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Massive brown tumors of the jaw: A case report

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Abstract

Brown tumors are non-neoplastic bone lesions caused by an abnormal remodeling of the bone that may occur with primary or secondary hyperparathyroidism. Their radiological aspect: lytic and aggressive can easily be misdiagnosed for a malignant origin hence the importance of knowing that diagnosis is to be considered through both clinical context and radiological semiology, which will be detailed via this case of a 32-year-old female patient with an end-stage kidney disease, admitted for facial disfiguration and palpable masses corresponding to brown tumors affecting the maxilla and the mandibular bone.

Keywords

Brown, tumor, hyperparathyroidism, kidney, disease, imaging, CT

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Introduction

Brown tumors are rare in developed countries and are present in only 3% of primary hyperparathyroidism (HPTH) above 10 times the normal limit.[1] This seldom incidence is due to the improved early on detection and the elaborate protocols and screening detection of HPTH. Usually, this tumor is slowly growing and can be localized in all bone locations, so the clinical manifestations depend on the latter.[2] In our case report, the lack of health infrastructure in remote countryside areas, in addition to the clinical neglect of the patient to go to a hospital, the brown tumor of the jaw was only diagnosed when admitted in the ward and was massive.

Case description

A 32-year-old woman, with medical history of end-stage kidney disease due to a hereditary polycystic kidney disease going through hemodialysis, was admitted for a massive facial disfiguration with maxillary and mandibular area swelling that appeared progressively for about 7 months. She didn't report any pain at first but suffered from a discomfort in chewing and eating. Since she lives in an area unequipped with health diagnostic centers, she only decided to come to the Mohammed V Military hospital of Rabat when her symptoms became unbearable. The initial clinical examination revealed a massive facial esthetic deformity with maxillo-mandibular palpable masses, hard and deeply implanted in the bone, with a mobility of the lower incisors. Biological tests showed a disturbed phosphocalcic balance with hypocalcemia at 80 mg/L (N = 84–102 mg/L), hyperphosphatemia at 73 mg/L (N = 23–47 mg/L), and increased parathormone (PTH) levels at 746 pg/mL (N = 10–50 pg/mL). These signs of secondary HPTH secondary to her kidney disease.

A facial computed tomography (CT) scan performed showed a massive hypertrophy with diffuse lytic and sclerotic lesion of the maxillary bone, mandibular bone, and cranial vault, without periosteal reaction or invasion of adjacent soft tissues. The lamina dura lysis of the alveolar bone gave the teeth a floating aspect (Figures 1 and 2).

Discussion

First described by Recklinghausen in 1891,[2] osteitis fibrosa cystica commonly known as brown tumors are solitary or multiple non-neoplastic bone lesions that often manifest due to HPTH either in its primary form commonly caused by a

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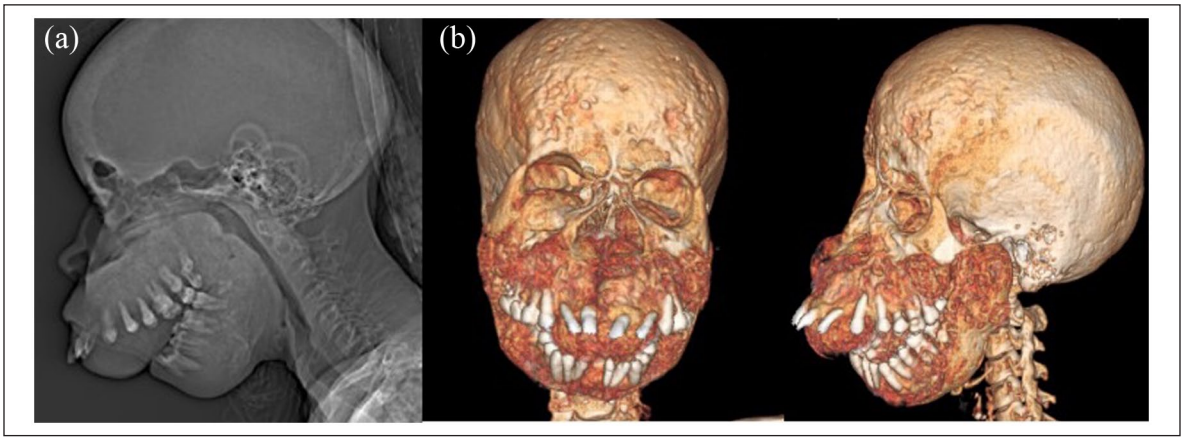

Figure 1. Side profile plain x-ray of the face (a) showing a massive hypertrophy of maxilla and mandibular bones showing a sclerotic aspect. Volume rendering CT scan of the face and skull (b) showing facial disfiguration with a massive hypertrophy and heterogenous aspect of both maxilla and mandibular bones corresponding to the brown tumors of the jaw.

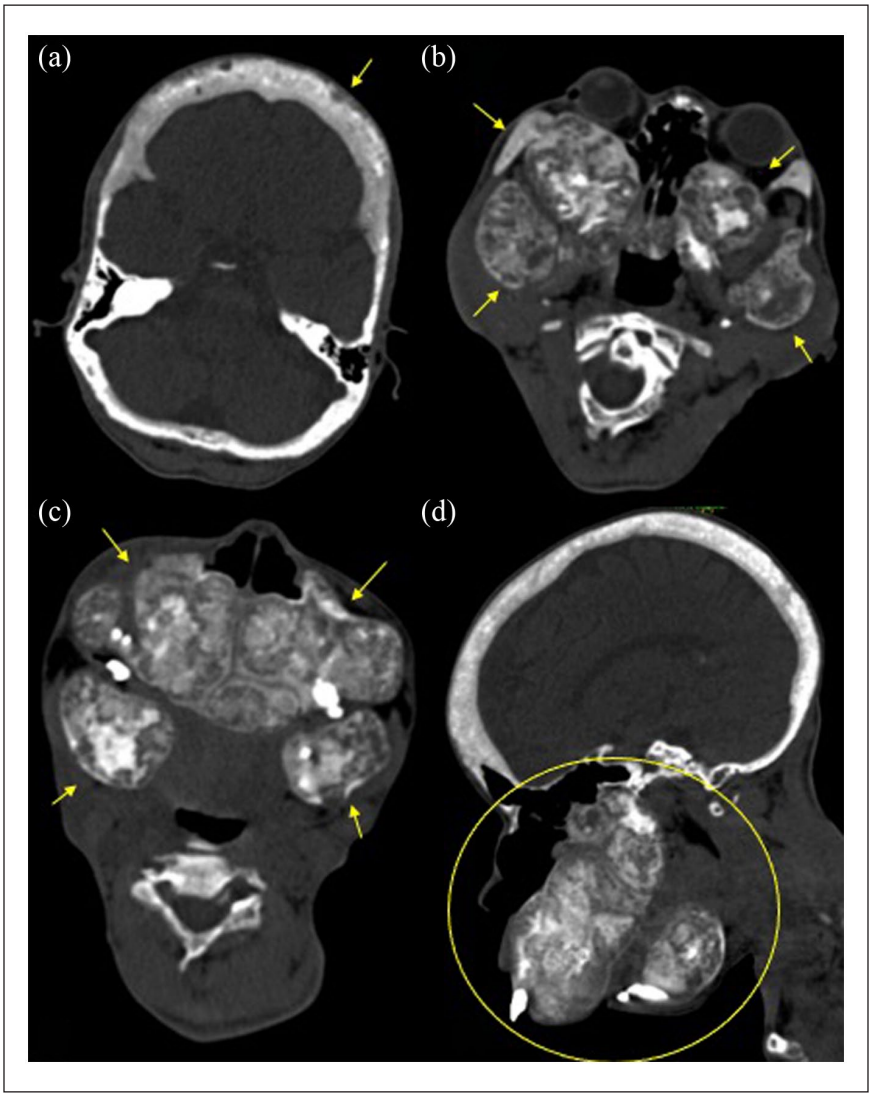

Figure 2. CT scan of the face and skull in bone window with both axial (a, b, c) and sagittal (d) images showing a massive hypertrophy with diffuse lytic and sclerotic lesions of the maxillary bone, mandibular bone, and cranial vault, without periosteal reaction or invasion of adjacent soft tissues. The lamina dura lysis of the alveolar bone gives the teeth a floating aspect.

parathyroid adenoma or its secondary form, usually the result of chronic kidney disease.[1] The term "brown" refers to the accumulation of hemosiderin pigments rendering the lesion brown macroscopically.[2]

It has a low prevalence of 0.1% with a feminine predominance. Its incidence in patients with chronic renal disease or end-stage renal disease is of 13% as opposed to 1.5% in case of primary HPTH. It can affect all bones such as the femur, sternum, ribs, and rarely facial regions (jawbone such as our case, palatinal bone, nasal, and orbital sinuses), and may be monostotic or polyostotic.[2,3]

Clinical symptoms depend on the topography of the lesions: Pain is usually caused by pathological microfractures caused by the lesion.[1] Patients with primary HPTH may also have gastrointestinal and neurological complaints, as well as kidney stones.[4]

Brown tumors form as a result from disturbances in the metabolism of phosphate, calcium, and vitamin D.[2] Chronic or end-stage kidney disease may be responsible for phosphate retention, with a decrease of 1,25-dihydroxyvitamin D and ionized calcium therefore stimulates PTH secretion, which explains the presence in blood tests of hyperphosphatemia levels with hypocalcemia. The excessive increase of the parathyroid hormone is responsible of a hyper osteoclastic activity and bone resorption explaining formation of brown tumors.[3] Those tumors, depending on their location, can cause facial deformity, pain, and difficulty to speak, eat, or even breathe.[5]

Blood markers evaluation is therefore essential to lead diagnosis. Brown tumors can also result as a paraneoplastic syndrome due to high levels of parathyroid hormone–related peptide mimicking the effect of PTH. They may also be taken then, for bone metastases.[4]

The first-line imaging examination is an ultrasound, especially if a parathyroid adenoma is suspected; it is shown as a hypoechoic homogeneous mass overlying the thyroid gland. It is mostly found inferior and posterior of the latero-inferior pole of the thyroid. The 99m Tc-sestamibi scintigraphy allows the confirmation of the parathyroid adenoma diagnosis and its location. A parathyroid adenoma keeps a tracer uptake, whereas thyroid gland and parathyroid glands have a delayed wash out.[4]

A plain x-ray of the affected bone can show a well-defined osteolytic lesion with thin peripheral bone shell and several internal bony bridges. Sclerotic lesions are more common, and lytic lesions can be seen in multiples bones and can be misdiagnosed for metastasis.[2]

The gold standard in such cases is the CT scan; it allows a 3D and multiplanar image study of the lesion, which can be single or multiple, sometimes multilocular, well-defined and osteolytic with bone hypertrophy, cortical thinning, and sometimes a ground glass pattern. It may be associated with bone destruction and pathological features.[2,3]

Despite the fairly obvious features, imaging can be variable and some tumors may be ill defined, mixed with both a lytic and sclerotic component, associated with adjacent soft tissue involvement, which makes it hard to differentiate with a malignant lesion.[4]

The main mimickers are fibrous dysplasia, metastatic carcinoma or aneurysmal bone cyst,[3] giant cell tumor, granuloma, ossifying fibroma, lytic metastasis from cancer and multiple myeloma when the clinical context is concordant and lesions are multiple.[2]

When lesions are not characteristic, diagnostic confirmation requires histological arguments showing an increased osteoclastic activity, fibroblast proliferation, hemosiderin deposition, and replacement of the connective tissue by cortical, trabecular bone, and giant cell tumors.[3] Those cell tumors contain numerous giant cells that have more than 12 nuclei each, which can be seen in giant cell tumors, giant cell granulomas, and brown tumors. Which makes them have the same histological features, but the association with HPTH allows diagnosis confirmation.[3,6] Immunohistochemical analysis may show mutations in H3F3A which occurs in more than 95% of the cases.[6]

Depending on the tumor and its functional associated problems, treatment first requires balance and normalization of calcium, and phosphate levels with phosphate binders, vitamin D analogs to control secondary HPTH and attenuate tumor mass growth, calcimimetics, and limiting dietary phosphorus intake.

If medical treatment fails, a parathyroidectomy is preferred,[3] and leads to regression of the tumor in most cases. For masses causing esthetic and functional problems in patients not responding to conservative therapy and parathyroidectomy, complete surgical removal of the tumoral masses may be necessary.[2]

Limitations of this case: No follow-up was available as our patient refused to be treated.

Conclusion

Brown tumors are non-neoplastic bone lesions that may occur in patients with primary, secondary, or tertiary HPTH. When neglected, the tumor may evolve into an aggressive pattern. Surgical intervention may be required in cases where lesions cannot be managed medically or when clinical manifestations are heavy. Increased awareness among physicians, multidisciplinary follow-ups, and a robust healthcare infrastructure in remote areas are all factors that contribute to diagnosis and prevent radical therapeutical approaches in patients with HPTH.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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