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Brown tumors revealing primary hyperparathyroidism: It still exists

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Abstract

Hyperparathyroidism is a frequent endocrinopathy, often diagnosed at the stage of subclinical hypercalcemia by systematic calcium measurement and the advent of new parathyroid hormone measurement techniques. Brown tumors represent a late-onset, locally aggressive but potentially benign non-metastatic bone complication secondary to an abnormality of bone metabolism within the context of hyperparathyroidism. It is usually a late complication, rarely revealing. We report 2 clinical cases of primary hyperparathyroidism revealed by a brown tumor, complicated in one case by a pathological fracture. The aim of our work is to underline the importance of looking for hyperparathyroidism in every lytic lesion, given the insidious nature of this endocrinopathy, thus preventing pathological fractures and improving functional quality of life.

Keywords: Brown tumors; Primary hyperparathyroidism; Parathyroid adenoma; Pathological fracture

1. Introduction

Hyperparathyroidism is the third most frequent endocrinopathy after diabetes mellitus and thyroid disease, and often diagnosed at the stage of asymptomatic hypercalcemia [1]. The occurrence of bone or kidney complications has become increasingly rare, essentially dominated by osteoporosis [2].

Brown tumors are non-neoplastic, focal osteolytic bone lesions giving a blown appearance to the cortices, exposing them to pathological fractures, secondary to hyperparathormonemia. They are most often associated with primary hyperparathyroidism [3].

We report 2 cases of primary hyperparathyroidism revealed by a brown tumor, complicated in one case by a pathological fracture.

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2. Observation 1

A 52-year-old female patient with 20 years of recurrent bilateral renal lithiasis was admitted for etiological assessment of bony lesions of the pelvic girdle discovered incidentally on a uroscanner (Figure 1, 2 and 3), without polyarthralgias, polyuropolydipsic syndrome, constipation, nausea, vomiting or fever. The diagnosis of primary hyperparathyroidism was based on corrected hypercalcemia of 119 mg/l, hyperparathyroidism of 1200 ng/l, normal phosphatemia of 25.34 ng/ml, high 24-hour calciuria of 268 mg/24h and the presence of a parathyroid adenoma on ultrasound. Sesta- mibi parathyroid scintigraphy was not performed due to the patient's lack of financial means. Standard X-rays were ordered in search of other locations, and revealed a lesion on the radial diaphysis with diffuse bone demineralization. The patient underwent surgery, with an intraoperative drop in IHPT to 62 ng/l and normalization of blood calcium levels on post-op day 1. Anatomopathological studies showed that the lesion was a parathyroid adenoma.

3. Observation 2

This is a 66-year-old patient with no previous pathological history, admitted for investigation of primary hyperparathyroidism revealed by a fracture of the femoral shaft on a pathological bone (Figure 4), pathological examination of which was in favour of a brown tumour. Other localizations were discovered incidentally in the contralateral femur (Figure 5), mandible (Figure 6) and right 5th finger (Figure 7), as well as a salt-and-pepper appearance of the skull typical of hyperparathyroidism (Figure 8). Biological findings included corrected hypercalcemia at 140mg/l, hyperparathyroidism at 1936 ng/ml, hypophosphatemia at 23.17mg/l and high 24-hour calciuria at 246mg/24h. Ultrasound and cervicothoracic CT showed a 2.7cm parathyroid adenoma, confirmed by histological study. Parathyroid scintigraphy with sestamibi was not performed due to difficulties in mobilizing the patient, given the hip fracture. The course of the disease was marked by the onset of bone starvation syndrome, which was treated with calcium supplementation and progressed well.

4. Discussion

Hyperparathyroidism (HPT) is a frequent endocrinopathy, discovered in 75-80% of cases by routine calcium measurement [4]. Sometimes revealed by renal or cardiovascular manifestations [5].

The bone complications of PTH, such as bone cysts, osteoporosis, subperiosteal resorption and brown tumours, have become rare and delayed. They occur in 5-15% of cases [4, 6]. These complications are secondary to increased bone remodelling and reduced bone mineral density (BMD), particularly in cortical bone (radius and femur), and are generally observed in patients over 60 years of age and in post-menopausal women [7].

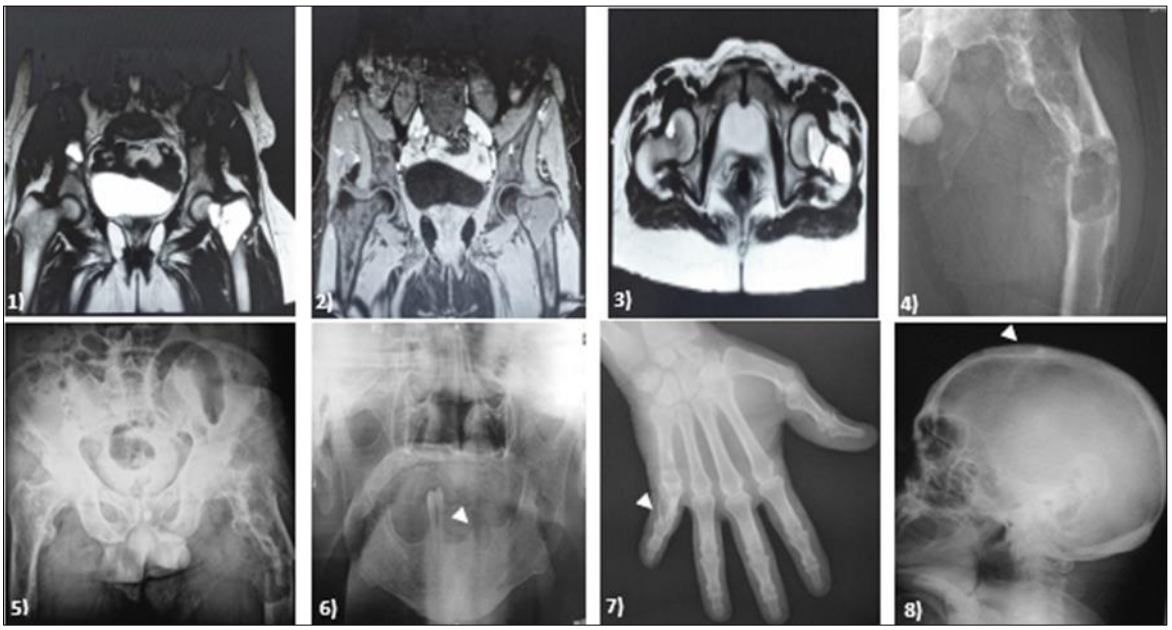
However, it is exceptional for a brown tumour to be the presenting feature of hyperparathyroidism. Its incidence, reported over the last twenty years, was 1.5 to 1.7% in secondary hyperparathyroidism and 3% in primary hyperparathyroidism [8]. These are fibrocystic lesions that appear as expansive osteolytic lesions giving a blown appearance to the cortical bone, affecting the ribs, pelvis and femur, with maxillo-mandibular involvement remaining very rare [9]. The brown nomenclature is derived from the characteristic brownish coloration of its macroscopic appearance. This appearance is due to the presence of numerous blood vessels, localized haemorrhages and secondary deposits of haemosiderin [4].

These are a locally aggressive tumors but potentially benign non-metastatic whose clinical presentation depends on the size and location of the process. They may be totally asymptomatic, and the diagnosis is made fortuitously following a systematic radiological examination, as in our first observation, or manifest as bone pain or pathological fractures [4, 10], as illustrated by our 2nd observation. In other patients, they may manifest as jugal, palatal and/or gingival bone swelling, with facial deformity and asymmetry, pain and mobility, or even tooth loss [11].

Radiologically, several non-specific aspects are possible, but the most evocative aspect is that of a mono-geodic bone lysis with blurred boundaries giving a blowing of the cortices simulating a malignant lesion, particularly for destructive lesions, making one think of multiple myeloma, osteosarcoma, bone metastasis of an osteophilic cancer, osteomyelitis or Paget's disease[11]. Differential diagnosis can also be made with true giant cell tumors (formerly myeloplastic tumors), central reparative giant cell granuloma (GRCCG), and aneurysmal cyst [12]. Distinguishing between brown tumor and other giant cell lesions is difficult, as they are confirmed only by histology. On the other hand, hyperparathormonemia and 99mTc-Sestamibi uptake at the level of the lesions help to orientate the diagnosis towards a brown tumor before any histological confirmation [13]. Our two patients did not benefit from this examination, the first due to lack of resources and the 2nd due to difficulty in mobilization, given the hip fracture.

The CT scan shows a tissue-dense, contrast-absorbing mass with no soft-tissue invasion or periosteal reaction [14]. Bone scintigraphy reveals a focus of hyperfixation that may simulate metastases on whole-body films [2].

Standard skeletal X-rays, which may also show renal lithiasis, nephrocalcinosis, a "salt-and-pepper" appearance of the skull or subperiosteal resorption, typically localized in the phalanges secondary to hyperparathyroidism, are necessary to search for other localizations [4,15].



Figures 1) and 2) Coronal section of a pelvic CT scan: lytic fibrocystic lesions of the left femoral neck; Figure 3): transverse section of a pelvic CT scan: lytic fibrocystic lesions of both femoral necks; Figure 4): radiograph of the left hip: fracture of the upper 1/3 of the femoral shaft over a fibrocystic lesion; Figure 5) : radiograph of the front pelvis: fibrocystic lesions of both femurs with fracture of the left femoral shaft; Figure 6): dental panramique: lytic mandibular lesion; Figure g): radiograph of the hand: lytic fibrocystic lesions of the 5th finger; Figure h): "salt-and-pepper" appearance

Imaging is of great help in making the diagnosis of bone lesions associated with hyperparathyroidism. Ultrasound, cervical CT and parathyroid scintigraphy with 99mTc MIBI are required to detect lesions of the parathyroid glands at the origin of hyperparathyroidism, in particular parathyroid adenomas. Difficulties are also encountered with ectopic adenomas and multiple parathyroid gland hyperplasia, hence the use of a MIBI/iodine-123 subtraction protocol which increases the sensitivity of the examination for the detection of ectopic adenomas, particularly when a complementary early SPECT/CT acquisition is performed[16]. In addition, this whole-body MIBI 99mTc acquisition could reveal both an intra-thoracic adenoma and any associated brown bone tumors [17], with better characterization of these lesions by highlighting intense focal hyperfixation and an osteolytic appearance, surrounded by osteosclerosis [2, 11].

Opinions on the treatment of brown tumors are divided [18]. Most studies confirm that the treatment of brown tumours is based on para-thyroidectomy, which is the treatment of choice, particularly for parathyroid adenomas, with rapid fall in blood calcium levels and regression of the lesion. This regression is achieved after replacement of the bone lesions with normal bone tissue, which is generally found six months after treatment of hyperparathyroidism. [19]. However, some authors have reported that the evolution of brown tumors is variable depending on their composition, and spontaneous bone regeneration may require several years before restoring its normal morphology [20]. In cases where lesions are extensive or largely destructive, or persist despite the achievement of normo-calcemia, or where they continue to grow, curettage of the tumour and its enucleation is recommended in order to halt bone destruction, sometimes in the first instance even before para-thyroidectomy aimed at normo-calcemia [17, 21].

5. Conclusion

Brown tumors remain a rare, but not exceptional, bone complication, which requires a search for primary hyperparathyroidism, because of the insidious nature of this endocrinopathy, while at the same time excluding an underlying malignant process.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

No conflict of interest.

Statement of informed consent

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

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