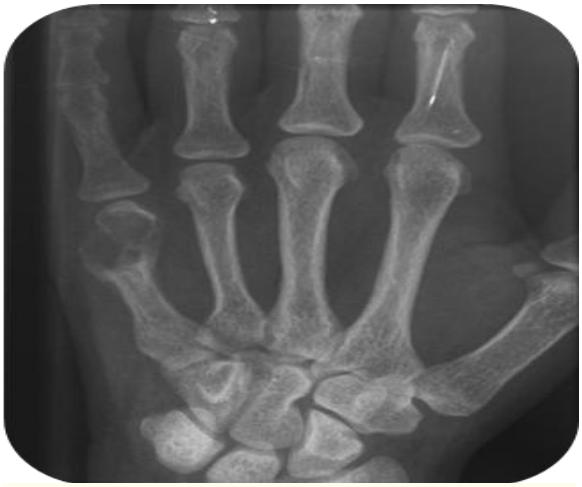

Figure4: radiograph of the left hand showing brown tumor of the fifth proximal phalange

No signs of dental infection were found. Vitality tests in teeth 33.34 and 36 were positive.

Table 1: Blood tests of the patient before and after metabolic control

Serum level	***Normal range***	***Initial value***	***Value after metabolic control***
PTH (pg/ml)	15–65	85	25.9
Vitamine D (25-OH) (ng/ml)	>(20–30)	17.1	21
Phosphorus (mmol/l)	0.87–1.45	59.33	

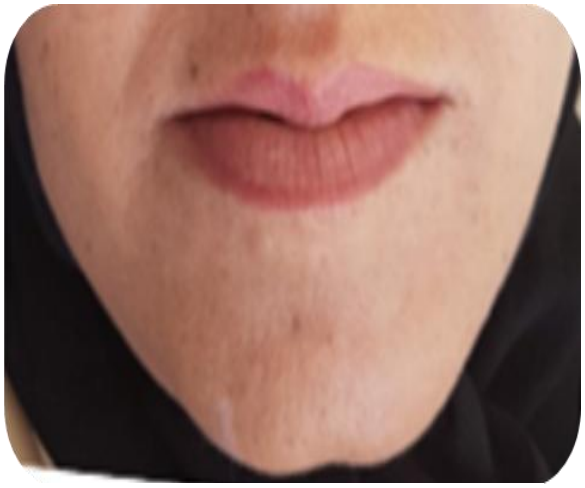

Figure 5 : extraoral examination of the patient was normal.

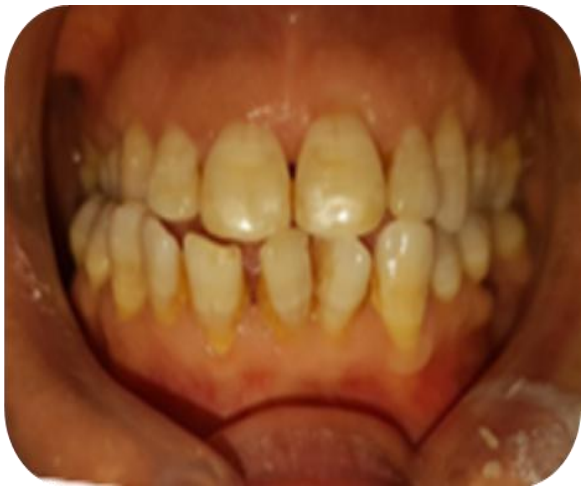

Figure6: intra-oral examination showed poor oral hygiene.

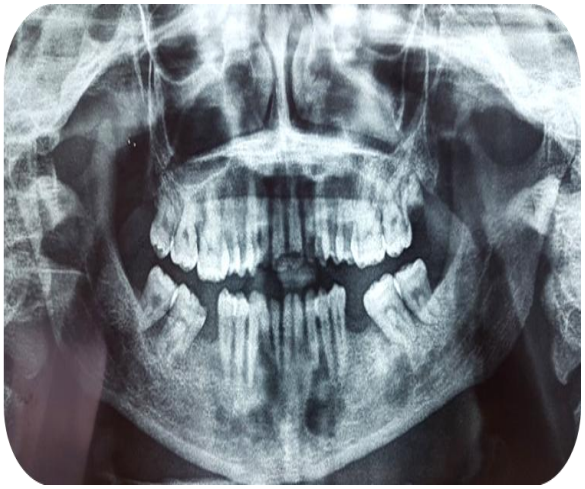

Figure7: Panoramic radiograph. Multiple diffused osteolytic mandibular lesions located on the right mandibular premolar region without evident contact with the adjacent teeth. A second lesion appended to the roots of the left mandibular canine and of the first premolar (teeth 33 and 34). A third radiolucent image detectable on the apical area of the left first mandibular molar (tooth 36) and a fourth well defined radiolucent image appeared on the left coronoid process

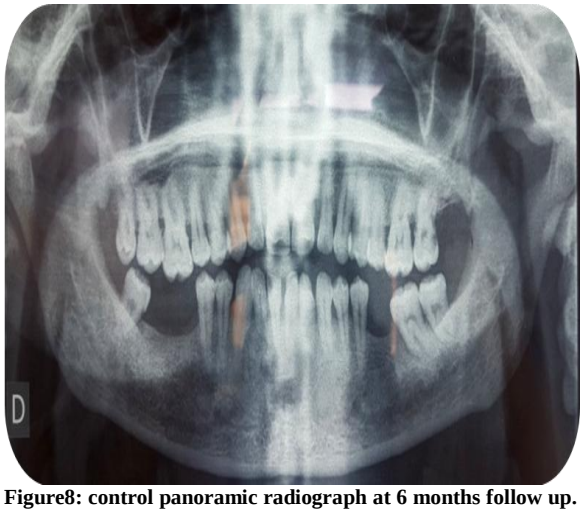

Figure8: control panoramic radiograph at 6 months follow up. No osteolytic lesions are noted.

Given the medical history of the patient, the diagnosis of a brown tumor of the mandible secondary to hyperparathyroidism was suspected. Laboratory investigations showed serum hypocalcemia and hyperphosphatemia and vitamin D deficiency. (table1)

The patient was referred to the department of endocrinology for further investigation and the correction of PTH level. All the dental procedures were scheduled after the correction of the hormonal unbalance. At Six months follow up all the lesions disappeared on control panoramic radiograph (Figure 8).

DISCUSSION

Osteolytic lesions of the jaw can be a diagnostic dilemma for oral surgeons, especially in the absence of characteristic clinical or radiological features. The brown tumor also known as osteitis fibrosa cystica or Von Recklinghausen's disease of bone is a rare yet existing lesion that should not be ignored by clinicians. It's a metabolic bone disease that develops in primary, secondary or tertiary hyper-parathyroidism and mostly affects women above the age of 50 years with a reported male to female ratio of 1/3. [3, 4]. The brown tumor has been reported in multiple skeletal bones mainly in ribs, clavicle, and pelvic girdle (2). The maxillofacial region is rarely affected, however, a predilection for the mandibular bone was reported. [5] In these two reported cases, both patients were female, and both had brown tumors of the mandible.

Clinical symptoms of brow tumors vary according to the size and the location of the lesion. It may be painful with hard bone swelling or asymptomatic and identified as part of routine investigations. [6]

In the first reported case, the lesion was painless but led to a severe deformity of the mandibular bone. In the second one, the lesions were totally asymptomatic and were discovered on the panoramic radiograph during a routine consultation. In both cases, no other systematic lesions or physical symptoms were found; however, the history of Osteitis fibrosa cystica of the fifth proximal phalange in the second patient oriented the diagnosis.

The radiographic appearance of brown tumors of hyperparathyroidism is often a well-defined radiolucent or hypodense image of the bone, without inflammatory signs, cortical disruptions or periosteal reactions. [6]

In the first case, the panoramic radiograph showed a well-defined osteolytic radiolucent image of the mandibular symphysis with no inflammatory signs. The patient was edentulous, thus an odontogenic origin of the lesion was ruled out. In this case, differential diagnosis included non-odontogenic cysts and tumors especially giant cell lesions, fibro-osseous lesions and eosinophilic granuloma. In the second case, the lesions appeared as radiolucent images of the entire corpus of the mandible and the diagnosis was easier to make giving the medical history of the patient. In all cases, the positive diagnosis was confirmed by biological analysis revealing hyperpara-thyroidism (HPT).

Hyperparathyroidism can be primary, secondary or tertiary. Primary HPT is diagnosed when increased or inappropriately normal PTH level is associated with elevated total serum calcium level. It appears as a result of abnormality in one or more of the parathyroid glands. The disease is most commonly caused by

adenoma (80–85%) followed by hyperplasia (10–15%) and carcinoma (less than 1%) [7]. This condition was found in the first patient where the mandibular lesion revealed hyperparathyroidism caused by the hyperplasia of the parathyroid gland which was revealed by parathyroid technetium scintiscan.

Secondary HPT is a result of hypersecretion of PTH as a response to decreased calcium level. It is generally associated with serum hypocalcemia and hyperphosphatemia. This condition is found in patients with chronic renal failure or vitamin D deficiency [6]. Our second patient has history of chronic renal failure which was complicated with HPT. Biological investigation showed hyperphosphatemia and high level of PTH which confirmed the diagnosis of secondary HPT.

In both forms, excessive PTH secretion increases osteoclastic activity resulting into bone resorption. In fact, as a result of excessive osteoclastic activity, bone resorption becomes faster in one or more location, with hemorrhage, reparative granulation tissue, and active, vascular, proliferating fibrous tissue. Finally, the hemosiderin deposition gives the brown color to the tumor. [8]

Histopathological findings are similar to other giant cell lesions especially giant cell reparative granuloma, fibrous dysplasia and true giant cell tumors. Biopsy often reveals giant cells and spindle-shaped cells, mixed with fibrous tissue and poorly mineralized woven bone. Biochemical analysis is necessary to confirm the final diagnosis of brown tumor of HPT. [9]

Treatment options have been controversial between conservative or surgical approach. Etiologic treatment of hyperparathyroidism has been proved to be an effective therapeutic option and to lead to complete regression of the lesions [10]. It requires patient cooperation with involvement of different specialists, in order to determine the right diagnosis and to give a close follow up of blood investigations of parathyroid hormone, calcium, phosphorous and vitamin D levels. In the other hand, it avoids the patient an unnecessary surgical procedure. However, in case of extensive lesions, some authors recommend surgical curettage and enucleation to prevent the risk of further damage of the adjacent structures, especially when the diagnosis is uncertain, or if the osteolytic lesions persist more than 6 months after the metabolic control [11]. In the reported cases, both patients were referred to the department of endocrinology to manage the HPT. In the first case, the treatment of the HPT was based on the management of hyperplastic lesion of the parathyroid gland. However, in the second patient the lesions totally disappeared after the correction of the hormonal unbalance: the PTH, Calcium and Vitamin D levels. No surgical treatment of the lesions was needed and the patient could benefit of this conservative approach without any further surgical procedures.

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