



Bilateral Brown Tumors of Jaw: A Case Report with Clinico-Radiologic Follow-Up

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Abstract

A brown tumor, also known as osteitis fibrosa cystica, is an osteolytic lesion of the jaw that occurs secondary to hyperparathyroidism. Although it can occur in monostotic or polyostotic forms, its presence in the jaws is considered rare. We present a case of a 55-year-old female who reported with multiple swellings in both jaws. Radiographs showed multiple osteolytic lesions in the mandible and maxilla. Other radiographic and blood investigations were also carried out, leading to the diagnosis of the brown tumor. The primary cause was found to be a parathyroid adenoma. After its surgical excision, further follow-ups were taken to assess the lesions of the jaw.

Keywords: Brown Tumor, Hyperparathyroidism, Osteitis fibrosa cystica, parathyroid adenoma.

Introduction

Parathyroid hormone (PTH) is produced and secreted by the parathyroid glands and its increased secretion leads to a complex of biochemical, anatomic, and clinical abnormalities, thereby causing hyperparathyroidism (HPT) It may occur in primary, secondary, and tertiary forms.^[1,2]

In primary hyperparathyroidism, excess PTH is secreted from one or more of the parathyroid

glands.^[3] It results from autonomous hyperplasia or tumor (adenoma).^[1,4] The increased PTH secretion results in hypercalcemia and hypophosphatemia. However, clinical manifestations of primary HPT are variable in terms of etiology and serum PTH levels. Most patients with primary HPT are asymptomatic, while some present with recurrent nephrolithiasis, osteoporosis, proximal muscle weakness, and

psychiatric symptoms.^[3] Secondary HPT is usually associated with chronic renal disease.^[4] Tertiary HPT results when the parathyroid glands gain an autonomous character, in long-standing secondary HPT. Some authors also report a fourth type of HPT which is thought to arise from increased PTH levels synthesized in patients with malignant disease.^[2]

The reported prevalence of brown tumors is 0.1% and those occurring in association with primary HPT is 4.5%.^[5,6] These develop in jaws, presenting as well-demarcated, circumscribed, osteolytic lesions and appear as a friable red-brown mass on tissue specimen, hence the term brown tumors.^[2]

Case Report

A 55-year-old female presented with a chief complaint of pain and swelling in her upper right back tooth region for the past 1 year and lower left back tooth region for 15 days. She gave a history of similar swelling in the lower right back tooth region before 1.5 years, for which she got her lower back teeth extracted. She visited a dentist and got her upper right back teeth extracted, but with profuse bleeding.

There was a history of asthenia and weight loss since few months, a past medical history of renal stones before 4 years, which she got treated conservatively.

On extra-oral examination single, diffuse swelling was noted on the right side of the face, which was tender on palpation (Figure 1). On intraoral examination, single, diffuse, dome-shaped swelling was noted in the maxillary buccal vestibule with marked erythema and a post-extraction scar on the maxillary ridge (Figure 2). It was tender and firm on palpation.

On extra-oral examination, another swelling was noticed on the left side of the face, measuring approximately 3×3cm (Figure 1). The left nasolabial fold was obliterated. On palpation, the swelling was tender, afebrile, and firm. On intraoral examination, a single, well-defined dome-shaped swelling was seen in the lower left vestibule (Figure 1). On palpation, it was tender, firm, and fluctuant. Grade II mobility of 34, 35 and grade III mobility of 36 was noted. No associated discharge or bleeding was noticed. The overlying mucosa was freely mobile and highly vascular. Right submandibular lymph nodes were round, enlarged (approximately 2cm), tender, firm, and mobile.

A provisional diagnosis of Odontogenic Keratocyst was made with a differential diagnosis of Ameloblastoma, glandular odontogenic cyst, brown tumor, and multiple myeloma.

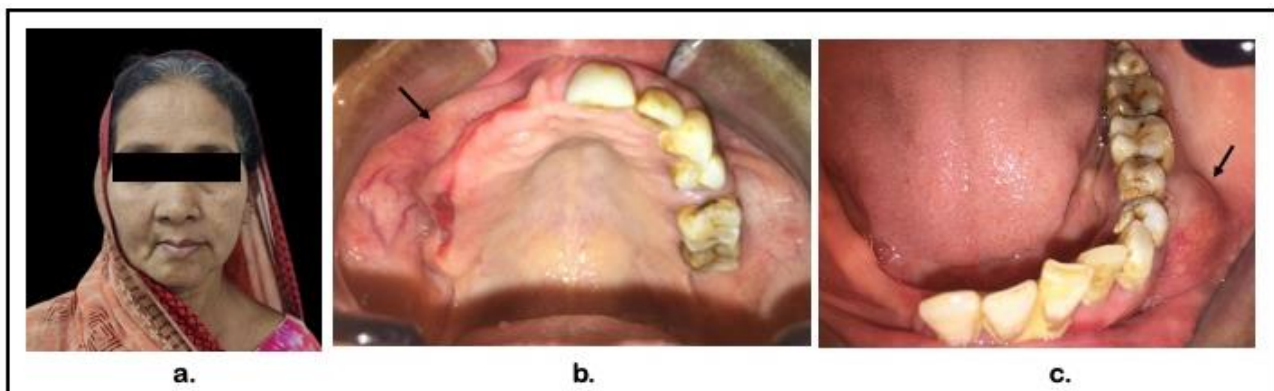


Figure 1: Clinical photographs of the patient

The periapical radiograph and orthopantomogram showed a single oval well-defined, multilocular radiolucency on the left side of mandible. Also, ill-defined focal regions of decreased bone density

were observed in the right body of the mandible. The maxillary lesion was not clearly discernible but revealed a diffuse radiopacity in the right maxillary

sinus. A generalized loss of lamina dura was noticeable on the radiograph (Figure 2).

Further, to rule out systemic disorders, PA Skull was taken, which showed a granular “salt and pepper” appearance, pathognomonic for hyperparathyroidism. Also, on close examination, a well-defined radiopacity was noted in the right frontal sinus. (Figure 2)

The CBCT scan confirmed the presence of these lesions, where the right maxillary lesion measured 29 x 19 x 31 mm with the presence of internal septations, and projected into the right maxillary sinus, and the inferior nasal turbinate. The left mandibular lesion measured 27 x 15 x 17 mm. There was a significant bony expansion with perforation of both the buccal and the lingual cortical plates (Figure 2).

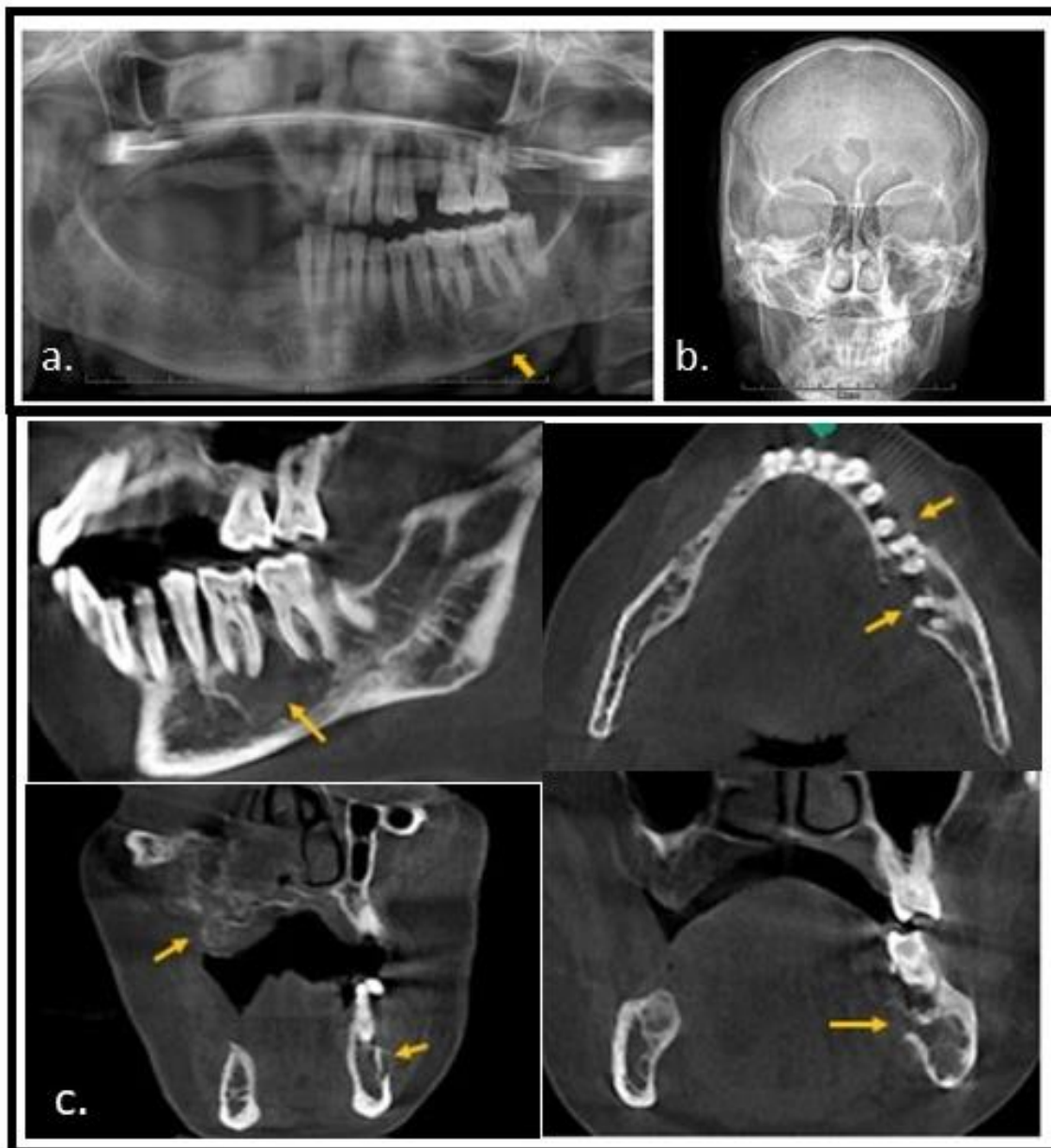


Figure 2: a. Orthopantomogram, b. PA skull, c. Sagittal, Axial and Coronal section of CBCT showing the lesion in the maxilla and mandible

With an opinion from the endocrinologist, the patient was advised a parathyroid scan (technetium-99m SESTAMIBI) which revealed an

area of increased uptake in the entire left lobe of the thyroid (corresponding to left parathyroid gland) suggestive of left parathyroid adenoma (Figure 3).

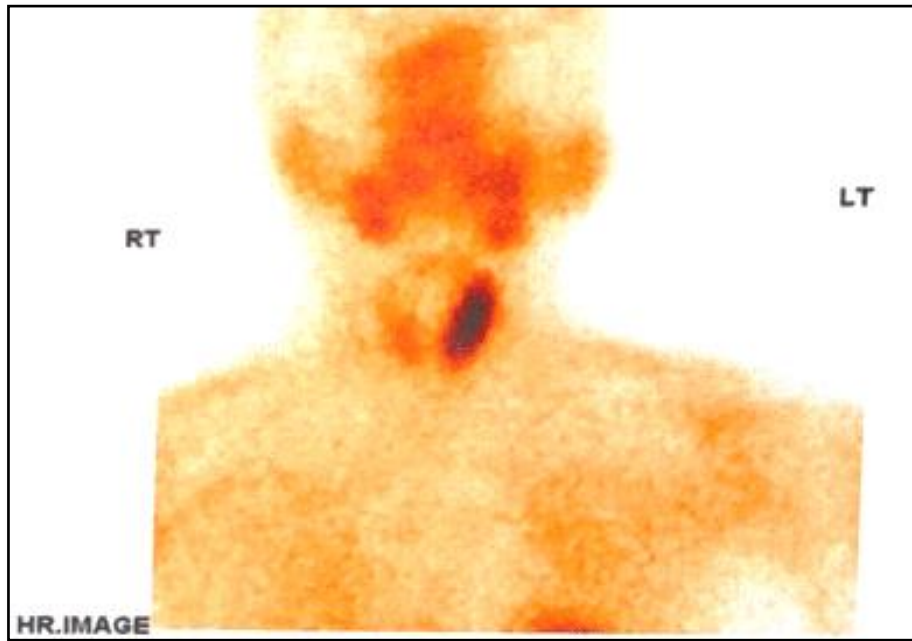


Figure 3: Parathyroid scan (technetium-99m SESTAMIBI) shows increased uptake in the entire left lobe of the thyroid (corresponding to the left parathyroid gland) with normal uptake by the thyroid gland

The patient underwent surgical resection of the left inferior parathyroid adenoma, after which the PTH levels dropped from 649.5 pg/ml to 91.10 pg/ml. On a 3-month post-operative follow-up, the swelling had regressed a bit with a change in

consistency from firm to bony hard. The radiographs revealed an abnormal increase in bone density with a unique disordered mineralization pattern in both the jaws (Figure 4).

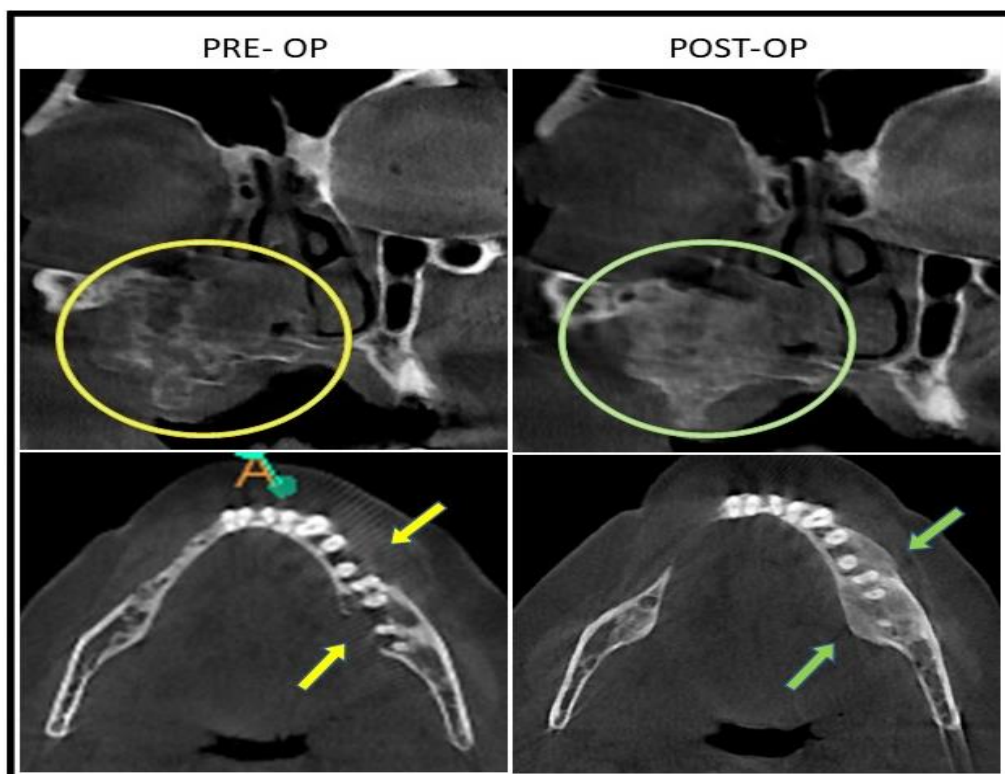


Figure 4: (a) pre-op radiograph (yellow) (b) 3 months post-op radiograph reveals slight regression of lesion with an increase in bone density due to disordered mineralization pattern (green)

Discussion

Sylvanus (1743) was the first to diagnose hyperparathyroidism. Recklinghausen (1891) first described the associated bone changes known as osteitis fibrosa cystica.^[6]

The most notable aspect of this case report was the infrequent finding of a brown tumor in the mandible as well as the maxilla as the first manifestation of an atypical parathyroid adenoma. It was the high levels of Alkaline Phosphatase and PTH that led us to the diagnosis of Brown tumor. Osteitis fibrosa cystica is a diffuse resorptive process of the bone resulting from hyperparathyroidism. Once common, it now occurs less frequently because hyperparathyroidism is diagnosed and managed earlier.^[7] The disease can manifest at any age, but it is more common among persons older than 50 years and is three times more common in women than in men; as is seen in our case.^[6]

Brown tumors can occur in monostotic and polyostotic forms. Bone lesions are among the main manifestations of HPT, ranging from generalized osteopenia, cystic lesions, and bone resorption to brown tumors affecting the mandible, clavicle, ribs, pelvis, and femur.^[6,8] Clinically significant lesions in the jaws, especially in maxilla are rare. Although Wood, Goaz, and Neville et al. implied that these lesions are most common in jaws, other literature does not agree with them.^[4] The presence of hemorrhage, hemosiderin, and hypervascularity lead to the brownish color, hence named so. These lesions are essentially cellular reparative processes and are non-neoplastic.^[9]

Various authors have reported the spontaneous regression of brown tumors after appropriate medical or surgical treatment of HPT,^[1,2,3,9] as seen in our case, while some have stated that remodeling is accelerated if the brown tumor is removed.^[5] Surgical/ operative intervention of the brown tumor is indicated if the brown tumor is large, disfiguring and causing persistent deformity or if the affected bone is weakened.^[1,5,6] Some authors have also treated the brown tumors initially with systemic corticosteroids to reduce the size, followed by

surgical excision of the residual lesion.^[8] However, recurrence of the brown tumor can occur because of persistent or recurrent Primary Hyperparathyroidism.^[9]

Conclusion

The report highlights the need to consider systemic abnormalities whenever diagnosing any osteolytic lesion of jaw. A history of kidney stones, abdominal groans, traumatic extraction with profuse bleeding, presence of unexplained bone pain, and swellings of jaw can be considered as the red flag signs of a possible brown tumor during clinical examination, warranting further investigations. Although the incidence of brown tumors is rare, their timely diagnosis is essential for proper treatment. Hence, it is important on the part of an oral physician to be aware of the oral manifestations associated with such systemic diseases and treat the underlying systemic condition rather than undergoing any unnecessary dental treatment or surgical intervention of the lesion.

Conflict of Interest

None

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