Medical and Research Publications
International Open Access

Case Report **Journal of MAR Oncology (Volume 5 Issue 3)**

Concomitant Maxillary and Mandibular Brown Tumor, Papillary Thyroid Carcinoma and Parathyroid Adenoma: A Case Report

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Received Date: February 01, 2023

Published Date: February 10, 2023

Citation: Dr Shaktisingh Deora, Dr Hastee Bhanushali, "Concomitant Maxillary and Mandibular Brown Tumor, Papillary Thyroid Carcinoma and Parathyroid Adenoma: A Case Report" MAR Oncology Volume 5 Issue 3

Abstract

Objective: *We discuss a rare case of brown tumour from primary hyperparathyroidism, presented as a radiolucent lesion in the jaw. It was treated by parathyroidectomy, which resulted in complete resolution of the osteolytic lesion without any surgery in the oral cavity. The crux of this paper is to make maxillofacial clinicians aware of endocrine causes of a common radiological sign to avoid unnecessary local surgical intervention.*

Case Report: *We discuss a case of a female patient, age 35, who came with a chief complaint of expansile growth, loosening of teeth, in upper and lower left region of jaw. She had a past medical history of papillary carcinoma of thyroid for which she was operated 10 years back.*

After performing an incisional biopsy on the oral lesion, histological analysis showed that it was a central giant cell lesion with areas of hemosiderin pigment, severe hemorrhagic exudate, and a lot of giant cells. Additionally, the patient had high levels of serum calcium and parathyroid hormone, hyperfunctioning parathyroid tissue. By imaging tests and histological analysis, synchronous parathyroid adenoma and papillary thyroid cancer were verified.

Conclusion: *The composition of all the clinical, pathological, and imaging findings led to the final diagnosis of brown tumour of hyperparathyroidism. The occurrence of parathyroid adenoma, papillary thyroid carcinoma, and brown tumors of hyperparathyroidism (osteitis fibrosa cystica) with oral manifestation involving the mandible and maxilla is extremely rare. The appropriate resection of the affected glands leads to the metabolic control of serum levels of calcium and PTH, thus avoiding any enucleation or resection in either of the jaws.*

Introduction

Severe parathyroid bone disease occurs rarely. The bone lesions are the result of a metabolic abnormality brought on by the altered parathyroid, which is mostly brought on by adenomas in 85% of patients **[**1**]** the imbalance in osteoclast activity brought on by the overproduction of parathyroid hormones (PTH) results in bone degradation **[**2**]**. Histologically, the lesions are distinguished by osteoclasts present in areas of erratic bone resorption, which are replaced by fibrous vascularized tissue (fibrous cysts) and large cells that resemble osteoclasts. The term "brown tumour" refers to tumours that have a reddish or brown colour due to an accumulation of red blood cells and their pigments **[**3**]**.

In today's scenario, brown tumours are uncommon because of advances in medical technology that allow for early detection and PTH level adjustment **[**4**]**. The classic bone lesions are most commonly found on the long bones, pelvis, and ribs. The face is only in exceptional cases, and when it is, it is usually the mandible. The occurrence of parathyroid adenoma, papillary thyroid carcinoma, and brown tumours of hyperparathyroidism involving the mandible and maxilla is extremely rare **[**5, 6**]**.

We report here a very rare case of concomitant maxilla and mandible brown tumours, thyroid carcinoma and parathyroid adenoma.

Figure 1: Extraoral Swelling seen on left side of the face

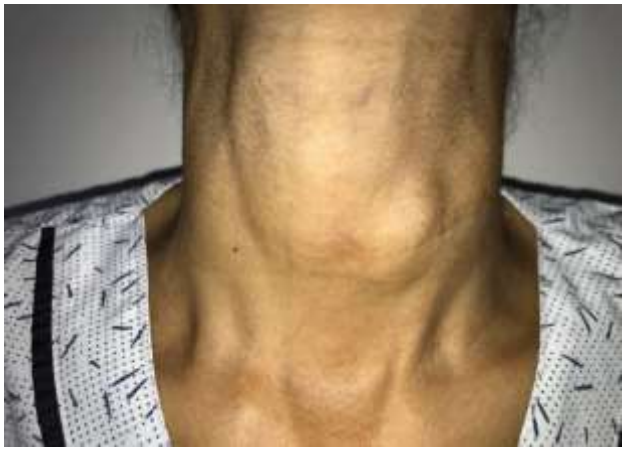

Figure 2: Expansile swelling seen on left side neck region

Figure 3: Exophytic growth extending from left mandibular premolar region to molar region

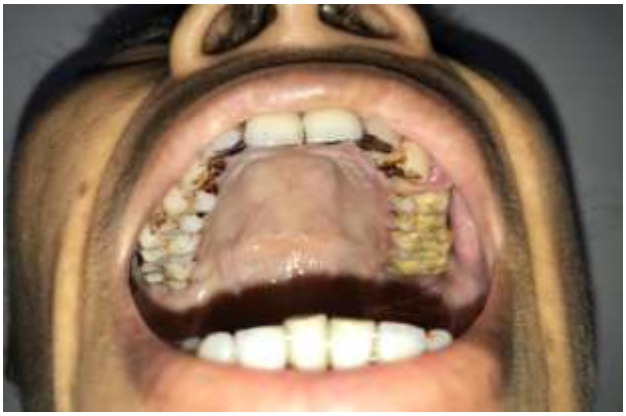

Figure 4: Exophytic growth seen in anterior maxillary region

The x-ray images showed an ill defined osteolytic lesion causing expansion of the affected mandible, loss of lamina dura, and root resorption of the anterior teeth (Figures 5,6). Computed Tomography (CT) was used to scan sites suspected to be affected by the brown tumours. Bone rarefaction focuses were also observed in the proximal phalanx of the third toe and the ribs. In the cervical region massive septated cystic-solid lesion was located posteriorly to the left thyroid lobe besides the presence of thyroid nodules.

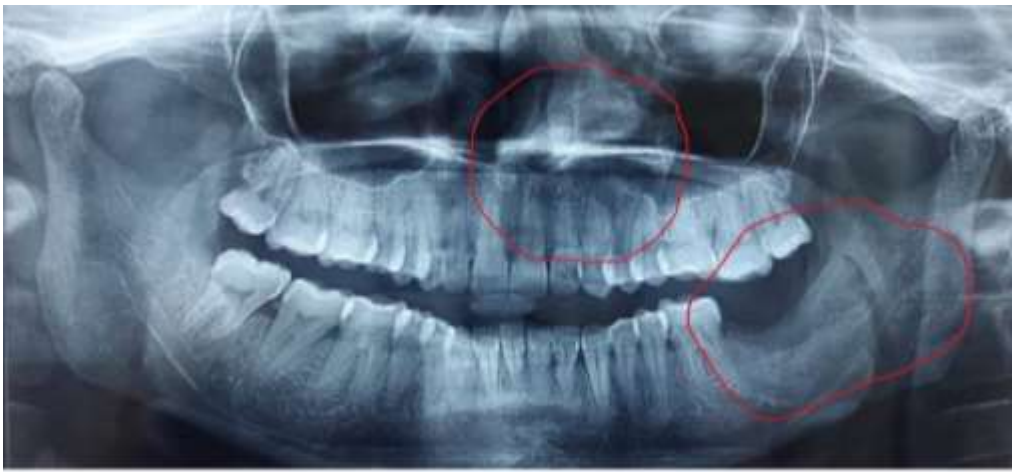

Figure 5: Panoramic radiograph of maxillary and mandibular lesions, an osteolytic area is visible from left mandibular premolar to molar region and in anterior region of maxilla

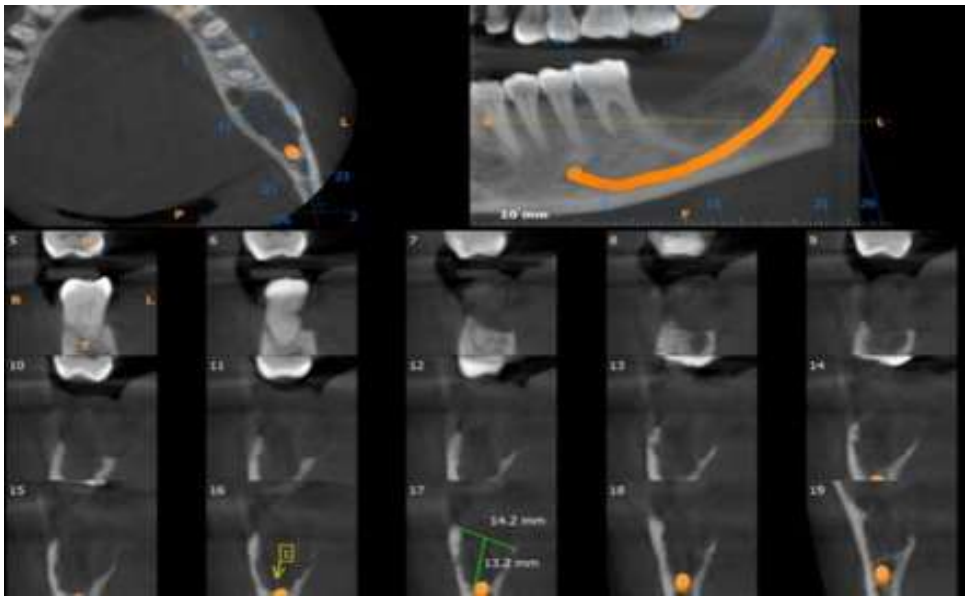

Figure 6: CBCT confirmed focal destruction extending from left mandibular premolar to molar region with destruction of lamina dura, part of mandibular canal wall, erosion of buccal and lingual cortex.

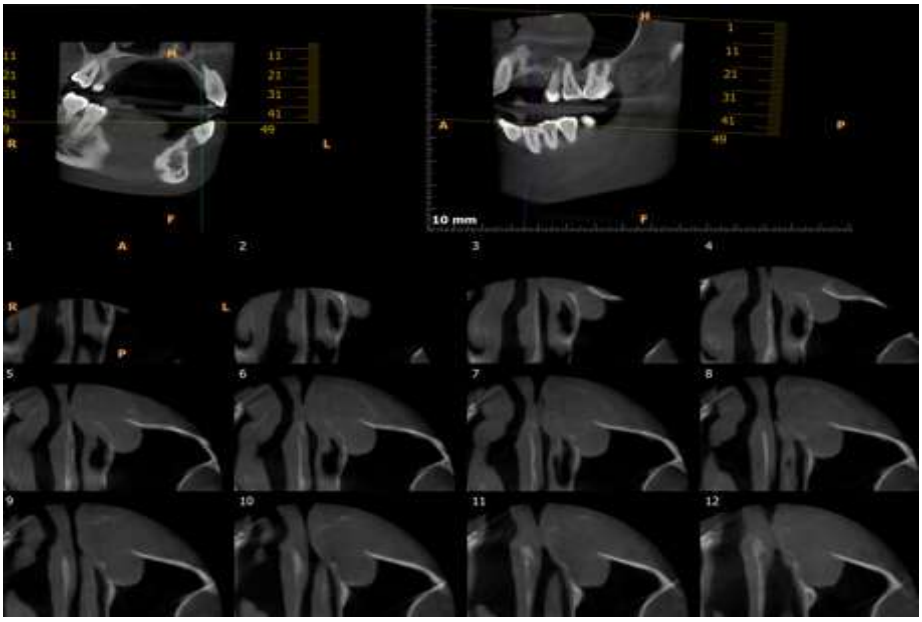

Figure 6 B: CBCT confirmed focal destruction in anterior region of maxilla.

An incisional biopsy was performed in the mandible lesion and histopathological examination was consistent with central giant cell lesions, composed of intense hemorrhagic exudate, abundant presence of multinucleated giant cells, and areas with haemosiderin pigment. A 99mTc-sestamibi imaging with SPECT (SINGLE PHOTON EMISSION COMPUTED TOMOGRAPHY) was performed and left lower parathyroid gland was identified. Neck ultrasonography was done which confirmed 1.2-1.4 cm heterogeneous nodule and central vascularity in the the left thyroid gland.

The biochemical investigations were carried out and showed marked hypercalcaemia (18 mg/dL) and an elevated parathyroid hormone (PTH) concentration 349 pg/mLin the blood.

A normal-appearing inferior parathyroid gland was identified and preserved in situ intraoperatively. An en bloc resection of the parathyroid mass and left thyroid lobe and central lymph node compartment dissection was performed. The PTH concentration taken 15-20 mins after the adenoma had been removed confirmed a level within the gross fall in PTH Level. The diagnosis of parathyroid adenoma was confirmed histologically. On the fourth postoperative day, the iPTH value was 80 pg/mL.

The serum calcium and inorganic phosphorus levels were estimated daily after surgery for next three days. The postoperative hypocalcaemia was administered by calcium carbonate orally in addition to vitamin D until stable levels. Thyroid hormone replacement therapy was instituted.

At 6-month follow-up, there was only a modest improvement of oral pain, but the oral lesions had remained. The patient interrupted her clinical follow-up despite the efforts of the medical team.

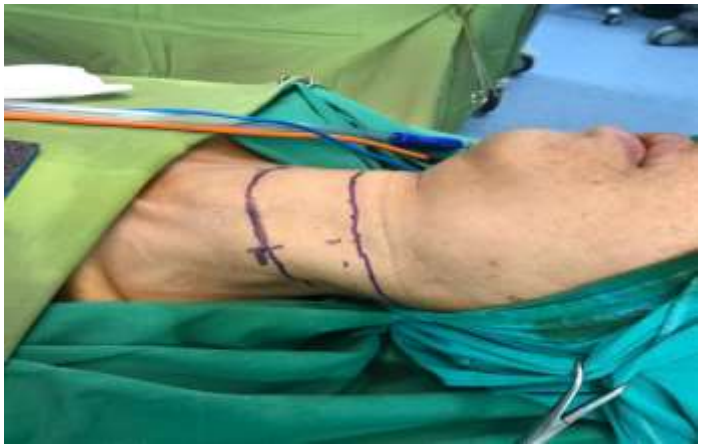

Figure 7: Incision marking for left parathyroidectomy

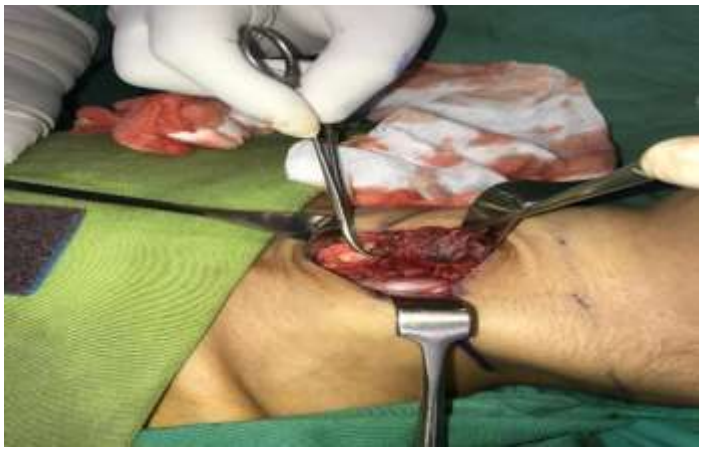

Figure 8: Parathyroidectomy with prevention of recurrent laryngeal nerve (as shown with artery forceps)

Discussion

The occurrence of brown tumours in both jaws concomitant with the presence of hyperparathyroidism, parathyroid adenoma, and synchronous papillary thyroid carcinoma is very rare according to the literature **[**7, 8**]**. The brown tumour of hyperparathyroidism results from a metabolic disorder that affects long bones, ribs, and pelvis. The progressive loss of bone mineral content due to the increased PTH secretion may produce radiolucent bone lesions of ill-defined margins, as those that were seen in our patient. The

destructive bone lesions of brown tumour show similar histological features to those viewed in central giant cell lesions, aneurysmal bone cyst, and cherubism and these are considered as differential diagnosis [4, 7].

The histopathological examination was consistent with a central giant cell lesion, but the composition of all the clinical, pathological, and imaging findings enabled us to reach the final diagnosis of brown tumour of hyperparathyroidism. The pathogenesis of this disease is caused by the marked increase in osteoclastic bone resorption followed by replacement of marrow spaces with fibrovascular tissue and increased formation of osteoid. The presence of brown tumours is recognized in less than 5% of these cases [1].

According to the literature, concomitant thyroid disease is present in 3.1% of patients with primary hyperparathyroidism[11], but the reason of this association remains unclear. Although this situation is unusual, the diagnosis of coexisting diseases should be considered. The early identification of cancer and appropriate resection of the affected glands quickly permitted the metabolic control, as noted in the assessment of serum levels of inorganic phosphorus and PTH after the surgery.

It was not possible to evaluate whether surgical excision of parathyroid adenoma and papillary thyroid carcinoma allowed the regression of oral lesions in our patient. After 6- month follow-up, she showed discrete improvement in pain.

Brown tumours should only be removed if they persist even after removal of parathyroid adenoma, when functional problems are detected [1], or if tumours are too large. Although the regression of brown tumours can take several months, conservative management can avoid more invasive procedure in facial bones, preventing further deformities. [12]

Conclusion

The composition of all the clinical, pathological, and imaging findings led to the final diagnosis of brown tumour of hyperparathyroidism. The occurrence of parathyroid adenoma, papillary thyroid carcinoma, and brown tumors of hyperparathyroidism (osteitis fibrosa cystica) with oral manifestation involving the mandible and maxilla is extremely rare. The appropriate resection of the affected glands leads to the metabolic control of serum levels of calcium and PTH.

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