

(CASE REPORT)



## Brown tumor of multiple facial bones associated with tertiary hyperparathyroidism in a patient on maintenance hemodialysis

Nezha El-bouhi \*, Halima Sobhi, Meriem Chettati, Wafaa Fadili and Inas Laouad

*Nephrology Department, Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Marrakech, Morocco.*

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### Abstract

A brown tumor is a non-cancerous bone lesion that develops due to excessive parathyroid hormone (PTH) levels and can result in bone resorption, ultimately contributing to skeletal deformities. We describe one rare case of a patient on Hemodialysis who develops a brown tumor of multiple facial bones due to Tertiary Hyperparathyroidism.

**Keywords:** Brown tumor; Tertiary Hyperparathyroidism; Hemodialysis

### 1. Introduction

The brown tumor is a rare non-neoplastic bone lesion due to rapid bone loss replaced by hemorrhage and reparative granulation tissue. The presence of hemosiderin causes the brownish color that gives the name to the lesion. (1,2) This lesion rarely involves the craniofacial region. The ribs, femora, and pelvis are brown tumors' most commonly seen sites. (3,4) It is present in only 0.1% of all cases of hyperparathyroidism (HPT)(5). Hyperparathyroidism (HPT) is a disorder characterized by excessive parathyroid hormone secretion by the parathyroid glands. Depending on the cause of this PTH production, HPT can be characterized into primary, secondary, and tertiary forms. (6)Chronic renal failure is the main cause of secondary HPT. (1,4) It results in hypocalcemia and the parathyroid glands over-function to compensate for this low serum calcium level. Tertiary HPT results from long-standing secondary HPT that the parathyroid glands gain an autonomous character. (2,4)

### 2. Case report

A 46-year-old female patient with a medical history of hypertensive heart disease has been undergoing dialysis twice a week for six years. The patient complained of painful swelling of multiple facial bones and exhibited a mild mouth-opening limitation. Physical examination revealed that she was short of stature and had a slight facial asymmetry and swelling of cheekbones. Intraoral examination showed anterior palatal enlargement.

Laboratory tests revealed an elevated serum calcium level of 101 g/L and a phosphorus level of 61.90 g/L. The serum alkaline phosphatase concentration was 980 IU/L. Subsequent examination revealed a parathyroid hormone (PTH) level of 4998.12 pg/mL.

Radiological examination showed multilocular radiolucency within the mandibular and maxillary bone, with decreased bone density affecting femoral and humeral bones.

Parathyroid scintigraphy with MIBI-99m Tc showed abnormally high uptake at the lower pole of the left lobe of the thyroid and the lower pole of the right lobe of the thyroid. These were interpreted as parathyroid hyperplasia.

\* Corresponding author: Nezha El-bouhi

Based on the physical examination, radiology, and laboratory findings, we concluded brown tumor was the diagnosis due to tertiary hyperparathyroidism. Subtotal parathyroidectomy was performed (the parathyroidectomy involved the left parathyroid glands, the lower right gland, and 2/3 of the upper right gland). This was followed by a significant PTH level drop (23.8 pg/ml) with a decrease in serum calcium level (68.3 g/l). intravenous calcium infusion was performed to correct the postoperative hypocalcemia.

The patient reported a marked absence of pain with normalization of serum calcium levels.

### 3. Discussion

Chronic kidney disease (CKD) that persists over time is linked to various metabolic disruptions, which can increase the secretion of parathyroid hormone (PTH). These disruptions include hyperphosphatemia, a deficiency of calcitriol, and hypocalcemia. Hyperphosphatemia directly stimulates the cells of the parathyroid gland, leading to nodular hyperplasia and an increase in PTH secretion. Additionally, prolonged hypocalcemia causes hyperplasia of the chief cells in the parathyroid gland, resulting in excess PTH. When there is an excessive secretion of PTH by the parathyroid glands, usually as a consequence of prolonged secondary hyperparathyroidism, it is referred to as tertiary hyperparathyroidism (HPT III)(2). HPT can lead to bone resorption and the development of microfractures in cortical bone. The resulting bleeding in the bone tissue triggers a response from multinucleated macrophages, leading to the ingrowth of granulation tissue. This pathological process ultimately culminates in the formation of a benign bone lesion known as a Brown tumor. (7). It is usually an uncommon lesion occurring with a frequency of 4.5% in primary hyperparathyroidism (HPT) and 1.5-1.7% in cases of secondary HPT. Prevalence in patients with tertiary HPT is unknown and has rarely been reported. (8) Only 4.5% of reported cases have involved the facial bones. Tumors of the maxillofacial region are more likely to affect women than men. Maxillofacial tumors usually involve solitary bones, with the mandible being the most frequently affected(9).

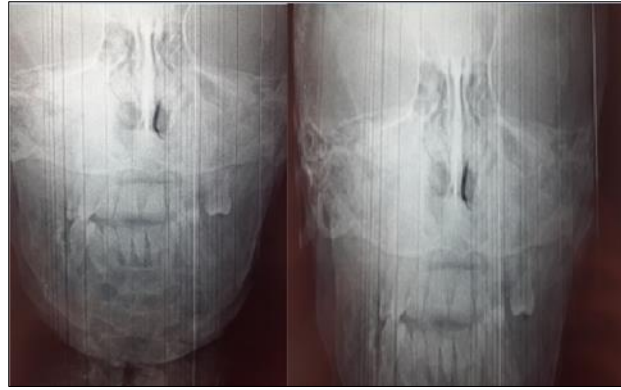
The case presented by our patient is of particular interest, as it involves a rare occurrence of Brown tumor resulting from tertiary hyperparathyroidism. Multiple facial bones were involved, which is rare (9).

The primary medical treatments for tertiary hyperparathyroidism (HPT) aim to manage hyperphosphatemia through dietary restrictions, phosphate binders, vitamin D replacement, calcimimetic agents (such as cinacalcet), and calcium supplementation. According to the National Kidney Foundation's Kidney Disease Quality Outcomes Initiative (KDOQI), parathyroidectomy should be considered for patients with severe HPT (PTH >800 pg/mL) who are unresponsive to medical interventions and have concurrent hypercalcemia and/or hyperphosphatemia. (8) In cases where the brown tumor causes anatomic complications such as facial pain or compromised vision, hearing, chewing, speaking, and breathing, surgical excision of the lesion may be a viable treatment option to consider (8).

Our patient presented with mild symptoms associated with the brown tumor, which did not require surgical intervention for the bone lesions. Following parathyroidectomy, the patient demonstrated some improvement in her symptoms.



**Figure 1** (a) Facial asymmetry (b) Palatal enlargement



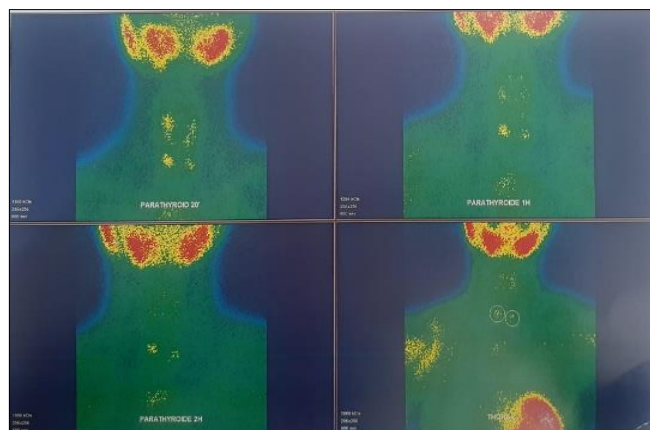
**Figure 2** Multilocular radiolucency within the mandibular and maxillary bone



**Figure 3** Decreased bone density humeral bones



**Figure 4** Decreased bone density affecting femoral bone



**Figure 5** Parathyroid scan showed increased uptake in the parathyroid glands

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#### 4. Conclusion

Brown tumor is a rare but important complication of hyperparathyroidism. The diagnosis of the brown tumor should be considered in patients with hyperparathyroidism who present with bone pain or deformity. Timely diagnosis and management can lead to the resolution of the lesion and prevent further bone loss.

## Compliance with ethical standards

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### *Disclosure of conflict of interest*

No conflict of interest is to be disclosed.

### *Statement of ethical approval*

The present research work does not contain any studies performed on animal/human subjects by any of the authors

### *Statement of informed consent*

Informed consent was obtained from all individual participants included in the study.

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