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Case Report

Multiple brown tumors in primary hyperparathyroidism ☆

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ABSTRACT

Brown tumors are benign bone tumors that rarely complicate hyperparathyroidism, manifesting as fibrous and erosive lesions secondary to rapid and localized osteoclast turnover. These lesions are typical of primary hyperparathyroidism, but they are not often observed. We present the case of a 72-year-old woman presenting with asthenia, bone pain, and hemiplegia. Biological analysis showed primary hyperparathyroidism, cervical ultrasound a right parathyroid adenoma that fixed on scintigraphy. When cross-sectional imaging was performed, it revealed multiple bone tumors of the axial and peripheral trunk with spinal cord compression which were diagnosed as brown tumors related to parathyroid adenoma. We illustrate through this case the importance of multidisciplinary imaging techniques before raising the diagnosis, especially in unusual pathologies such as brown tumors.

Introduction

Brown tumors are benign, osteolytic lesions giant cell secondary to the effects of parathyroid hormone on bone tissue, complimenting only 4.5% of primary hyperparathyroidism cases [1]. The most common sites are ribs, clavicles, pelvic bones, and the mandible [2]. In asymptomatic hyperparathyroidism, the discovery of these lesions is often incidental, and their imaging patterns are similar to metastatic osteolytic lesions, considered the first differential diagnosis [3]. Different bone imaging techniques are necessary to ensure the diagnosis of these bone tumors and must be correlated to those allowing the etiological diagnosis of hyperparathyroidism.

Observation

A 72-year-old woman with no specific medical history presented functional impotence of the lower extremities for 6 months evolving to paraparesis, consulted an internist prior to the onset of symptoms and was prescribed phosphate-calcium analysis, which revealed hypercalcemia (144 mg/L), very high PTH (1815 units) and calciuria (317 mg/24 hours). On

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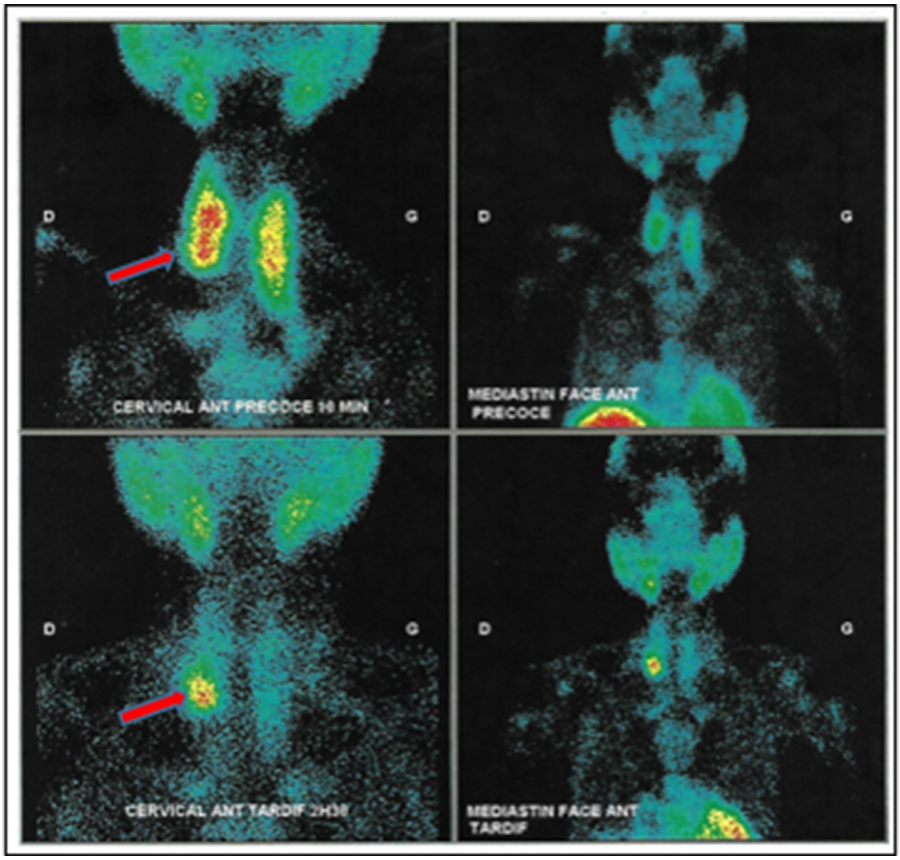

Fig. 1 – MIBI scan showing a projecting focus at the base of the right thyroid lobe compatible with a pathological parathyroid.

admission, the patient presented with an acute hypercalcemic crisis, made of dehydration and vomiting associated to a very high rate of blood calcium 158 mg/L a.

The patient also benefited from a cervical ultrasound that showed a right parathyroid nodule measuring (27 × 21 mm), with a left thyroid nodule graded EUTIRADS 4 (16 × 13 mm), completed by a MIBI scan showing a projecting focus at the base of the right thyroid lobe compatible with a pathological parathyroid (Fig. 1).

Because of long immobility history and edema of the lower extremities, D-dimer dosage was positive, and at chest CT angiography there was a right lower lobar pulmonary embolism, with multiple irregulars contoured osteolytic lesions involving the scapulae, the right clavicle, ribs bilaterally and dorsal vertebrae, of which the most pejorative were located at the level of D3, D4 and D5 associated to endocanalar extension responsible for spinal cord compression (Fig. 2).

An abdomino-pelvic CT scan with multiplanar reconstructions and diffuse bone lesions with similar characteristics, were found in the spine and the pelvis, with bilateral renal microlithiasis (Fig. 3).

Spinal magnetic resonance imaging showed osteolytic lesions in the dorsal spine in hyposignal T1, hypersignal T2, and STIR, enhanced after injection, complicated by spinal cord compression, suggesting brown tumors considering the context of primary hyperparathyroidism (Fig. 4).

Subsequently the patient underwent a bone biopsy for a giant cell tumor, which could be classified as a brown tumor (Fig. 5), and the right parathyroid mass was removed, revealing a parathyroid adenoma (Fig. 6).

Discussion

Brown tumors (BT) are benign osteoclastic lesions with a rare (1.5%-4.5%) and late manifestation of primary hyperparathyroidism that can affect the entire skeleton. They may manifest as bone pain, pathological fractures, or remain asymptomatic. They are mistaken for metastases when they present as diffuse osteolytic lesions [4]. BT results from bone resorption secondary to excessive osteoclastic activity and replacement by fibrotic tissue and giant cells [5].

They affect women more frequently and occur between the ages of 50 and 60 years, due to the hypersecretion of parathyroid hormone (PTH) by a parathyroid adenoma [6].

Most of the cases published in the literature describe an isolated affected site, with a review of the literature illustrating the different types of involvement of brown tumors [7]. In our case, we report diffuse involvement of the skeleton secondary to primary hyperparathyroidism.

The diagnosis of BT is therefore based on a combination of clinical history, laboratory findings, imaging data, and histology

The clinical manifestations of these tumors are variable and nonspecific, such as weakness, weight loss, bone pain,

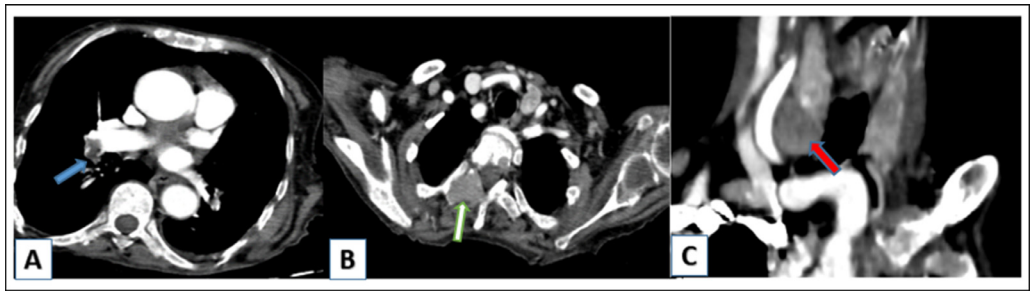

Fig. 2 – Chest CT angiography showing a right lower lobar pulmonary embolism (A), with irregular osteolytic lesion located at the level of D3 with endocanalar extension (B), a lesion opposite the lower pole of the right thyroid lobe, hypodense, related to the parathyroid adenoma (C).

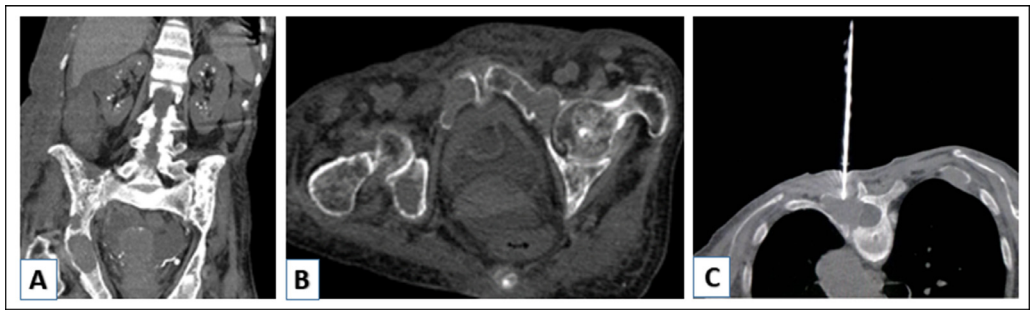

Fig. 3 – Abdominal pelvic CT scan coronal section (A) showing renal lithiasis with diffuse osteolytic lesions of the pelvis. Axial section bone (B) showing lytic ischio pubic lesions with cortical rupture. (C) CT-guided bone biopsy of D5.

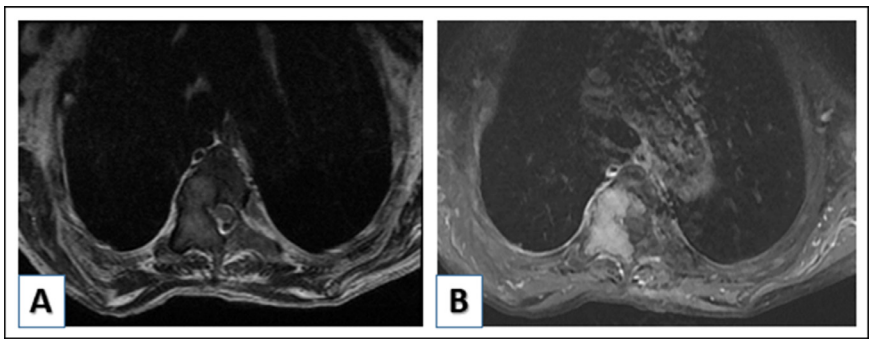

Fig. 4 – Spinal magnetic resonance imaging shows osteolytic lesions in the dorsal spine in intermediate signal in T2, enhanced after injection, complicated by spinal cord compression.

or pathological fractures, revealed sometimes as progressive bone swelling or urinary lithiasis [8].

Biologically, the diagnosis of primary hyperparathyroidism is based on the association of hypercalcemia and hyperparathyroidism (PTH 1-84), and hypophosphatemia and hypercalciuria [9].

Radiologically, in case of clinically suspected or biologically confirmed hyperparathyroidism, cervical ultrasound and 99mTc-sestamibi scintigraphy are the most commonly used methods. Ultrasonography identifies the adenoma as a lesion at the lower pole of the thyroid, circumscribed, hypoechoic, homogeneous, covering the thyroid gland. It may not be visible if it is small, hence the interest of analyzing the lower pole of the thyroid [10]

The thyroid and parathyroid glands show tracer uptake in early images on 99mTc-sestamibi scintigraphy; however, the parathyroid gland retains a later uptake in delayed images about 2 hours, and the parathyroid adenoma is characterized by persistent and avid uptake due to its delayed tracer removal compared to the thyroid gland [11].

The diagnosis of brown tumors is often made incidentally on imaging performed for another indication, as in the case of our patient, in which a CT scan was requested on suspicion of a pulmonary embolism. CT or MRI is performed to evaluate the extent of the lesion and guide a bone biopsy. Preferential sites for brown tumors are the clavicle, jaw, ribs, and pelvis with less frequency at the cranial level [12,13]. In imaging, brown tumors are presented as lytic bone lesions, circum-

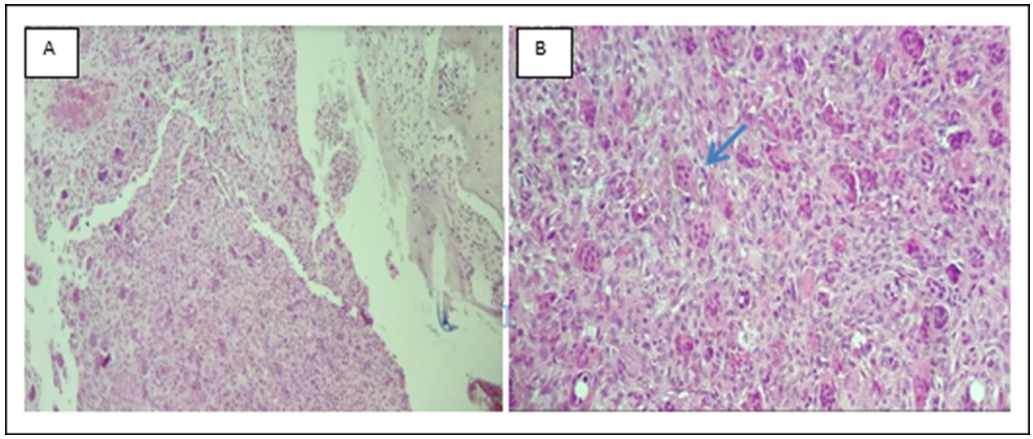

Fig. 5 – (A) Histological image of a bone benign tumor location. (B) Numerous multi-nucleated giant cells are shown (arrow) HE x200.

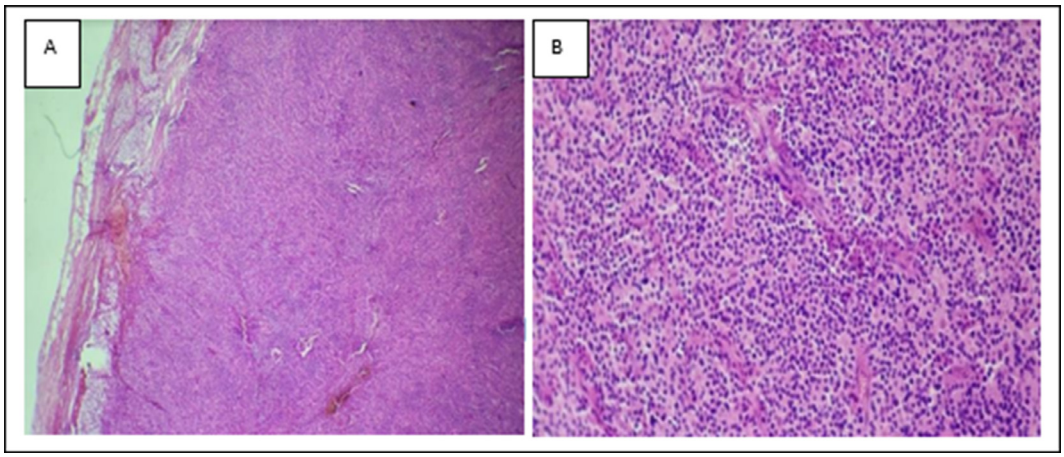

Fig. 6 – (A) Histologically, para-thyroidal parenchyma is compressed by a well circumscribed adenomatous proliferation without capsular effraction or vascular emboli. HE x40. (B) Less regular cells with eosinophilic cytoplasm HE x200.

scribed, single or multiple, simple or poly lobular, sometimes with bone expansion and extension to adjacent structures responsible in some cases for pathological fractures. These lesions can present a mixed appearance of sclerotic and lytic lesions, with ill-defined contours [14], capable of being confused with a malignant origin, which requires histological confirmation; however, the distinction between a brown tumor and giant cell tumor in histology is difficult, so a confrontation to hypercalcemia and hyperparathyroidism is important [15].

The main differential diagnoses are bone metastases, multiple myeloma, sarcomas, giant cell tumors, tonsilloid cysts, chondromas, and aneurysmal bone cysts. Treatment of this condition is etiologic in most cases (80%-85%). After surgical resection of the parathyroid adenoma there is regression of the bone lesions [5].

Conclusion

Although brown tumors are rare, this case emphasizes the importance of a complete investigation of the context in cases of osteolytic bone involvement based on: medical history, physical examination, laboratory tests, imaging data and histological confirmation when in doubt.

Patient consent

I confirm that patient has given her consent.

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