

*Case Report*

## **Brown Tumour of Maxilla Presenting As First Manifestation of Primary Hyperparathyroidism Due to Parathyroid Adenoma - Case Report with Radiological Review**

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### **ABSTRACT**

Brown tumour is rarely the first symptom of hyperparathyroidism. It is a localized form of osteitis fibrosa cystica. Primary hyperparathyroidism is characterized by hypersecretion of parathormone in which bone loss occurs due to resorption of bone. Now-a-days diagnosis of hyperparathyroidism is done in asymptomatic or minimally symptomatic stage. In this case report, we present to you a rare case of brown tumour of maxilla and mandible in a 27-year-old male patient who presented with multiple painful left maxillary and mandibular swellings suspicious of neoplasia. These were diagnosed on Computed tomography (CT) as brown tumors. Parathyroid adenoma was diagnosed retrospectively on ultrasonography as cause of hyperparathyroidism.

**Key Words:** Brown tumour, Maxilla, Hyperparathyroidism, Osteitis Fibrosa Cystica, Parathyroid adenoma.

### **INTRODUCTION**

Hyperparathyroidism is the commonest etiology of hypercalcemia in outdoor patients. The incidence is 2-3 per 1000 in women and 1 per 1000 in men. [1, 2] In patients with primary hyperparathyroidism, 80-85 % have parathyroid adenoma, 15-20 % have parathyroid hyperplasia while less than 0.5 % have parathyroid carcinoma. [3]

Brown tumours are rare sequelae of hyperparathyroidism. The reported prevalence of brown tumours is 0.1 %. They can be presentation of both primary as well

as secondary hyperparathyroidism. The reported occurrence is 4.5 % in primary disease and 1.5 % in secondary hyperparathyroidism. The common sites of brown tumour are the long bones, pelvic girdle, clavicle, ribs and mandible. Tumours involving the maxillae are rare. They are seen more commonly in mandible than maxilla. [4, 5]

They manifest as reactive lesions in the bone, very seldom seen during the course of hyperparathyroidism and are secondary to localised rapid osteoclastic bone turnover resulting from direct effect of

parathormone. The normal marrow tissue is replaced by haemorrhage, vascular fibrous tissue and eventually granulation tissue. These tumours have a brown or yellow hue. The bone expansion results due to localised accumulation of fibrous tissues and giant cells. All these changes described above are reversible with removal of the parathyroid adenoma or all four glands in hyperplasia. [1,2]

## CASE REPORT

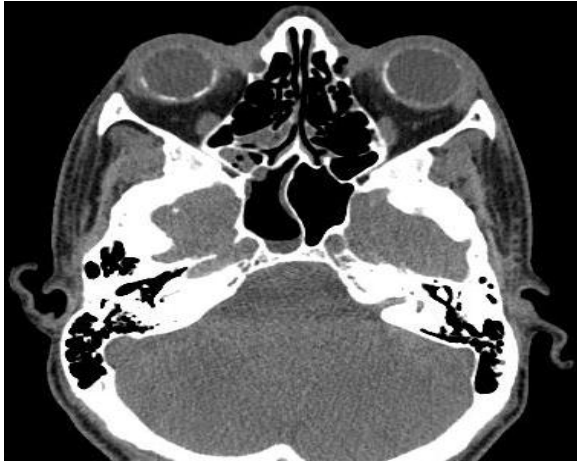
A 27-year-old gentleman presented to the Oral-Maxillofacial Department with pain and swelling over the upper and lower jaw. He noticed the swelling 4 months prior to presentation, which had been gradually increasing in size. He reported no recent dental problem or dental surgery or trauma to the face. He was otherwise systemically well, and reported no weight loss, fevers or rigors. The patient had no other significant medical history and was not on any regular medications.

On physical examination, he was afebrile and systemically well, with no cervical lymphadenopathy. The left cheek was visibly more swollen than the contralateral side, which exhibited no evidence of infection on clinical assessment. Intraorally, there was cortical expansion of mandible and maxilla it was present in the buccal vestibule and hard palate. The swelling was noted to be hard and tender. Maximal mouth opening was normal for the patient. The floor of mouth was soft on palpation. Initial differential diagnoses included odontogenic cyst or ameloblastoma.

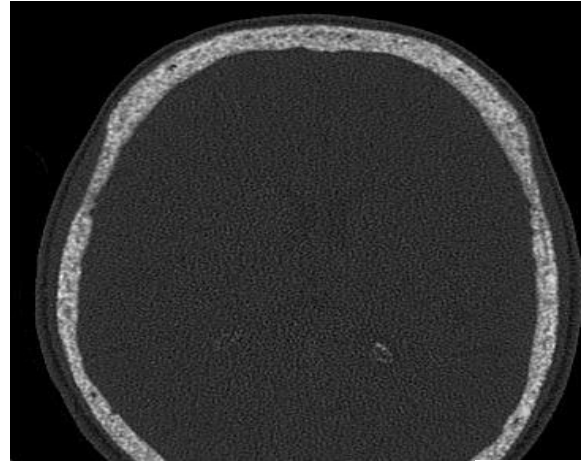
Computed tomography (CT) PNS revealed: Sclerochoroidal calcification noted in both eyeballs (Fig.1). Entire skull vault showed granular appearance with loss of distinction of inner & outer table - "Pepper pot skull" appearance (Fig.2). Well defined expansile lesion measuring approx. 4.7 x 2.7

x 3.2 cm was noted involving anterior, posterolateral, medial walls and roof of left maxillary sinus having both solid and cystic components. Solid component showed moderate enhancement in contrast study. Medially, it was extending into adjoining left nasal cavity. Laterally, it was extending into left retroantral fat. Superiorly, it was eroding through left orbital floor and extending in left extraconal compartment (Fig.3a-j). Expansile osteolytic lesion measuring approx. 3.3 x 2.8 x 2.4 cm was noted in anterior portion of bony palate with thinning and erosion of overlying cortex at places. It was filled with soft tissue component showing moderate homogenous enhancement in contrast study (Fig.3a-d, f, h, i). Expansile osteolytic lesion filled with soft tissue showing homogenous enhancement on contrast measuring approx. 1.4 x 1.1 cm was noted in anterior wall of right maxillary sinus (Fig.3c, d) and 3 x 1.7 cm noted in body of left mandible (Fig.3j). Ultrasound (US) revealed a large well defined hypoechoic solid lesion measuring 2.1 (AP) x 2.75 (T) cm posterior to lower pole of left thyroid gland. No calcification or cystic areas were noted. On Doppler study, the lesion showed moderate vascularity. Both lobes of thyroid gland and isthmus were normal with no focal lesion. No cervical lymphadenopathy was noted on either side. In view of characteristic location and appearance, a diagnosis of parathyroid adenoma was made (Fig.4a-d).

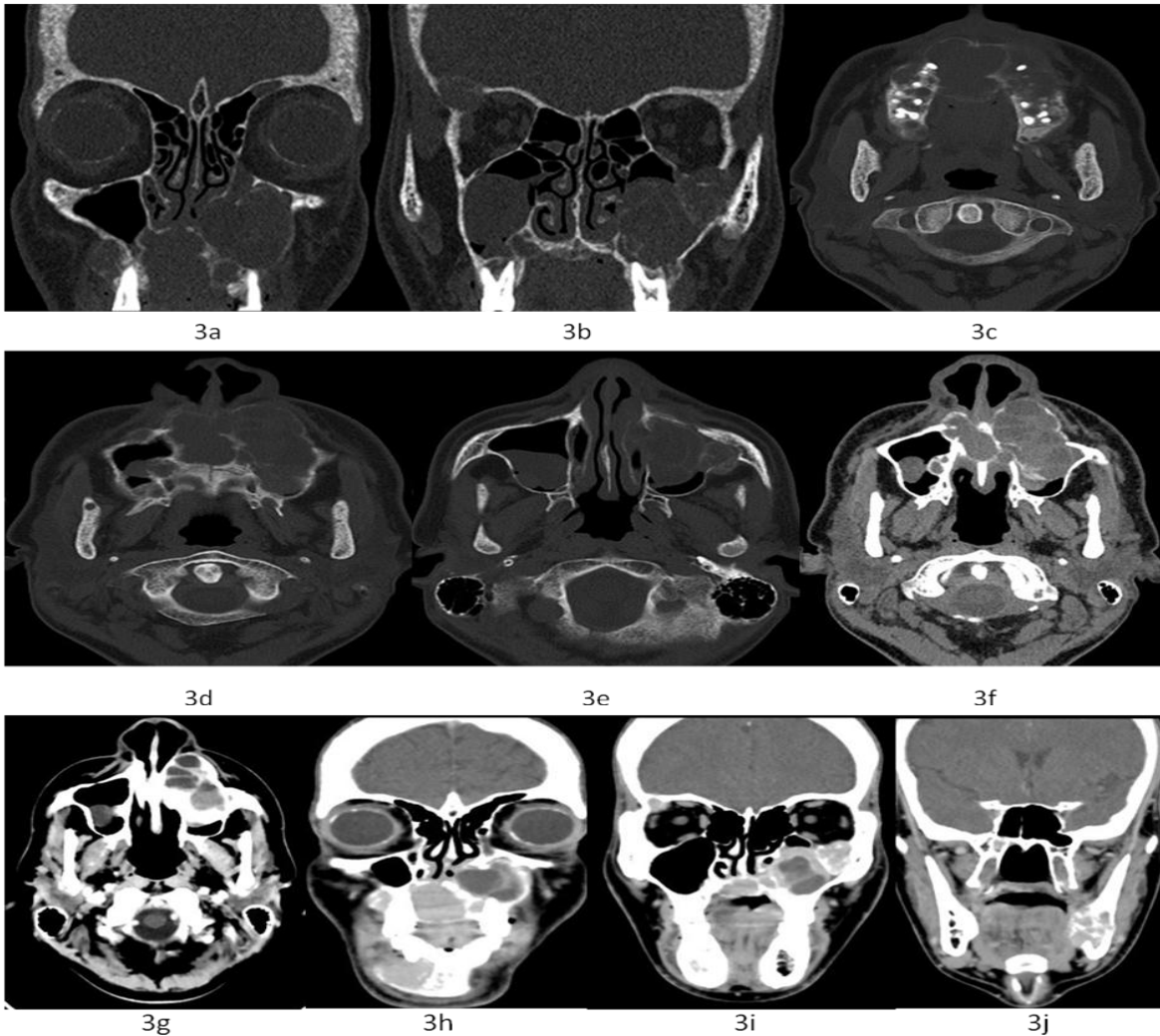
Serum biochemistry revealed a serum calcium level of >15 mg/dl (normal range 8.8-10.6 mg/dl), PTH level of 1198 pg/ml (normal range 15-65 pg/ml) with alkaline phosphatase, renal and liver profiles within normal limits. FNAC from maxillary lesion and neck lesion confirmed diagnosis of brown tumor and parathyroid adenoma respectively (Fig. 5a and 5b).



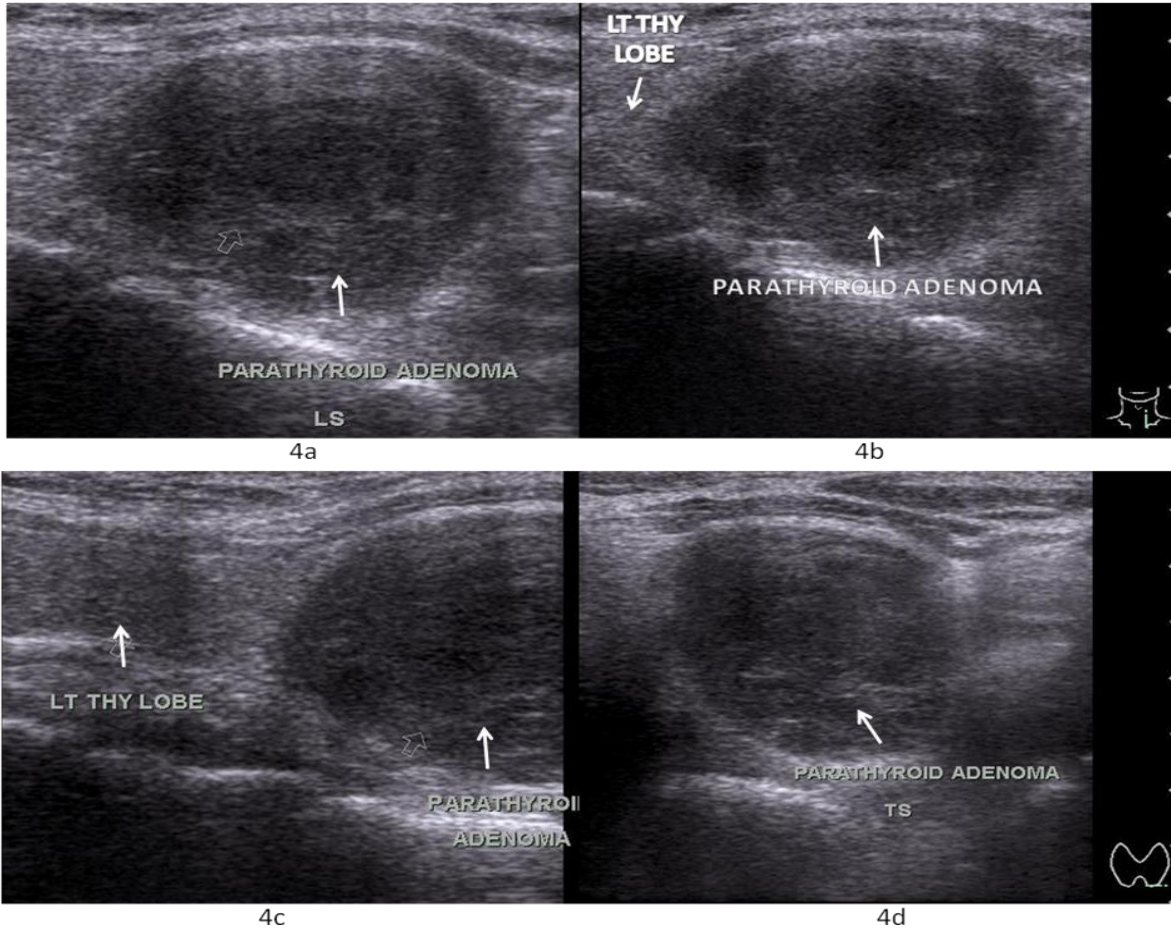
**Fig. 1:** CT Orbit: shows sclerochoroidal calcification in both eye balls.



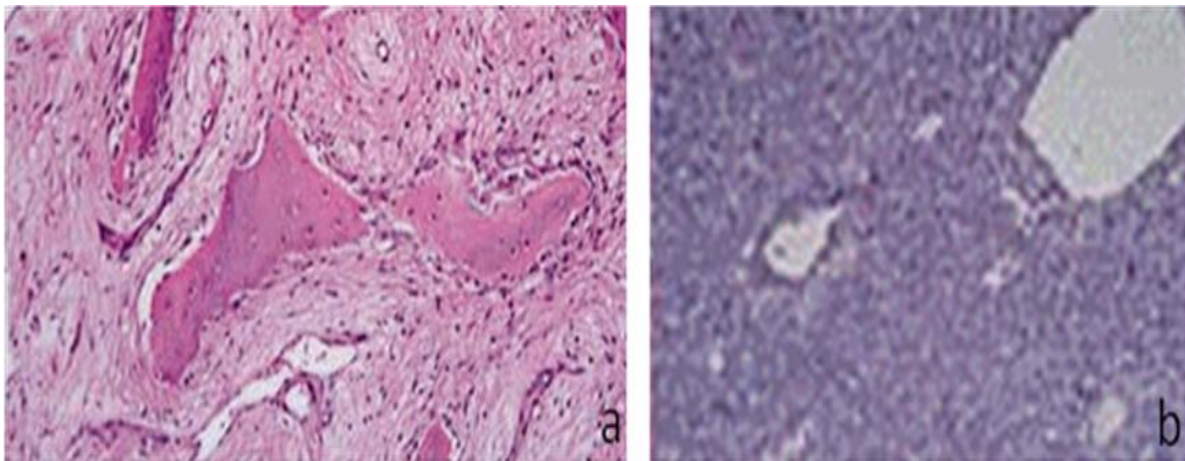
**Fig. 2:** CT skull vault shows granular appearance with loss of distinction of inner & outer table - "Pepper pot skull" appearance.



**Figure 3 (a-j):** Axial and Coronal CT scan of paranasal sinuses revealed well defined expansile lesion involving anterior portion of bony palate filled with soft tissue component showing moderate homogenous enhancement on CECT, well defined expansile lesion involving walls of left maxillary sinus having both solid and cystic component extending in left nasal cavity with moderate enhancement of solid component on CECT. Erosion of left orbital floor with extension in extraconal compartment of left orbit is seen. Expansile osteolytic lesion in anterior wall of right maxillary sinus filled with soft tissue, involving body of mandible on left side.



**Figure 4 a-d:** USG: longitudinal and transverse section of left thyroid lobe showing a well defined hypochoic solid lesion posterior to lower pole of left thyroid lobe.



**Figure 5 a:** Maxillary lesion showing multinucleated giant cells, osteoblastic and osteoclastic areas, mononuclear stromal cells and haemosiderin deposits.

**Figure 5b:** Parathyroid adenoma showing proliferation of principle cells with replacement of normal fat areas of parathyroid gland.

## DISCUSSION

The function of PTH is to maintain a balance in calcium and phosphate levels

between extracellular fluid and bones. Diagnosis of hyperparathyroidism is based on elevated serum calcium and parathyroid

hormone levels. Early diagnosis of hyperparathyroidism in asymptomatic period can be done in early stages due to advances in blood analysis methods. Subsequently, the incidence of bone lesions in patients with hyperparathyroidism has reduced from 80% to current 15%.<sup>[4]</sup>

Hyperparathyroidism can be primary, secondary or tertiary. Primary hyperparathyroidism occurs due to excessive parathyroid hormones secretion by an autonomous gland with resultant hypercalcemia. Secondary hyperparathyroidism occurs in hypocalcemia or vitamin D deficiency, which act as stimulus for parathyroid production. Tertiary hyperparathyroidism occurs in renal failure and results from autonomous functioning glands in patients with long standing secondary hyperparathyroidism. Fourth type of hyperparathyroidism occurs due to ectopic hyperparathyroidism which arises from increased parathyroid hormones levels synthesized in patients with malignant disease.<sup>[6]</sup> Primary hyperthyroidism occurs due to single adenoma (90%), multiple adenomas (4%), nodular hyperplasia (5%) and carcinoma (1%). Secondary hyperthyroidism occurs in chronic renal failure, malabsorption, osteomalacia and rickets. Parathyroid adenoma varies in size from few mm to several cm in diameter.<sup>[7]</sup> A single parathyroid adenoma is seen in sporadic disease while hyperplasia of all four glands is suggestive of familial disease (MEN-1 or 2A).<sup>[8]</sup>

Primary hyperparathyroidism is the third most common endocrinological condition following diabetes mellitus and thyroid diseases. Its annual incidence has been reported as 28/100,000 in the USA.<sup>[9]</sup>

Many systems are affected in primary hyperparathyroidism. Most pronounced changes are observed in bone. Presence of well defined osteolytic

expansile bone lesions in facial bones in case of hyperparathyroidism is highly suggestive of brown tumours.<sup>[10]</sup> Brown tumour is fairly an uncommon lesion associated with hyperparathyroidism.<sup>[11]</sup> Approximately 0.2% patients develop osteitis fibrosa cystica and 0.8% develop brown tumour suggestive of very low frequency of brown tumour in primary hyperthyroidism.

Brown tumour is a localized form of osteitis fibrosa cystica. Axial skeleton is rarely involved. Occasionally, it may cause pathological fracture.<sup>[8]</sup> Brown tumour is more commonly found in ribs, clavicles, pelvis, femur and facial bones. In craniofacial region, mandible is more frequently involved than maxilla.<sup>[12]</sup> Simultaneous involvement of both jaws is extremely rare.<sup>[13]</sup> Amongst osseous manifestation of hyperparathyroidism, brown tumours are seen in 10% of cases and advanced stages of this disease. Mandibular involvement is seen in 4.5%.<sup>[14]</sup>

Parathyroid hormone causes direct effect on bones. As a result there is conversion of osteogenic potential of the cell, changing from osteoblasts to osteoclasts, with bone resorption predominating over the formation of new bone tissue. This leads to intraosseous bleeding and tissue degeneration. As a result cysts may develop. Groups of hemosiderin-loaded macrophages, giant cells and fibroblast fill these cystic lesions. Vascularization, hemorrhages and hemosiderin deposits give rise to the characteristic colour of the lesions and the term brown tumours.

In absence of hyperparathyroidism, other differentials considered are odontogenic cyst and tumours (radicular cyst, periodontal cyst, ameloblastoma); infectious disease (bone abscess, localized osteomyelitis); metastasis from known and unknown primary site (lung, breast, simple

bone cyst, eosinophilic granuloma, giant cell lesions, odontogenic keratocyst, myxoma, odontogenic fibroma. Bone expanding giant cell lesions arising in jaw bones include - central giant cell tumour, giant cell reparative granuloma, cherubism and brown tumor. Differentiation between these giant cell lesions based on radiological and histological examination is difficult. Brown tumours do not exhibit any pathognomonic histological changes. Diagnosis of brown tumour can only be made with evidence of hyperparathyroidism. [4]

Primary treatment of brown tumour is surgical removal of parathyroid pathology following which brown tumours usually regress spontaneously overtime. Regression of brown tumour may take many years. [15] During follow-up, parathyroid hormone, calcium levels, potassium levels and increase in bone mineral mass are looked for, [16] Treatment of brown tumour in jaw includes enucleation and curettage, radical resection and reconstruction, radiation therapy and chemotherapy. [17]

## CONCLUSION

This case highlights that brown tumour should be taken into consideration as a rare differential diagnosis of a bone-destroying lesion of the facial bones. Clinicians should always bear in mind atypical presentations of parathyroid adenomas, which need to be excluded in the presence of hypercalcaemia.

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