Intraosseous calcaneal lipoma with subtalar perforation through cystic degeneration

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Intraosseous lipoma is a benign tumor that originates from proliferating mature lipocytes. It often occurs in the metaphysis of long bones of the lower extremity, and also in the calcaneus, humerus, mandible, sacrum and rib bones. Frequently, it involutes spontaneously through a process of infarction, calcification and cyst formation. It can either present as pain or be asymptomatic and only discovered through an incidental radiological finding. In our case, the patient presented to us with heel pain. Intraoperatively, it was found that the intraosseous cavity was filled with fat along with an adjacent but separate area of cystic degeneration. There was also a cortical perforation at the cystic lesion which was communicating to the subtalar joint. This cortical breach is most likely the cause of the subtalar pain experienced by our patient, and such pathological fracture due to intraosseous lipoma has never been reported before.

Key words: intraosseous lipoma, calcaneal lipoma.

Intraosseous lipomas are rare benign lesions that account for about 0.1-2.5% of all bone tumors. It is, however, the most common lipogenic tumor in bone. It can present in an extremely wide age range (5 to 85 years) although the peak age at discovery is usually in the fourth to fifth decades of life. There may be a slight male predilection [1].

Case Report

A 51 years old female presented to our clinic with right heel pain for the past 8 months. The pain was insidious in onset on the lateral aspect of heel and was described as dull aching type of pain. Initially, it was aggravated only by walking. However, this time the patient complained of pain during rest, which had begun to progress significantly within the last month. There was no diurnal variation in the pain. There was no history of trauma or any systemic symptoms. On examination, the skin over the heel appeared to be normal. There was tenderness presented on the sinus tarsi area without any swelling or signs of inflammation. There was full flexion and extension of ankle joint, which also appeared to be pain free. However, inversion and eversion of the foot was restricted due to pain. The hematological and biochemical studies did not show any abnormality.
Figure 1 Axial radiologic view showing cystic degeneration of calcaneus.

Figure 2 Lateral view of the calcaneus showing superior and anterior extension of cystic change up to the subtalar joint and calcaneocuboid joint respectively.

Figure 3 MRI showing T1-(top) and T2-(middle) weighted image in sagittal and coronal plane (bottom).
An axial radiographic view of the ankle showed three well-defined cystic lesions in the body of the calcaneus. One was near the lateral aspect of the calcaneus while the other two were just above it divided by septa. There was no reactive sclerosis around the cystic wall (Figure 1). On the lateral view, a large cystic lesion was presented over the calcaneal neutral triangle just below the subtalar joint extending the calcaneocuboid joint (Figure 2).

An MRI of the right foot suggested a well-margined mass in the right calcaneus with high signal intensity on T1-weighted image. There were two cystic lesions inside the mass which showed low signal intensity on the fat suppression image (Figure 3).

The patient was scheduled for the curettage of the cystic lesion and bone grafting. A well-padded tourniquet was placed on the proximal aspect of the right thigh, and a lateral approach was used with the patient in the left lateral decubitus position. Intraoperatively, the center of the lesion was found to be exactly in between the two peroneal tendons (Figure 4). A window was made under fluoroscopic guidance over the lateral calcaneal wall.

Intraosseous deposition of fat almost in a liquefied state extending all the way up to the calcaneocuboid joint was found inside the cyst. Superiorly, the cavity extended up to the subtalar joint, causing a pathological fracture and breakage of calcaneal articular surface (Figure 5). Once the fat was curetted, a cystic cavity was seen exactly over the window touching the medial aspect of the calcaneal wall and two more cavities were found on the posterior aspect of the calcaneus, which was separated by a septum.

All cysts were curetted, the walls were cauterized and the margin of the cysts was debrided with a diamond burr. Following a thorough irrigation, the cavity was packed with cancellous allograft and the window was fixed with two 2.7 mm screws (Figure 6). The wound was closed and a short leg splint was applied. The tissues taken out from the bone was sent for the pathologic determination and the report came back consistent with that of lipoma. The patient was allowed to do partial weight-bearing exercise with the splint for four weeks and then full-weight bearing was tolerated thereafter.
At 15 months postoperatively, the patient was symptom-free and complete bony union was identified on radiographs. All devices were removed without any associated complications (Figure 7).

Discussion

Intraosseous lipoma has an incidence of 0.1-2.5% of all primary bone tumors [2]. The etiology of the tumor still remains unknown. Three theories have been put forward: 1) traumatic origin and later fat degeneration, 2) infection or osseous fat infarction with metaplasia, 3) primary tumor of marrow fat. The third theory seems to be the most probable one [3].

Intraosseous lipoma has no gender predilection and can occur at any age, most commonly occurring in the fourth decade [3]. It affects long bones of the lower extremity more frequently than the upper extremity by 6:1 ratio. The most common occurrence is in the calcaneus, which is only 8%, followed by the skull, jaw and then ribs [4-7]. Clinical presentation varies significantly, leaving almost two thirds of the patient asymptomatic. The most frequent symptom is calcaneal pain, which is often related to prolonged standing or exercise [3]. When asymptomatic, most of the diagnoses are incidental.

Radiologically, intraosseous lipoma is characterized by cystic, radiolucent lesion with thin, sclerotic and well-defined borders. In long bones, the lesion is more expansile without any periosteal reaction or cortical breach in the metaphysis or epiphysis of the bones. In short tubular bones, the lesion shows a geographic
pattern with sclerotic rim. In the calcaneus, it appears as a radiolucent cystic image with sclerotic borders and a central calcification (Bull’s eye image), which is almost always located in the neutral triangle at the base of the neck of calcaneus whereas bone infarcts occur in the dorsal part of calcaneus [8,9]. Milgram [5,9] and Chow [2] classified a lipoma into three separate categories: Stage 1 contains mature lipocytes without necrosis, and osteolytic lesions are seen radiologically. Stage 2 contains partial fat necrosis (due to pressure on capillaries) with focal calcification, live lipocytes still present, and radiolucent lesion with central calcification seen on radiograph. Stage 3 contains extensive fat necrosis with variable degrees of cysts formation, calcifications and formations of reactive bone.

Computed tomography demonstrates a well-defined lytic lesion with negative Hounsfield unit equivalent to those of fat. MRI shows high signal intensity on T1- and T2-weighted images similar to that of subcutaneous fat. Thus, surgical biopsy for the diagnosis of such tumors is not significant [11,12].

Differential diagnosis includes solitary bone cyst, fibrous bone infarct, enchondroma, fibroma, aneurismal bone cyst, chondrosarcoma, osteoid osteoma, liposarcoma and eosinophilic granuloma. Solitary bone cyst occurs at the base of the calcaneal neck but is typically seen in adolescence. Aneurysmal bone cyst is extremely rare before the age of five and after the age of thirty. In the case of liposarcoma, CT scan shows less homogenous lesion without typical negative Hounsfield units [1]. Enchondroma typically shows endosteal scalloping and calcification that ranges from punctuate to rim type lesions. Osteoid osteoma is characterized by the typical night pain and dense sclerotic central nidus on the radiographic images. Bone infarct in the calcaneus is always seen at the dorsal aspect, with calcification usually seen in the periphery. On the contrary, there is almost always a central calcification in lipoma [8,9].

The treatment most frequently used in consists of debridement of the lesion through an ample bone window and includes filling of the defect with autologous bone which consist of a frozen, dried allograft of hydroxyapatite or polymethylmethacrylate cement.

Malignant transformation of pre-existing bone lipoma in the femur and tibia has been reported but there have been no studies on calcaneal lipoma [13]. Pathological fracture or cortical perforation has not been reported to date but in this case, cortical perforation opening into the subtalar joint was one of the intraoperative findings. This pathological fracture might be the reason for the heel pain.

In summary, calcaneal intraosseous lipoma with cystic degeneration is a very infrequent benign tumor. Such lesions can be asymptomatic or may present with heel pain. Diagnosis is usually made incidentally on radiographic images, and CT or MRI scans are used to confirm the fatty nature of the lipoma. This condition has a very good prognosis and its symptoms improve with rest and the use of analgesics. Long standing cases may complicate into pathological fractures and become symptomatic. At the very least, these cases require surgical intervention at the earliest.

Conclusion

Intraosseous lipoma has been thought of as an infrequent occurring benign tumor. In the past, pathological fracture secondary to intra-osseous calcaneal lipoma has never been reported most likely because the tumor occurs in the non-weight bearing areas of the calcaneus such as the neutral triangle [5]. In this case, a cortical perforation through the cystic degeneration of the tumor was identified, which merits special attention.

References