

How to Cite:

Arya, S., Pandey, A., Gulia, S., Dwivedi, P., Zeeshan, M., & Singh, S. (2022). Brown tumours of maxilla and mandible as index presentaion of primary hyperparathyroidism. *International Journal of Health Sciences*, 6(S1), 919-929. <https://doi.org/10.53730/ijhs.v6nS1.4838>

Brown Tumours of Maxilla and Mandible as Index Presentaion of Primary Hyperparathyroidism

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Abstract--It is rightly said that oral cavity is a mirror that may reflect and reveal many of the human body's internal secrets. Some of oral manifestations are disease specific and help in raising high degree of suspicion for an alert clinician. Hyperparathyroidism is one of such systemic disorder which can present in craniofacial regions as erosive bony lesions known as brown tumours resulting in a local destructive phenomenon. As histological examination alone is not sufficient for differentially diagnosing it, clinical, radiological correlation and laboratory data are necessary for its definitive diagnosis. It is essential that while dealing with such clinical presentations, the underlying systemic disease is rightly recognized using a high index of suspicion

International Journal of Health Sciences ISSN 2550-6978 E-ISSN 2550-696X © 2022.

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Manuscript submitted: 27 Nov 2021, Manuscript revised: 18 Feb 2022, Accepted for publication: 09 March 2022

and performing targeted investigations to provide correct diagnosis and referral for proper treatment avoiding unnecessary orthopaedic intervention. Here, we report a case of brown tumors involving maxilla and mandible resulting from primary hyperparathyroidism in a young female patient with history of recent femur fracture and orthopaedic treatment.

Keywords--brown tumor, hyperparathyroidism, maxilla, primary hyperparathyroidism.

Introduction

Hyperparathyroidism is an endocrinal disorder having various systemic complications. Raised parathormone levels in hyperparathyroidism cause an increase in osteoclastic activity and peritrabecular fibrosis.¹ Hyperparathyroidism is of three types, namely, primary, secondary, and tertiary. Primary hyperparathyroidism is caused by pathology in parathyroid glands most commonly being adenomas or carcinomas. Secondary hyperparathyroidism is in response to hypocalcemia caused by chronic renal disease, liver disease, intestinal disease, and vitamin D deficiency; and tertiary hyperparathyroidism is a result of long standing case of secondary hyperparathyroidism. Clinical manifestations of primary hyperparathyroidism include pain, fatigue, and muscular weakness; cortices of bones are more affected with associated subperiosteal bone resorption.² Here, we present a case report of primary hyperparathyroidism with the oral manifestations.

A 17 year old female reported in the department of dentistry with the chief complaint of bleeding and ulceration in left upper back region of jaw since 3 days. On taking history of present illness, patient reported that she was apparently well 3 days before, when she got tooth brush trauma in her left maxillary posterior region of jaw. She also reported swelling in the right mandibular region for 3 months due to which she had developed asymmetry in her facial appearance. In medical history, she reported that she had got a fracture of her left thigh bone while walking 3 months before and reportedly undergone the treatment it.

On extraoral examination, a oval shaped well defined swelling was noted involving the right mandible region extending anterioposteriorly from the right parasymphysis to right body region and lateromedially from right inferior border of mandible towards right submandibular region. Size was approximately 2×1 cm. On palpation, it was bony hard and nontender. [Figure 1,2]. On intraoral examination, a sessile oval swelling wrt left posterior maxillary molar region wrt 26,27. Size was approx 2×2 cm extending anteroposteriorly wrt 26,27,28 and lateromedially from buccal aspect towards palatal region upto 1cm away from midpalatine suture. Overlying mucosa was ulcerated with partial healing, yellow granulation tissue on base and surrounding erythema. Tooth migration wrt 26,27 was evident. Overlying mucosa on palatal side of lesion was normal. On palpation the lesion was hard and nontender. (figure 3,4,5)



Figure 1



Figure 2



Figure 3



Figure 4



Figure 5

IOPA wrt 26,27 showed hazy bony trabeculae with overall lack of sharpness.(figure 6). At alveolar crest level, bone loss was apparent wrt 26,27. Pathological migration was present with loss of contact interproximally wrt 26 and 27. Tapering of roots, loss of lamina dura and pdl space widening were apparent wrt 24,25,26,27 and 28. There was diffuse, irregular shaped mixed lesion wrt 26 and 27 with more of radiolucency wrt palatal root of 26 extending backwards. It appeared osteolytic in nature and pathological migration of 26 and 27 was observed.[figure 6] Orthopantomogram showed generalized hazy appearance of bone trabeculae in maxilla with radiolucent to mixed appearance of the region in left quadrant in relation to 25, 26, 27 and complete loss of cortical and cancellous bone evident. Irregularity in floor of maxillary sinuses bilaterally was evident. Multilocular radiolucencies extending from inferior cortex of symphysis to right body region with subperiosteal bone resorption at right angle of mandible. There was generalized loss of lamina dura [figure 7]. Skeletal radiographs showed multiple cystic lesions in bones [Figure 8].

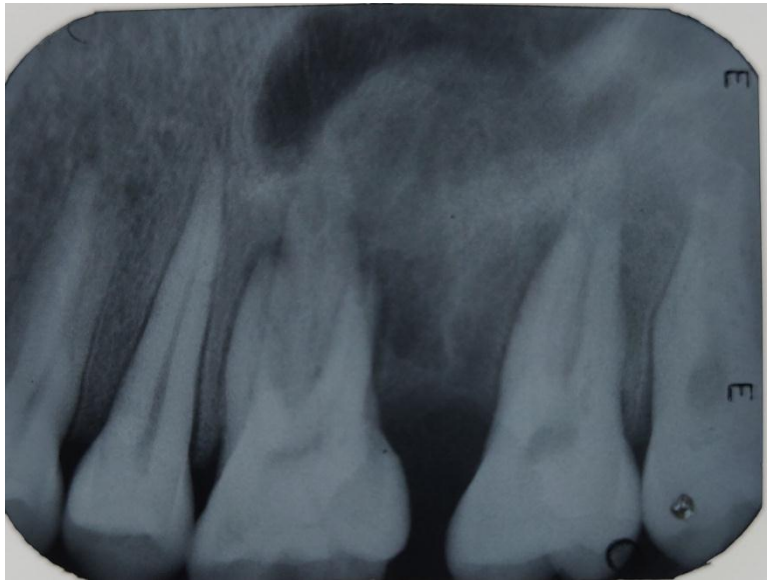


Figure 6

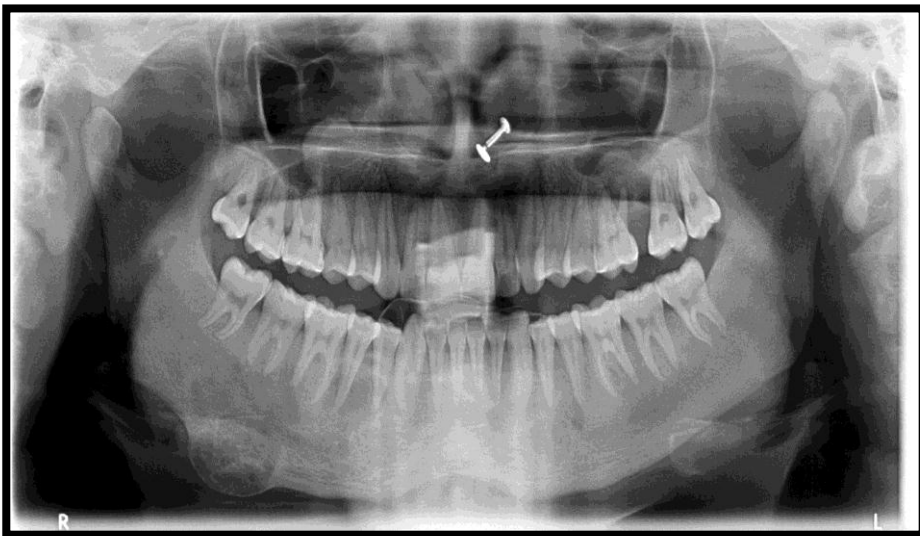


Figure 7

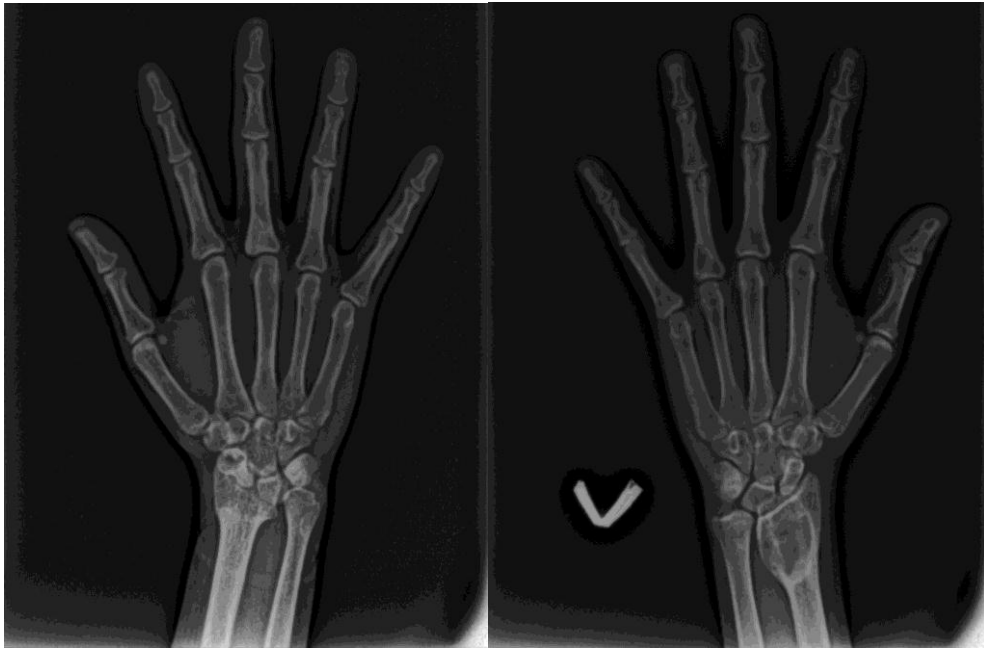


Figure 8

Computed tomography of maxilla revealed generalized appearance of cancellous bone in maxilla is with invariably enlarged marrow spaces in the anterior palatal region and complete loss of cortical and cancellous bone evident in left quadrant in relation to 25, 26, 27 regions with radiolucent appearance of the region. (Figures 9,10,11). Scan of mandible showed generalized granular appearance of cancellous bone evident in the entire mandible with more pronounced lesion in the right quadrant and with expansile mixed radiolucent-radiopaque lesion evident in right body of mandible extending from incisor region to the angle. There were perforations of cortical bone in mandible near inferior border at symphyseal and body region. displacement of inferior alveolar canal in superior direction was also evident. (Figure 9,10,11)

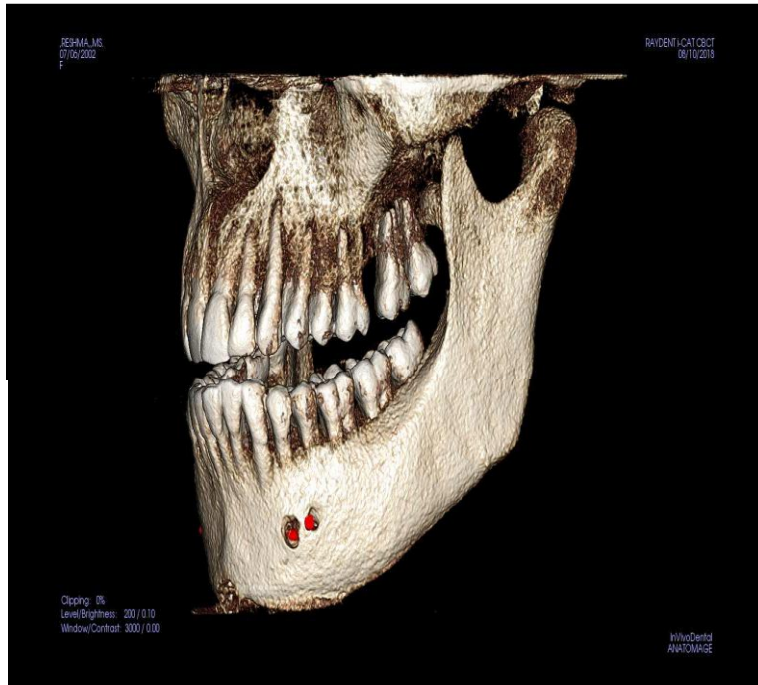


Figure 9

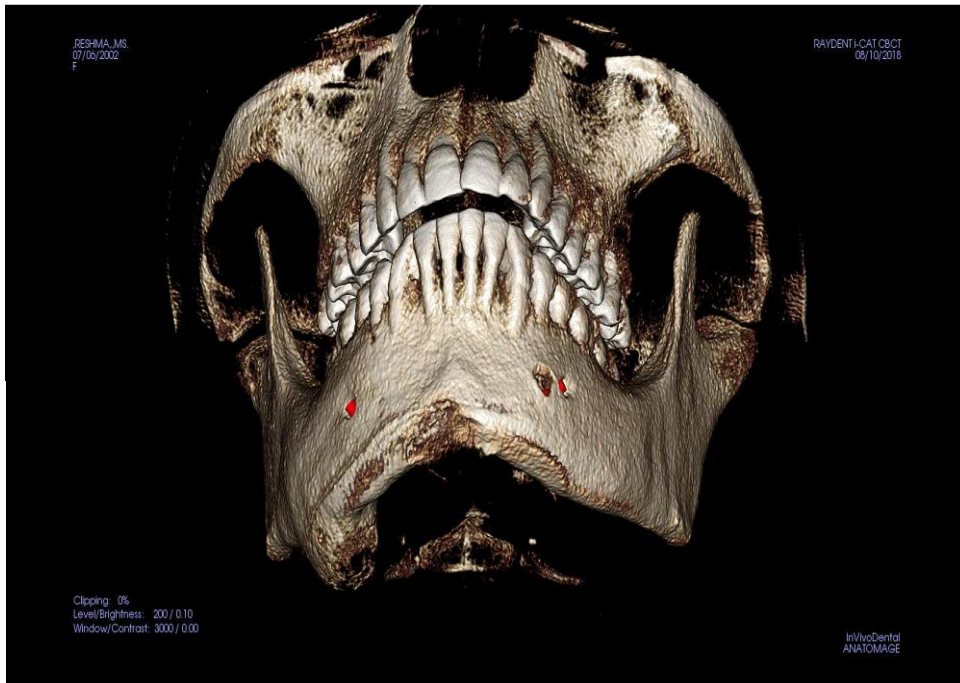


Figure 10

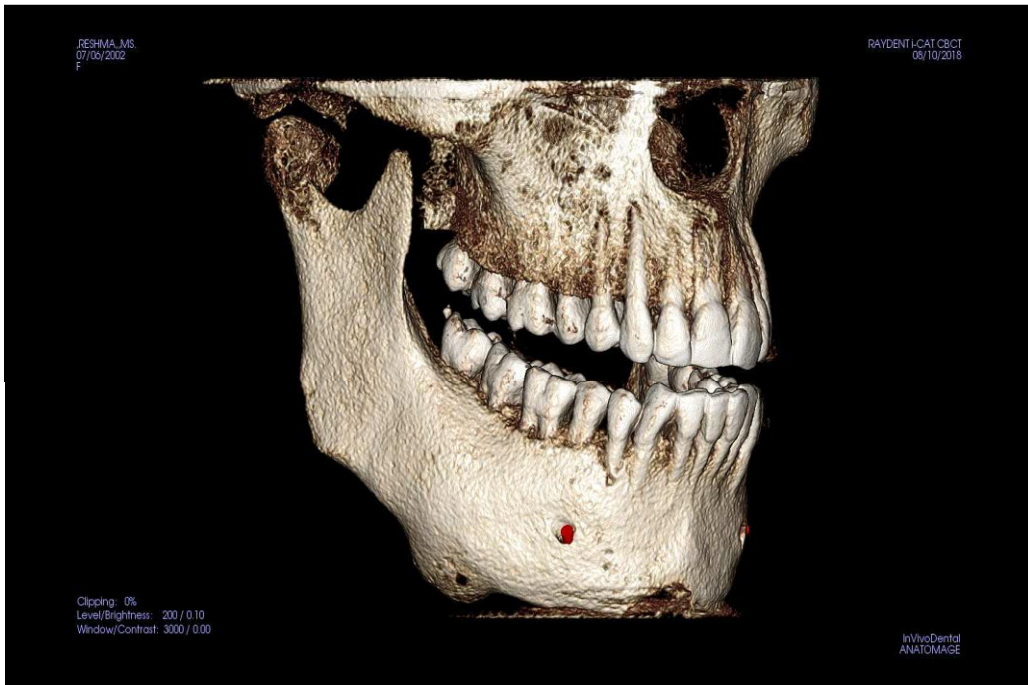


Figure 11

Patient had raised parathormone 1132 pg/ml (Normal: 10–60pg/ml) and serum alkaline phosphatase levels 968.9 IU/L (Normal: 30–120U/L) with elevated serum calcium level 12.60 mg/dl (Normal: 8.5–10.5 ml/dl). High resolution usg showed well defined iso echoic lesion measuring approx 19×14×11 mm attached to inferior pole of right lobe of thyroid where lesion is less vascular to thyroid and separate from it. Parathyroid scintiscan showed small focal tracer uptake below the inferior pole of right lobe of thyroid images acquired after 2 hrs showed wash out of tracer from thyroid but focal uptake below the right lobe of thyroid is persistent and more intense was suggestive of right inferior parathyroid adenoma.(figure-12). She was diagnosed with right parathyroid gland adenoma and parathyroidectomy was performed. After surgery both ionic and serum calcium were found to be in normal range.

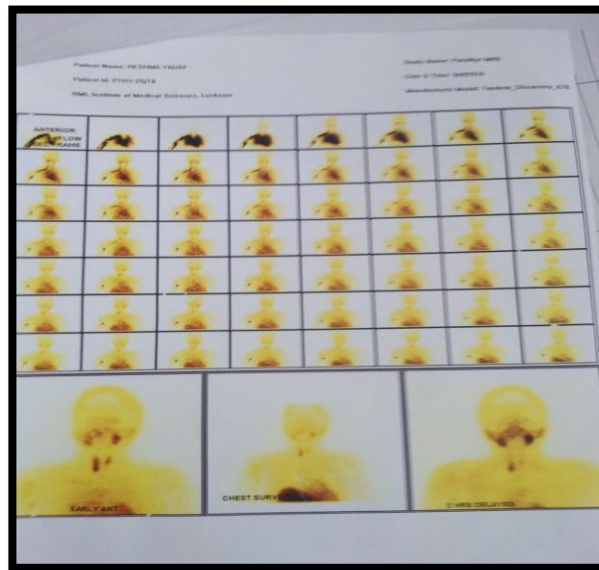


Figure 12. Parathyroid nuclear imaging scan showing intense uptake on right inferior parathyroid suggestive of right inferior parathyroid adenoma

Discussion

Hyperparathyroidism is a condition when there is excess production of parathyroid hormone by parathyroid glands¹ which are two pairs of glands usually positioned behind the left and right lobes of the thyroid. Each gland is a yellowish-brown flat ovoid that resembles a lentil seed, usually about 6 mm long and 3 to 4 mm wide, and 1 to 2 mm anteroposteriorly.² The two parathyroid glands on each side which are positioned higher are called the superior parathyroid glands, while the lower two are called the inferior parathyroid glands.³

The function of the parathyroid glands is to maintain calcium and phosphate levels of body within the normal range. The parathyroid glands secrete parathyroid hormone (PTH)⁴ also known as parathormone which is a protein that contributes in maintenance of calcium and phosphate homeostasis, as well as bone physiology. Parathyroid hormone has effects antagonistic to the calcitonin hormone.⁵ Sylvanus (1743) was the first to diagnose hyperparathyroidism. Recklinghausen (1891) gave the first description of the associated bone changes known as osteitis fibrosa cystica also known as brown tumour⁶ which is one of the important complications of primary hyperparathyroidism. It must be differentiated from other true giant cell tumors of bone, and it represents reparative granuloma rather than a true neoplastic process.^{7,6} The reported prevalence of brown tumor is 0.1% and can occur in mandible, maxilla, clavicle, ribs and pelvic bones. Furthermore the frequency of occurrence is more among persons older than 50 years of age with a male to female ratio of 1:3.^{8,9,10} In the present case, the patient was 17-year-old female and brown tumors of maxillary and mandibular bone were found to be the important clinical manifestation of PHPT.

Clinically, brown tumors may be observed as small, asymptomatic swelling or symptomatic exophytic mass in the jaw bone in hyperparathyroidism. They can be found in the facial bones, pelvis, ribs and femur. They may cause swelling, pathological fracture, and bone pain. Early findings as suprapariosteal erosion may be seen on radiographs of bones of the hands. They may be multiple in the terminal stage of hyperparathyroidism or in the parathyroid carcinomas. Radiographically, involvement of jaws in hyperparathyroidism can manifest as brown tumors appearing as well-defined marginated expansile lytic lesions, causing cortical expansion with other bony changes associated, such as generalized demineralization of the medullary bones, loss of lamina dura around the roots of teeth, and subperiosteal erosion of mandibular angle^{11,6}. All these findings were evident in our case. Histologically, brown tumors are characterized by several osteoclast-like multinucleated giant cells often interspersed with hemorrhagic infiltrates and hemosiderin deposits and vascular fibroblastic stroma¹²

In hyperparathyroidism, when brown tumors are observed, the diagnosis must be confirmed by establishing increased serum calcium and PTH levels because histological features alone are insufficient as it may resemble giant cell tumor. Ultrasonography (USG) is one of the most common primary imaging methods used for neck evaluation in the hyperparathyroidism assessment as done in our case. On USG, parathyroid adenoma is seen typically as a round or oval homogenous, hypoechoic or isoechoic nodule localized behind the thyroid gland and at the lower aspect of paratracheal or paraesophageal region. The parathyroid technetium scintiscan is one of the most preferred imaging modality to localize diseased parathyroid glands prior to surgery as done in our case.¹¹

The initial step in the management PHPT involves control of PTH and a partial parathyroidectomy is considered effective in spontaneous regression of small osteolytic jaw lesions. However, surgical excision may be indicated in large symptomatic lesion usually done after parathyroid surgery. Post-operative hypocalcemia may occur in patients who undergo a partial parathyroidectomy. Therefore, calcium supplements may be required. In our case patient was found to be normocalcemic post surgery.

Conclusion

The dentists and oral physicians should be alert of the possible occurrence of brown tumors in the jaws of the previously undiagnosed and diagnosed patients of hyperparathyroidism because the success of treatment largely depends upon the correction of underlying systemic disease in such cases. Careful history taking, proper physical examination and investigations with proper knowledge are must for assessment and correct diagnosis of such oral manifestations of systemic diseases. Hence we can say that mouth is the mirror of our health.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients

understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial Support and Sponsorship: Nil.

Conflicts of Interest: None.

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