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Case Report

Cervical Brown Tumor Revealing Primary Hyperparathyroidism

Nabil Raouzi[1*], Mohamed Dahamou[1], Fahd El Ayoubi[2], Noureddine Oulali[1], Fayçal Moufid[1]

[1]Neurosurgery department, CHU Mohammed VI, Mohammed Premier University, Oujda, Morocco
[2]ORL department CHU Mohammed VI, Mohammed Premier University, Oujda, Morocco

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***Corresponding author:** Nabil Raouzi

Abstract

Brown tumors are benign osteolytic bone lesions, they are rare, caused by the disorder of the phosphocalcic metabolism that typically complicate primary hyperparathyroidism (PPHT) in 4.5% and secondary hyperparathyroidism (HPTS) in 1.5 to 1.7%. Brown tumors are located mainly in the ribs, the pelvis, femurs and mandibles. Spinal locations are very rare, they especially affect the thoracic region. Very few cases of cervical brown tumors have been reported in the literature. To date, there have been only 12 cases, the last in 2018 by M. Sánchez-Calderón et al. Reporting the case of a 65-year-old woman treated in our department for cervical C4-C5-C6 brown tumor responsible for spinal compression, revealing primary hyperparathyroidism. We note the interest of the suspicion of a brown tumor in front of any osteolytic vertebral lesion, especially in patients suffred from end stage renal disease, and hypercalcemia. Early diagnosis and setting up a multidisciplinary management prevent complications and functional aftereffect, despite the benign histological characterof her injuries.

Keywords: Brown tumor, Hyperparathyroidism, End-stage renal failure, Spinal cord compression.

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INTRODUCTION

Brown tumors or osteoclastomas, are benign osteolytic bone lesions, first described in 1891[1]. They are caused by a disorder of bone metabolism observed in primary and secondary hyperparathyroidism [2-3]. Their frequency has significantly decreased thanks to early diagnosis of HPTP and improved management of the chronic renal insufficiency. Vertebral locations are rare compared to other locations, including the mandible, the clavicles, pelvis and femur, which are common [1-4]. The first case of Spinal brown tumor was reported in 1978 by Ericsson et al [22]. Reporting to us the case of a brown cervical spine tumor, in a patient followed for inflammatory rheumatism, who has a cervical spinal cord compression revealing HPTP by a parathyroid adenoma.

CASE REPORT

A 65-year-old woman with a history of high blood pressure and chronic seronegatif, inflammatory rheumatism, under metotrexate 15mg/week. Since a year, she had an asymmetrical tetraparesis predominant on the left. The radiological examination shows an osteolytic cervical lesion, of the fifth cervical vertebra, whose bodyseems completely lysed with infiltration of the posterior elements, reducing the cervical canal and compressing the spinal cord, this lesion takes contrast to the injection, and partially infiltrates the vertebral bodies C4 and C6 (fig1). The biological assessment reveals an hypercalcemia at 120 mg/l, with a slight hyperparathyroidism: 73 pg/ml. The ultrasound and cervical CT scan shows a right lower parathyroid adenoma of 16/7 mm (fig2). The patient did not present other complaint, radiological examination did not objectified other locations. On his data we strongly suspected a brown cervical tumor, complicating an adenomatous HPTP.The patient benefited of a double surgical approach (fig4), at first a decompression was carried out by the posterior way carrying the infiltration C4-C5-C6 slides with C2 Pars screws, and C3-C7 lateral masses screws (fig3 and fig4: B). The macroscopic appearance (Fig. 4: C) shows an osteolytic lesion with irregular polycyclic to cortical contourthin erased or blown. In a second delayed time, a double team cervicotomy was performed for ablation of the right parathyroid nodule followed by a C4-C5 and partial C6 corporectomy within place of an Iliac crest bone graft fixed anterior cervical plate (Fig. 4: A-D). The patient presented an Improvement of the deficit, with a total recovery in one month. Postoperatively, with normalization of calcemia and parathyroid numbers. Histological examination finds cells that match in layers, diffuse, mixed with many giant cells, multinucleated osteoclastic type in a fibrous stroma

andhemosiderin deposits, this histological aspect is compatible with a brown Tumor of hyperparathyroidism.

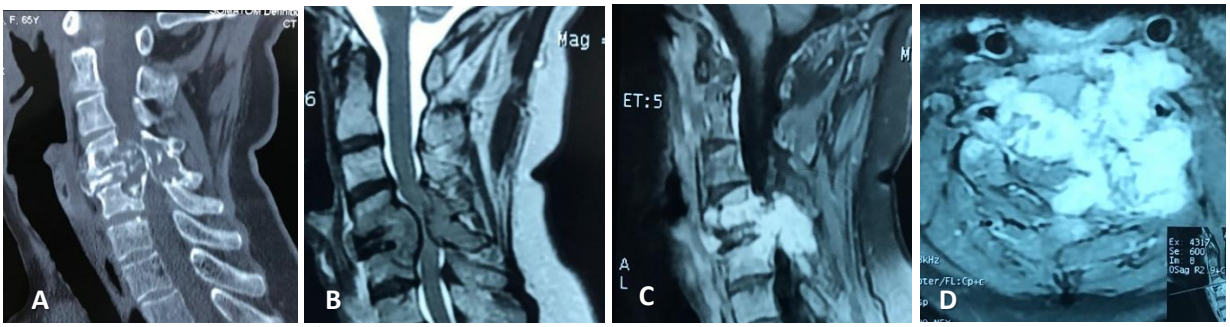

Fig -1: A) Sagittal reconstruction of cervical spine CT scan: osteolytic mass of C4, with cortical erosion and spinal canal compromise. B) Sagittal T2-weighted MRI. C) sagittal T1-weighted MRI with gadolinium, and D) axial T1-weighted MRI with gadolinium: osteolytic C4 - C5 and C6 vertebral mass causing severe canal stenosis and spinal cord compression.

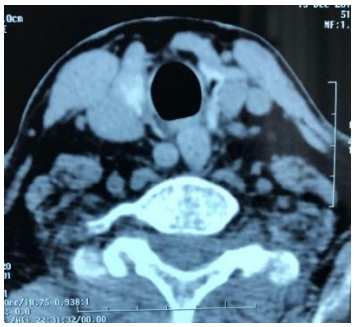

Fig-2: cervical CT scanner: right parathyroid adenoma

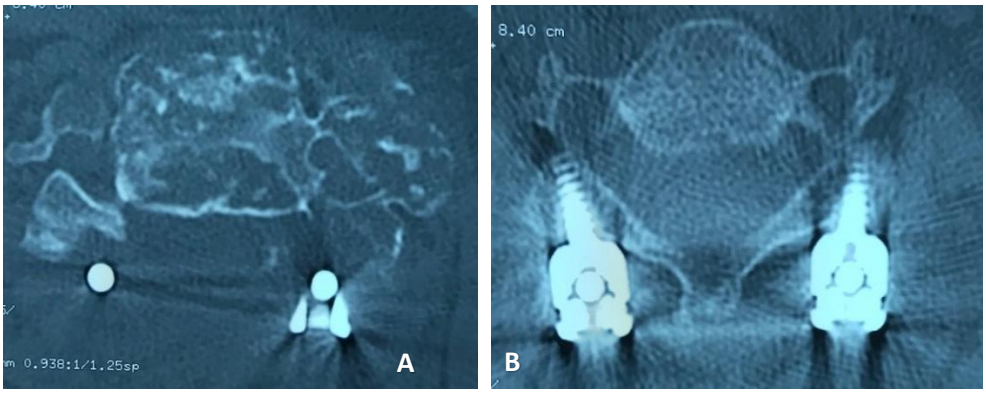

Fig-3: A) Postoperative CT scan: spinal cord decompression, B) screws fixation of cervical lateral masses

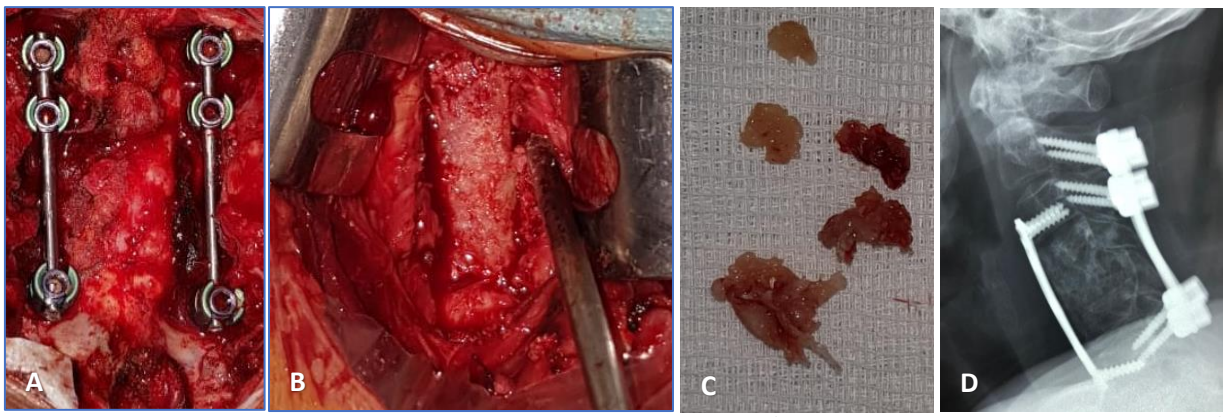

Fig-4: Intraoperative image: A) posterior approach: spinal cord decompression, screws of the lateral masses, B) corpectomy and bone graft, C) Macroscopic appearance of the brown tumor, D) postoperative X-ray

DISCUSSION

Brown tumors (BT), also known as osteoclastoma, were first described in 1891 [1]. Are Benign osteolytic bone lesions, reflecting abnormal bone metabolism, leading to hyperactivity of the osteoclasts in the hyperparathyroidism. Primary hyperparathyroidism is defined by hypercalcemia with an increase in the parathyroid hormone level, it results in more than 80% of the cases of a parathyroid adenoma, hyperplasia (20%) and exceptionally parathyroid carcinoma (2%) [5-6]. Secondary hyperparathyroidism is in patients on hemodialysis due to the end-stage renal failure. In these patients, 1,25-dihydroxy cholecalciferol causes hypocalcemia and phosphate retention, resulting in hypersecretion of the PTH by the parathyroid glands [8-10]. While tertiary hyperparathyroidism is defined as a secondary hyperparathyroidism which lasted a long time and led to the formation of autonomic parathyroid glands [1-9]. The brown tumor was reported in 4.5% of patients with primary hyperparathyroidism (PPHT), and 1.5 to 1.7%of those with secondary hyperparathyroidism (HPTS) [7], this incidence has decreased significantly with systematic serum calcium [17-18]. However, recently Fargen *et al* [10] can be seen in recent decades, an increase in the number of cases of brown tumors observed in hemodialysis patients with End-stage renal failure, which may simply reflect an increase in the reported cases and / or survival in this population [14]. Imen Gorsane *et al* [3i] reported a series of 20 chronic hemodialysis patients presenting a brown tumor during the course of their disease. Hypersecretion of PTH leads to an increased osteoclastic activity leading to bone resorption with subsequent reactive fibrosis [10-19]. These metabolic disturbances, ead to osteolytic lesion formation, which fuse during the progression of focal resorption of the bone marrow and fibrosis to form a brown tumor [20-21]. Histologically, these tumors exhibit hemosiderin (which gives the brown appearance of the tumor) and multinucleated giant cells, resembling osteoclasts in a fibrovascular stroma consisting of fleshy fibroblasts and a rich vascular network [19]. Macroscopically, there are solid, brownish-red lesions associated with cyst formation [21] (Fig. 4). Lesions can be single or multiple. The most common localization of TB is the mandible, ribs, clavicles, femurs and pelvis. Spinal localization is extremely rare [10-11]. The first case of brown tumorcervical was reported in 1993 by I.W. Barlow, et al [11], a total of 12 observations of cervical brown tumors have been reported in the literature until 2018 [12]. Bahareh *et al* [13] reported 37 cases, with female predominance (61.1%). The average age of patients with brown tumors in secondary hyperparathyroidism was 43.67 ± 14.9 years. The dorsal spine is affected in (58.3%) [10] The clinical presentation is nonspecific, lesions may be asymptomatic it means an accidental finding [15], or during a fracture with compression of the spinal cord [23-29]. 77.8% of the brown spinal tumors are manifested by slow spinal cord compression at different evolutionary types [14]. The imaging is not specific, these are lytic lesions with a non-precise limit leading to ablistering of cortices [23-24].

Computed tomography confirms osteolytic appearance without cortical disturbance of the periosteal reaction [25]. In MRI, hemosiderin results in visible signal loss in gradient echo images [13]. The bone scintigraphy has a better sensitivity than 99mTc-Sestamibi for brown tumors [26] is useful for the diagnosis of pathological parathyroid and the search for ectopic parathyroid glands. This has agreat interest in guiding the surgical procedure which remains the only curative treatment. Positron emission tomography (PET) is considered, is a non-invasive tool that allows in some cases the diagnosis and monitoring of brown tumors [27]. Our patient was not able to benefit from her last two investigations seen that we do not have in our center. Brown tumors should be considered in the differential diagnosis of neoplastic lesions of the spine particularly in patients with end-stage renal disease. [10-21] In HPTP, the treatment of choice is parathyroidectomy with parathyroid adenoma, which allows normalization of serum levels of parathyroid hormone, is calcemia leading to progressive and complete regression tumor mass [28-29]. In secondary hyperparathyroidism. Medical treatment is based on an adjustment of the calcium intake, and active vitamin D derivatives based on phosphocalcic balance data and PTH values with a low phosphate diet [30-31]. The spinal localization remains particular because of the neurological considerations. Surgery is the treatment of choice, it allows spinal decompression and stabilization of the spinal cord. The intervention is urgently needed in case of neurological disorders. Our patient benefited from double approach with parathyroidectomy allowing a good neurological and metabolic evolution [09-32] (fig3). Brown tumors have a good prognosis if the diagnosis is made at the beginning stage. The systematic dosage, and taking into adequate loading of end-stage renal failure, is the optimal way to prevent the development of these tumors which remains serious, especially in case of spinal cord localization that may be responsible for neurological disorders.

CONCLUSION

Brown tumors are benign osteolytic bone lesions, secondary to the metabolic disorders seen inprimary and secondary HPT, their radiological appearance is nonspecific, they must be evoked before any spinal lesions, and seek disorders of phosphocalcic metabolism. Vertebral localizations are often responsible for serious neurological disorders, early diagnosis allows medical and surgical multidisciplinary management to avoid complications. Brown tumors are benign lesions of good prognosis, the prevention of the occurrence of its lesions is the main objective of the care of patients at risk.

Conflits of interest

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

Disclosures

None

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