Unusual Presentation of Brown Tumor in Lateral Malleolus And Talus, A Case Report

Lateral Malleol ve Talusta Olağandışı Yerleşim Gösteren Brown Tümörü, Vaka Sunumu

Brown Tümör / Brown Tumor

Abstract
Excess production and secretion of parathormone (PTH) from parathyroid glands causes primary hyperparathyroidism (PHPT). Orthopaedic surgeons should be aware of skeletal manifestations, laboratory abnormalities, and the treatment options of hyperparathyroidism (HPT). Elevated serum calcium or pathognomonic findings of HPT on plain radiographs should alert the orthopaedic surgeon. In these cases serum intact PTH and additional diagnostic tools should be obtained for proper diagnosis. We report a 43 year-old patient with PHPT who developed two Brown Tumors including one at the talus and second in the lateral malleolus. The present case is the first report of Brown Tumor of the lateral malleolus and talus in the literature. In addition we reviewed literature pertaining to HPT, from orthopaedic surgeons' aspect.

Keywords
Brown Tumor; Parathormone; Hyperparathyroidism

Özet

Anahtar Kelimeler
Brown Tümör; Parathormon; Hiperparatiroidizm

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Introduction
Excess production and secretion of the parathormone (PTH) from the parathyroid glands causes primary hyperparathyroidism (PHPT) which is caused by adenoma or hyperplasia of the parathyroid gland [1]. Von Recklinghausen was the first to describe the characteristic bone disease of hyperparathyroidism (HPT). The Brown Tumors develop mainly in the facial bones, pelvis, ribs, and femur, and can be multifocal [2]. In this case we report a patient with PHPT who developed two Brown Tumors including one at the talus and the second in the lateral malleolus. To our knowledge, the present case is the first report of a Brown Tumor in the lateral malleolus and talus.

Case Report
A 43 year-old woman reported a gradual onset of pain in her left ankle lasting for 5 months. After evaluations such as plain radiographs and magnetic resonance imaging (MRI), an excisional biopsy was performed before. After one month, the patient described some pain and discomfort in her ankle. Also, there was no swelling, erythema or tenderness in the area of the previous incision. Range of motion (ROM), stability, strength, and neurovascular examination were normal in the ankle joint.

The patient was otherwise healthy except for a history of pulmonary tuberculosis, which, according to the patient, had no further complications.

After a physical examination, evaluations such as plain radiographs, and a Tc-99 bone scan were completed. Laboratory tests were normal except for calcium, phosphorous and alkaline phosphatase levels. The calcium, at 11mg/dl, was high (8.1-10.4 mg/dl), the phosphorous was slightly low at 2.3 mg/dl (2.5-5.0 mg/dl) and alkaline phosphatase was reported at 250 U/L (28-125 U/L).

AP and lateral views of the cruris showed a mildly expansile lytic lesion at the distal part of the fibula close to the lateral malleolus (Figure 1). Although the margins of the lesion were slightly irregular, the border between the native bone and the lytic lesion could be easily depicted. Sagittal T1 and axial T2 weighted MRI images revealed another lesion in the talus with the signal intensity identical to the fibular lesion seen on the radiography (Figure 2). Lesions had well-defined margins but were expanding to the peripheral soft tissues by destroying the cortex. They did not show enhancement after contrast administration, but marked perifocal enhancement was present (Figure 3).

A Tc-99 bone scan demonstrated increased uptake on the distal fibula, talus and 4th -7th ribs.

The histologic study revealed that in the osseous sections multinucleated giant cells which are distributed in the stroma consisting of fusiform cells showing clusters and swirls are observed. Osteoid formations, some of which circumscribed with osteoblastic rim and pigmented histiocytes were seen in the focal stromal areas (Figure 4). Because of the histologic findings, the differential diagnosis was modified to Brown Tumor of HPT (osteitis fibrosa cystica) and giant cell tumor of the bone. A serum intact PTH was ordered, and was reported as 973 pg/mL (12-72 pg/mL). After this finding, the diagnosis was confirmed as Brown Tumor of HPT. Tc 99m MIBI scans of the parathyroid glands were performed and revealed a parathyroid adenoma.
located in the inferior right side of the thyroid gland, a multinodular guatr and autonomous hyperactive nodule of the thyroid gland.

Discussion

PHPT is a relatively common disease. However, the frequency of Brown Tumor, pathognomonic skeletal form in this disease, is declining. Primary or secondary bone lesions characterized by destructive process should be differentiated from Brown Tumor of HPT [3]. The incidence of Brown Tumor is 3% in PHPT. In secondary HPT, the incidence of Brown Tumor is 1.5% to 1.7% [3,4].

In recent years, the most commonly seen skeletal manifestation of HPT is simple diffuse osteopenia, resembling osteoporosis. The reason for the changing pattern of skeletal involvement is unknown [5]. Although often the symptoms of osteitis fibrosa cystica are severe, the affected bone undergoes extensive remineralization and healing after the removal of the parathyroid adenoma [5].

In HPT, the histologic findings of the affected bone demonstrate great variations. The abnormalities of the bone include osteitis fibrosa cystica (Brown Tumor) with replacement of marrow elements by vascular fibrous tissue. The other abnormalities include osteoporosis and osteomalacia [6].

In HPT, most studies indicate cortical loss rather than trabecular loss [7,8]. PTH seems to be catabolic at cortical sites and may have anabolic effects at cancellous sites of the bone. In some patients, cancellous bone density of the lumbar spine can be markedly reduced [9].

In HPT, there is a process of either bone resorption or bone formation and bone resorption is usually a dominant factor. Bone resorption is generally periaricular and is classified as subperiosteal subchondral, trabecular, endosteal, intracortical, subligamentous and subendinous. Subperiosteal resorption is pathognomonic sign of HPT. It may be seen at any part of the body but the commonly involved parts are hands and feet [8]. The second mostly affected body part is the skull. Trabecular resorption produces a characteristic salt and paper appearance in the diploic space of the skull. Other sites of subperiosteal resorption include the medial aspects of the proximal tibia, femur and humerus [6,8].

Bone resorption in subchondral locations are mostly in the major articulations in the axial skeleton, particularly the sacroiliac joints, sternoclavicular joints, symphysis pubis and discovertebral junction [10].

In several studies, differential effects of the PTH on cancellous and cortical bone have been evaluated. It was found that the PTH affects mostly the cortical bone and the most significant postoperative improvements in bone mineral density occurs in cancellous bone [2,11].

Brown Tumors’ histopathological diagnosis include; extravasated blood cells areas of hemorrhage, histiocytosis including hemosiderin, trabeculation of unmineralized new bone and a mixture of osteoblasts, mononuclear cells and multiloculated giant cells [3].

With current patient, plain radiographs and MRI showed an expansile cystic lesion on the lateral malleolus and talus, and an open biopsy was performed at an outside institution. One month after this procedure, the patient visited the authors’ institution. A parathyroid adenoma, multinodular guatr and an autonomous hyperactive nodule on the thyroid gland was diagnosed. Total excision of the parathyroid adenoma, and right total and left subtotal thyroidectomy were performed on the patient. We performed curretage and grafting for the lesions in the talus and lateral malleolus in the same session.

2 years after surgery, the patient was doing well and had fully recovered. Repeat radiographs of the ankle joint showed full healing. Labarotory values showed a normal serum calcium and intact PTH levels. The patient was able to move her ankle joint with no stiffness and had normal sensation and normal function.

Competing interests

The authors declare that they have no competing interests.

References


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