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A rare case of intraosseous lipoma of distal tibia – A case report and review of literature

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ABSTRACT

Introduction: Intraosseous lipoma is a very rare neoplasm accounting for < 0.1 % of primary bone tumors. Pain is the leading symptom in majority of the reported cases but it can be asymptomatic. There is slight male predominance in occurrence of this lesion. The plain radiological findings are not specific and requires differential diagnosis. The lesion is mostly diagnosed by histopathological examination. **Case report:** A 31-year-old male presented with localized pain around the medial aspect of right ankle joint for one week. Examination revealed mild tenderness over the distal part of the right tibia over the medial aspect. Plain radiograph of the right leg showed well-defined expansile osteolytic lesion with sclerotic rim and calcified matrix at metaphysis of distal tibia. MRI showed enhancing T1-weighted hypo intense and T2-weighted hyperintense lesion. For this case, radiological impression was giant cell tumor with differential diagnosis of aneurysmal bone cyst and fibrous dysplasia. However, the histopathological examination showed intraosseous lipoma, consistent with stage II of Milgram's classification. **Conclusions:** Although the diagnosis of intraosseous lipoma can be very challenging due to its rarity and indistinct plain radiograph findings, combination of computed tomography or magnetic resonance imaging may be useful by being able to show the presence of fat within the lesion. However, the clinicians, surgeons and radiologist should be familiar and be aware of these findings to be able to come to a correct diagnosis since not all cases need surgery and can be managed conservatively.

Keywords: Benign; Case report; Distal tibia; Intraosseous lipoma.

INTRODUCTION

Intraosseous lipoma is a very rare neoplasm accounting for < 0.1 % of primary bone tumors[1-3]. Pain is the leading symptoms in majority of the reported cases[3-5]. However, 30% of the cases are known to be asymptomatic and are detected incidentally while investigating for other unrelated reasons[3]. It has no gender or ethnic predilection but some reports found slight male predominance[1,3-6]. It frequently occurs in lower limb[4], particularly in os calcis, femur and fibula in decreasing order[4,7]. It is especially very rare in distal tibia with only few reported cases[2,7].

Radiographically, the intraosseous lipoma presents as a well-defined expanding lucent lesion with (or without) central calcification[8] but the findings are not very specific for this entity and such findings can mimic giant cell tumor (GCT), aneurysmal bone cyst (ABC), simple bone cyst (SBC) and fibrous dysplasia among others. Due to its rarity and non-specific clinical and radiological features, it is frequently misdiagnosed. The definite diagnosis of this entity is mostly made by histopathological examination.

There is dichotomy of therapeutic options in the management of intraosseous lipoma. Surgical management consists of curettage and packing either with autologous bone graft or allograft or synthetic bone substitute[9]. However, if there is no suspicious feature of malignancy or pathological fracture, patients can be managed conservatively with clinical and radiological follow up[9,10].

Herein, we report a rare case of intraosseous lipoma at unusual location, which was not clinically suspected but was diagnosed by histopathological examination. The main aim of this report is to create awareness among the clinician about the occurrence of this rare lesion in our clinical practice. We also aimed to discuss the clinical and radiological findings and the preferred management of this lesion based on literature.

CASE REPORT

A 31-year-old male presented with localized mild pain (VAS 3 - Visual Analogue Scale 3) around right ankle joint for one week duration. He denied any history of trauma to the site and he did not have any constitutional symptoms or night pain. His past medical and surgical history were unremarkable. Local examination revealed mild tenderness over the distal part of the right tibia over the medial aspect. There was no swelling or erythema or deformity and the range of motion at ankle and knee joints were normal. His

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general and systemic examination was unremarkable. His blood investigations including serum uric acid level were all within normal range.

Plain radiograph of the right leg showed well-defined expansile osteolytic lesion with sclerotic rim and calcified matrix at metaphysis of distal tibia (Figure 1a). No fracture or peri-osteal reaction or soft tissue swelling or bony destruction was noted. Magnetic resonance imaging (MRI) of the right ankle joint showed enhancing T1-weighted hypo intense (lesion appearing dark on MRI) and T2-weighted hyperintense (lesion appearing white on MRI) lesion measuring 4x2.1cm (Figure 1B). T1 and T2 are technical terms used in different MRI imaging methods. T1 and T2 refers to the time taken between magnetic pulses and the image taken. T1 weighted has Short TR (Repetition time) and Short TE (Time to Echo) whereas T2 weighted has long TR and TE. The radiological impression was GCT with differential diagnoses of ABC and fibrous dysplasia.

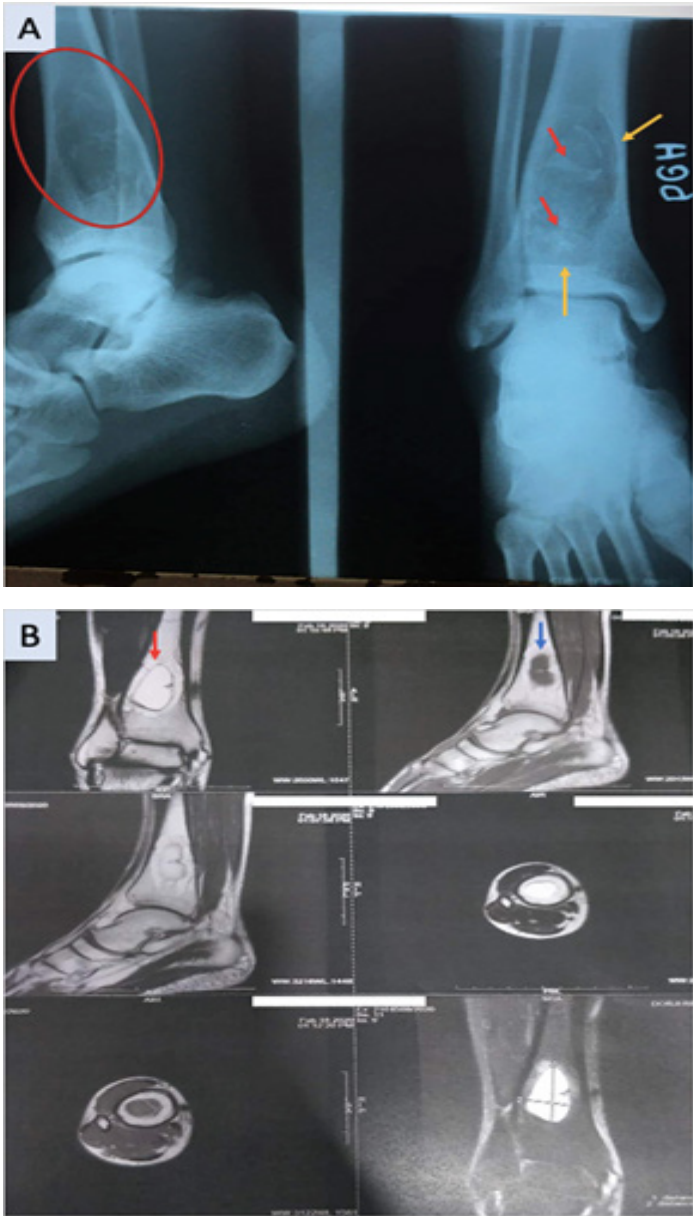

Figure 1. a) Plain radiograph of the right leg showing well-defined expansile osteolytic lesion (red circle, lateral-view) with sclerotic rim (yellow arrow, AP-view) and calcified matrix (red arrow, AP-iew) at metaphysis of distal tibia;
b) MRI showing enhancing T1-WT-hypointense (blue arrow) and T2-WT-hyperintense lesion (red arrow).

Patient was planned for extended curettage with the preoperative diagnosis of GCT of right distal tibia. Through medial longitudinal incision, a bone window was created from medial cortex using drill bit (Figure 2A) and the tissue content (Figure 2B) from the lesion was removed completely and sent for histopathological examination. The cavity was then filled with bone cement (poly methyl methacrylate) and it was covered with the same cortical bone.

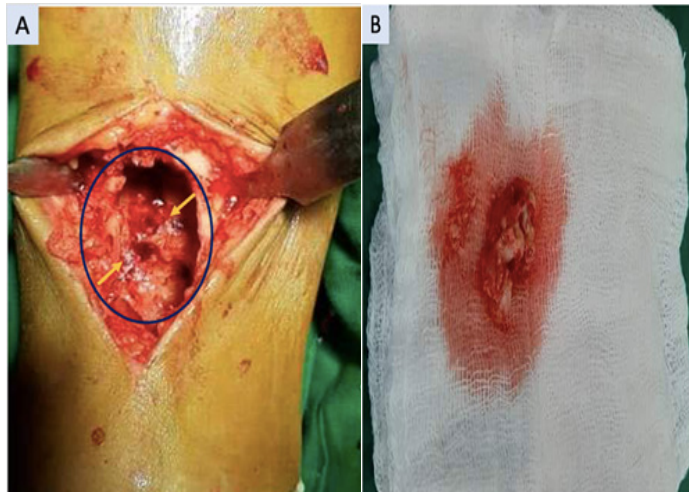

Figure 2. a) Intramedullary lesion (yellow arrow) as seen from bone window (blue circle);
b) Gross image of intramedullary lesion

Macroscopic examination of the curettage specimen showed pieces of lobulated adipose tissue admixed with small bone fragments, measuring 4x2x1.5 cm in aggregation.

Microscopically, the lesion showed lobules of mature-appearing adipocytes replacing the marrow and focally encasing the bony trabeculae (Figure 3a). One of the fragments showed a thin fibrous capsule (Figure 3b). Areas of dystrophic calcification (Figure 3c) and focal fat necrosis were noted. No reactive new bone formation or sclerosis was present. These findings were consistent with intraosseous lipoma, stage II of Milgram's classification.

The immediate post-operative period was uneventful. Patient was discharged after 3 days and was followed up in 3 months at orthopaedic-OPD for clinical assessment, which was unremarkable. Patient was then followed up after 1 year with plain radiograph, which showed no evidence of recurrence (Figure 4a-b).

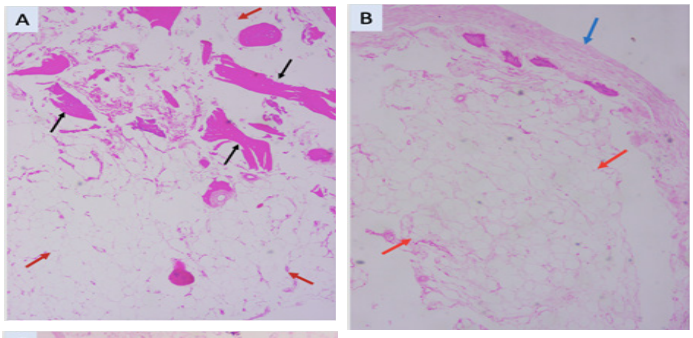

Figure 3. a) Microscopic images showing viable mature adipocytes (red arrows) replacing the marrow and encasing the bony trabeculae (black arrows) (H&E; 40X).
b) Presence of thin fibrous capsule (blue arrow) (H&E; 40X).
c) Dystrophic calcification (green arrow) (H&E; 40X)

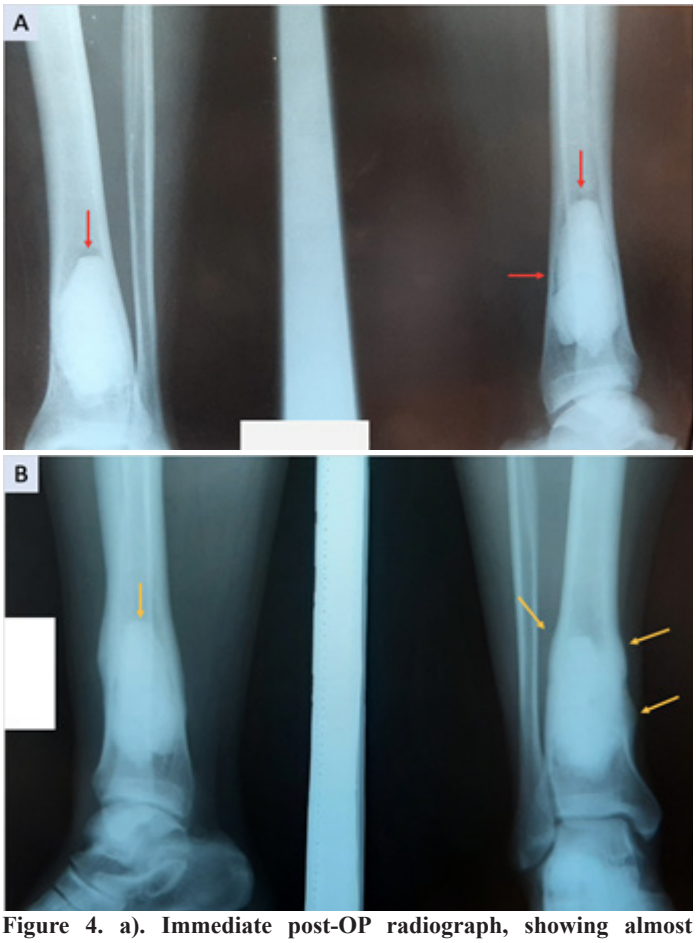

Figure 4. a). Immediate post-OP radiograph, showing almost complete filling of the cavity by bone cement. A small amount of radiolucency (red arrow) is seen at the superior part of bone cement. b). Post-Op-one-year radiograph, showing healing, represented by complete consolidation of the cavity (with callus formation, yellow arrow).

DISCUSSION

Intraosseous lipoma is a considered as a rare benign bone lesion accounting for <0.1 % of all primary bone tumors[1-3]. Although, it can occur at any age, the highest incidence is reported to be in 4th, 5th and 6th decades of life[3]. Males are more frequently affected than the females[1,3-6] but some reports showed no sex predilection[3].

Plain radiograph findings can be non-specific but majority (75%) of the cases present as a cystic lesion with increased radiolucency and surrounded by sclerotic rim. Foci of central calcification can be appreciated, especially in the lesion corresponding to stage II and III of Milgram classification[1,4,11]. The computed tomography (CT) and MRI are helpful in detection of fat within the lesion, allowing for more precise diagnosis and morphological evaluation [2,11]. The adipose tissue is known to have low accentuation coefficient in CT[11]. The adipose tissue signal in both T1- and T2 weighted images of fast spin-echo sequences of MRI is known to be high, which becomes extinguished in fat-suppression sequences[11]. In addition, the CT and MRI are useful in extensive evaluation the lesion to rule out malignancy. Though intraosseous lipoma is a benign lesion, malignant transformation has been reported[12].

Histologically, intraosseous lipoma is composed of mature adipose tissue devoid of hematopoietic elements with (or without) variable areas of fat necrosis and calcification[2]. The pathogenesis of this lesion is controversial and has not been clarified yet. However, there are several postulated hypothesis including secondary bone reaction to trauma, healing reaction of osteonecrosis and primary benign tumor[1,3,13-15].

In 1988, Milgram et al postulated this lesion into 3 stages based on degree of involution in histological examination[1]. Stage I consists of viable lipocytes. Stage II has partial fat necrosis and focal calcification with regions of viable lipocytes. Stage III has complete fat necrosis with variable degree of calcification, cyst formation and reactive new bone formation. Our patient had mostly viable mature adipocytes with focal fat necrosis and dystrophic calcification (Figure 3c), which corresponded to stage II of Milgram classification. The nature of intraosseous lipoma is still controversial without any adequate genesis on this lesion. Few studies found similar genetic findings of the soft tissue lipoma and parosteal lipoma, including the translocation of t (3;12)(q28; q14) [2,16,17].

Patient with intraosseous lipoma can be symptomatic or asymptomatic. According to the literature, 70% of the patients were found to be symptomatic with pain[3,4]. This lesion can be found anywhere within the skeleton but lower limb accounts for majority of the cases (71%), followed by spine and pelvis (12%), upper limb (7%) skull and mandible (7%) and ribs (2.5%) (4, 18). In the lower limb, calcaneum (32%) and femur (20%) accounts for the majority the cases[4,18] and it is rare to occur in distal tibia with very few reported cases. It is usually a solitary lesion but multiple site involvement has also been reported[1,11].

Our patient presented with mild pain and discomfort around the medial aspect of his right ankle joint for one week.

Plain radiography of his right leg showed a well-defined expansile osteolytic lesion with sclerotic rim and calcified matrix at metaphysis of distal tibia. MRI showed enhancing T1-weighted hypo intense and T2-weighted hyperintense lesion at distal tibia. Although it may be very difficult to diagnose intraosseous lipoma from plain radiographs due to its extensive differential diagnoses but combination with either CT or MRI was found to be useful in detection of fat within the lesion allowing for more precise diagnosis[2].

Due to rarity in its incidence, the pre-operative diagnosis of intraosseous lipoma can be very challenging especially if we are not familiar with this entity and its manifestations. Moreover, it has a very indistinct clinical and radiological features. Nevertheless, the clinician, surgeon, radiologist and pathologist should be aware of this lesion during their clinical practice and get familiarized with its clinical, radiological and pathological features for the correct diagnosis.

In our case, patient presented with mild pain and discomfort around the medial aspect of his right ankle joint for one week, for which he was investigated with plain radiograph and MRI.

Although radiological features were similar to previous reports, we failed to provide correct diagnosis pre-operatively. The patient was treated with extended curettage with provisional diagnosis as GCT. The histopathological diagnosis was consistent with intraosseous lipoma. Both GCT and intraosseous lipoma (when surgery is needed) have similar approach of treatment, i.e. with curettage. The lesson we learn from this case is to consider the rare lesions in differential diagnoses and the importance of pre-operative biopsy in uncertain cases on clinical presentation. Final histopathological examination will aid in confirming the diagnosis. The need for surgery is controversial. However, most of the cases with intraosseous lipoma are managed conservatively with closer follow up[7]. Surgery for intraosseous lipoma is usually indicated if there is a risk of pathological fracture or malignant transformation[1,3,19]. We followed up patient for one year. The patient was symptom free and there was no evidence of recurrence radiologically.

CONCLUSIONS

Although intraosseous lipoma is a rare benign neoplasm which is very challenging to diagnose based on clinical and plain radiograph findings, combination of CT or MRI and pre-operative biopsy would be useful by being able to show the presence of fat within the lesion. Nevertheless, the clinicians, surgeons, radiologist and pathologist should be familiar and aware of these findings to be able to come to a correct diagnosis.

CONSENT

Informed written consent was obtained from the patient.

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