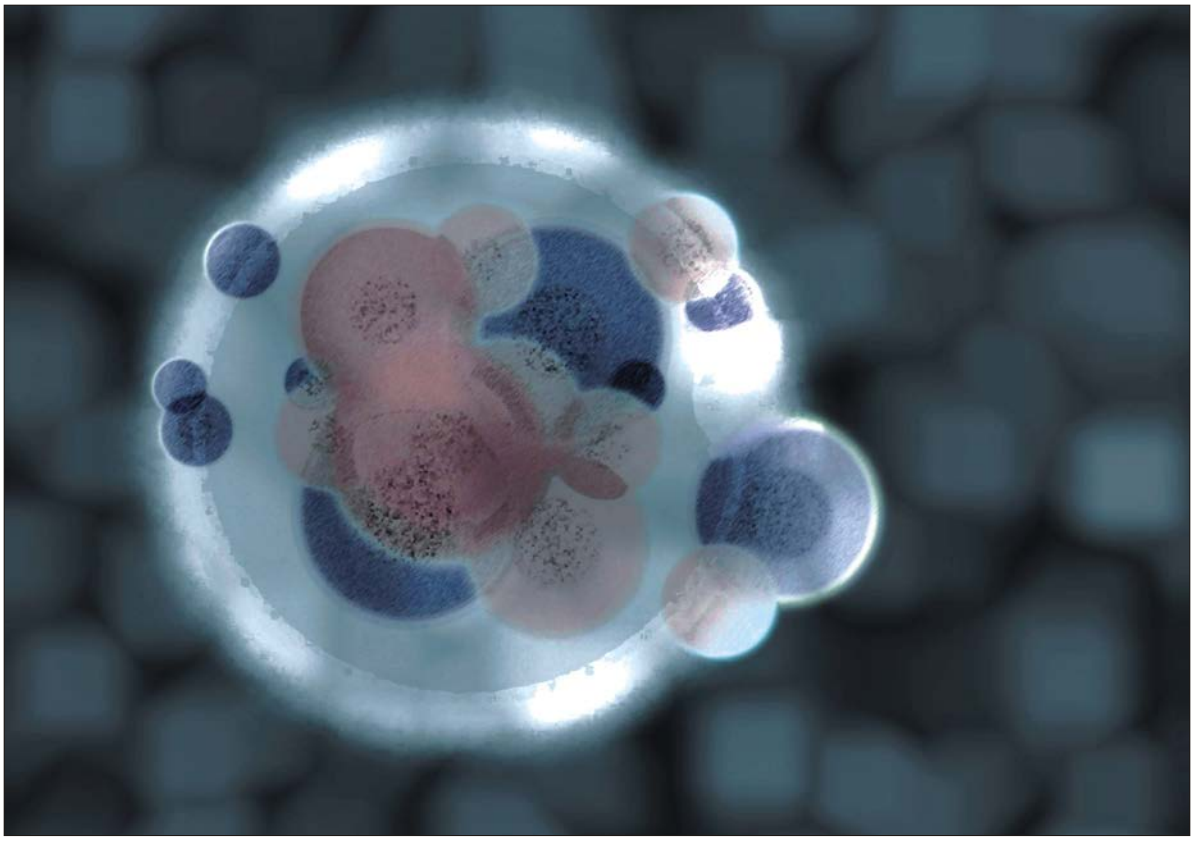

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Case Report
Unifocal Brown Tumor of Tibia: An Unusual Presentation

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Abstract

Brown tumour is a non-neoplastic lesion resulting from abnormal bone metabolism in hyperparathyroidism. These are rare focal giant cell lesions that arise as a direct result of the effect of parathyroid hormone on bone tissue in some patients with hyperparathyroidism and can affect the mandible, maxilla, clavicle, ribs and pelvic bone. The term brown tumour comes from the colour of lesion, which results from the vascularity, haemorrhage and deposits of hemosiderin. The main objective of this study was to investigate the peripheral brown tumour associated with secondary hyperparathyroidism simulating a peripheral giant cell lesion of the tibia. Experiment was conducted on a 12 years old male who presented with pain and deformity in the right leg with a globular bone-expanding lesion in the mid of the tibia. Fine needle aspiration cytology of mass showed multinucleated giant cells admixed with plump to spindle shaped cells. Histological sections showed regions of discrete bone resorption with osteoclast activity, areas with newly formed bone tissue and a loose fibrillar matrix with deposits of hemosiderin and multinucleated giant cells.

Key words: Brown tumour, hyperparathyroidism, histopathology, giant cells, tibia, maxilla, clavicle, pelvic bone, ribs

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Data Availability: All relevant data are within the paper and its supporting information files.

INTRODUCTION

Brown tumors are rare sequelae of hyperparathyroidism (HPT). The lesions localize in areas of intense bone resorption and the bone defect becomes filled with fibroblastic tissue. These tumors have a brown or yellow hue[1]. Brown tumors arise secondary to both primary and secondary hyperparathyroidism and have been reported to occur in 4.5% of patients with primary hyperparathyroidism and 1.5-1.7% of those with secondary disease[1]. The name "tumor" is a misnomer because the lesion, although invasive in some instances does not have a neoplastic potential and should be differentiated from true giant cell tumors of bone[2].

The so called brown tumor, which is one of the giant cell lesion of the bone is caused by hyperparathyroidism. It often causes an expansile osteolytic lesion of the bone, commonly in the mandible, pelvis, ribs and the femur. Brown tumors, with prevalence of 0.1% are more commonly seen in the mandible than in the maxilla[1]. The disease can manifest at any age, but it is more common among persons older than 50 years and is three times more common in women than in men[2].

Most patients with hyperparathyroidism are asymptomatic. Hypercalcemia is often discovered incidentally during routine laboratory testing; hypophosphatemia and increased alkaline phosphatase levels in blood may also be seen[2]. Any of the skeletal bones may be affected, including the cranio-maxillofacial ones. Brown tumors may be the first clinical sign of hyperparathyroidism. Brown tumors are a localized form of fibrous cystic osteitis found in the presence of hyperparathyroidism[1,2].

Histologically, brown tumors are made up of mononuclear stromal cells mixed with multinucleated giant cells, among which recent hemorrhagic infiltrates and hemosiderin deposits (brown color) are often found[3]. Whenever a round, radiolucent and bone expanding lesion in the facial bones of a patient with hyperparathyroidism is presented, a brown tumor must be considered to be the most likely diagnosis[1,2]. However, when the same type of lesion is found in patients without hyperparathyroidism, a more complex differential diagnosis procedure must be followed[2,3].

So, the main purpose of this study was to report a rare and unusual case of peripheral brown tumour associated with secondary hyperparathyroidism simulating a peripheral giant cell lesion of the tibia in a 12 years old male who presented with pain and deformity in the right leg with a globular bone-expanding lesion in the mid of the tibia.

CASE SUMMARY

This study was conducted in the Department of Pathology at Jawaharlal Nehru Medical College, Aligarh in February, 2019 on a 12 years old male, who presented with pain and deformity in the right leg with a globular bone-expanding lesion in the mid of the tibia. There was no family history of peptic ulcer, parathyroid disease or any other endocrinopathy. There was no history of increased thirst, urinary frequency, constipation, weight loss, urolithiasis, fracture, peptic-ulcer, use of vitamin or calcium supplements, Paget's disease, exposure to ionizing radiation or industrial toxins, or use of tobacco or alcohol.

On physical examination, the patient appeared well. His head and neck were normal; no cervical masses were palpated. His lungs, heart, breasts and abdomen were normal, as was his urine. Hematocrit was 39.8%, white cell count was 6800 μL^{-1}, with a normal differential count, platelet count, prothrombin time and partial-thromboplastin time. Blood analysis demonstrated PTH level of 223.0 pg mL^{-1} (normal range: 2-72 pg mL^{-1}). Calcium, phosphorus and alkaline phosphatase levels were within normal limits but his kidney function tests were deranged. A provisional diagnosis of brown tumour associated to a secondary HPT was made. The patient had no familiar history of HPT or other endocrine disorder and he has been prepared to be submitted to kidney transplant.

Fine needle aspiration cytology of mass showed multinucleated giant cells admixed with plump to spindle shaped cells (Fig. 1). The mass was excised and grossly, it was 10×8 cm in size, with a solid grey white cut surface (Fig. 2, 3).

Fig. 1: Fine needle aspiration cytology of mass showed multinucleated giant cells admixed with plump to spindle shaped cells
Hematoxylin and Eosin×40X

Fig. 2: Grossly a globular bone-expanding lesion in the mid of the tibia

Fig. 3: Cut surface of the growth showed solid grey white areas with specks of haemorrhage

Fig. 4: Histological sections showed regions of discrete bone resorption with osteoclast activity, areas with newly formed bone tissue and a loose fibrillar matrix with deposits of hemosiderin and multinucleated giant cells Hematoxylin and Eosin ×40X

Histological sections showed areas of discrete bone resorption with osteoclast activity, areas with newly formed bone tissue and a loose fibrillar matrix with deposits of hemosiderin and multinucleated giant cells (Fig. 4). A final diagnosis consistent with brown tumor of hyperthyroidism was given.

RESULTS AND DISCUSSION

Brown tumours are non-neoplastic lesions resulting from abnormal bone metabolism in HPT[4]. The committed sites are facial bones, clavicle, ribs, pelvis or femur[5]. In contrast to secondary HPT, jaw bones are commonly affected by brown tumours in primary HPT[6]. Oral radiographic manifestations of HPT include a generalized loss of lamina dura surrounding the roots of the teeth, loss of cortication around the inferior alveolar canal and maxillary sinus and of the trabecular pattern of the jaws [6]. Peripheral manifestation of brown tumour on the oral cavity is rare. As it can be observed in the present case, the clinical appearance simulates peripheral giant cell granuloma.

The term brown tumour comes from the colour of lesion, which results from the vascularity, haemorrhage and deposits of hemosiderin[7]. Therefore, the brown tumour is actually a kind of giant cell lesion and often appears as a multiple and expansive osteolytic lesion of the bone. Because it is difficult to distinguish histopathologically brown tumour from other giant cell lesions, a clinical diagnosis is made based on the association with HPT[8]. Histologically, all giant cell lesions have two main components: Mononuclear stromal cells and multinucleated giant cells.

There is no single test that establishes the diagnosis of secondary HPT. However, the diagnosis is made based on the high level of PTH associated with low or normal serum calcium[8]. In the present case, the patient presented with chronic progressive renal failure together with PTH level 5 times higher than normal and did not show alteration in calcium level.

The treatment for brown tumour in the jaws includes enucleation and curettage, radical resection and reconstruction, radiation therapy and chemotherapy[9]. Surgical excision of the brown tumours is indicated for large and disfiguring lesions and in case that the affected bone is weakened[10]. However, initial treatment of this lesion was done with systemic corticosteroids and, when it reduces its size, they make its surgical excision[5,11,12]. In conclusion, the present report describes a rare case of peripheral brown tumour associated with secondary HPT simulating a peripheral giant cell lesion of the jaw, which was surgically resected.

In brown tumors, however, there is a combination of osteoblastic and osteoclastic activity. Brown tumors are mainly the result of secondary hyperparathyroidism in patients with renal insufficiency, but they have also been described as a rare manifestation of calcium malabsorption and some forms of osteomalacia[4-6]. Brown tumors as a manifestation of primary hyperparathyroidism are extremely rare. In such cases, the primary hyperparathyroidism usually results from the overproduction of parathyroid hormone by a parathyroid tumor[7-9]. In our patient, the histological findings were indicative of a giant cell bone expanding lesion with brown tumor being the most probable diagnosis.

CONCLUSION

Primary or secondary HPT may be recognized by the presence of an osteolytic lesion with giant cells, a condition referred as "brown tumor". The most useful therapy for patients with brown tumors is surgical excision of bone lesions and therapy (surgical or medical) for primary or secondary HPT.

SIGNIFICANCE STATEMENT

Brown tumor may be the first clinical sign of hyperparathyroidism. It can manifest at any age and though more common in females, it can also affect the males. This study will help the researchers to uncover the critical areas of brown tumor, regarding the etiopathogenesis with regards to age, gender, site, serum calcium levels, that many researchers were not able to explore. Thus a new theory that, "whenever a round, radiolucent and bone expanding lesion with hyperparathyroidism is presented, a brown tumor must be considered to be the most likely diagnosis" may be arrived at.

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