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Intraosseous Lipoma: An Elusive Cause of Long Bone Pain: A Case Series

MaKenzie M Chambers[1], Justin B Ungerleider[1], Tae Won B Kim[2], Christina J Gutowski[2]

Learning Point of the Article:

Intraosseous lipoma is an underrecognized, underdiagnosed benign bone tumor that should be included in a differential diagnosis of dull, chronic bone pain as prompt surgical treatment provides symptom resolution and return to function.

Abstract

Introduction: Intraosseous lipomas are benign tumors composed of mature fat that reside within the intramedullary canal. Although many cases are asymptomatic, some patients report pain that interferes with daily living. For patients presenting with refractory pain, surgical excision may be performed. These tumors were once thought to be rare, but a recent increase in awareness and diagnostic capability may contradict this.

Case Presentations: A 27-year-old female presented with a 3-month history of deep and aching pain in her left shoulder. The second patient was a 24-year-old female who presented with a 3-year history of pain in her right tibia. The third was a 50 year old female who presented with a 4-month history of deep pain in her right humerus. The fourth patient was a 34-year-old female who presented with a 6-month history of the left heel pain. All were found to have intraosseous lipomas and were treated with excisional curettage, achieving resolution of symptoms.

Conclusion: These cases may help orthopedists better understand the presentation and treatment of intraosseous lipomas due to several shared characteristics. We also hope that this report may lead clinicians to include this pathology in their differential diagnosis when patients present with similar symptoms. As the prevalence of these tumors appears to be increasing, efficient diagnosis and treatment will become increasingly valuable to orthopedists and patients.

Keywords: Intraosseous lipoma, orthopedic oncology, bone tumor, curettage.

Introduction

Intraosseous lipomas are rare and benign bone tumors that are derived from mature adipocytes existing within the intramedullary canal. They account for <0.1% of primary bone neoplasms and usually manifest in long bones [1, 2, 3]. They are thought to be idiopathic, with unexplained etiology and unknown pathogenesis.

Diagnosis of an intraosseous lipoma must combine radiographic imaging findings with histopathologic features consisting of varying degrees of viable lipocytes, calcification, marrow necrosis, and bony trabeculae [4]. Symptoms include vague, throbbing bone pain that may or may not radiate or be related to activity. Recent advanced imaging techniques may explain the increased incidence of reported intraosseous lipomas, revealing that these tumors may not be as rare as once thought [5, 6].

Treatment for an intraosseous lipoma is usually conservative, as these lesions are often asymptomatic and do not carry a malignant degeneration risk [7]. Operative treatment is indicated for refractory pain or if the lesion is large enough to risk pathologic fracture [2] and involves corticotomy with excisional curettage of the lipomatous tumor. Prophylactic fixation may also be included in treatment [5]. There are few reports in the literature that thoroughly describe this entity, its presentation and natural history, treatment strategies, and outcomes after

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Author's Photo Gallery


Dr. MaKenzie M Chambers


Dr. Justin B Ungerleider


Dr. Tae won B Kim


Dr. Christina J Gutowski

[1]Cooper Medical School of Rowan University, 401 Broadway, Camden, New Jersey USA 08103 ,
[2]Department of Orthopaedic Surgery, Cooper University Healthcare, Cooper Plaza Suite 400, Camden, New Jersey USA 08103

Address of Correspondence:
Dr. MaKenzie M Chambers, Cooper Medical School of Rowan University, 401 Broadway, Camden, New Jersey.
E-mail: chambe42@rowan.edu

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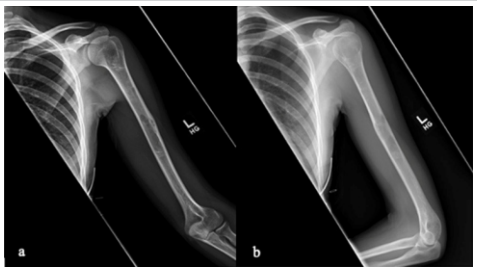

Figure 1: AP radiograph (a) and lateral radiograph (b) of left humerus. These X-rays demonstrate intraosseous lipoma of the humeral shaft.

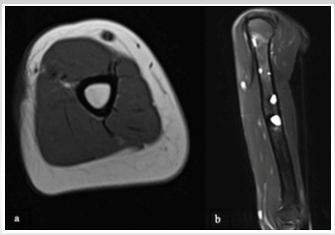

Figure 2: Axial T1 MRI (a) and sagittal STIR MRI (b) of the left humerus. These MRI images demonstrate obliteration of bony trabeculae due to intraosseous lipoma (a) along with intralesional cystic change (b).

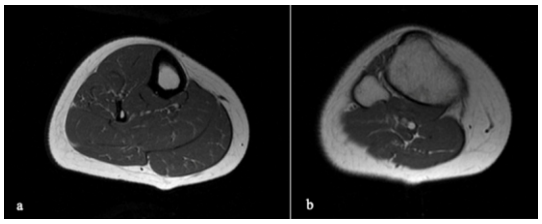

Figure 3: Axial T1 MRI of right tibia, at two levels. (a) demonstrates loss of trabeculae at the level of intraosseous lipoma, in comparison to (b) demonstrating normal trabecular markings of cancellous bone at an unaffected level of the tibia.

treatment [1, 2, 3, 4, 8] (Table 1). Due to this paucity in the literature, we aim to describe four cases of intraosseous lipomas that required surgical intervention to alleviate pain and prevent impending pathologic fracture.

Case Report

Case Presentation: #1

A 27-year-old female complained of a 3-month history of throbbing activity-related pain in her left anterior shoulder. The left humerus radiographs revealed a lytic bone lesion within the diaphysis with loss of trabecular markings (Fig. 1a, 1b). Magnetic resonance imaging (MRI) revealed fatty replacement of the normal marrow, with areas of cyst-like fluid signal and inconsistent contrast-enhancement (Fig. 2a, 2b). Due to these discrete areas of cystic change, open biopsy was performed. Pathology revealed adipose tissue and normal marrow elements, confirming the diagnosis of intraosseous lipoma.

Intralesional excision was performed through the anterolateral approach to the left humerus. A 10 mm diameter trephine reamer was used to create a round cortical window in the anterolateral cortex to gain access to the intramedullary canal. Curved curettes were utilized to excise the tumor and a flexible suction tip was inserted to achieve thorough excision of its proximal- and distal-most extents. The tumor cavity was packed with cancellous allograft bone chips and the cortical window was replaced. The patient's arm was placed in a sling and range of motion/weight bearing was progressed as cortical healing and incorporation of the graft was demonstrated on x-ray. She had returned to full function without pain by her 6-month post-operative appointment.

Case Presentation: #2

A 24-year-old female patient complained of 3 years of aching and constant pain in the right proximal tibial metaphysis that was aggravated by activity. She reported a history of motor vehicle accident 2 years before, when she sustained a non-displaced right tibial plateau fracture treated nonoperatively. In the months before presentation to our clinic, she was diagnosed at an outside facility with tibial stress fracture as the cause of her pain and underwent anterior tibial cortical drilling without relief of symptoms. Radiographs were unremarkable, but MRI demonstrated subtle loss of trabecular markings within the tibial marrow space, replaced by homogeneous fat signal (Fig. 3a, 3b). There was also bone edema around the site of drilling, concerning for stress fracture. An open biopsy was performed and pathologic examination revealed marrow elements with mature adipose tissue, confirming the diagnosis of intraosseous lipoma.

Excision of the intramedullary tumor was initiated through a longitudinal incision over the medial tibial metaphysis. A 3 cm oval cortical window was created and the lipomatous tissue was excised using curettes. There was considerable sclerotic bone found at the site of chronic stress reaction, concerning for incomplete healing, indicating prophylactic fixation. The surgeon then proceeded with uneventful prophylactic

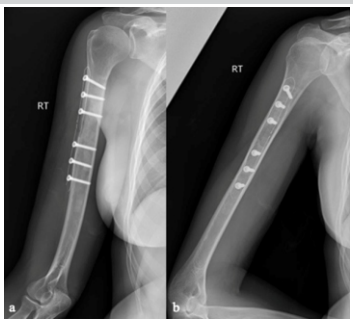

Figure 4: AP radiograph (a) and lateral radiograph (b) of the right humerus. These x-rays demonstrate carbon fiber plating after corticotomy, curettage, and grafting of intraosseous lipoma.

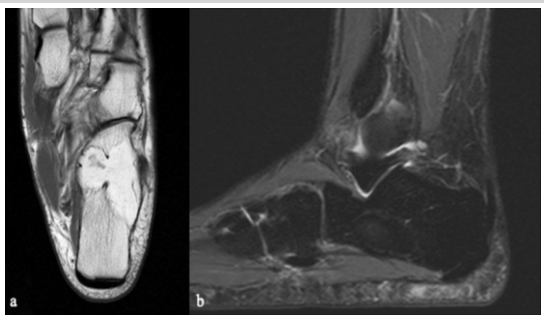

Figure 5: Axial T1 MRI (a) and sagittal STIR MRI (b) of the left foot. These images demonstrate intraosseous lipoma of the calcaneus. The fatty tumor is hypointense on STIR, confirming diagnosis.

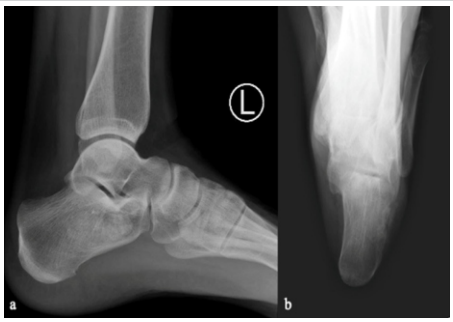

Figure 6: Lateral radiograph (a) and Harris view radiograph (b) of calcaneus. These X-rays demonstrate incorporation of allograft after curettage of the intraosseous lipoma.

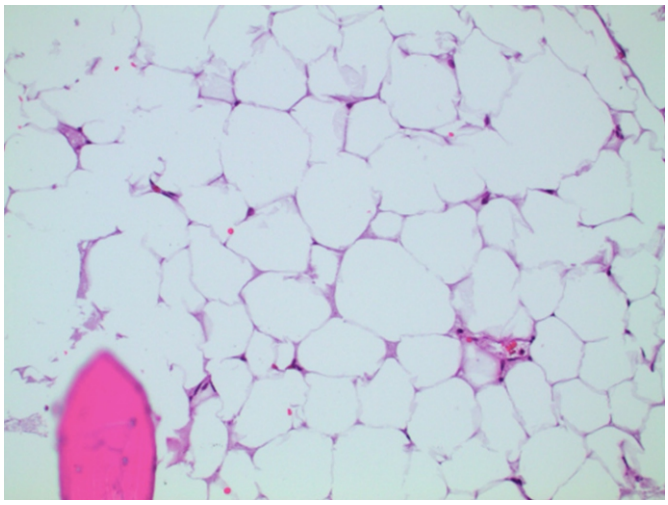

Figure 7: 20× H&E stained histology slide demonstrating mature adipocytes.

intramedullary nailing of the tibia through a medial parapatellar approach. The reamers served to secondarily excise the tumor. Finally, the cortical window was packed with allograft. Postoperatively, the patient was weightbearing as tolerated and reported complete resolution of symptoms a month later.

Case Presentation: #3

A 50-year-old female presented with a 4-month history of a constant and deep aching pain in her right proximal humerus that was aggravated by weather changes and sleeping on the affected side. Radiographs were unremarkable, but MRI revealed fatty replacement of the normal trabecular bone for nearly the length of the humerus.

Tumor excision was performed through an anterolateral approach to the humerus. A 10 cm incision centered over the junction of the proximal and middle thirds of the diaphysis was made, dissection was carried down to the anterolateral cortex of the humeral shaft, and a burr was used to create a 4 cm oval cortical window. Curettes and flexible suction tips were used to excise the tumor from the humeral head down to the epicondyles. After thorough excision, the canal was packed with allograft cancellous bone chips. Due to the length of the cortical window acting as a stress riser, the humerus was prophylactically stabilized using a carbon fiber plate (Fig. 4a, 4b). The patient's arm was placed in a sling and she initiated immediate physical therapy. She had regained full range of motion and strength 3 months from surgery. Histopathologic examination revealed mature adipose tissue and bone marrow elements, compatible with intraosseous lipoma.

Case Presentation: #4

A 34-year-old female patient presented with a 6-month history of activity-related left heel pain. On initial assessment of a left foot CT scan, the lesion was thought to be a large fluid-filled cyst within the calcaneus. However, MRI revealed that the lytic bone lesion was composed of fat, as opposed to fluid (Fig. 5a, 5 b). No pathologic fracture was present, but there was slight remodeling of the adjacent cortical bone at Boehler's angle and thinning of the cortex in some areas. The tumor filled the marrow space completely in the coronal plane.

Excision of the intramedullary tumor was performed through a laterally-based incision over the hindfoot, using the longitudinal limb of the classic L-shaped extensile approach to the lateral calcaneus. The "no-touch technique" was employed. An ultrasonic bone scalpel (Sonopet Ultrasonic Aspirator, Stryker, Kalamazoo, MI) was used to create a 3 × 1 cm cortical window in the lateral calcaneus to access the marrow space. The tumor was excised using a pituitary, curettes, and suction. Once complete excision of the mass was confirmed, the tumor cavity was packed with allograft cancellous chips, and the excised cortical window fragment was replaced into the cortical defect. Postoperatively, the patient was placed in a CAM boot and was non-weight bearing on the lower left extremity for 7 weeks. At her most recent follow-up appointment, 9 weeks from surgery, X-rays revealed excellent incorporation of the bone graft and cortical fragment, and she had returned to normal function with residual mild soreness after extended periods of weight bearing

Table 1: Compilation of the current literature on intraosseous lipomas

Title	Author	Year	Clinical Presentation	Imaging Characteristics	Treatment	Conclusions
Multiple intraosseous lipomatosis - a case report and a review of the literature.	Dammerer *et al.*	2021	Pathological compression fracture of right tibial plateau and history of multiple other fractures	Radiographs showed well-defined, radiolucent lesions forming osteolytic-appearing lesions in the proximal tibia and distal femur. MRI showed multiple intraosseous lipomatosis	ORIF of tibial fracture	A history of multiple fractures with unexplained causes or minimal trauma and subtle changes on imaging should raise suspicion for a diagnosis of intraosseous lipoma
Threatening fracture of intraosseous lipoma treated by internal fixation	Frangez *et al.*	2019	Chronic unilateral heel pain accompanied by swelling of the lateral ankle with no history of trauma to the area	Radiographs showed a lytic lesion in the calcaneus. MRI and CT confirmed a diagnosis of intraosseous lipoma	Curettage of the tumor was performed, followed by stabilization with a plate due to impending fracture	Symptoms of vague, continuous pain with no history of trauma or other pain- generating phenomena should raise suspicion for an intraosseous lipoma
Calcaneal intraosseous lipoma: a case report and review of the literature	Narang *et al.*	2011	Chronic, dull, aching pain in the left heel that was aggravated by activity	Radiographs showed a radiolucent lesion in the anteroinferior portion of the left calcaneus. CT showed a cyst in the same location with expansion of the inferior cortex	Operative decompression of the cystic lesion was performed followed by packing with bone graft	Chronic, dull pain that is aggravated by activity and is accompanied by non-diagnostic imaging findings should raise suspicion for an intraosseous lipoma
Intraosseous lipoma of the rib	Sivrikoz *et al.*	2019	Right sided chest pain	Radiographs revealed a large expansive lesion in the eighth rib. CT showed a paravertebral localized well-defined expansive lytic lesion	Wide en bloc excision of the seventh and eighth rib was performed	Non-cardiogenic chest pain associated with no abnormal physical examination findings, normal biochemical parameters, and radiographic evidence of a bone lesion should raise suspicion for an intraosseous lipoma.
Intraosseous lipoma of the calcaneus	Bertram *et al.*	2001	Chronic right heel pain aggravated by activity and relieved by rest	Plain radiographs of the right heel showed a sharply defined lytic lesion with sclerotic margins at the base of the calcaneus neck. MRI revealed a cystic lesion of the calcaneus without interruption or permeation of the cortex. Homogenous high signal intensity images with T1 and T2 weighting were produced by the lesion.	Curettage of the whole lesion was performed followed by autologous bone grafting	On diagnosis of an intraosseous lipoma, curettage of the lesion results in complete resolution of symptoms
Intraosseous lipoma. A clinical, radiologic, and pathologic study of 5 cases	Eduardo *et al.*	2007	Two out of five cases had pain localized to the lesion. Three out of five cases were incidental findings	Plain radiographs varied among cases but consisted of the presence of a lytic lesion, well-defined radiolucency, and mild expansion of the bone. MRI and CT in four of the five cases both showed the presence of fat within the lesion	Curettage of the lesion was the treatment for four out of five cases while one case required excision	In the presence of a well-defined radiolucent lytic lesion on plain radiographs accompanied by no symptoms or vague pain, MRI and/or CT should be ordered to investigate the possibility of an intraosseous lipoma diagnosis

(Fig. 6a, 6b). Final pathologic examination revealed mature adipose tissue, compatible with the diagnosis of intraosseous lipoma (Fig. 7).

Discussion

Intraosseous lipomas are unique intramedullary lesions that can pose challenges in identification, diagnosis, and treatment. Due to their limited occurrence, non-specific symptom presentation, and subtle changes on radiographs, many intraosseous lipomas may be misdiagnosed or overlooked resulting in delayed treatment [2]. However, with advancing imaging techniques and improved physician understanding [1], we are able to highlight four cases of confirmed intraosseous lipomas treated at our institution.

Vague, deep and throbbing pain are the most common symptoms that the patient will note. The presence of an intraosseous lipoma can lead to increased intraosseous pressure, which likely explains this sensation. Radiographic diagnosis remains difficult, as the majority of patients have X-rays that appear normal, yet some images do show subtle changes such as loss of trabecular markings. MRI can also show loss of bony trabeculae and replacement with homogenous fat signal, but this can be difficult to detect. Due to the subtle changes in imaging, physicians should have a high index of suspicion for an intraosseous lipoma when a patient presents with characteristic symptoms and other pain-generating phenomena such as trauma, rotator cuff pathology, radiculopathy, and degenerative joint disease have been ruled out. In general, the authors recommend having a low threshold for obtaining an MRI of a long bone that is painful for several weeks in the setting of normal X-ray findings, as marrow-replacing lesions may be undetectable on plain radiographs but obvious on MRI.

Treatment in all four cases involved intralesional excision of the mass through a cortical window utilizing curettes and flexible suction tips. As it was determined necessary by the surgeon in two of our four cases, prophylactic fixation can also be performed. Risks of pathologic fracture secondary to the cortical thinning from the tumor itself, or secondary to the cortical window created iatrogenically, are indications for prophylactic fixation and should be considered on a case-specific basis. Postoperatively, all patients experienced resolution of their symptoms and demonstrated a return to full range of motion and radiographs consistent with healing. We hypothesize that excision of the tumor allows the intraosseous pressure to normalize, facilitating these patients' symptomatic improvement. To date, no patient has experienced local recurrence or pathologic fracture, suggesting that intralesional excision is appropriate and successful when a patient meets surgical indications.

Although there is limited literature detailing the pathogenesis, progression, and resolution of intraosseous lipomas (Table 1), it is important to note that our findings and approach to treatment are consistent with the current understanding of these lesions [1, 2, 3, 4]. Frangez et al. described a similar case of a calcaneal intraosseous lipoma that required surgical intervention and yielded post-operative improvements in symptoms and full rehabilitation time consistent with our fourth patient. Our cases all demonstrated similar histopathologic changes, specifically the presence of mature adipose tissue, which is also consistent with the current literature [8, 9, 10]. The authors hope that contribution of these four cases to the sparse available literature that exists will allow for increased awareness and understanding of these tumors.

Conclusion

Once thought to be extremely rare bone lesions, the incidence of intraosseous lipomas has increased due to the widespread nature of advanced imaging and more clinicians including this condition on their differential diagnoses. Proper diagnostic workup should be initiated for patients who present with symptoms of throbbing bone pain with subtle radiographic changes to evaluate for this diagnosis once other likely pathologies have been ruled out. If conservative treatment fails, intralesional excision with or without prophylactic fixation provides pain relief and return to function.

Clinical Message

Intraosseous lipomas can cause vague and persistent bone pain and may be difficult to diagnose on physical examination and imaging studies. Pain control is achieved through excision of the tumor, occasionally paired with prophylactic fixation as indicated.

Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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