Intraosseous lipoma of the calcaneus: A rare case report

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ABSTRACT

Aim: The aim of this study is the presentation of a rare case of an intraosseous lipoma of the calcaneus in a middle-aged woman, the therapeutic surgical treatment and the post-operative results.

Material and Methods: A 49 years old female patient presented with pain in the area of the calcaneus for the past three years, localized at the plantar region. The patient had no previous history of trauma, and described the pain as progressively increasing. Furthermore, she reported tenderness at the calcaneus region during the clinical examination and soft tissue swelling. The patient was subjected to a routine radiographic control of the foot (anteroposterior and lateral views) and a computed tomography (CT) scan of the calcaneus.

Results: The CT scan showed an osteolytic lesion with density equal to that of adipose tissue, marginal sclerosis without cortical breakthrough and a central nidus of calcification, giving the diagnosis of an intraosseous lipoma of the calcaneus. The patient underwent curettage, surgical debridement and bone grafting of the lesion in the operating room. The histological findings included the presence of fatty tissue with various areas of fat necrosis, consistent with a grade 2 lesion according to Milgram. Three months postoperatively the bone graft had been fully incorporated and the heel pain had resolved.

Conclusion: Intraosseous lipoma is one of the rarest benign primary bone tumors. The lesions are often asymptomatic, but if symptoms occur, as in our case, mild pain and swelling are described. The potential of the lesion for a pathological fracture made the surgical intervention necessary with excellent short- and medium-term functional and roentengraphic results.

KEY WORDS: Intraosseous lipoma, calcaneus, bone grafting
Introduction
Intraosseous lipomas are benign lesions, accounting for less than 0.1% of all bone tumors [1]. They are composed mainly of mature adipocytes and atrophic bone trabeculae and may contain varying areas of necrosis [2]. They are mainly found in the lower limb, with the calcaneus being involved in approximately 30% of the cases [3]. In this report we present a case of this rare tumor, the therapeutic treatment used and the short- and medium-term post-operative results.

Case report
A 49-year-old female patient visited our outpatient department complaining of left heel pain, localized at the plantar region. The pain had started approximately 3 years before and was progressively getting worse, making her unable to perform her everyday work. She reported no previous history of trauma in the aforementioned area. The clinical examination revealed tenderness at the region of the calcaneus with concomitant soft tissue edema. The patient had no familial history of malignancy, was a recreational smoker and was receiving tibolone and paroxetine for the last 3 years. She was subjected to routine anteroposterior and lateral radiographs and a computed tomography (CT) scan of the calcaneus. The radiographs showed a 3.2cm x 2.2cm osteolytic lesion of the calcaneus with well-defined borders containing a central sclerotic nidus with a diameter of 5mm (Fig. 1). The CT scan revealed sclerotic margins demarcating the lesion from the surrounding healthy bone tissue, without any signs of cortical perforation. The density within the lesion was consistent with adipose tissue, which led to the diagnosis of an intraosseous lipoma (Fig. 2). Due to the duration of the symptoms and the risk of a pathological fracture the patient underwent curettage and surgical debridement of the lesion, which was subsequently packed with heterologous bone grafts. The histological examination revealed the presence of adipose tissue with varying areas of necrosis. Three months postoperatively the graft had been fully incorporated and the patient was able to full weight bear without pain (Fig. 3). Eight years postoperatively the patient continues to be free of symptoms.

Discussion
Intraosseous lipomas are considered to be amongst the rarest primary bone tumors with an incidence of approximately 0.1% [1]. They are most commonly found in the lower limb (72%), and especially in the os calcis (30%) and femur (20%) [3]. Other sites include the fibula, the upper limb, the mandible, the spine and the pelvis [4-6]. They are mainly observed between the ages of 40 and 60 and show a slight male predominance [2]. They usually present with pain, which is extravagated by walking, tenderness and soft tissue edema [3]. Pathological fractures have also been reported [7]. However, almost one third of the cases is asymptomatic and is diagnosed incidentally on radiographs performed for other pathologic modalities. This has led many researchers to believe that intraosseous lipomas are actually not so rare, as many cases are undiagnosed due to lack of symptoms [8].
The pathogenesis of this tumor is poorly understood and certain theories have been proposed. One hypothesis suggests that these tumors arise as a result of bone reaction secondary to trauma [9]. However, less than 10% of the patients report injury and more than 30% of the cases are asymptomatic. Another hypothesis suggests that lipomas arise during the healing of a bone infarct. This is possible, although unlikely, as calcification in bone infarcts is located usually in the periphery, in contrast to lipomas where it is always found centrally [10]. Also up to 50% of lipomas show bone expansion, which is impossible for infarctions, as they cannot expand beyond their original location [11]. Last but not least many researchers believe bone lipomas to be true primary benign tumors, which seem to be the most plausible explanation [9].

The lesion usually appears on plain radiographs as a well circumscribed osteolytic lesion with a sclerotic rim which may contain a central nidus of calcification [9]. In CT scans the lesion displays negative Hounsfield units (ranging for -110 to -40) equivalent to those of fat [12]. Bone expansion is frequently seen and areas of pathological fractures may be noted [8]. Magnetic resonance imaging (MRI) studies demonstrate high signal intensity in both T1-weighted and T2-weighted images, which becomes extinguished in fat-suppression sequences [13]. The sclerotic margins and the central areas of calcification appear with a low intensity signal in both T1- and T2-weighted images. Fluid filled cavities are found in over 60% of the cases and appear with a low to medium signal intensity in T1-weighted and a very high signal intensity in T2-weighted images. No contrast enhancement is observed within the lesions [8].

Milgram has categorized intraosseous lipomas in 3 stages according to radiological and histological findings [2]. Stage 1 is characterized by pure osteolytic lesions containing viable adipocytes intermingled with bony trabeculae which appear thin due to pressure atrophy. Stage 2 lesions appear as osteolytic areas containing sites of increased density due to central calcification and ossification. Histologically both viable adipocytes as well as areas of necrosis and calcification are observed. Stage 3 lesions are characterized by a reactive ossified rim and contain central cysts and calcified areas. Histologically extensive fat necrosis is seen throughout the specimen.

The differential diagnosis of an intraosseous lipoma is wide and depends on the stage of the disease. In stage 1 one should consider other benign osteolytic lesions like pseudocysts or simple bone cysts [14]. Even...
though these lesions appear similar in plain radiographs, studies with CT or MRI can quite accurately differentiate between them, making surgical biopsy usually unnecessary [15]. Stage 2 and 3 lesions can appear similar to both benign and malignant conditions. Benign ones include bone infarcts, enchondromas, non-ossifying fibromas, giant-cell bone tumors, chondromyxoid fibromas and fibrous dysplasia, while malignant ones include mainly liposarcoma and malignant fibrous histiocytoma (MFH) [7,16]. Even though malignant lesions usually appear less homogenous in CT or MRI studies, biopsy is many times needed in order to establish the correct diagnosis [9].

As a general rule asymptomatic lesions are best left untreated with a close follow-up, as a number of them have been shown to regress spontaneously [17,18]. Care though has to be taken when they affect weight-bearing areas in order to avoid a pathological fracture [19]. Symptomatic lesions on the other hand are usually treated with an extensive curettage and a tight bone grafting [20]. Both homologous and heterologous grafts have been used with equal great results [9,21]. Recurrence rate after surgical excision is unknown, but it appears to be extremely low [3].

Malignant transformation of intraosseous lipomas is a matter for discussion. Milgram is the only one having published 4 cases, in which intraosseous lipomas were transformed to either MFH or liposarcoma [22]. Given the fact that bone infarcts, which as mentioned before mimic intraosseous lipomas, have a potential for transformation to tumors of mesenchymal origin, one should consider Milgram’s report with skepticism [23,24]. Were those cases true malignant transformation, or were they actually misdiagnosed bony infarcts? This question remains to be answered.

**Conclusion**

Intraosseous lipomas are rare benign tumors usually found in the lower limbs. Their diagnosis can be made quite accurately using CT or MRI studies and surgical treatment with curettage and bone grafting is usually reserved for symptomatic or suspicious lesions with excellent postoperative results. Whether these lesions have any malignant potential remains still a mystery.

**Conflict of interest**

The authors declare no conflicts of interest.

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