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## Case Report

# Primary hyperparathyroidism presenting as a brown tumor in the mandible: A case report ☆☆☆

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## ABSTRACT

A brown tumor is a non-neoplastic lesion resulting from an abnormality of bone metabolism in the context of hyperparathyroidism.

We report the case of a 51-year-old woman who initially consulted for edentulism and a growing mandibular mass. She benefited from a radiological and biological assessment which made the diagnosis of primary hyperparathyroidism combined with a parathyroid adenoma.

We remind through this observation the difficulty to establish a correct diagnosis in patients with an osteolytic process of the maxilla and the necessity to look for hyperparathyroidism in front of a giant cell lesion given the insidious character of this endocrinopathy.

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## Introduction

The term "brown tumor" is derived from its brownish appearance due to its vascularization and hemosiderin deposits [1].

Unlike the name suggests, brown tumors are not the result of a neoplastic process. They are focal bony lesions as a consequence of bone remodeling from either hyperparathyroidism or paraneoplastic syndrome

## Case report

A 51-year-old woman who initially consulted for edentulism and a growing mandibular mass for the past 4 months. The

patient reports a history of generalized weakness, lethargy, and weight loss noted over the past few months. Her previous family and medical history was unremarkable.

On palpation, the swelling was soft to firm and not painful.

The patient was referred for blood tests, which revealed alkaline phosphatase and PTH levels of 823 IU/L (Normal: 36–14 IU/L) and 2500 pg/ml (Normal: 12–65 pg/ml), respectively. Serum calcium level was 13.5 mg/dl (Normal: 8.5–10.5 mg/dl).

Because of the mandibular mass, a CT scan was performed and revealed incidental parathyroid nodules at the lower pole of the thyroid, bilaterally (Fig. 1).

The CT scan also showed an osteolytic bone injury in the left costal and humeral areas (Fig. 2), and a large masse of mandible and maxilla (Fig. 3).

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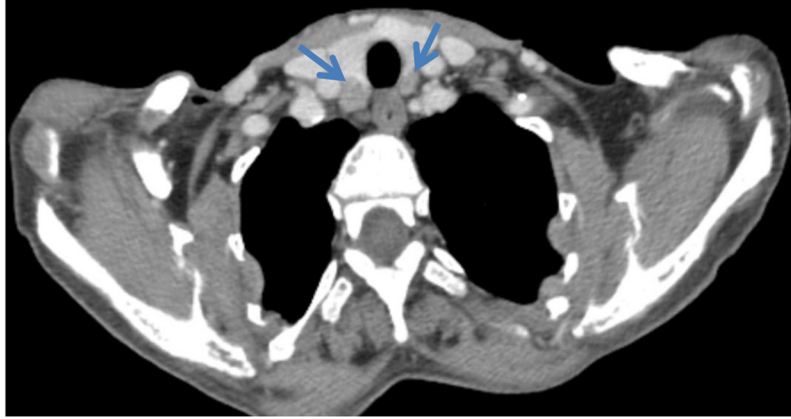
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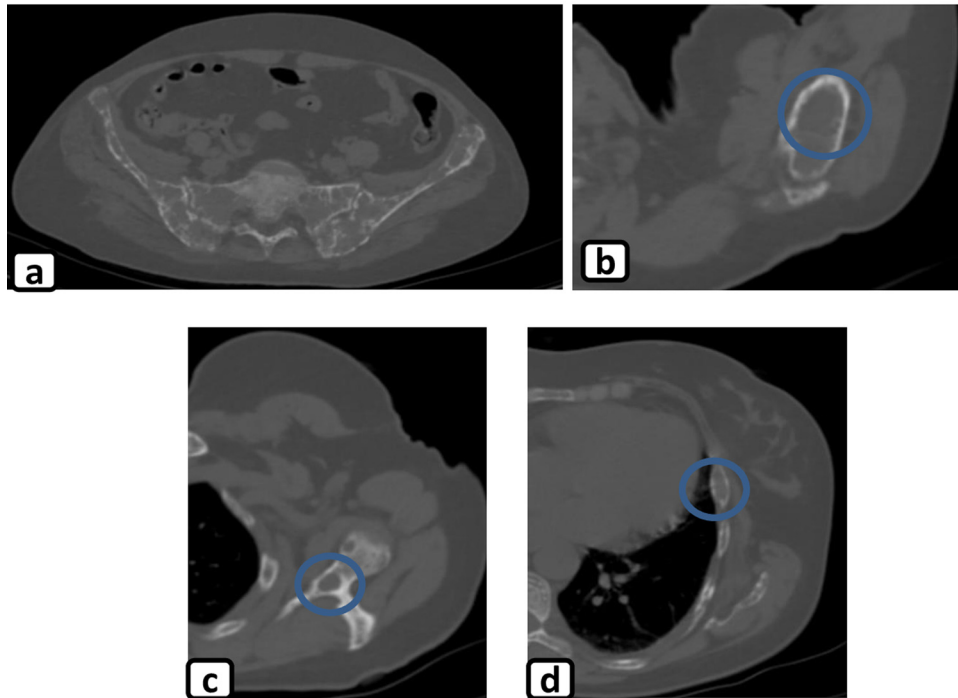
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**Fig. 1 – Cervical CT scan in axial section through the thyroid shows two parathyroid nodules at the inferior pole of the thyroid bilaterally (blue arrow).**



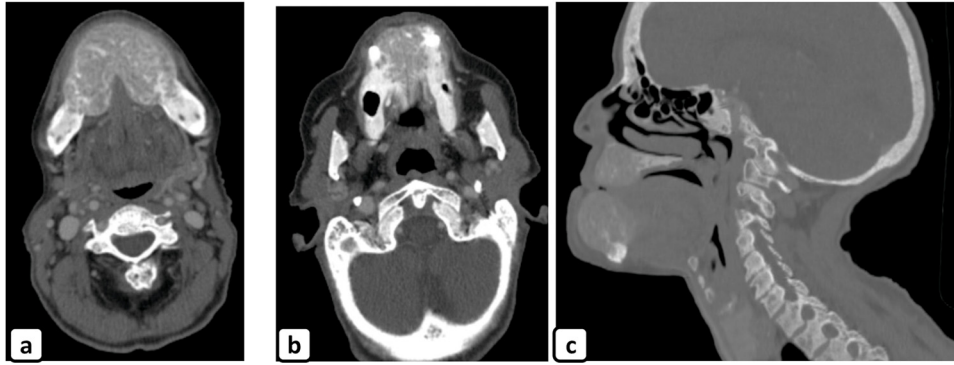
**Fig. 2 – CT scan showed osteolytic lesions in: (A) iliac bone and sacrum; (B) humerus; (C) scapula; (D) ribs.**

The patient proceeded to a cervical ultrasound, which revealed two solid, well-defined, heterogeneous Hypoechoic lesions of the lower thyroid pole, highly vascularized on color Doppler, and measuring approximately 16 mm × 7 mm on the right and 10 mm × 6 mm for the left.

Parathyroid adenoma was removed and histologically it corresponded to hyperplasia. Patient was given oral calcium supplementation in addition to vitamin D3 for possible post-operative hypocalcaemia.

## Discussion

Primary hyperparathyroidism is a common endocrine disorder that is characterized by hypercalcaemia and elevated or inappropriately normal serum levels of parathyroid hormone [1]. Single benign parathyroid adenoma is the most common cause of this disease (about 80% of the patients), whereas four gland parathyroid hyperplasia accounts for ap-



**Fig. 3 – Bone window CT scan, (A) axial section through the maxillary bone; (B) axial section through the mandible bone, (C) sagittal facial section: Two osteolytic tissue masses of the prominence of the chin and the palatal process, following their shapes, well limited, of mixed density associating dense areas and areas of osteocondensation, enhanced after contrast, combined with edentulism, without invasion of the soft tissues, or any periosteal reaction.**

proximately 15%-20% [2], multiple parathyroid adenomas for 5% and parathyroid cancer for <1% of cases [1]. Brown tumor of bone, a rare non-neoplastic lesion resulted from abnormal bone metabolism in hyperparathyroidism, mostly affects mandible, maxilla, clavicle, ribs, pelvis, and femur [3]. When brown tumors associate with primary hyperparathyroidism, they are most frequently caused by adenomas [4].

The reported prevalence of brown tumor is 0.1%. Furthermore the frequency of occurrence is more among persons older than 50 years of age with a male to female ratio of 1:3 [5,6].

Clinically, brown tumors present as a slow growing palpable bony swelling and may cause bone pain or pathological fractures. It is also possible to present with symptoms of weakness, weight loss, polyuria and recurrent stone formation associated with the hyperparathyroidism

Ultrasound is commonly used for patients with suspected parathyroid adenoma. The adenoma is frequently identified as a homogeneously hypoechogenic lesion overlying the thyroid gland. The lower pole of the thyroid should be closely examined, as the parathyroid adenoma is commonly found inferior, posterior or lateral to this position. The entire thyroid gland should be reviewed as the superior parathyroid gland can be found in the upper or mid pole of the thyroid [7].

<sup>99m</sup>Tc-sestamibi scintigraphy can help confirm the presence and location of a parathyroid adenoma.

The radiological features of brown tumors consisted of single or multiple well-defined osteolytic lesions, single or multi-lobular, usually with bone expansion and can demonstrate bony destruction and be associated with pathological fractures. There is also variability in its radiological characteristics, which can include an ill-defined lesion, mixed lytic/sclerotic lesion or margin, with adjacent soft tissue involvement [8]. In these situations, a malignant lesion should be considered in the differential diagnosis and histological confirmation is recommended. Further correlation with hypercalcaemia and hyperparathyroidism is also important because histological differentiation between brown tumor and other giant cell-containing lesions can be difficult.

On x-ray, solitary, or multiple expansible lesions with sharp outlines can be seen. Sclerotic margins, invading tissue, and sclerotic rim are rare. CT scan reveals expansible lytic lesions, with various bone erosion and cortical attenuation. The lesions are relatively well demarcated. On MRI, Brown tumors show hypo intense on T1-weighted images, hypo intense or hyper intense on T2-weighted images. Spinal MRI can show compression fractures of vertebra and intraspinal soft tissue masses with neural compression [9]. PET/CT can also be sensitive in detecting brown tumors [10].

Definitive treatment of primary HPT depends on surgical removal of the underlying cause. Treatment of brown tumors relies on management of hyperparathyroidism. Osseous lesions usually shrink and ossify, however, rarely when the pain persists, surgical enucleation is needed.

## Conclusion

Brown tumor is a rare bony disorder as a consequence of hyperparathyroidism. Apart from biochemical investigations for hyperparathyroidism, both US and sestamibi scintigraphy should be used to confirm the location of the active parathyroid gland. The radiologic features of brown tumor can be variable. CT and/or MRI should be performed to help differentiate from other bone tumors, which should be considered even in the context of hyperparathyroidism. If there is any doubt in the diagnosis, biopsy of the bone tumor is recommended for definitive diagnosis.

## Patient consent

Oral and signed consent was obtained from the patient concerned. The study was conducted anonymously.

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