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Case Report

Tertiary hyperparathyroidism with multiple Brown tumors mimicking malignancy

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Abstract: Brown tumors are an uncommon type of benign osteolytic bone lesions that occur in hyperparathyroidism, which is an endocrine disease characterized by excessive secretion of the hormone parathormone. They occur more in primary hyperparathyroidism and are seen to regress after removal of parathyroid adenomas. In our report, we describe a a case of tertiary hyperparathyroidism with multiple lytic lesions of the right great toe, left fibula, twelfth posterior rib on right side and base of middle phalanx of left hand. This report illustrates the importance of taking brown tumor in differential diagnosis in patients with multiple lytic lesions, occurring in the backdrop of chronic kidney disease, where a failure to establish an accurate diagnosis may lead to further unnecessary and painful diagnostic procedures and even extensive surgery.
Keywords: Brown tumors, endocrine disease, osteolytic bone lesions, primary hyperparathyroidism, parathyroid adenomas, and chronic kidney disease.

INTRODUCTION

Brown tumors are seen in severe untreated primary and Tertiary hyperparathyroidism [1]. They are infrequently seen today due to early diagnosis and management. A classical description of clinical presentation and radiographic images of a patient with chronic kidney disease, on chronic hemodialysis, presenting with brown tumors due to Tertiary hyperparathyroidism is presented.

In view of the parathyroid hyperplasia and adenoma in setting of CKD with mildly elevated serum calcium level, diagnosis of tertiary hyperparathyroidism was considered.

CASE REPORT

Clinical History

A 41 year-old woman of Indian origin was admitted to the orthopedic emergency department. Clinically she presented with pain in her right great toe, without a significant history of trauma. The patient had a history of pain in the right toe while walking and on weight bearing. She was referred to the radiology department for a plain radiograph of the right foot.

Examination and lab parameters

On physical examination of the patient, tenderness was found to be present in the right toe. No other significant clinical abnormality was observed. On laboratory analysis, serum calcium level was 11.1 mg/dl (normal 8.4-10.7 mg/dl), serum albumin level was 5 g/dl (normal 3.4-4.8 g/dl), serum alkaline phosphatase level was 787 IU/l (normal 50-240 IU/l), serum parathyroid hormone level was 1627.4 pg/ ml (normal 7-53 pg/ml), vitamin D3 (1,25-dihydroxy cholecalciferol) was 43 pg/ml (normal 25-45 pg/ml) and vitamin D3 (25-hydroxy cholecalciferol) was 24 ng/ml (normal 15-80 ng/ml).

Radiological features

The following investigations were done-

(A) Radiograph right foot AP/ lateral view - showed a geographic lytic expansile lesion involving the proximal phalanx of the great toe.

(B) MRI of the right foot was done

It revealed an expansile lytic lesion involving the entire proximal phalanx of right great toe. The cortex was significantly thinned and not visible at all, along the lateral margin of the lesion. The lesion was seen extending till the proximal as well as distal articular margins of the proximal phalanx, with focal breach in the proximal articular surface. The lesion showed intermediate signal on T1 and T2 weighted images. Minimal soft tissue component was noted along the lesion, abutting the underlying flexor tendon.

C) Further evaluation of the patient was done to rule out malignancy and elucidate the diagnosis. Plain digital radiographs were recommended to the patient including those of both the hands, skull, and feet. The radiographs all showed similar lytic expansile lesions in the twelfth posterior rib on right side and base of middle phalanx of left index finger.
However, there was no evidence of calcification in the lesions.

(D) Similar lesion was also seen in the mid shaft of left fibula
(E)The radiograph of the skull was found to be normal.

(F) An ultrasound scan of the neck was also done which showed hyperplastic parathyroid glands, posterior to the thyroid gland.

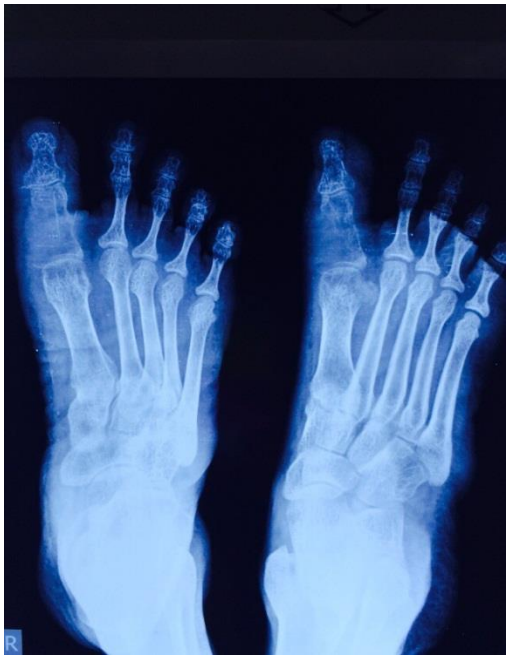

Fig-1: Radiograph right foot AP/ lateral

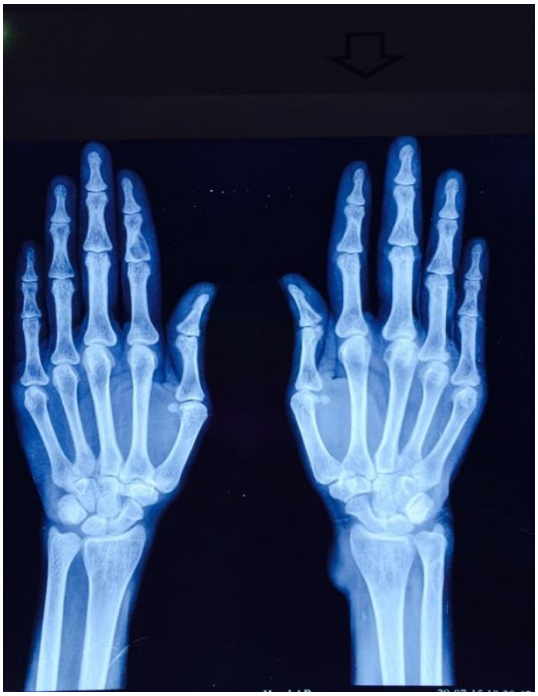

Fig-2: Radiograph both hands AP view

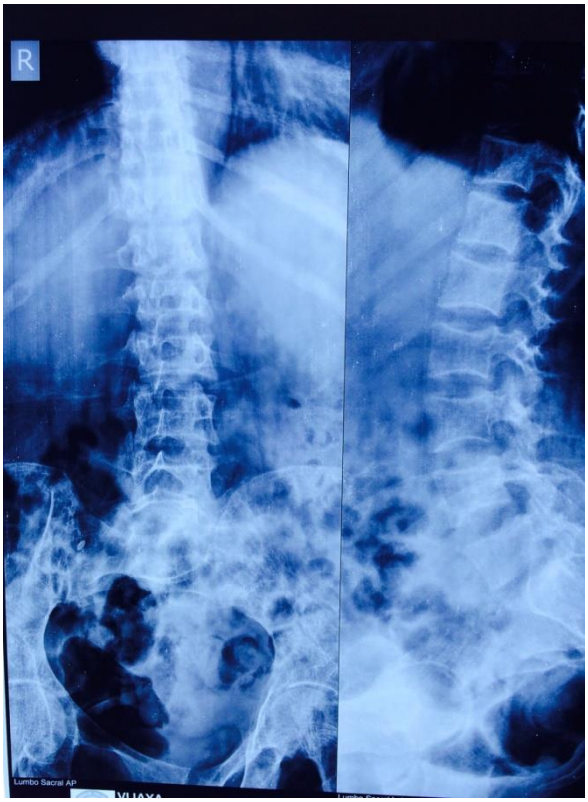

Fig-3: Radiograph thoracic lumbar spine AP/lateral view

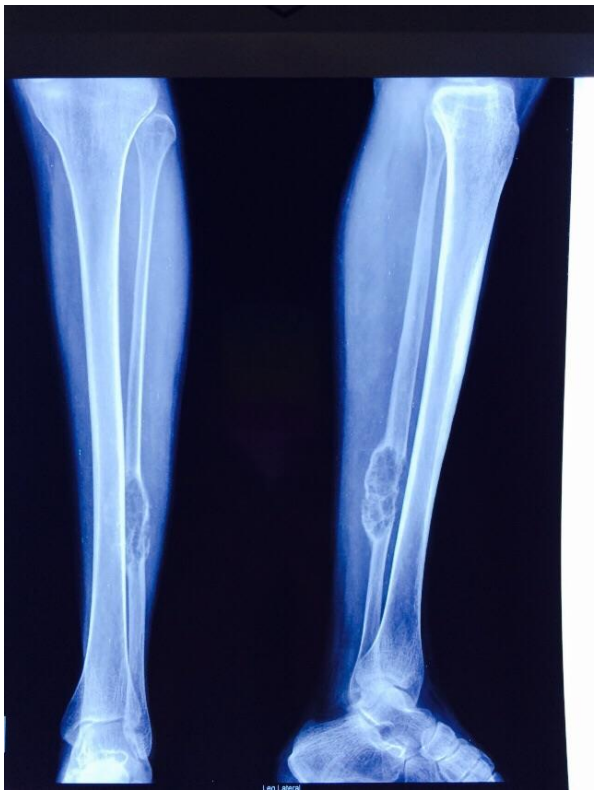

Fig-4: Radiograph left leg AP/lateral view

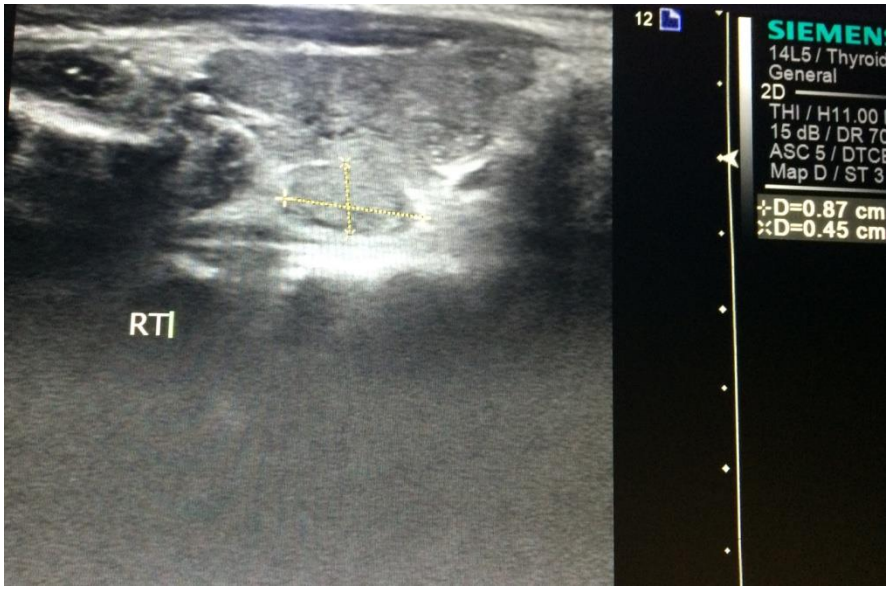

Fig-5: USG neck right side

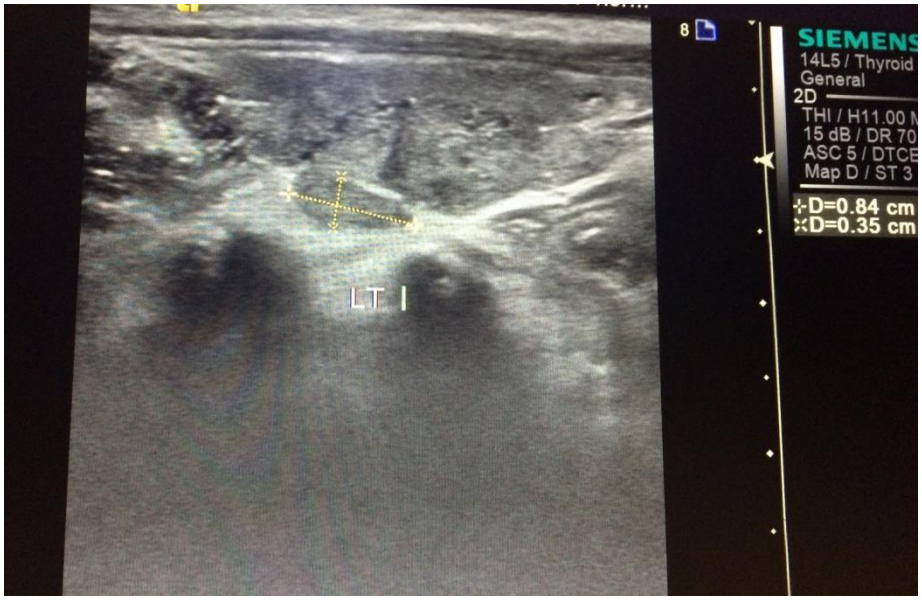

Fig-6: USG neck left side

Diagnosis

To arrive at conclusive diagnosis, pathological analysis was essential. Hence trucut biopsy of the soft tissue and bone from proximal phalanx of right great toe was done. This was followed by fine needle aspiration cytology of the parathyroid lesions.

Histopathological examination and results

(a)True cut biopsy of soft tissue and bone obtained from proximal phalanx of right great toe reveled a lesion composed of numerous osteoclastic giant cells scattered throughout. However there was no evidence of malignancy. The pathological diagnosis was given as BROWN TUMOR OF HYPERPARATHYROIDISM.

Fig-7: Trucut biopsy of right toe

(b) Fine needle aspiration cytology of the parathyroid lesions reveled typical hyper cellular nodules. These nodules were surrounded by a thin capsule and showed chief cell hyperplasia; with the pattern of growth being acinar / diffused and pseudo papillary. The pathological diagnosis was given as PARATHYROID ADENOMA.

Intervention and Conclusion

Following pathological diagnosis of Brown tumor, successful internal fixation was performed by the orthopaedician and the patient was able to ambulate with normal gait and without pain.

Ten days later, bilateral para thyroidectomy was done. Three weeks postoperative the serum levels of calcium and PTH had declined to normal levels. The patient is currently on regular follow up, which is uneventful so far.

DISCUSSION

Secondary hyperparathyroidism is an endocrine disorder, where there is excessive functioning of the parathyroid glands. There is hyperplasia of the gland and excessive secretion of the parathyroid hormone. This is done in order to compensate long standing hypocalcemia, especially seen in patients with long standing chronic renal failure, as is seen in our case. Chronic renal diseases tend to cause deterioration of nephrons, with gross decline in the glomerular filtration, leading to deficient vitamin D synthesis which thereby leads to deficient calcium absorption. Consequently, elevated levels of serum phosphate are seen in the body. As phosphate is responsible for bone mineralization, this excessive serum phosphate causes excessive serum calcium to be deposited in the bones, ultimately leading to reduced serum calcium levels. In reaction to low serum calcium, there is hyper stimulation of the parathyroid gland, leading to excessive parathyroid hormone production and resulting in hyperparathyroidism. Beyond a certain point, the hyperplastic / adenomatous parathyroid gland that occurs due to secondary hyperparathyroidism becomes autonomous, leading to tertiary hyperparathyroidism.

The bone changes occurring in primary hyperparathyroidism were known as osteitis fibrous cystic [5] and were first described by Recklinghausen. Such lesions, although benign may mimic malignancy and appear as a challenge for the clinician to arrive at a definite diagnosis, due to a wide range of differentials. Lytic lesions caused by hyperparathyroidism are called Brown tumors and multiple Brown tumors associated with primary hyperparathyroidism were initially reported by Joyce *et al.;* [2, 4]. The term "Brown tumor" is a misnomer as it is not really a true neoplastic lesion. Although Brown tumors are seen in both primary and secondary hyperparathyroidism, they are more common in primary hyperparathyroidism in the back drop of chronic renal failure. Brown tumors may be seen in the facial bones, mandible, pelvis, ribs, phalanxes and femur. Initially, the earliest findings are seen in the small bones of hands. They are exponentially more in numbers in later stage of hyperparathyroidism or in parathyroid carcinomas. Clinically they manifest as redness, swelling, pathological fracture, and bone pain. The lesions appear as smooth expansile masses causing bone destruction [7]. They are known to cause bone expansion. Radiologically, they are present as well defined expansile radiolucent areas and may be multiloculated [Fig 3.0]. Histologically they are composed of intensely vascular fibrotic stroma with multiple osteoclastic cells.

The presence of hemorrhage, hemosiderin and hyper vascularity leads to the brownish color and hence the name.

About 90% of the patients with skeletal metastases present with multiple lesions [6]. Hence in the presence of renal failure and hypercalcemia, brown tumors of hyperparathyroidism should always keep as a differential diagnosis once metastasis is excluded. Treatment of brown tumor is mainly pharmalogic and gets corrected by treating the underlying hyperparathyroidism. Trianta fillidou et al. advocate curettage of the lesion and wound packing allowing for secondary healing in addition to adjunctive treatment of underlying disease [8]. Hyperparathyroidism is commonly treated with calcium, vitamin D, and controlled dialysis, although renal transplant or subtotal para thyroidectomy may also be options. As is seen in our case. Since bone healing in these patients is compromised, hyperparathyroidism must be controlled prior to successful surgical bone reconstruction [9, 10].

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