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Case Report

Primary Hyperparathyroidism Having Multiple Brown Tumours Mimicking Metastatic Bone Disease

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Abstract

Primary Hyperparathyroidism (PHPT) is a disease characterized by excessive secretion of parathyroid hormone (PTH), leading to the resorption of parts of the skeleton, bone loss and elevated calcium levels. Brown tumor (BT) aka Von Recklinghausen's disease of bone is a rare skeletal manifestation of PHPT. This multiple benign bone lesion may simulate a metastatic bone disease, posing a great diagnostic challenge for the clinician. A high index of suspicion is needed for diagnosis in its early stage. Clinical suspicion, laboratory and radiological investigations are required to diagnose this condition. In a resource poor setting like ours, unavailability and unaffordability of these facilities, poses a greater challenge. We present a 39 year old lady with multiple bone lesions, initially considered to be a metastatic bone disease, but later turned out to be BT. In all cases, with multiple osteolytic lesions, a possibility of "BT" must be kept in mind.

Keywords: *Primary Hyperparathyroidism; Brown Tumour; Metastatic Bone Disease*

Introduction

BT has never ceased to be a subject of great interest in orthopaedics. A rare and late manifestation of prolonged hyperparathyroidism (HPT).

PHPT is a common disease, which affect 0.1 - 2% of populations with 3:1 female preponderance [1]. Frequency of various parathyroid lesions underlying the hyperfunction are adenoma (80 - 95%), primary hyperplasia (5 - 19%), parathyroid carcinoma (1%) [2].

HPT is characterized by elevated PTH secretion, excessive calcium reabsorption, phosphaturia, increase vitamin D synthesis and bone resorption.

Its name was derived from the characteristic brown coloration, due to hemosiderin deposition into bone cysts.

We present a 39year old lady with multiple bony lesions, earlier considered a metastatic bone disease.

Case Presentation

38yr old female, referred with complaints of low back pain, recurrent severe pain on the right thigh and knee for one year. There was history of weight loss and anorexia. No history of fever, drenching night sweats, trauma or contact with adult with chronic cough.

Examination revealed chronically ill patient with an antalgic gait, swelling on the right side of the neck, nil lymphadenopathy, however, had tenderness over the right thigh and knee. Neurologic status was intact. Prior to presentation, she had earlier been evaluated as a case of metastatic bone disease.

Investigations showed elevated serum calcium, 13.9 mg/dl (8.1 - 10.4) mg/dl. Alkaline phosphatase 204 (9 - 35) ul, decreased serum phosphate 2.2 (2.5 - 5.0) mg/dl, potassium 3 (3.5 - 5.5) mg/d, uric acid 6.2 (2.4 - 5.7) mg/dl. Serum vitamin D and parathyroid hormone levels were not done due to serious financial constraints. Serum protein electrophoresis and Urine for Bence Jones proteins were negative.

Neck ultrasound revealed a well circumscribed, ovoid, solid, hypo echoic mass lesion (3.11 x 2.97 x 2.53) cm, on the anterolateral aspect of the neck adjacent and inferior to the right thyroid lobe. Positron electron tomography (PET) scan was not done for financial reasons.

Abdominal Ultrasound revealed multiple calcifications of the renal parenchyma. Plain X-rays of the hands revealed multiple lucencies affecting some carpal and metacarpal bones of both hands and penciling of the terminal digits of the right fifth finger (See figure 1). Femur and pelvis- lucent expansile lesions on the left pubic ramus and ischium and the entire right femur without reactive sclerosis (See figure 2). She could not afford magnetic resonance Imaging (MRI).

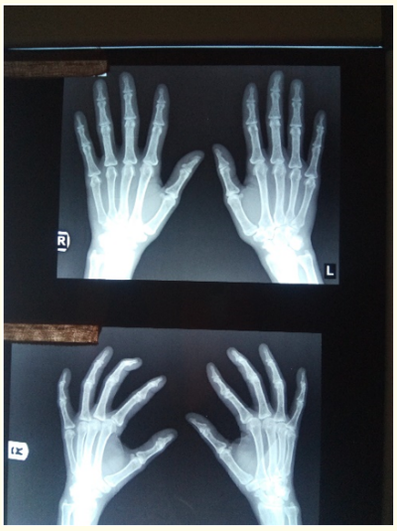

Figure 1: *Plain radiographs of both hands.*

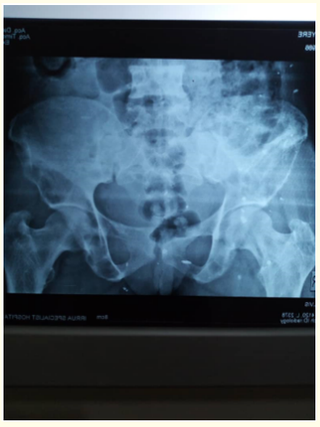

Figure 2: *Plain radiographs of the pelvis.*

A diagnosis of "BT" due to hyperparathyroidism was made.

Neck surgery revealed normal thyroid with right parathyroid mass. Right lobectomy with partial parathyroidectomy was done. Histology showed confined hypercellular parathyroid consistent with Adenoma.

Tablet cholecalciferol 60,000 units weekly for 8 weeks along with other symptomatic therapy was prescribed. She responded well and symptoms abated. Two weeks later, calcium and phosphate levels normalized. Hormone levels were unaffordable. Cast application to protect the femur was declined.

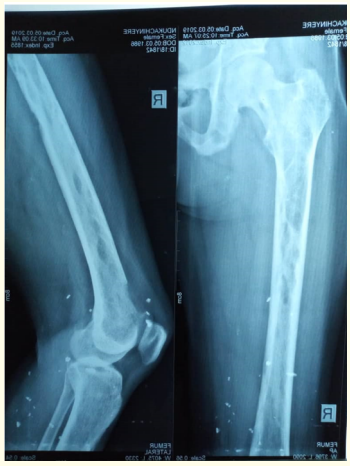

Figure 3: *Plain radiographs of the femur.*

Discussion

Fibrous dysplasia, simple bone cysts, eosinophilic granuloma, enchondroma, non-ossifying fibroma, infection, chondromyxoid fibroma, chondroblastoma, aneurysmal bone cyst, fibrous cortical defect, multiple myeloma, metastasis, giant cell tumour, osteoblastoma are some differential diagnosis of BT.

It is a disease of adulthood, seen mostly in those less than 50 years [3].

Multiple BT cases associated with PPHT were initially reported by Joyce., *et al*. in 1994 [4]. Actual global estimate is unknown. Such rare and multiple benign lesions may pose a great challenge for the clinician, hence awareness of this condition is necessary for early diagnosis. They manifest with uni or multilocular radiolucencies with expansion and thinning of the cortices on radiographs [5]. Serum PTH is often significantly elevated and may be associated with low serum vitamin D levels. Computerized tomography (CT), MRI, PET are also required to confirm diagnosis, particularly in precarious situations. Albeit requested, they were unaffordable. Biopsy is considered the gold standard, often times they are inconclusive. Microscopically, these lesions appear as multinucleated giant cells in a spindle cell matrix containing hemosiderin deposits.

BT had been classically described as "bone, stone, abdominal groan and psychic moan" [6]. These symptoms are attributed to elevated serum calcium, excessive bone resorption and mental agony that ensue.

BT are primarily observed on the face, mandible and neck region, pelvis, femur and ribs. Pathological fractures are uncommon.

There is a rapid osteoclastic bone resorption resulting from PTH. Hemorrhage, vascular, fibrous tissue and granulation tissue replace existing bone. Localized accumulation of fibrous tissue and giant cells resulting in bone expansion [7].

Total or subtotal parathyroidectomy is the gold standard for the treatment of PHPT [8], which was performed in this case. She improved tremendously, both in clinical and biochemical parameters. Pathological fractures may need stabilization or surgical correction [9].

Limitations

Unaffordability of crucial diagnostic tools like hormonal assays (PTH and vitamin D), radiological (CT, MRI, PET isotope bone scans) investigations were limitations in the management of this case. Our major strength was the involvement of multiple specialists in the application of care. This multidisciplinary approach prevented us from miss diagnosis and performing unnecessary surgeries. Working in a specialist center and having a co-operative patient were other assets.

Conclusion

Prevailing serious financial and unavailability of some key laboratory and radiological investigations in resource poor setting like ours poses a great challenge in diagnosing PHPT and BT. A high index of suspicion is required. In cases of hypercalcemia and radiographic evidence of multiple lytic lesions, PHPT should be kept in mind.

Conflict of Interest

None declared.

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