



Brown Tumor a Rare Presentation of Maxillary Cyst with Pathological Fracture of Femur

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Abstract

Brown tumor is a giant cell lesion associated with hyperparathyroidism. It is a benign condition and represents the terminal stage of the remodeling process in the hyperparathyroid state. This severe parathyroid bone disease is a rare clinical presentation of primary hyperparathyroidism which is most often due to a parathyroid adenoma, secreting Parathormone (PTH). Elevated PTH levels cause bone resorption, the formation of polyostotic lesions and a reduction in bone mineral density, predisposing to pathological fractures [1]. Here we report the case of middle aged female having primary hyperthyroidism due to a parathyroid adenoma with osteolytic cystic lesions of the pelvis, the femur, and right maxillary bone. She was treated with surgical excision of the affected parathyroid gland, followed by open reduction and internal fixation with intra medullary nailing of the fractured femur.

Keywords: Primary hyperparathyroidism; Brown tumor; Excision

Introduction

Primary hyperparathyroidism most often is due to a parathyroid adenoma secreting PTH. The severity of hypercalcemia is proportional to the size of the adenoma. In patients with primary hyperparathyroidism, 85% are caused by solitary parathyroid adenoma, 13% have hyperplasia, 1% to 2% has double adenoma and 1% has carcinoma [2]. The PTH hypersecretion causes excess calcium reabsorption from kidneys, phosphaturia, and increased vitamin D synthesis and bone reabsorption. PTH increases osteoclastic activity which predisposes to pathologic fractures [1,2-4]. Accumulation of erythrocytes and their pigments give a reddish/brown hue to the lesions, hence the name "brown tumor". These may be the first sign of hyperparathyroidism [2]. In this report we have discussed the rare presentation of a patient having parathyroid adenoma with maxillary cyst, femoral pathological fracture and pelvic bone skeletal lesions. Brown tumors are more commonly seen in the mandible than in the maxilla. The reported prevalence of brown tumors is 0.1%.

Case Presentation

A 53-year old female came with complaints of pain over the left thigh following trauma for the past 2 weeks, with no co-morbidities. On examination: The right lower limb appeared externally rotated with painful range of motion over the right hip joint, restricted mobility and a swelling over the right maxilla which was firm in consistency (Figure 1). No nodule was palpable over the neck. Other systems were normal. Laboratory analysis revealed serum calcium - 11.5 mg/dl, alkaline phosphatase - 956 IU/l, phosphorus- 1.5 mg/dl, serum T3 - 3.43 ng/ml, serum T4 - 4.7 µg/dl, serum TSH - 2.45 µIU/ml and serum parathyroid hormone level was 1900 pg/ml.

USG neck: A large heterogeneously hypoechoic lesion measuring 4.4 cm × 2.2 cm × 2.5 cm with increased vascularity seen posterior to the right lobe of the thyroid - neoplastic lesion of the parathyroid may be considered.

X-ray pelvis: Comminuted and displaced fracture seen involving the proximal 1/3rd of the right femur (Figure 2).

CT pelvis: Diffuse osteoporotic changes with thinning of the cortex and the trabeculae involving all the pelvic bones and the visualized portion of both the femurs. Multiple mildly expansile lytic lesions seen involving the left iliac bone, both the femurs in the proximal aspect and the transverse process of L5 vertebrae on the right side-suggestive of brown tumor (Figure 3).

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Figure 1: RT-Maxillary cyst.



Figure 2: RT-Femur fracture.



Figure 3: CT-Pelvis: Comminuted and displaced fracture seen involving the proximal 1/3rd of the right femur with over-riding of the bony fragments-suggestive of pathological fracture.

Surgical treatment

Findings: right superior parathyroid was well encapsulated measuring about 5 cm × 3 cm which was displacing the thyroid medially. Right recurrent laryngeal nerve was spared, and the adenoma was excised into (Figure 4).

Repeat PTH levels: 14.9 pg/ml. Following this she was posted for ORIF, with intramedullary nailing of the fractured femur 2 weeks after surgery. The patient was given oral calcium and vitamin D for 2 months. Histopathological report confirmed the diagnosis of a parathyroid adenoma. We followed up the patient every month for 6 months, and there were no other complications till the last follow up.

Discussion

The prevalence of primary hyperparathyroidism may range from 1% to 4%, with a male:female ratio of 1:3. In most cases (around 80%) a solitary adenoma is the cause and in 20% of cases, it is due to a glandular hyperplasia. Serum calcium, ionized calcium and PTH levels



Figure 4: Intra operative picture, RT-para thyroid adenoma.

must be obtained to confirm the diagnosis [3,5]. Brown tumors are relatively rare benign lesions, resulting in abnormal osteoclastic and osteoblastic activity resulting in resorption of the bone. The incidence of brown tumors in patients with primary hyperparathyroidism is 1.5% to 1.7%. At present, hyperparathyroidism is usually treated before such lesions develop; therefore they have become extremely uncommon [6,7]. Radiological examination showed osteopenia of the whole skeleton and multiple localized lytic lesions with a benign aspect. Hyperparathyroidism affects mainly cortical bone. The bone marrow in these cysts may be replaced by vascularized fibrous tissue and giant cell reaction. Brown tumors represent foci of hemorrhage within an enlarged fibrotic marrow space. Organization of these lesions results in the release of hemosiderin and the accumulation of macrophages, fibroblasts and giant cells. In our case the patient had multiple cystic lesions in the skeleton [3]. Sestamibi scan, however, is indicated if ectopic PTH producing adenoma is suspected or if the CT scan and the USG failed in localizing the PTH producing lesion [3,8]. After parathyroidectomy, the serum PTH level falls dramatically, and bone resorption stops, resulting in hungry bone syndrome [3,5,8]. Brown tumors are the late manifestation of hyperparathyroidism. They should only be removed if they persist even after the removal of the parathyroid adenoma, when functional problems are detected, or if the tumors are too large. However the regression can take several months.

Conclusion

Any patient suspected of having a pathological fracture should undergo serum PTH level assessment, along with whole skeleton screening. Parathyroidectomy is the treatment of choice, coupled with other orthopedic interventions to address the pathological fractures.

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