Intraosseous "Lipoma" of the Calcaneus Developing in an Intraosseous Ganglion Cyst

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ABSTRACT

Intraosseous lipomas are rare primary bone tumors, most common to the calcaneus. There are several proposed theories of the natural etiology of intraosseous lipomas; however, all lack definitive support. In this report, an 18-year-old man presented with radiologic evidence of a simple bone cyst of the calcaneus. Over a 4-year period, the patient was followed with interval magnetic resonance imaging. The cyst demonstrated progressive development of peripheral intralesional fat with final magnetic resonance imaging features characteristic of an intraosseous lipoma. To our knowledge, this is the first longitudinal study that shows gradual peripheral fat deposition within an intraosseous ganglion cyst, illustrating a potential cause of intraosseous lipomas of the calcaneus.

CASE REPORT

CASE REPORT

An 18-year-old man with a past medical history of Gilbert syndrome and asthma initially presented with pain that developed in the distal portion of his leg while he was playing basketball. He reported the initial pain as 9 out of 10. When he sought medical care, the clinical diagnosis was Achilles tendinitis. The presenting radiographs showed a lytic lesion in the body of the calcaneus (Figure 1). The lesion margins had a nonaggressive, narrow zone of transition with a nonsclerotic margin and no internal calcifications or calcified matrix formation.

One month after the initial radiographs, magnetic resonance imaging (MRI) showed a bilobed benign cystic lesion in the calcaneus, which was thought to be a unicameral bone cyst (Figure 2). Given the size of the lesion, the patient was referred to an orthopedic surgeon for evaluation 5 months after the initial symptoms. On presentation to the orthopedic surgeon, the patient reported improvement in the Achilles tendon pain with only minimal residual symptoms and avoidance of strenuous activity. Physical examination findings were unremarkable; specifically, the patient had no pain with palpation of the calcaneus. The surgeon recommended that the patient gradually return to his usual activities with follow-up MRI to confirm the stability of the calcaneal lesion. Given the size of the lesion, the patient was followed with yearly MRI examinations.

The cystic lesion was stable in size on multiple MRI examinations over 4 years with no findings of pathologic fracture. Follow-up MRI showed a narrow neck connecting the cyst to the subtalar joint, which was more consistent with an intraosseous ganglion cyst than a unicameral bone cyst. Both portions of the bilobed cyst demonstrated gradual peripheral filling in with fat (Figure 3). The final MRI demonstrated a lesion more compatible with the MRI appearance of a calcaneal lipoma with central cystic change than an intraosseous ganglion cyst. The patient was instructed to follow up as needed if he became symptomatic.

DISCUSSION

Intraosseous lipomas (IOLs) are thought to be rare primary bone tumors and are most frequently found in the calcaneus [1-3]. Lesions are generally asymptomatic and commonly discovered as an incidental finding on imaging studies [3-6]. IOLs typically occur in the neutral triangle of the calcaneus; the only specific radiographic feature is a lytic lesion with coarse central calcification. Computed tomography (CT) may show internal fat attenuation for the diagnosis, but MRI is the imaging modality of choice. Given the characteristic location of IOLs in the calcaneus, multiple theories have been proposed about the relationship of IOL either secondary to or independent of other lesions [7]. The most widely accepted theory, proposed by Milgram, suggested that IOLs are primary neoplasms [5]. Despite many theories, the true cause of IOL of the calcaneus is not entirely clear. This case report illustrates the cause of a lesion with final MRI features that were most suggestive of an IOL of the calcaneus arising within an intraosseous ganglion cyst that was demonstrated longitudinally with MRI.

Etiology & Demographics:

IOLs have an unknown etiology and are frequently found in the lower extremities, specifically the calcaneus and femur [3-5,8]. The calcaneus is the most commonly reported site (32% of IOLs) [3]. IOLs have been reported as rare bone tumors (0.1% to 2.5% of all primary bone tumors) [1,2]. This incidence likely underrepresents the true frequency of the lesions, which can be asymptomatic, incidental, and misdiagnosed and, therefore, underreported [3-5,9].

In a meta-analysis, the age range of presentation was 4 to 85 years (average, 43 years) [3]. In addition, gender distribution is mostly equal, with a slight male predominance [10]. Interestingly, this is drastically different from soft tissue lipomas, which are disproportionately found in females [3]. Overall, there is no age or gender predilection.

Clinical & Imaging Findings:

Incidental asymptomatic lesions are found in 30% to 40% of patients [3,4,6]. When lesions are symptomatic, dull pain is the most common symptom [3]. Soft tissue swelling and tenderness have also been reported [7]. Campbell et al suggested, however, that symptoms are likely from an unidentified source because after surgical excision, spontaneous recovery or recurrence of symptoms is frequently noted [3].

The Milgram staging system is used to characterize the sequential histopathologic progression of IOLs [11]. Stage 1 lesions have the gross and microscopic characteristics of normal lipocytes with thin bony trabeculae. Stage 2 lesions resemble stage 1 lesions but show partial necrosis with or without secondary calcification. Stage 3 lesions lack bone trabeculae with complete necrosis of lipocytes and a variable degree of cystic, calcific, and reactive bone formation.

Milgram also suggested that each stage had specific radiographic features that correlated with histopathologic findings [11]. A stage 1 lesion is a well-circumscribed osteolytic area with trabecular resorption and a narrow zone of peripheral sclerosis [11,12]. Stage 2 lesions are lucent, with peripheral sclerosis, evidence of necrosis and calcification with subtly increased radiodensity, and possibly expansion. A stage 3 lesion is even more radiodense, with a thick sclerotic border with or without central calcification. The nonspecific radiographic features are equivocal in determining the degree of necrosis, and recent studies indicate greater radiographic variability compared with advanced imaging [12].

Advanced imaging of IOLs with CT shows hypoattenuating fat that corresponds to the radiographic area of lucency and a narrow zone of peripheral sclerosis [12,13]. Stage 2 lesions have diffusely scattered areas of dystrophic calcification and fat necrosis. Features of stage 3 include fat necrosis, dystrophic calcification, and cystic formation with central or peripheral (or both) reactive ossification [3,12].

MRI of stage 1 lesions shows isointensity to subcutaneous fat on T1-weighted and hypointensity on T2-weighted fat-saturated sequences [12]. Reactive sclerosis is evident by a narrow circumferential hypointense border on both T1- and T2-weighted sequences. Again, stage 2 lesions retain features of stage 1 and possess areas of T1 and T2 hypointensity indicating dystrophic calcification. Stage 3 lesions retain aforementioned characteristics with a thick border of peripheral sclerosis, adjacent fat, and large central calcification [12]. The T1-weighted sequence has variably low signal intensity in representing fat necrosis and cystic degeneration, whereas T2-weighted sequences are hyperintense [8,12]. Because of the additional detail, the use of advanced imaging is needed to accurately diagnose and characterize the stage of an IOL [14]. Biopsy is not necessary as was once believed.

Treatment & Prognosis:

Recommended treatment of symptomatic IOL of the calcaneus is largely conservative. The use of anti-inflammatory medication, cold wrap, heel cushion, non–weight-bearing devices, and activity limitation can be beneficial [15]. Surgical intervention has been debated [16-18]. Some authors have suggested that surgical intervention may be unnecessary because lesions have a low malignancy potential and can regress. Ulucay et al considered curettage and autogenous bone grafting for patients who were overtly symptomatic or at risk for pathologic fracture [15]. Risk of pathologic fracture of an IOL of the calcaneus is suggested by a critical size cyst occupying more than 30% of the anteroposterior length and extending the full width of the calcaneus [9]. However, pathologic fracture of the calcaneus has been reported in only 2 patients [10].

Differential Diagnoses:

Radiographically, the differential diagnosis of IOL of the calcaneus is relatively narrow [3,9,14]. Ganglion cyst, simple bone cyst, and giant cell tumor may occur in this location and have similar radiographic features. Additionally, with the relative decrease in central trabeculation, a lesion may be simulated in the body of the calcaneus centrally.

Simple Cyst/Ganglion Cyst

Simple cysts are often found centrally within long bones of younger patients. Classically, a pathologic fracture through a simple cyst can contain a dependent cortical fragment, known as the fallen fragment sign. Radiographically, simple cysts are lytic with a narrow zone of transition with or without cortical thinning. CT findings are similar to the radiographic findings, with the addition of a hypoattenuating fluid density within the lesion. MRI shows a homogenously low to intermediate T1 signal and a high T2 signal. Ganglion cysts demonstrate similar imaging characteristics with the addition of a small fluid filled cleft extending to the articular surface.

Giant Cell Tumor

On radiographs and CT imaging, giant cell tumors are classically described as being eccentric, with a nonsclerotic narrow zone of transition, and adjacent to the articular surface in the skeletally mature patient. MRI shows a heterogeneous, intralesional, low to intermediate T1 signal and a generally heterogeneous, high T2 signal.

Conflicting Data

In our patient, the initial radiographic features were compatible with a stage 1 lesion; MRI showed that the lesion was completely cystic, though, rather than being a homogeneously fat-containing lesion. Subsequent MRI showed a stage 2 IOL with a gradual, peripheral filling with fat. Although microscopic evaluation was not performed, the lesion did have central cystic change without regions of necrosis or dystrophic calcification within the fat-rimmed lesion. This finding is at odds with the Milgram hypothesis, which states that cystic change is present only in stage 3 lesions and that cystic degeneration occurs only when lesions are still viable because lesions require living osteoclasts to remodel bone [11]. Older and more advanced stages, however, should be the most necrotic. In addition, Campbell et al stated that older patients do not have a preponderance of advanced-stage lesions as would be expected in a slowly progressive, involuting lesion [3]. In our patient, consecutive images suggested a gradual, peripheral filling with fat rather than a lesion containing pure fat that developed central necrosis. This lesion lacked sequential involution as suggested by Milgram.

The true neoplastic nature of IOLs has been challenged by multiple authors who have proposed many theories on the etiology of IOL of the calcaneus [3,5,9,19]. These theories include causes related to bone infarction, posttraumatic reaction, benign neoplasm, and cyst involution [7]. It has been noted previously that calcaneus cysts have a higher cholesterol and fibrous content in comparison with simple humeral and femoral cysts [20]. Hatori et al reported a case of an IOL of the calcaneus identified with imaging and surgical pathology, which had been a biopsy-proven simple bone cyst 8 years earlier [19]. Additionally, Wada and Lambert reported the involution of a simple bone cyst with fat replacement after alcohol irrigation [21]. As in our patient, Wada and Lambert observed fatty replacement beginning peripherally. Despite these findings, to our knowledge no report has depicted the natural evolution of IOL of the calcaneus from a simple intraosseous ganglion cyst.

Campbell et al reviewed 35 IOLs and made several interesting observations related to those occurring in the calcaneus [3]. They found that all calcaneal IOL occurred in the exact location within the calcaneus, a finding that is also substantiated in numerous published case reports [3]. IOLs of the calcaneus occur plantar to the critical angle of Gissane in an area lacking a trabecular network called the neutral triangle [9,10]. Campbell et al also found that cysts were more common in IOLs of the calcaneus than in IOLs occurring in other locations [3]. Authors have long debated the evolution of IOLs in the calcaneus [3,19-21]. In particular, simple cysts are known to occur in the exact location as calcaneal IOLs, and the evolution of an IOL from a simple cyst has been a proposed mechanism.

Cystic lesions at the angle of Gissane occur in 40% to 75% of patients [22,23]. As with lipomas, patient age is not associated with the presence of a lesion [23]. This hyperintense focus on T2-weighted MRI sequencing corresponds to the approximate anatomical location of vascular penetration of the calcaneus [22]. Small linear foci arising from the anterior margin of the posterior facet of the subtalar joint space may be dilated vascular segments extending into the relatively rarefied anterior calcaneus; however, larger foci are more lobulated and are likely intrusions of synovial fluid forming a cyst [23]. Given the spectrum of lesion size and similar anatomical location, Elias et al speculated that these lesions arise from the same disease process [23]. On magnetic resonance arthrography of the subtalar joint, a patient of theirs had contrast medium communicating between the joint space and the lesion and thus had an intraosseous ganglion cyst. Therefore, vascular penetration of the calcaneus at the angle of Gissane may be related by providing a conduit for the formation of these cysts.

Previously, authors have suggested cystic transformation to IOL based on specific moments in time and could not definitively determine whether the lesions were distinct or a process of maturation [3,19-21]. Elias et al and others speculated that cystic hemorrhage, which has been reported in surgically excised specimens, could serve as a sclerosing agent inducing cyst involution and subsequent lipomatous formation [23,24]. With the evolution of the lesion occurring in reverse order, this would again refute the staging system proposed by Milgram. Although the lesion in our report did not show evidence of hemorrhage, it may have occurred between imaging studies or it may have been radiologically occult.

Although IOLs are considered rare entities, the incidence is likely greater than reported because many are asymptomatic and discovered incidentally. Despite multiple theories previously proposed, the present case report illustrates radiologic evidence of the formation of an IOL of the calcaneus as a maturational continuum of a simple cyst. The lack of sequential involution of this lesion as proposed by Milgram suggests that the staging system should be abandoned. The application of this report of IOL to sites other than the calcaneus is unknown.

Companion Case

A 58-year-old man with a history of diffusely metastatic esophageal adenocarcinoma presented after tripping at home and abrading the lateral aspect of his left foot. Radiography showed a large lytic lesion in the body of the calcaneus with a sclerotic, narrow zone of transition and without internal calcification or matrix formation (Figure 4). Given the patient's history, MRI was performed to exclude metastasis. MRI showed a predominately cystic lesion containing a thick peripheral rim of fat with a central cystic cavity (Figures 5 and 6).

TEACHING POINT

Intraosseous lipomas most commonly occur in the calcaneus and contain intralesional fat with variable central cystic changes and calcifications. The Milgram staging system is based on sequential histopathologic and radiographic features of intraosseous lipomas; however, the usefulness of this staging system for the calcaneus should be questioned because IOLs of the calcaneus may belong to the maturational continuum of an intraosseous ganglion cyst.

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FIGURES

Figure 1 (right): 18-year-old man with an intraosseous calcaneal cyst.
FINDINGS: A lateral radiograph of the ankle demonstrates a round lytic lesion in the body of the calcaneus with a nonsclerotic, narrow zone of transition and no internal calcifications or matrix formation.
TECHNIQUE: Lateral radiograph.

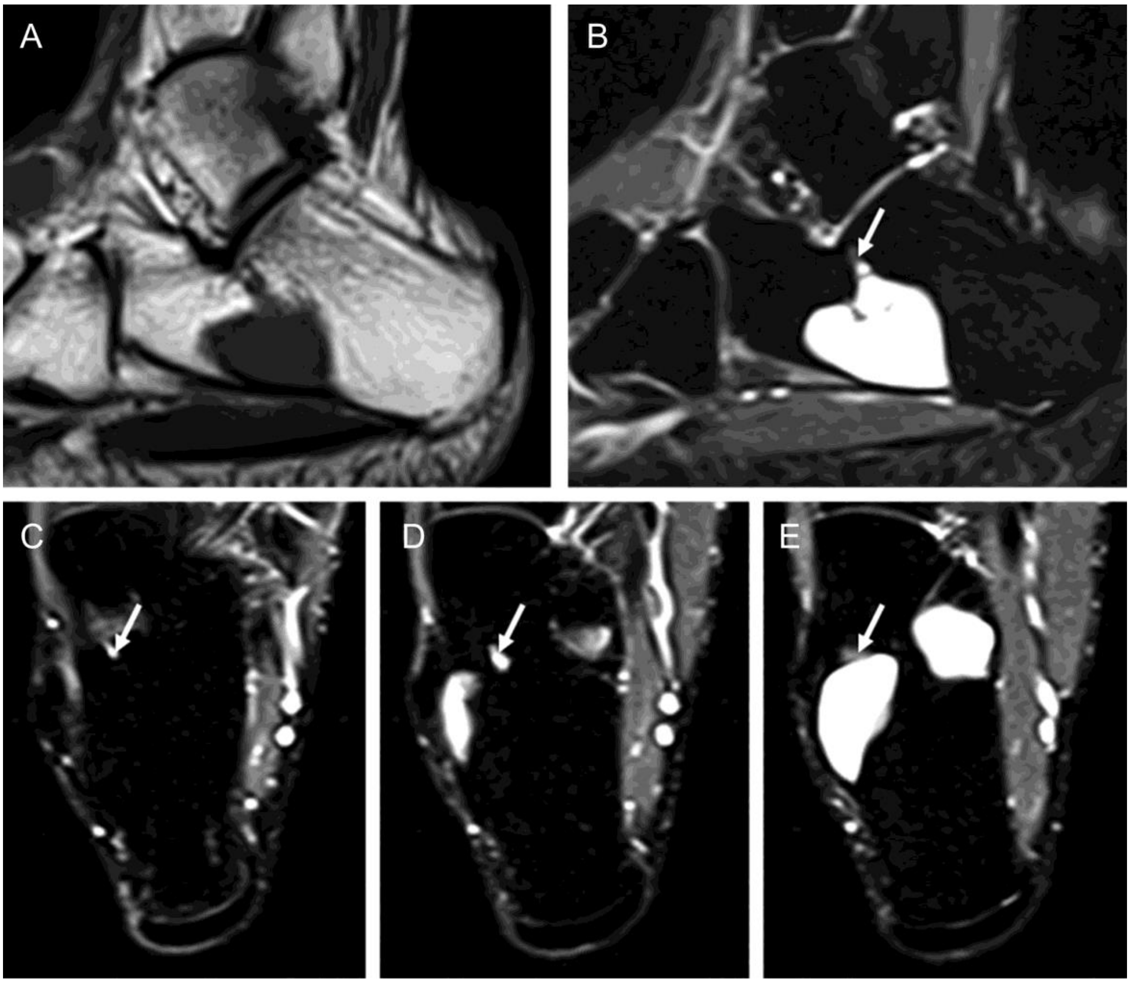

Figure 2: 18-year-old man with an intraosseous ganglion cyst in the calcaneus.
FINDINGS: Sagittal T1 (A) and short tau inversion recovery (STIR) (B) magnetic resonance (MR) images of the calcaneus demonstrate an intraosseous ganglion cyst in the calcaneus. In retrospect, a narrow neck is identified extending from the subtalar joint to the cyst (B, arrow). This connection is best demonstrated on sequential axial STIR images (C-E, arrows).
TECHNIQUE: Sagittal and axial 1.5-T MR images; T1 (repetition time [TR], 457 ms; echo time [TE], 11 ms) and STIR (repetition time [TR], 5020 ms; echo time [TE], 32 ms; inversion time, 160 ms).

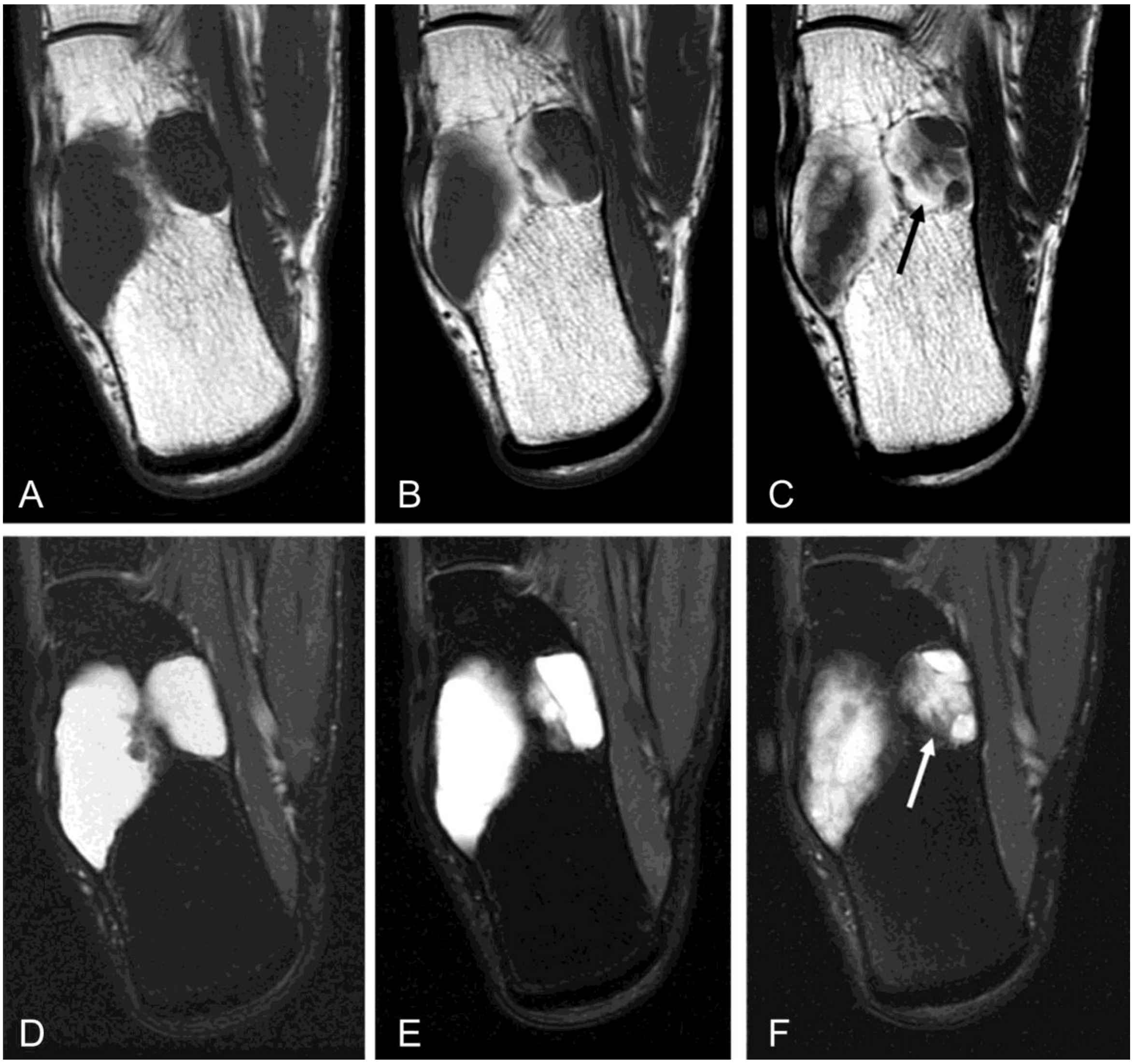

Figure 3: 18-year-old man with longitudinal development of peripheral lipomatous change in an intraosseous ganglion cyst of the calcaneus.
FINDINGS: Axial T1- and T2-weighted fat saturated magnetic resonance (MR) images through the calcaneus at 6 months (A and D), 30 months (B and C), and 42 months (E and F). Internal fat developed over time (C and F, arrows).
TECHNIQUE: Axial 3-T MR images; T1 (repetition time [TR], 700-900 ms; echo time [TE], minimum) and T2 fat saturated (repetition time [TR], 3,000-6,000 ms; echo time [TE], 45 ms).

Figure 4 (left): 58-year-old man with an intraosseous calcaneal cyst with lipomatous change.
FINDINGS: A lateral radiograph of the foot demonstrates a round lytic lesion in the body of the calcaneus with a sclerotic, narrow zone of transition and no internal calcifications or matrix formation.
TECHNIQUE: Lateral radiograph.

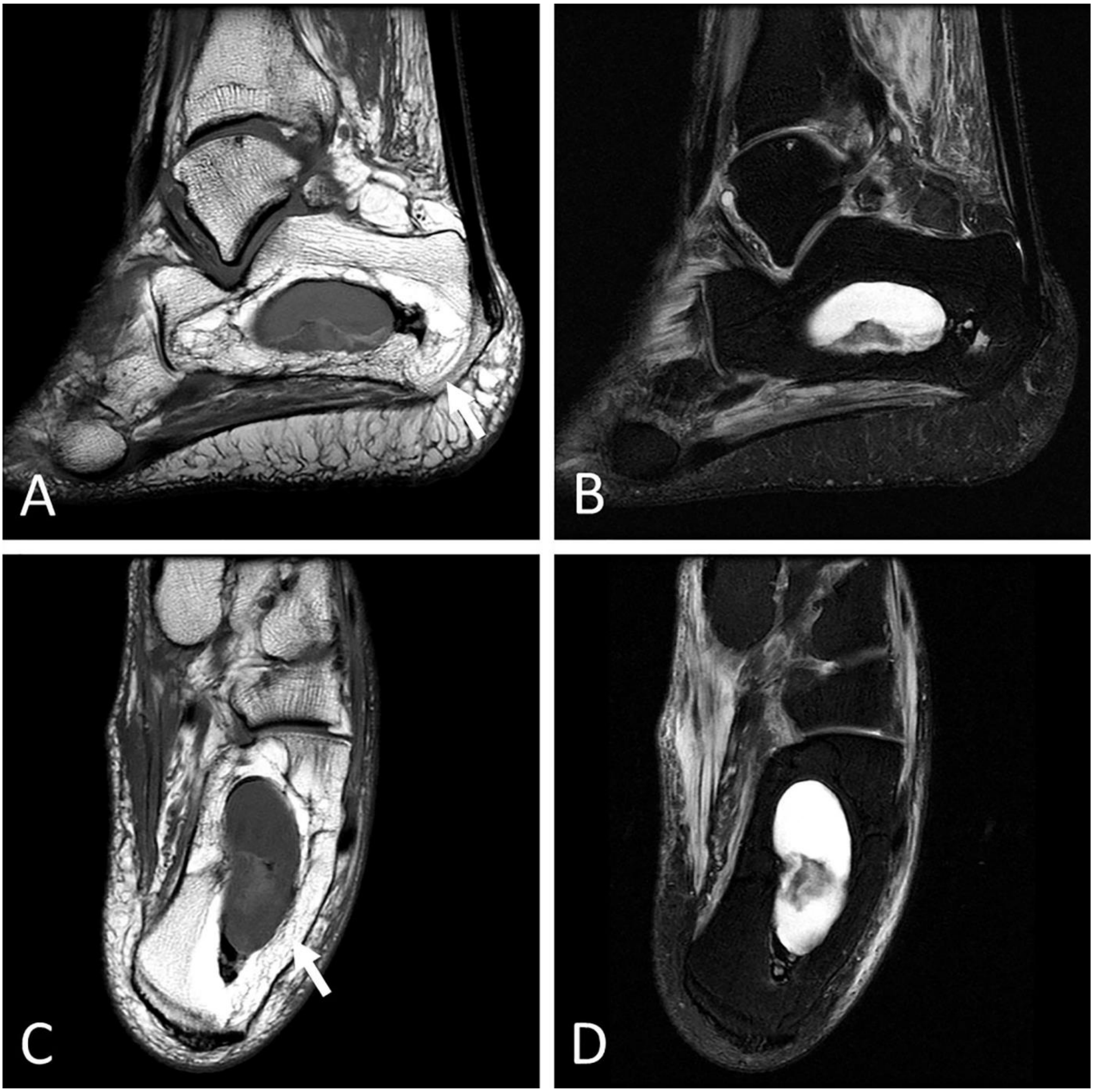

Figure 5: 58-year-old man with an intraosseous calcaneal cyst with lipomatous change.
FINDINGS: Sagittal T1 (A), sagittal T2 fat-saturated (B), axial T1 (C), and axial T2 fat-saturated (D) magnetic resonance (MR) images of the calcaneus demonstrate a cystic lesion that has a thick peripheral rim of fat (C, arrow) with a central cystic cavity.
TECHNIQUE: Sagittal and axial 3-T MR images; T1 and T2 fat-saturation (quad knee coil; field of view, 14 cm; slice thickness, 3 mm; echo train length, 8; repetition time [TR], 5,308 ms; echo time [TE], 49 ms).

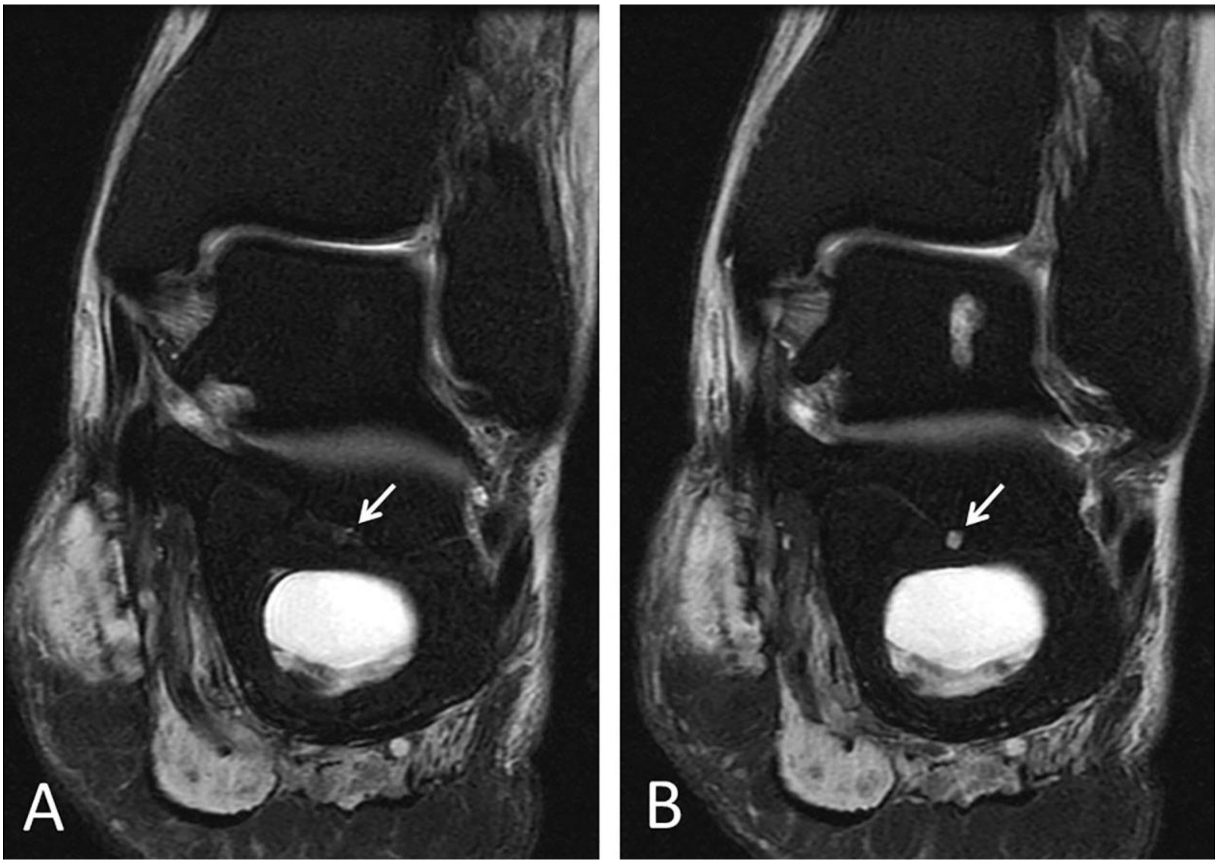

Figure 6: 58-year-old man with an intraosseous calcaneal cyst with lipomatous change. FINDINGS: Coronal oblique T2-weighted fat-saturated magnetic resonance (MR) images demonstrate the large central area of cystic change. A small fluid signal neck extended toward the subtalar joint (A and B, arrows).
TECHNIQUE: Coronal oblique 3-T MR images; T2-weight fat saturated (quad knee coil; field of view, 14 cm; slice thickness, 3 mm; echo train length, 8; repetition time [TR], 5,308 ms; echo time [TE], 49 ms).

Etiology	Unknown
Incidence	0.1%-2.5% of all primary bone tumors [3]
Gender ratio	Slight male predominance [10]
Age predilection	Range, 4-85 y; average age, 43 y [3]
Risk factors	Unknown
Treatment	• Largely conservative, including anti-inflammatory medication, cold wrap, heel cushion, non–weight-bearing devices, and activity limitation [22] • Surgical treatment can be considered if lipoma reaches critical size [9].
Prognosis	Excellent
Findings on imaging	• Radiography: Lytic lesion with a narrow zone of transition and with or without internal calcification. • Computed tomography: Intralesional hypoattenuating fat with or without fat necrosis • Magnetic resonance imaging: Intralesionally isointense to subcutaneous fat on T1 and hypointense on T2 with fat saturation; fat necrosis and cystic degeneration indicated by hypointense signal on T1 and hyperintense signal on T2

Table 1: Summary table for Intraosseous Lipoma.

	XR	CT	MRI
Intraosseous lipoma	• Lytic • Narrow zone of transition • With or without internal calcification	• Similar to radiography with hypoattenuating fat density • With or without fat necrosis	• Isointense to subcutaneous fat on T1 and hypointense on T2 with fat saturation • Fat necrosis and cystic degeneration indicated by hypointense signal on T1 and hyperintense signal on T2
Simple bone or intraosseous ganglion cyst	• Lytic • narrow zone of transition • cortical thinning	• Similar to radiography with hypoattenuating fluid density	• Homogeneous, low to intermediate signal on T1 and high intensity on T2 • Cystic neck to subtalar joint differentiates intraosseous ganglion cyst from simple bone cyst
Giant cell tumor	• Lytic • Eccentric, cortical scalloping • Nonsclerotic • Narrow zone of transition • Extends to the end of long bones	• Similar to radiography	• Heterogeneous, hypointense to isointense on T1 and heterogeneously hyperintense on T2

Table 2: Differential diagnoses table for Intraosseous Lipoma.

ABBREVIATIONS

CT - computed tomography
IOL - intraosseous lipoma
MRI - magnetic resonance imaging
XR – radiography

KEYWORDS

Lipoma; Intraosseous; Calcaneus; Ganglion; Cyst; MRI

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