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Brown Tumor of the Facial Bones in a Patient with Hyperparathyroidism Due to Chronic Renal Failure: A Case Report

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Abstract **Case Report**

Brown tumors are benign, non-neoplastic lesions found in 1.5% to 1.7% of patients with end-stage chronic kidney disease. They are classic manifestations of hyperparathyroidism (HPT): primary due to parathyroid disorders, secondary from non-parathyroid causes leading to chronic hypersecretion, and tertiary due to autonomous secretion in longstanding secondary HPT. These lesions can be monostatic or polystatic, primarily affecting the ribs, pelvis, femur, and mandible, with a higher prevalence in young women. Maxillary involvement is rare, occurring in about 4.5% of cases. We report the case of a 22-year-old female patient with a history of chronic kidney disease since the age of 7, who presented a bilateral painless mandibular swelling. Laboratory evaluation revealed that the patient had secondary hyperparathyroidism. Due to the mandibular swelling, a facial CT scan was performed that revealed multiple lytic lesions that were expansive, causing cortical bone blowout and rupture, with visible intralesional trabeculations in the frontal bone, the outer wall of the left orbit, right temporal region, condyles, mandibular angles, and the maxillomandibular dental arches. There was diffuse bone involvement predominantly showing ground-glass opacity, with cortical-medullary dedifferentiation and bone hypertrophy affecting the facial mass, skull vault, and base of the skull. After clinical and radiological evaluation, the patient underwent an excisional biopsy of the hemimandibular right lesion, and it was diagnosed histologically as a brown tumor. In the first year of postoperative follow-up, the patient reported rib pain, prompting a cervical-thoracic CT scan. The scan showed multiple lytic bone lesions with cortical rupture in the clavicles, scapulae, right humeral head, and ribs, along with diffuse bone changes indicating dedifferentiation and hypertrophy in the vertebral and rib areas. Brown tumors pose a diagnostic challenge due to their association with hyperparathyroidism and their resemblance to aggressive bone tumors. Further research is needed to enhance diagnostic and therapeutic strategies for brown tumors associated with hyperparathyroidism.

Keywords: Brown Tumor, Osteitis Fibrosa Cystica, Recklinghausen Disease of Bone, Hyperparathyroidism.

INTRODUCTION

Brown tumors, also known as von Recklinghausen's fibrous osteitis, an uncommon focal giant-cell lesion. They are rare, affecting between 1.5% and 1.7% of patients with end-stage chronic kidney disease. Brown tumors are part of the classic bone manifestations of hyperparathyroidism (HPT) but are rarely revealing, and arise as a direct result of the effect of parathyroid hormone on bone tissue in some patients with hyperparathyroidism [1]. Brown tumors occur most commonly in ribs, clavicle, long bones and pelvis and are uncommon in other facial bones except the mandible. Radiographic and histologic changes associated with the presence of this lesion may be very similar to those of other bone lesions of the face. Therefore, the diagnosis requires a systemic investigation for lesion differentiation [2].

CASE REPORT

We report the case of a 22-year-old female patient with a history of chronic kidney disease since the age of 7, who presented a bilateral painless mandibular swelling that had been affecting her chewing for one year. Clinical examination revealed facial deformity, with two firm, hard, and fixed masses palpated along both mandibles (figure 1). The phosphocalcic assessment showed abnormalities, with normal calcium levels (76 mg/l), hyperphosphatemia (66 mg/l), and elevated parathyroid hormone levels (1932 pg/ml). Due to the mandibular swelling, a facial CT scan was performed

without contrast injection. The scan revealed multiple lytic lesions that were expansive, causing cortical bone blowout and rupture, with visible intralesional trabeculations in the frontal bone, the outer wall of the left orbit, right temporal region, condyles, mandibular angles, and the maxillomandibular dental arches. There was diffuse bone involvement predominantly showing ground-glass opacity, with cortical-medullary dedifferentiation and bone hypertrophy affecting the facial mass, skull vault, and base of the skull (figure 3). The patient underwent a cervical ultrasound, which revealed a large right solitary parathyroid adenoma. She underwent an excisional biopsy of the hemimandibular right lesion, and it was diagnosed histologically as a giant-cell tumor.

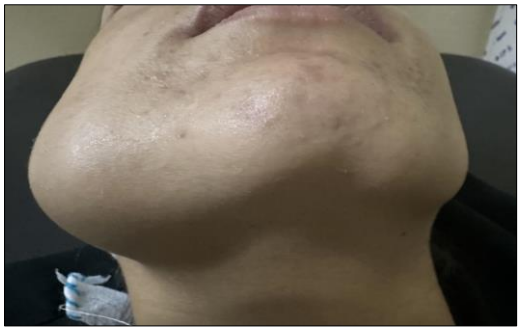

Figure 1: Bilateral mandibular swelling, more pronounced on the right side.

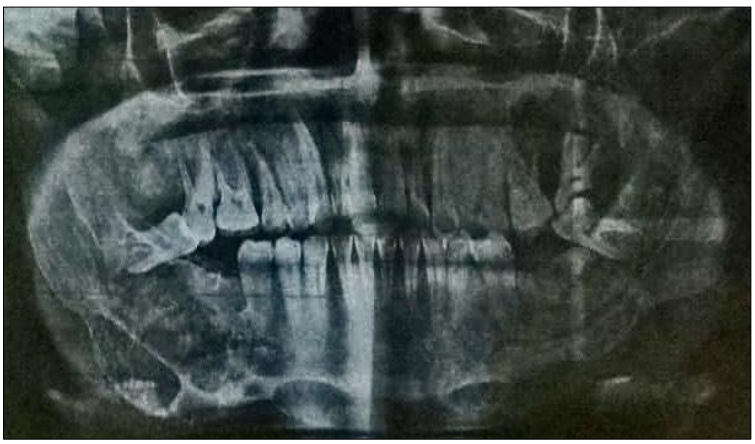

Figure 2: Orthopantamogram showing a sharply demarcated and lobulated radiolucency in the right hemimandible and the left alveolar bone, related to the root of the right and left molars (27, 26, 47 et 46 teeth).

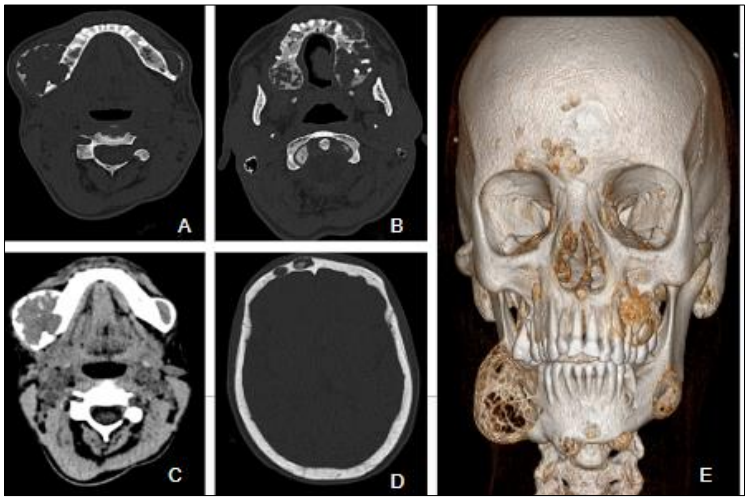

Figure 3 : Facial CT scan in axial sections with bone (a, b, d) and parenchymal (c) windows, along with 3D reconstruction (e), showing multiple expansive bone lesions with cortical disruption in places, featuring intralesional trabeculations visible in the frontal bone, maxillomandibular arches, and mandibular angles, with ground-glass attenuation and cortico-medullary dedifferentiation of the cranial vault.

During the first year of postoperative follow-up, The patient reported rib pain, prompting a request for a cervical-thoracic CT scan revealing Multiple expansive lytic bone lesions with blowout and rupture of the cortical bone visible in areas such as the bilateral clavicles, scapulae, right humeral head, and layered rib regions, as well as diffuse bone involvement predominantly showing condensing patterns with cortical-medullary dedifferentiation and bone hypertrophy in the vertebral and rib areas (figure 4).

Subsequently, the treatment consisted of surgical removal of the parathyroid mass. The lesion was histopathologically diagnosed as a parathyroid adenoma. Medical treatment by bisphosphonates was associated. No surgical treatment for the brown tumor lesions has been performed. Regression of the swelling was noticed months later.

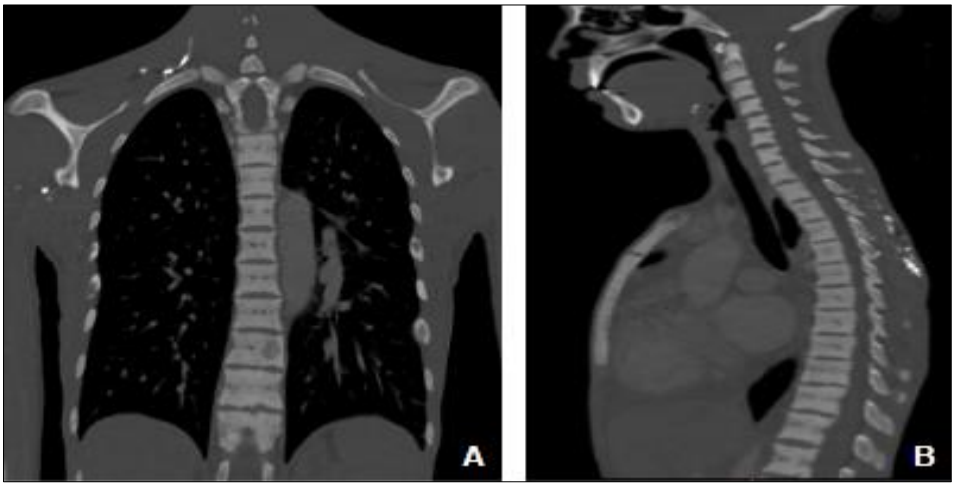

Figure 4: Chest CT in bone window with coronal (a) and sagittal (b) sections showing a lytic bone lesions with blowout and rupture of the cortical bone visible in areas such as the bilateral clavicles, scapulae, and layered rib regions, as well as diffuse bone involvement predominantly showing condensing patterns with cortical-medullary dedifferentiation and bone hypertrophy in the vertebral and crib areas.

DISCUSSION

Hyperparathyroidism is classified into three prominent entities; primary, secondary, and tertiary hyperparathyroidism. Primary hyperparathyroidism has increased parathyroid hormone (PTH) levels along with high calcium and low phosphate levels, usually secondary to parathyroid adenoma, parathyroid hyperplasia or parathyroid carcinoma [3]. Secondary hyperparathyroidism has high PTH levels along with low calcium and high phosphate levels, usually due to chronic kidney disease or vitamin D deficiency. Lastly, tertiary hyperparathyroidism has high PTH along with high calcium and high phosphate levels and this usually manifests itself in the setting of long standing secondary hyperparathyroidism [4]. First described by Recklinghausen in 1891 [5], osteitis fibrosa cystica commonly known as brown tumors are solitary or multiple non-neoplastic bone lesions with prevalence of about 4.5% in primary hyperparathyroidism and 1.5-1.7% in secondary hyperparathyroidism. The term “brown” refers to the accumulation of hemosiderin pigments rendering the lesion brown macroscopically. It has a low prevalence of 0.1% with a feminine predominance. 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 [6]. Clinically, brown tumors manifest themselves as localized palpable, tender, and hard masses usually in the mandible, sternum, ribs, pelvis, and long bones. Additionally, patients usually complain of other symptoms secondary to hypercalcemia; like abdominal pain, nausea, vomiting, constipation, renal stones, and fatigue. Radiological manifestation varies according to the extent of the hyperparathyroidism; nonetheless, they are typically well-circumscribed unilocular or multilocular lytic masses with either sclerotic margins at the periphery or infiltration into surrounding soft tissue with cortical damage. Diagnosis can be difficult sometimes as radiologically it can mimic metastatic lesions or primary tumors like giant cell tumors, hence to differentiate between them, the diagnosis is made based on the laboratory findings with calcium, phosphate, parathyroid hormone level, alkaline phosphatase; histologic results; and radiological results all together [7, 8]. Treatment is mainly aimed at resolving the primary problem, which is hyperparathyroidism. Since parathyroid adenoma is one of the most common etiologies for brown tumors, surgery to remove the adenoma can result in almost complete remission with remineralization of the lytic bone portions [9]. Nonetheless, for large disfiguring lesions surgical excision of the brown tumor alone may be indicated [6]. In hyperparathyroidism, the mainstay of the treatment is aiming to control the calcium level [10]. However, in cases of primary and tertiary hyperparathyroidism, surgical management is first line therapy which is achieved by performing a parathyroidectomy [13]. Parathyroid adenoma

localization can be done by several ways; invasive and non-invasive methods. Non-invasive methods consist of imaging studies like: ultrasonography, CT, MRI, or scintigraphy using technetium (Tc-99m) sestamibi [11]. Scintigraphy has a high sensitivity reaching around 80% in localizing a single adenoma, however this sensitivity drops to 25% when there are multiple adenomas. Surgical management of parathyroid adenoma is by surgical exploration of the neck with subsequent removal of the adenoma, biopsy of another parathyroid gland is indicated if another adenoma or multiple gland hyperplasia is suspected [12]. There are several ways to check if the adenoma is successfully removed during and after the surgery. Intraoperatively a rapid PTH assay can be used and post-operatively a calcium and PTH levels can be checked which would show a downward trend if the pathological gland was removed [13]. Clinically, a brown tumor may present similarly to other maxillary tumors, with bony swelling in the zygomatic, palatine, and/or gingival regions, accompanied by facial deformity, asymmetry, pain, and mobility issues, including potential tooth loss. Sometimes, the lesion may be asymptomatic, and the diagnosis is made incidentally during routine radiological examinations. Additionally, patients usually complain of other symptoms secondary to hypercalcemia; like abdominal pain, nausea, vomiting, constipation, renal stones, and fatigue [14]. Biologically, the disruption of the phosphocalcic balance is very suggestive, with elevated parathyroid hormone (PTH) levels confirming the diagnosis.

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 [14].

Radiologically, 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 [15].

Radiographic features are compatible with bone resorption caused by activated osteoclasts. 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 multiple bones and can be misdiagnosed for metastasis [16]. Special feature of maxillomandibular location includes dental changes such as abnormally narrow pulp chambers, resorption of the lamina dura around the roots of teeth, and demineralization of the marrow bones of the jaws causing a characteristic “ground glass” appearance [13, 14]. 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. The MRI appearance depends on the relative proportion of its components. The lesions, therefore, may be solid, cystic, or mixed. Solid components are intermediate to low intensity on T1- and T2-weighted images, while the cystic components are hyperintense on T2-weighted images and may have fluid-fluid levels. The solid component and septa can be intensely enhanced after contrast injection [17]. 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. 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. 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 [6]. The treatment of hyperparathyroidism is the first step in the management of the brown tumor [18]. There is general consensus that the treatment of primary hyperparathyroidism is parathyroidectomy, but opinions are divided about the management of the bony lesions [5-12]. Most authors believe that brown tumor regression and healing are expected after the correction of hyperparathyroidism]. The time necessary for bone regeneration varies from several months in young patients to several years in older patients. In the case reported here, no treatment of the palatal brown tumor has been done. However, several cases of brown tumor that grew after parathyroidectomy or normalization of hyperparathyroidism have been reported. In these cases, many authors have reported the surgical resection of remaining brown tumor [4-18].

CONCLUSION

Any lacunar lesion in the maxillae should raise suspicion of a brown tumor and should prompt systematic investigation for hyperparathyroidism. Imaging plays a crucial role in diagnosing and monitoring brown tumors; it also aids in identifying the parathyroid nodule for etiological diagnosis. Recognizing primary hyperparathyroidism can prevent unnecessary surgery on the brown tumors in the

maxillae, as these lesions tend to regress following the removal of the parathyroid lesion.

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