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Case Report

# Brown Bone Tumors Mimicking Metastases: A Case Report

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## Abstract

Brown tumours are rare non-neoplastic osteolytic lesions that occur in the context of hyperparathyroidism. Parathyroid carcinoma is an extremely rare etiology of hyperparathyroidism, with an incidence of 0.74%.

We report a 28-year-old woman operated for parathyroid carcinoma, who consulted 2 months later with diffuse bone pain. Complementary investigations revealed invasive osteolytic lesions of the bilateral iliacs and left clavicle, with hyperfixation on scintigraphy and hypercalcaemia. The diagnosis of malignant bone lesion was suggested, but pathological examination showed a brown tumour. After excluding a bone malignancy, the decision was taken to continue the treatment of hypercalcaemia and evolution was marked by regression of the bone lesions with normalisation of the calcaemia after 9 months.

This case highlights the importance of thinking about a brown tumour in the presence of an aggressive bone lesion mimicking a malignant lesion and evolving in the context of primary hyperparathyroidism to avoid needless intervention.

***Keywords:*** *Brown Tumour; Metastasis; Hyperparathyroidism; Parathyroid Carcinoma; Scanner; Scintigraphy*

## Abbreviations

CT: Computed Tomography; SPECT: Single Photon Emission Computed Tomography

## Introduction

Brown tumours or cystic fibrous osteitis are rare pseudotumourous osteolytic lesions resulting from a disorder of bone metabolism in response to the direct action of parathormone on the bone structure. They occur in both primary and secondary hyperparathyroidism. They are seen more frequently in primary hyperparathyroidism but with a higher incidence in parathyroid adenoma 80% [1]. Parathyroid carcinoma is a rare type of cancer with an incidence of less than 0.005% [2], and only manifests as primary hyperparathyroidism in 0.74% of patients [3]. Parathyroid carcinoma is a slow-growing tumour with a high recurrence rate (60%) and a high mortality rate (35%) [4]. Hypercalcaemia, nephrocalcinosis, cystic fibrous osteitis, brown tumours, reduced bone structure and fractures may be seen as a result of high levels of the hormone parathormone. Bone metastases have also been reported in patients with parathyroid carcinoma [5], making it a challenge for the radiologist and clinician to distinguish between them in the absence of characteristic clinical or imaging arguments. It should be highlighted that it is very important to distinguish between the two pathologies because therapeutic attitudes differ: brown

tumours regress after treatment of primary hyperparathyroidism, whereas bone metastases are much more difficult to treat. We report here the case of a young 28-year-old patient with a history of parathyroid carcinoma operated on 1 month ago, who presented with lytic bone lesions mimicking secondary lesions, and whose anatomopathological examination showed brown tumours.

## Case Report

We report here the case of a 28-year-old female, whose history of illness dates back to two months ago, with the appearance of a tumefaction on the left cervical region, evolving in the context of an altered general condition, which prompted a consultation in a peripheral hospital. The initial clinical examination revealed an asthenic, haemodynamically stable patient with a firm left basi-cervical tumefaction that was fixed to the superficial and deep planes. Biological tests revealed hypercalcaemia at 3.50 mmol/L with an elevated parathyroid hormone level of 1530 pg/mL (N < 52). Cervical ultrasound and Cervical CT showed a left parathyroid mass measuring: 28 x 20 x 14 mm (Figure 1) and the diagnosis of primary hyperparathyroidism was accepted. The patient underwent surgical resection of the left parathyroid mass. Pathological examination showed a parathyroid carcinoma stage p T2 with vascular invasion and focal damage of resection limits.

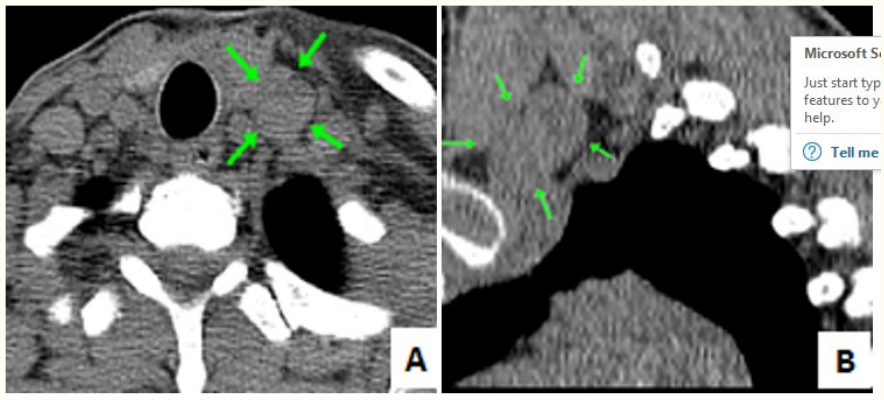
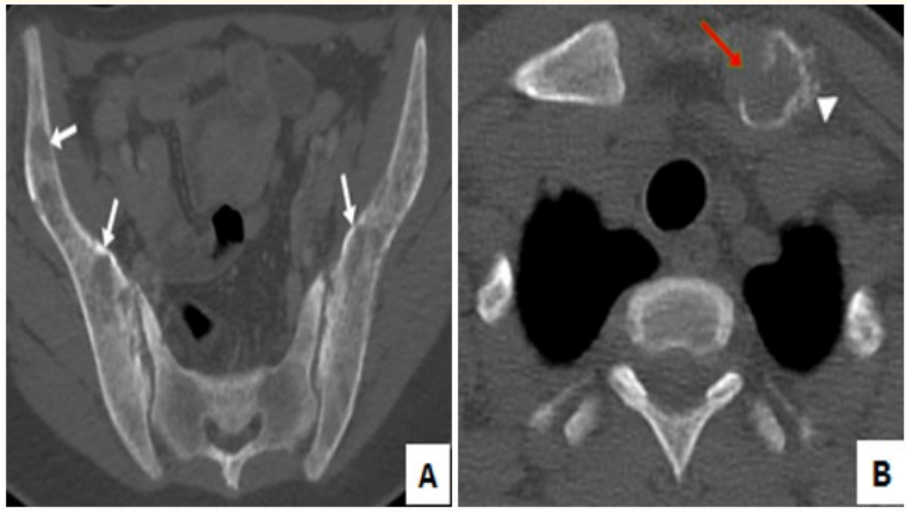



***Figure 1:*** *CT images in axial (A) and sagittal (B) sections passed by the cervical level showing a left inferior parathyroid mass (green arrow) with tissue density, oval, well limited, regular contours, measuring: 28 x 20 x 14 mm.*

Two months later, the patient developed diffuse bone pain with hypercalcaemia of 2.53 mmol/L and a normal parathyroid hormone level. The patient was referred to our department for management. A thoracic-abdominal-pelvic CT scan performed as part of the evaluation revealed lytic bone lesions involving the iliacs bilaterally (Figure 2) and the medial extremity of the left clavicle (Figure 3). Bone scintigraphy showed hyperfixation of the iliacs, the medial extremity of the left clavicle, the facial bones and the cranial vault (Figure 4). In view of these radiological and scintigraphic signs of malignancy, a scannoguided bone biopsy (Figure 5) with anatomopathological study was imperative for histological proof, which showed a giant cell tumour with no signs of malignancy, in favour of a brown tumour. The therapeutic decision was to continue symptomatic treatment of the hypercalcaemia, and to monitor progress, which was marked by clinical improvement, regression of bone lesions on CT scan and normalisation of hypercalcaemia after 9 months of treatment.



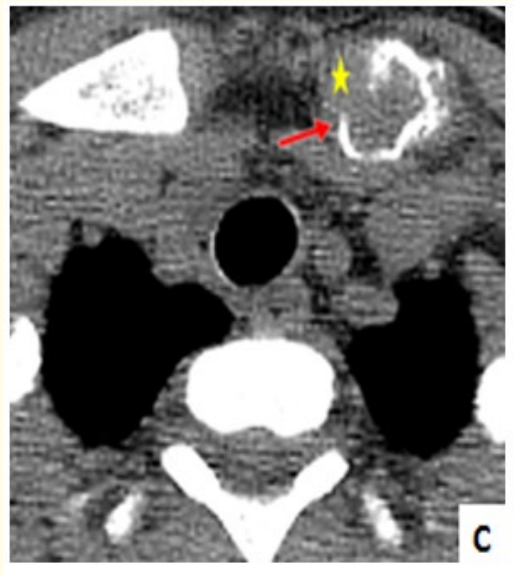




***Figure 2:*** *Computed tomography images, in axial sections, with bone (A and B) and parenchymal (C) windows.*

*- Section through the pelvis (A) showing lytic lesions of the iliac wing bilaterally without osteosclerosis or rupture of the bone cortex (white arrow).*

*- Sections through the clavicle (B and C) showing a lytic bone lesion at the médiale extremity of the left clavicle, blowing and rupturing the cortical bone (red arrow) associated with a periosteal reaction (arrowhead) and invading the adjacent soft tissue (yellow star).*

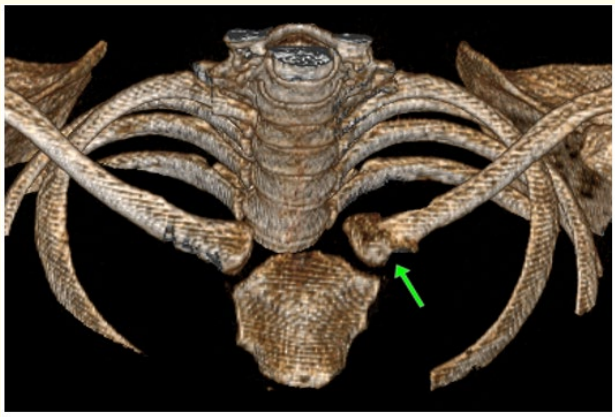


***Figure 3:*** *Volume reconstruction showing the lytic lesion at the mediale extremity of the left clavicle.*

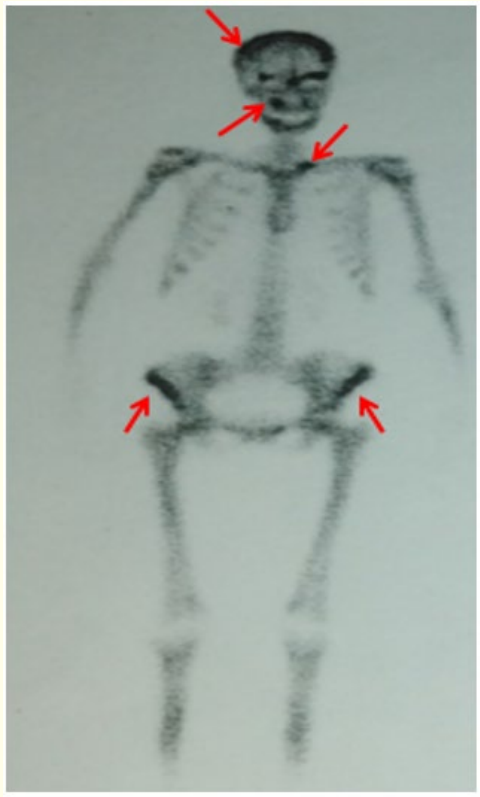


***Figure 4:*** *Anterior view of a bone scan showing multifocal hyperfixing foci (red arrow) involving the bones of the arch of the skull and the face, the medial extremity of the left clavicle and the iliac wings bilaterally.*

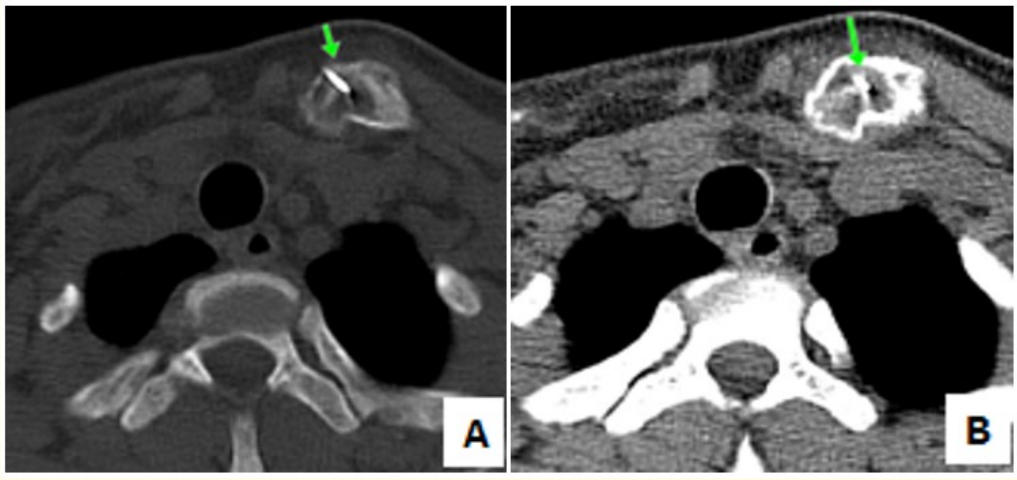



***Figure 5:*** *Axial-slice CT images in the bone window (A) and parenchymal window (B), passing at the level of the clavicle, showing the distal end of the trocar inside the lesion (green arrow).*

## Discussion

Brown tumours are rare lesions of hyperparathyroidism, with an incidence of 3% in primary hyperparathyroidism and 1.5 to 1.7% in secondary hyperparathyroidism [6,7]. They are often related to primary hyperparathyroidism, essentially to parathyroid adenoma (80%) [1] but much more rarely to parathyroid carcinoma (0.74%) [3]. Primary hyperparathyroidism mainly affects patients over 50 years of age, with a predominance of post-menopausal women. Our patient is a 28 year old female with primary hyperparathyroidism secondary to parathyroid carcinoma.

Hyperparathyroidism is currently discovered accidentally in 75 to 80% of cases following asymptomatic hypercalcaemia revealed by blood tests [9], but it can also be revealed by renal lithiasis or cardiovascular disorders [10].

The bone manifestations of hyperparathyroidism are the late expression of the disease, occurring in 5 to 15% of cases [11,12] and include bone cysts, osteoporosis, endosteal resorption and brown tumours. On the other hand, it is exceptional for a single brown tumour to be the revealing sign of parathyroid hyperfunction [10]. Brown tumours can affect the entire skeleton, with a predilection for the pelvis, ribs, mandible and hands [8]. The diagnosis of a brown tumour can be difficult, mimicking a primary malignant or secondary bone tumour, especially if the lesion is unique, accompanied by inflammatory bone pain and evolving in a context of altered general condition [13], or in the presence of a history of neoplasia. Our patient had primary hyperparathyroidism secondary to parathyroid carcinoma, which made the diagnosis more difficult.

On standard radiography, the appearance of brown tumours is variable and non-specific; the most common appearance is of single or multi-locular bone lysis, with unclear margins responsible for blowing or even rupture of the cortices, in which case the appearance may suggest malignancy in aggressive lesions. The appearance on CT is also variable, with either osteolytic lesions of high tissue density, taking contrast, with no invasion of the soft tissues or periosteal reaction [15] or lesions invading the soft tissues and giving a pseudotumoral appearance [16]. Other radiological manifestations include subperiosteal resorption, typically located in the phalanges [17]. Ultrasound and cervical CT scans are performed to look for lesions of the parathyroid glands. Bone scintigraphy shows multiple foci of hyperfixation,

most often localised in the skull, facial bones, pelvis, ribs and femur [18]. The use of SPECT/CT allows better characterisation of the lesions, which combine intense focal hyperfixation with an osteolytic, sometimes expansive appearance surrounded by osteosclerosis [19]. In our patient, both the radiological and scintigraphic appearance were in favour of a malignant bone tumour, justifying a scan-guided bone biopsy. Pathological examination showed a giant cell tumour but no signs of malignancy.

Brown tumours are part of giant cell tumours, which include true giant cell tumours (formerly known as myeloplastic tumours), central reparative giant cell granulomas and aneurysmal cysts [9,14]. It is difficult to distinguish between these entities on the basis of histological data alone. Brown tumours are manifested by non-pathognomonic histological changes which may be found in the other entities [9,20]. The diagnosis of brown tumours is based on a combination of clinical, biological and, above all, hormonal factors. However, if there is evidence of a disturbance in the phosphocalcium balance, hyperparathyroidism is suspected, and if parathyroid hormone level rises, the diagnosis is made [9]. It is essential to distinguish between brown tumours, which are benign non-neoplastic bone lesions with no potential for malignancy, and which are treated medically, and true giant cell tumours, which carry a risk of malignant transformation with the possibility of developing pulmonary metastases, making radical surgical treatment necessary.

Treatment of hyperparathyroidism is the first step in the management of brown tumours. Indeed, there is a general consensus that parathyroidectomy is the treatment of choice for primary hyperparathyroidism, whereas opinions differ on the treatment of secondary bone lesions. The evolution of brown tumours after parathyroidectomy is variable and depends on their composition [21]. Most authors believe that brown tumours can regress spontaneously after correction of hyperparathyroidism [9], with replacement of bone lesions by normal bone tissue. According to some authors [22], disappearance of the lesion is possible six months after treatment of hyperparathyroidism, and others believe that spontaneous bone regeneration may take several years before normal morphology is restored [23]. In the case of destructive lesions affecting organ function, the tissue damage caused cannot be repaired despite the restoration of normal blood calcium levels. In these situations, or when lesions persist despite treatment of hyperparathyroidism or continue to grow despite hormonal treatment, Yamazaki [24] recommends curettage and enucleation of the tumour. For Cicconetti [14], the first step is surgical resection of the tumour to arrest bone destruction, followed in a second stage by parathyroidectomy to reverse parathyroid hormone secretion. In our patient, the diagnosis of the brown tumour was made two months after para-thyroidectomy and after the initiation of treatment for hypercalcaemia. Given the decrease in blood calcium levels, the therapeutic decision was to continue the medical treatment of hypercalcaemia and to monitor. After two CT scans at 6 and 9 months, the tumour regressed with normalisation of blood calcium and parathyroid hormone levels.

## Conclusion

Parathyroid carcinoma is an extremely rare cause of primary hyperparathyroidism, which may manifest as non-neoplastic osteolytic tumours. The invasive nature of the bone lesion on CT scan and the hyperfixation on scintigraphy make a secondary localisation most likely. Anatomopathological examination showing a giant cell tumour which, in association with primary hyperparathyroidism, confirms the diagnosis of a brown tumour. It is essential to distinguish between a brown tumour and a secondary bone lesion, in order to avoid unnecessary surgery, given the spontaneous regression of brown tumours after treatment of primary hyperparathyroidism.

## Author's Contributions

All the authors contributed to study concept, data analysis and writing the paper. All authors read and approved the final version of the manuscript.

## Conflicts of Interest

The authors declare that there are no conflicts of interest regarding the publication of this manuscript.

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