

Osteosclerotic and osteolytic manifestations of hyperparathyroidism in a case of Tc99m SestaMIBI positive parathyroid adenoma

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ABSTRACT

Primary hyperparathyroidism is the first differential diagnosis when a patient presents with asymptomatic hypercalcemia. The symptoms of hyperparathyroidism can be as grave as skeletal, cardiovascular, and neuropsychological changes. Skeletal manifestations are relatively common, and patient may present with generalized or focal bone pains, fragility fractures, subperiosteal bone resorption, and osteolytic lesions like brown tumors and salt and pepper appearance of the skull. However, focal osteosclerotic lesions of the skull are rare findings in hyperparathyroidism. Only a few cases of associated osteosclerosis are reported in the literature. Here, we report a case of Tc99m SestaMIBI positive parathyroid adenoma with coexisting osteolytic and osteosclerotic skull lesions on Tc99m methylene diphosphonate bone scan.

Keywords: Brown tumor, methylene diphosphonate bone scan, osteosclerotic skull lesion, parathyroid adenoma, primary hyperparathyroidism, Tc99m SestaMIBI

INTRODUCTION

Hyperparathyroidism is characterized by increased synthesis and release of parathyroid hormones (PTHs). Primary hyperparathyroidism (PHPT) is caused by an abnormal parathyroid gland; adenoma being the cause in >85% of cases.^[1] Secondary hyperparathyroidism is due to parathyroid gland hyperplasia resulting from end-organ resistance to PTH; the most important cause being chronic renal insufficiency. The major physiologic effects of PTH on skeletal tissue are generally catabolic in nature.^[2] Bony manifestations include diffuse osteopenia, brown tumors, subperiosteal bone resorption, pathological fractures, terminal acro-osteolysis, and salt-and-pepper appearance of the skull. Though osteolytic lesions have been widely documented, osteosclerosis (“dense bones” or hyperostosis) has been observed rarely in hyperparathyroidism. The exact incidence of osteosclerotic lesion in hyperparathyroidism is not

known. Its rarity can be speculated from the fact that only a handful of literature has reported osteosclerotic lesion in cases of hyperparathyroidism. Osteosclerotic lesions have been reported in some patients with secondary hyperparathyroidism; particularly in those with chronic renal failure.^[3,4] Focal osteosclerotic lesion usually involves pelvis, spine, skull, and metaphyseal end of the bone with simultaneous destructive or erosive lesions in other parts of the skeleton. The exact mechanism for the focal hyperostosis remains elusive. In very rare cases of PHPT, similar focal hyperostotic lesions have been described. We report one such case of focal osteosclerosis in skull in a patient with PHPT.

CASE REPORT

A 35-year-old male with pathological fracture in intertrochanteric region of right femur was referred to our department for a ^{99m}Tc-methylene diphosphonate (MDP) bone scan. The patient had a history of trivial injury at the site of fracture 1-year back. Investigations revealed hypercalcemia (12.8 mg/dL [8.5–10.1 mg/dL]), hypophosphatemia (1.2 mg/dL [2.5–4.9]), raised alkaline phosphatase (180.9 IU/L [37–147]), and low 25-OH Vitamin D levels (18.05 ng/ml [21–100]). Serum intact PTH (480.00 pg/ml [14–72]) level was raised. His kidney function tests were within normal limits negating the possibility of secondary hyperparathyroidism.

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His skeletal survey showed a lobulated lesion in the right proximal femur with a malunited intertrochanteric fracture and a well-defined lytic lesion in the mid-shaft of right femur [Figure 1]. His skull X-ray showed multiple round to oval focal sclerotic changes, which is an unusual radiological presentation in PHPT [Figure 2]. Magnetic resonance imaging (MRI) pelvis and both femori revealed a well-defined lobulated lesion involving intertrochanteric area and proximal shaft of right femur with associated expansion and cortical thinning. Also rounded to oval lesion with similar magnetic resonance morphology was noted in the mid-shaft region of right femur. Suggested differential diagnosis was polyostotic fibrous dysplasia and brown tumors. ^{99m}Tc-MDP bone scintigraphy revealed multiple hot spots over the frontal and parietal region of the skull [Figure 3]. Increased tracer accumulation was also noted in the trochanteric region of right femur (consistent with site of fracture) and along medial border

of mid-shaft of right femur (known lytic expansile lesion as on MRI) [Figure 3].

An ultrasound of the neck region done to localize the culprit parathyroid adenoma revealed no abnormality. Subsequently, a ^{99m}Tc-SestaMIBI planar [Figure 4] and single photon emission computed tomography/computed tomography scan [Figure 5] was done, which localized the parathyroid adenoma in the lower pole region of right thyroid lobe.

DISCUSSION

The diagnosis of hyperparathyroidism is made clinically. An elevated PTH with hypercalcemia is diagnostic of PHPT. Excess PTH results in an increase in bone breakdown (osteoclastic resorption) with subsequent fibrous replacement and reactive osteoblastic activity.^[5] This process results in characteristic radiologic features. The most common radiologic finding in PHPT are osteopenia, bone resorption (most common is subperiosteal reabsorption-virtually pathognomonic sign of hyperparathyroidism), acro-osteolysis, brown tumors, erosion of the dental lamina dura, and pathologic fractures. Osteosclerotic changes are an uncommon feature of hyperparathyroidism. It has been described in secondary hyperparathyroidism. While there are reports of diffuse or patchy osteosclerotic lesions in patients with hyperparathyroidism, discrete osteosclerotic lesions in the skull are reported rarely in these patients. Fujino

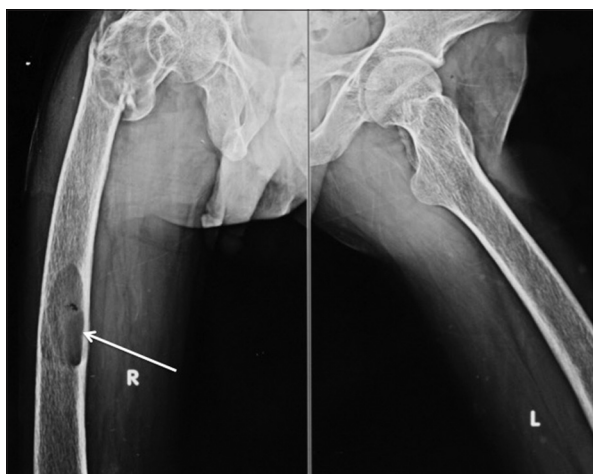


Figure 1: X-ray hip joint and bilateral femur: A lobulated lesion in the right proximal femur with a malunited intertrochanteric fracture and a well-circumscribed lytic lesions in mid-shaft of right femur that represent the osteoclastic resorption of bone

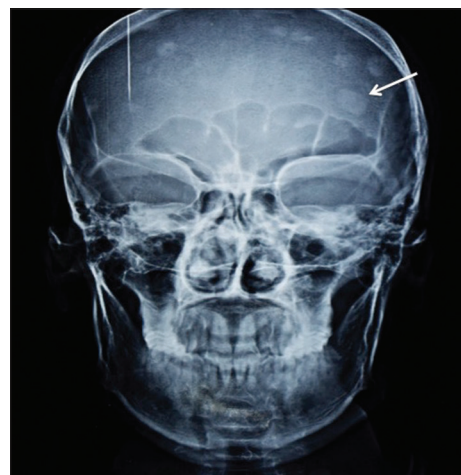


Figure 2: Antero-posterior view skull radiograph shows multiple well-defined round to oval sclerotic lesions

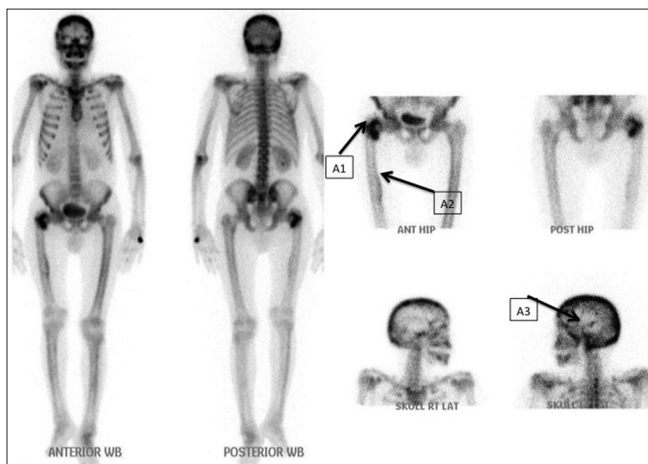


Figure 3: ^{99m}Tc-methylene diphosphonate whole body and spots bone scintigraphy revealed multiple hot spots over the frontal and parietal region of the skull. Increased tracer accumulation was also noted in the trochanteric region of right femur (consistent with the site of fracture) and along the medial border of mid-shaft of right femur

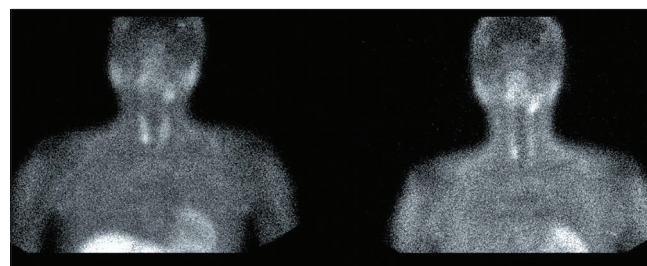


Figure 4: ^{99m}Tc-SestaMIBI planar images at 20 min and 2 h demonstrating parathyroid adenoma in the lower pole region of right thyroid lobe

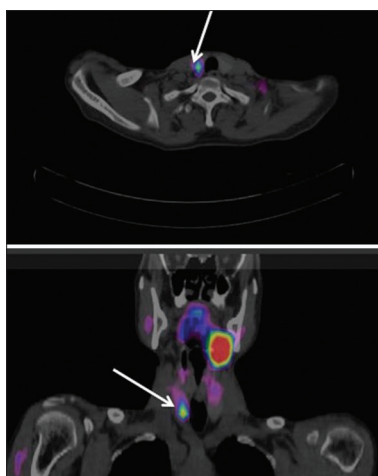


Figure 5: 99mTc-SestaMIBI single photon emission computed tomography/computed tomography scan localized the Parathyroid adenoma in the lower pole region of right thyroid lobe

et al.^[5] reported multiple osteosclerotic lesions of the skull in a 26-year-old man with PHPT. Boechat *et al.*^[6] reported two children with PHPT, who radiologically manifested with prominent osteosclerosis in the tibia. Chopra *et al.*^[7] report two females patients with hyperparathyroidism and multiple osteosclerotic lesions of the skull with co-existing Vitamin D deficiency. Our patients presented with discrete osteosclerotic lesions in the skull (a rare finding) along with a lytic lesion in the mid-shaft of femur (brown's tumor). The exact mechanism responsible for the focal hyperostosis in hyperparathyroidism remains unknown. Disproportionate osteoblastic activity after prolonged osteoclastic activity, an anabolic effect of supraphysiologic amount of parathormone on bone metabolism and high bone turnover in young patients are some of the plausible mechanisms responsible for the osteosclerosis.^[2,5,7,8] The same mechanism can be also applied to our case. The PTH level in our case is 480 pg/ml [14–72 pg/ml]. The age when the pathological fracture occurred was at 34 years, so the patient may have remained undiagnosed for a long time. Literature reports that in some patients with hyperparathyroidism, particularly in those with chronic renal

failure, osteosclerosis has been observed.^[9,10] This possible association was not observed in our patient as his renal parameters were within normal limits. The young age of the patient and supraphysiologic amount of PTH may have been a main factor in our case. Though the detection of focal osteosclerotic lesion does not alter the management of hyperparathyroidism *per se*. The most common differential diagnosis of multiple focal osteosclerotic lesion of the skull is metastasis. Therefore, its association with hyperparathyroidism should be brought to light as it may lead to unnecessary aggressive metastatic evaluation/search for a second diagnosis.

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