Facial Dysmorphism Due to Multiple Brown Tumors Secondary to Large Parathyroid Adenoma, Diagnosed on ^{99m}Tc-Sestamibi Parathyroid Scintigraphy

Abstract

Brown tumors are an uncommon entity associated with hyperparathyroidism. Initially described with primary hyperparathyroidism, with the widespread use of dialysis, they were also seen to be associated with secondary and tertiary hyperparathyroidism. These are lytic, expansile lesions causing bony pains, and depending on the size and extent, skeletal dysmorphism. The present case illustrates the detection of parathyroid adenoma with multiple brown tumors on ^{99m}Tc-sestamibi parathyroid scintigraphy in a patient presenting with facial dysmorphism and multiple bony pains.

Keywords: *^{99m}Tc-sestamibi, brown tumors, facial dysmorphism, parathyroid adenoma*

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A 32-year-old woman presented with progressive facial dysmorphism for the last 3 years. She had a past history of a left forearm fracture, following trivial trauma, 4 years ago, which was conservatively managed. Since then, she reported having multiple bony pains of varying intensity. Her biochemical workup revealed elevated serum calcium (13.8 mg/dL), parathyroid hormone (1562 pg/mL), and alkaline phosphatase (2744 U/L) levels. In view of the clinical features and biochemical parameters, she was scheduled for a ^{99m}Tc-sestamibi parathyroid scintigraphy with suspicion of primary hyperparathyroidism. The planar image in the anterior view showed focally increased tracer uptake at the upper pole of the left lobe of thyroid gland, persisting till 2 h [Figure 1a, thick arrow]. Increased tracer uptake was also noticed in the skull [Figure 1a, arrowhead], maxilla, and mandible [Figure 1a, thin arrow]. Single photon emission computed tomography/computed tomography revealed tracer uptake in the solid component (~2.2 cm × 2.4 cm × 2.1 cm) of a solid-cystic mass, posterior to the superior aspect of left lobe of thyroid [Figure 1b and c, thick arrow] suggestive of left superior parathyroid adenoma, and multiple tracer avid expansile, lytic lesions in the skull [Figure 1b and c, arrowhead], maxilla and mandible [Figure 1b and c, thin arrow], likely brown tumors. The patient underwent parathyroidectomy and experienced clinical and biochemical recovery. The surgical histopathology was consistent with parathyroid adenoma. Multiple expansile skeletal lesions in the facial bones were causing progressive facial dysmorphism, which eventually led to ^{99m}Tc-sestamibi imaging and detection of the parathyroid adenoma.

Primary hyperparathyroidism occurs due to excessive production of parathyroid hormone leading to calcium resorption from the bones, osteopenia, and eventually pathologic fractures and decreased serum phosphate levels due to phosphaturia. Parathyroid adenoma is the most common etiology of primary hyperparathyroidism.[1] Brown tumors are focal skeletal lesions seen in hyperparathyroidism due to aberrant bone turnover and do not represent true tumors. The name stems from the hemosiderin content giving a brownish tinge and their lytic expansile "tumor-like" nature.[2] Brown tumors are often accompanied by generalized osteopenia, salt and pepper appearance of the skull, and subperiosteal cortical erosions. Brown tumors are an uncommon entity, with an estimated prevalence of 2% in cases with primary hyperparathyroidism, showing variable uptake on ^{99m}Tc-sestamibi

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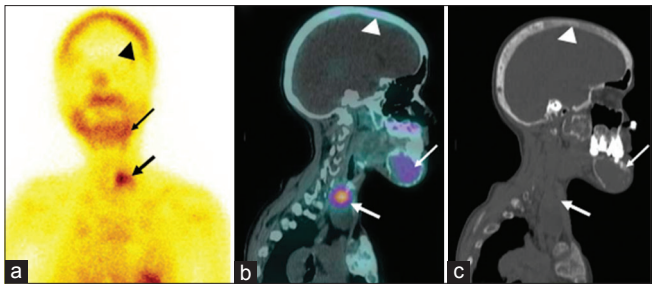

Figure 1: ^{99m}Tc-sestamibi imaging, (a) planar delayed anterior image, (b) sagittal-fused single photon emission computed tomography/computed tomography, and (c) computed tomography images showing focally increased tracer uptake in a soft-tissue lesion at the superior aspect of the left lobe of the thyroid gland (thick arrow), cortical lytic lesions in skull (arrowhead), and mandible (thin arrow)

imaging.[3,4] The extensive skeletal involvement, as in the present case, often leads to dysmorphic features, which reflects the need to identify and treat such patients early in the disease course. Brown tumors secondary to parathyroid adenoma show variable response to parathyroidectomy, with larger lesions often requiring surgical resection.[5,6] The onus thus lies on the treating physicians to investigate patients presenting with features of skeletal dysmorphism, multiple bony swellings, and bony pains on the lines of hyperparathyroidism because of the definitive cure that can be offered by removal of the primary adenoma.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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