

Calcaneal cysts and lipomas: a common pathogenesis?

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Abstract Calcaneal cysts and lipomas are relatively rare, benign bone lesions. They are similar in many ways, including in their location and radiological appearance, but their content differs. Cysts contain fluid whereas lipomas contain fat, although some lesions may exhibit a mixed content. The pathogenesis of the two entities is the subject of controversy. The theory that calcaneal cysts may result from lipomatous necrosis has been widely suggested in the literature, but no such progression has ever been shown. The contrary hypothesis has also been considered, i.e., that the content of regressing cysts may be replaced by fatty marrow, leading to a lipoma-like appearance. This second theory is based on indirect arguments, notably that lipomas have a similar location to intraosseous calcaneal ganglion cysts that arise from the subtalar joint and that patients are older in cases of calcaneus lipomas than in cases of calcaneus cysts. We offer an additional argument in favor of this hypothesis. We present an original observation of a calcaneal cyst incidentally discovered in a 15-year-old male whose spontaneous evolution 3 and 7 years later revealed progressive replacement of its fluid content by fat.

Keywords Bone cyst · Intraosseous lipoma · Pathogenesis · Benign bone tumors · Calcaneus

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Introduction

Simple (solitary or unicameral) bone cysts of the calcaneus, which are fluid-filled cavities, are benign lesions that have been described for many years [1]. The calcaneal location is considered to be relatively rare [2–5]. Calcaneal cysts represent 2 to 14% of simple bone cysts [6, 7]. Cysts in the long bones are generally discovered during the first 2 decades of life [8–10]. Calcaneal cysts are seen a little later, usually during the 2nd and 3rd decades of life [3, 5, 7, 11–17]. In a recent review of 10 studies in 171 patients, mean age was 25.7 ± 8.1 years, and males were clearly most often affected (two thirds of cases) [17].

Calcaneal lipomas, which are cavities filled with fatty tissue, are also benign lesions that have long been described [18], and they too are considered rare [5, 19–21]. Of all intraosseous lipomas of the skeleton (1/1000 primary bone tumors [22]), the calcaneal location is proportionally the most common (one third of 110 cases in a literature review [23]). Calcaneal lipomas are observed later than solitary calcaneal cysts, generally between 30 and 60 years of age [5, 20, 21, 24–26], and male sex clearly predominates (two thirds of cases) [21, 23].

Calcaneal cysts and lipomas are probably more common than the cases reported: they are very often discovered incidentally, and their unalarming radiological appearance does not call for further exploration to determine their nature [15]. Their natural histories are poorly known, probably because their appearance is so clearly benign that they require no long-term follow-up or because follow-up is only performed by plain radiography, which cannot differentiate them.

It has been hypothesized that calcaneal cysts and lipomas are a single entity and that a pathogenic continuity exists

between them: either cysts result from the liquefaction of lipomas or lipomas result from the regression of cysts and their replacement by fatty marrow [5, 6, 15, 27, 28]. This point will be the subject of the discussion that follows.

Similarities between calcaneal cysts and lipomas

Both lesions are characterized by the same location under the calcaneal sulcus, which is situated on the anteroinferior margin of the posterior articular surface [1, 3, 5, 11, 15, 16, 21, 26, 29]. Their radiological appearance is also identical, forming a more or less triangular area pointing upwards, devoid of trabecular structure, well delineated and sometimes surrounded by a sclerotic margin that is usually thin [more visible on computed tomography (CT)]. The defect may display signs of moderate chronic expansion, most often predominating inferolaterally, with cortical thinning and bowing (visible on tangential axial radiographs) and no periosteal reaction. This appearance has been described for both cysts and lipomas of the calcaneus [1, 5, 11, 13–15, 20, 22, 23, 26, 29–32]. Central calcifications are also present in more than half of lipoma cases (73% [23]). They are often irregular, sometimes with a chondroid appearance, or suggestive of the periphery of a bone infarct [5, 20, 26, 32], or it has a regular circular appearance (the bull's-eye appearance) [22, 30].

Another point that cysts and lipomas have in common is that they are usually discovered incidentally or associated with generally moderate pain that may relate to microfractures [1, 3, 7, 23, 26, 28]. Pathological fractures are very rare with cysts [4, 15, 16] and even more rare with lipomas [20, 33]. The anterior calcaneal location of cysts and lipomas corresponds exactly to a triangular anatomic area in which the trabecular

network is naturally much sparser (Fig. 1). This area is more or less visible on 70% of lateral radiographs and has a downright pseudo-cystic appearance in 7% of cases [34]. The almost total lack of spongy structure in this area is due to the absence of locally transmitted forces, which pass instead

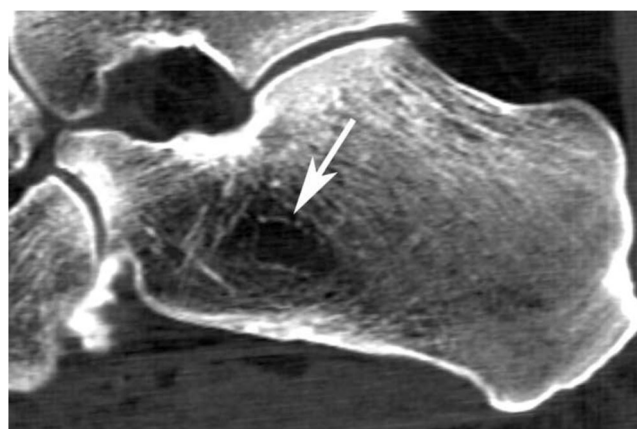


Fig. 1 Calcaneal pseudo-cyst: note the persistence of some spongy bone within the margins of the pseudo-cyst (arrow)

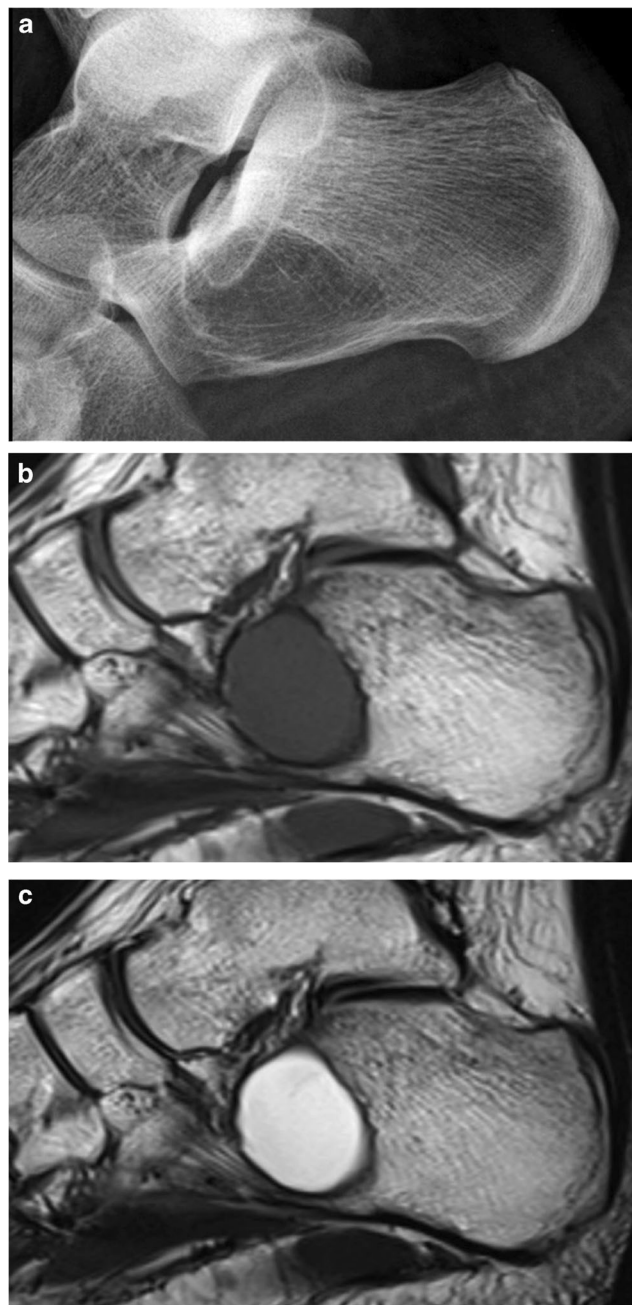


Fig. 2 Calcaneal cyst in a 17-year-old male. Radiography (a) shows a defect in the same area as a pseudo-cyst. The defect contains fluid that has low signal intensity on T1-weighted MRI (b) and high signal intensity on T2 weighting (c)

through the thick trabeculae that surround this area, forming arches between the joints, tuberosity and inferior aspect of the calcaneus [5, 29, 35]. However, unlike cysts and lipomas, pseudo-cysts contain a sparse trabecular structure, and there is no peripheral sclerotic rim, sign of expansion or central calcification.

Differentiating cysts and lipomas on imaging

On CT and magnetic resonance imaging (MRI), the content of cysts and lipomas appears entirely different, which makes differentiating them easy. The fluid content of cysts displays water density on CT [0 to 30 Hounsfield units (HU)]. On MRI, signal intensity is low on T1-weighted images, shows no enhancement after intravenous gadolinium and is high on T2-weighted images (Fig. 2). The peripheral rim has higher density on CT and very low signal intensity on MRI. The fatty tissue contained within lipomas has very negative density values on CT (−60 to −130HU), high signal intensity on T1-weighted MRI images, relatively high signal intensity on T2-weighted images and very low intensity on fat-suppressed

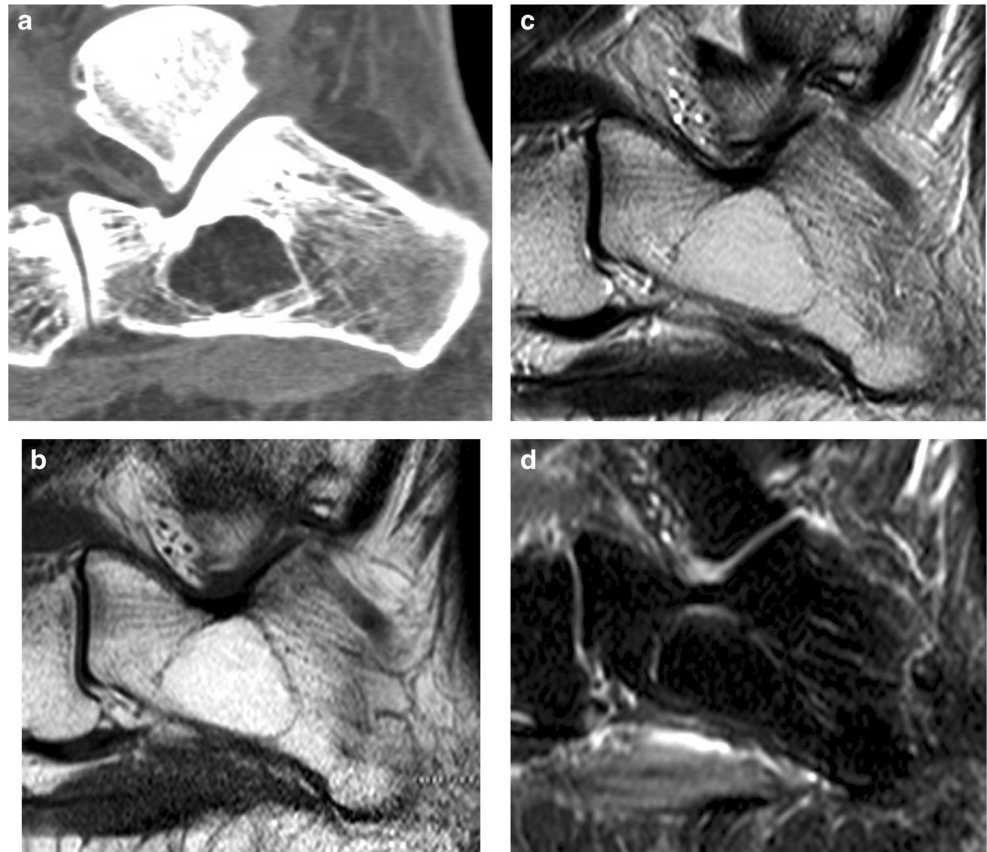
imaging sequences [5, 7, 22, 23, 26, 30, 36] (Fig. 3). The central portion of lipomas may be more heterogeneous and comprise myxoid, cystic and calcified areas (Fig. 4) with CT densities and MRI signals corresponding to these different components [5, 22].

There are some ambiguities in descriptions in the literature. First, some old descriptions are based exclusively on histologically proven cases [31], whereas others include cases diagnosed by plain radiography alone, which cannot differentiate between the two entities [4, 11, 12, 26, 29]. Second, cases whose content includes both fat and fluid have been illustrated in both articles describing lipomas [5, 20, 22, 37] and in others describing cysts [6, 10, 15].

Pathogenesis

The pathogenesis of calcaneal cysts probably differs from that in the long bones because calcaneal cysts are not adjacent to growth plates. Many explanations have been put forward for this particular location, such as that they develop from a congenital remnant in the region of the primary ossification center

Fig. 3 Calcaneal lipoma in a 51-year-old female: CT image (a) shows very-low-density content resembling the sub-calcaneal hypodermic regions. Another lipoma, here in a 61-year-old male: MRI images show the defect's content to have intense signal on T1 weighting (b), relatively high signal intensity on T2 weighting (c) and no signal on fat-suppressed image sequences (d), as with subcutaneous fat



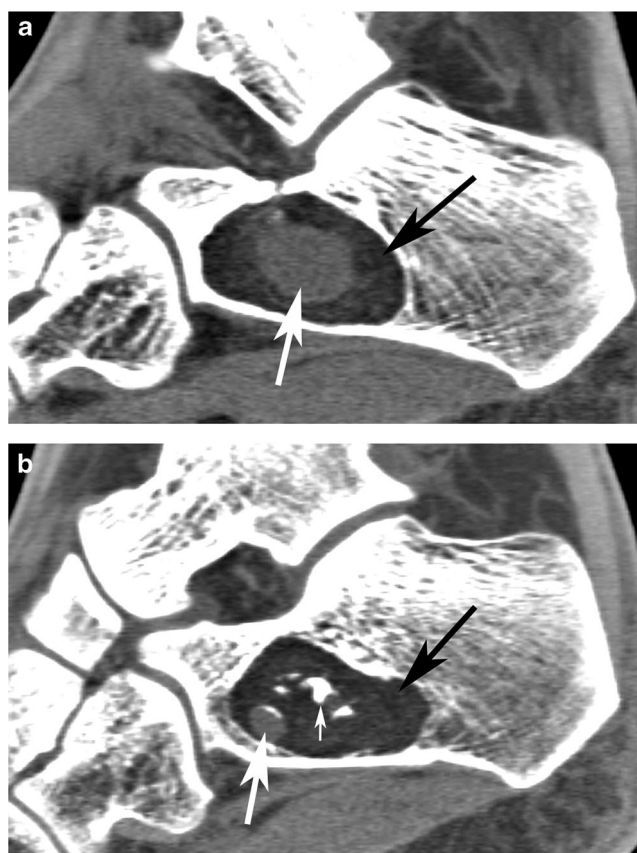


Fig. 4 Lipomas from both calcanei in a 33-year-old male: CT images show very low density content of fatty nature in the cavities (black arrows in a and b). The center of the lesions contains areas of intermediate density compatible with fluid (large white arrows) (in a and b) as well as very-high-density calcific areas (small white arrow in b)

[11], or because of a degenerative process in an area of bone rarefaction [10], or under the influence of mechanical factors (although their typically unilateral nature argues against this theory [1]) or because of organized hemorrhages as suggested by the presence of hemosiderin, cholesterol deposits and histiocytic reactions in the walls [13]. The hypothesis that cysts may result from ischemic degeneration of lipomas has also been widely discussed, especially following Milgram's descriptions (see below) [22, 23, 31]. Recently, Elias et al. [28] suggested that calcaneal cysts may result from ganglion cysts arising from the subtalar joint. They base their theory on the frequent observation in MRI examinations of the calcaneus of small cyst-like lesions just below the anterior margin of the posterior calcaneal articular surface. These cyst-like lesions probably correspond to ganglion cysts communicating with the joint (as demonstrated by MRI arthrography in one of their cases). Such small formations are located in the same area as the upper portion of simple calcaneal cysts, and the calcaneal

cysts may therefore result from ganglion cysts' expanding into the locally very loose bone space underneath [28].

The pathogenesis of calcaneal lipomas has also been widely debated [6, 10, 20, 22, 23, 37]. Many authors believe them to be true benign tumors, the result of adipocyte proliferation [31, 36, 38]. According to Milgram, the changing appearance of their content corresponds to progressive stages of the primary lipoma, namely stage 1: solid tumor composed of mature fat cells without cytological abnormalities; stage 2: appearance of ischemic necrosis due to the lipoma's expansion into a rigid bone cavity, producing mixed areas of viable adipocytes, infarcted fat and calcification; stage 3: more diffuse necrosis, extension of calcifications, myxoid degeneration and formation of cystic cavities [31, 32]. However, the stages described relate to the histological appearance of different cases, and the natural transition from one stage to the next has never been demonstrated. For Murphey et al., it may not be a tumor in the strict sense but rather a hamartoma resulting from a focal excess conversion of hematopoietic marrow into fatty marrow [22]. Other hypotheses have been put forward, including previous ischemic infarction, the final stage of infection, and the result of elevated venous pressure or mechanical stress leading to hypertrophy of marginal trabeculae and central resorption [23, 26]. Lastly, we must mention the hypothesis that calcaneal lipomas may be caused by the involution of cystic lesions [23, 28, 36].

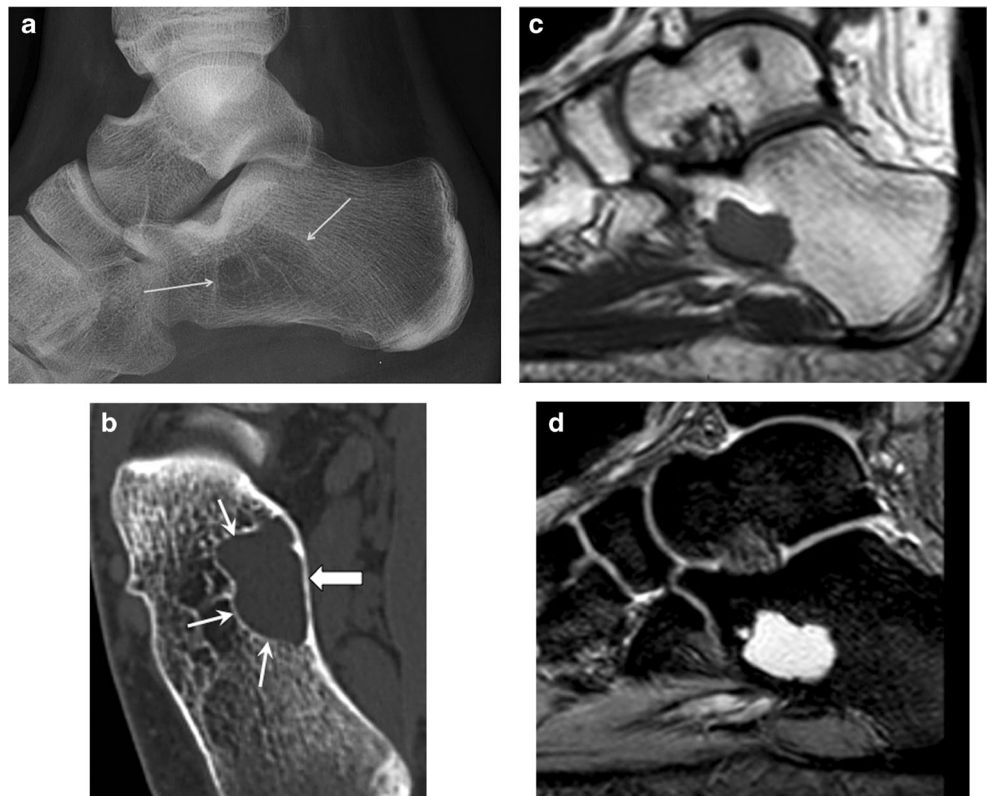
Discussion of the pathogenesis of these two entities is, thus, confused, especially since, as we have pointed out, calcaneal cysts have been described as lipomas and vice versa. Lagier believes that lipomas and cysts of the calcaneus may constitute two types of response to a single mechanical or vascular stimulus [27], while for Mirra, the two terms are even used to describe a single entity with a varying cyst- or lipoma-like histology [6].

Possible relationship between calcaneal cysts and lipomas

Thus, the possibility that the one may evolve into the other has been considered in both directions. Milgram's hypothesis that lipomatous ischemia causes myxoid degeneration and the appearance of fluid content [31] has been taken up by other authors [5, 22, 30], but transformation of this kind of a calcaneal lipoma into a cyst has never been shown [5], while the stability of the fat content of a lipoma was observed by MRI for 18 years in one case, with progression of central calcifications [39].

Reverse progression, from cystic to fatty content, has also been proposed. Hatori et al. described a calcaneal lipoma (on

Fig. 5 Radiograph of the ankle of a 15-year-old male taken during examination of a metatarsal tumor shows a defect (arrows in a) in the anterior portion of the calcaneus. The axial CT image (b) shows the well-delineated appearance of the defect, surrounded by a very thin bony wall (thin arrows). The medial cortex (thick arrow) displays moderate bowing and thinning. Sagittal MRI images show fluid content with low signal intensity on T1 weighting (c) and high signal intensity on T2* weighting (d)



CT, MRI and histological examination) in a 22-year-old female within the site of a cyst (not shown) punctured 8 years earlier [36], although progression was not truly spontaneous. Elias et al. hypothesized that cysts communicating with the joint may regress and that such an involution may be followed by the replacement of the fluid content with fatty marrow [28]. Such a progression from cyst to lipoma would offer a good explanation for their respective prevalences. Calcaneal cysts are observed in adolescents and young adults, whereas lipomas are seen in older subjects. It should also be noted that the appearance of fatty marrow in place of a regressing intraosseous lesion may be observed in other types of disease, such as lymphoma, metastasis or rheumatic sacroiliitis [40–43]. The replacement of a calcaneal cyst's content with fat is exactly what happened in the observation described below.

Personal observation

A defect was incidentally discovered in a 15-year-old male in the anterior portion of a calcaneus (Fig. 5a) during exploration of a metatarsal tumor (florid reactive periostitis, not shown). On CT, the defect had well-defined margins and there were signs of moderate chronic expansion (Fig. 5b). The

attenuation coefficient measured in the lesion on CT was 25 HU. On MRI, the defect's content displayed low signal intensity on T1 weighting (Fig. 5c) with no enhancement after intravenous injection of gadolinium and high signal intensity on T2* weighting (Fig. 5d). Hence, the lesion had the typical appearance of a simple calcaneal cyst. An MRI follow-up conducted 4 years later to check the operated metatarsal tumor showed that a portion of the calcaneal defect had changed and was displaying high signal intensity on T1-weighted images similar to the subcutaneous fat signal (Figs. 6a and 7b). A further MRI follow-up conducted 3 years later (or 7 years after the first examination) revealed that almost the entire defect displayed an extensive fat-like signal intensity on T1 weighting accompanied by a few residual non-fat areas (Figs. 6b and 7c). The content of the initial calcaneal cyst gradually came to resemble that of a lipoma. To our knowledge, such a spontaneous progression has never been shown in the literature.

Conclusion

Several characteristics specific to calcaneal cysts and lipomas argue in favor of a common pathogenesis. The discovery of cystic areas within cavities containing fatty tissue has led to

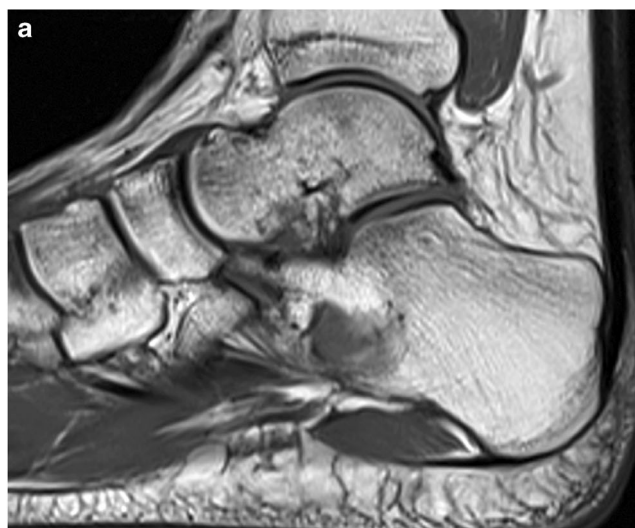


Fig. 6 Sagittal T1-weighted MRI images respectively 4 (a) and 7 (b) years after initial examination: the low signal intensity of the cystic area is gradually replaced by a high signal intensity similar to subcutaneous fat signal

the suggestion that lipomas liquefy because of necrotic processes, but to date no observations have corroborated this hypothesis. The contrary hypothesis, that fat-containing cal-caneal cavities called lipomas may result from the involution of pre-existing cysts, finds indirect arguments in the analogy with intraosseous ganglion cysts and in the respective ages at which lesions are discovered, i.e., later for lipomas than for cysts. Our observation provides an additional argument in favor of the latter hypothesis that a continuum exists between

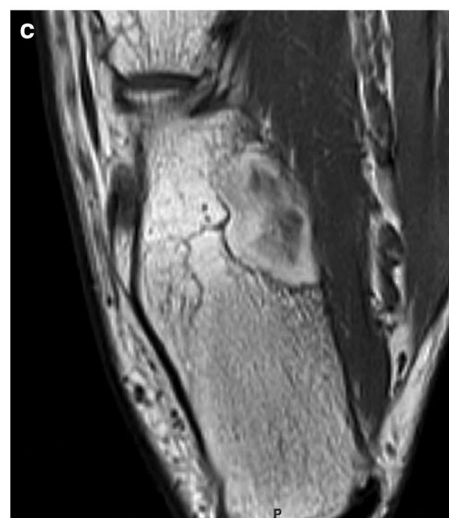
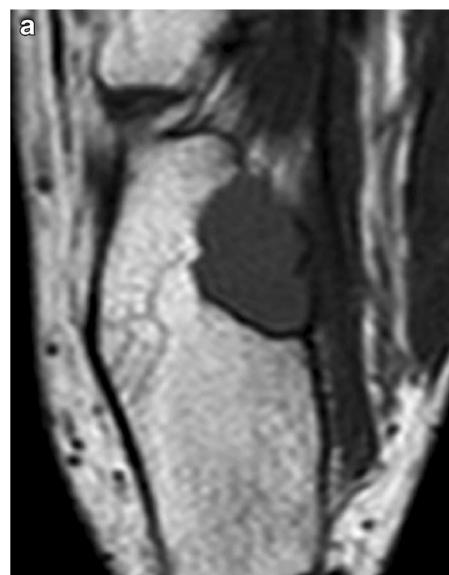


Fig. 7 Recap of the progression on axial T1-weighted MRI. The lesion displays low signal intensity on initial examination (a). Some 4 years later, high signal intensity appears in the peripheral portion of the lesion (arrow in b). Then, 7 years after the initial examination, most of the lesion displays high signal intensity on T1 weighting (c) similar to the signal of subcutaneous or medullary fat

calcaneal cysts and lipomas. Long-term follow-up of other cases will be required to determine whether such progression happens occasionally or is rather the norm.

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Compliance with ethical standards

Conflicts of interest The authors have no conflicts of interest to declare.

Ethical statement The ethics committee of our institution gave its approval for retrospectively presenting our observation.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

References

- Copleman B, Vidoli MF, Crimmings FJ. Solitary cyst of the calcaneus. *Radiology*. 1946;47:142–8.
- Moreau G, Letts M. Unicameral bone cyst of the calcaneus in children. *J Pediatr Orthop*. 1994;14:101–4.
- Glaser DL, Dormans JP, Stanton RP, Davidson RS. Surgical management of calcaneal unicameral bone cysts. *Clin Orthop Relat res*. 1999;360:231–7.
- Hanna SJ, Dasic D, Floyd A. Simple bone cysts of the calcaneus: a report of five cases and a review of the literature. *Foot Ankle Int*. 2004;25:680–4.
- Diard F, Hauger O, Moinard M, Brunot S, Marcet B. Pseudo-cysts, lipomas, infarcts and simple cysts of the calcaneus: are there different or related lesions? *JBR-BTR*. 2007;90:315–24.
- Mirra JM. Calcaneal bone cyst. In: Mirra JM, editor. *Bone tumors clinical, radiologic, and pathologic correlations*. Philadelphia, London: Lea & Febiger; 1989. p. 1235–61.
- Polat O, Sağlik Y, Adigüzel HE, Arikan M, Yıldız HY. Our clinical experience on calcaneal bone cysts: 36 cysts in 33 patients. *Arch Orthop Trauma Surg*. 2009;129:1489–94.
- Dahlin DC, Unni KK. Simple cysts. In: Dahlin DC, Unni KK, editors. *Bone tumors, general aspect and datas of 8,952 cases*. Springfield: Charles C Thomas; 1986. p. 440–1.
- Mirra JM. Simple bone cyst. In: Mirra JM, editor. *Bone tumors clinical, radiologic, and pathologic correlations*. Philadelphia, London: Lea & Febiger; 1989. p. 1325–34.
- Campanacci M. Simple bone cysts. In: Campanacci M, editor. *Bone and soft tissue tumors*. Wien, New York: Piccin Nuova Libreria & Springer-Verlag; 1999. p. 798–811.
- Smith RW, Smith CF. Solitary unicameral bone cyst of the calcaneus. A review of twenty cases. *J Bone Joint Surg am*. 1974;56:49–56.
- Norman A, Schiffman M. Simple bone cysts: factors of age dependency. *Radiology*. 1977;124:779–82.
- Van Linthoudt D, Lagier R. Calcaneal cysts. A radiological and anatomico-pathological study. *Acta Orthop Scand*. 1978;49:310–6.
- Södergard J, Karaharju EO. Kystes du calcaneum, diagnostic et traitement. *Rev Chir Orthop Reparatrice Appar Mot*. 1990;76:502–6.
- Pogoda P, Priemel M, Linhart W, Stork A, Adam G, Windolf J, et al. Clinical relevance of calcaneal bone cysts: a study of 50 cysts in 47 patients. *Clin Orthop Relat res*. 2004;424:202–10.
- Takada J, Hoshi M, Oebisu N, Ieguchi M, Kakehashi A, Wanibuchi H, et al. A comparative study of clinicopathological features between simple bone cysts of the calcaneus and the long bone. *Foot Ankle Int*. 2014;35:374–82.
- Levy DM, Gross CE, Garras DN. Treatment of unicameral bone cysts of the calcaneus: a systematic review. *J Foot Ankle Surg*. 2015;54:652–6.
- Child PL. Lipoma of the os calcis; report of a case. *Am J Clin Pathol*. 1955;25:1050–2.
- Appenzeller J, Weitzner S. Intraosseous lipoma of os calcis. Case report and review of literature of intraosseous lipoma of extremities. *Clin Orthop Relat Res*. 1974;101:171–5.
- Weinfeld GD, Yu GV, Good JJ. Intraosseous lipoma of the calcaneus: a review and report of four cases. *J Foot Ankle Surg*. 2002;41:398–411.
- Aumar DK, Dadjo YB, Chagar B. Intraosseous lipoma of the calcaneus: report of a case and review of the literature. *J Foot Ankle Surg*. 2013;52:360–3.
- Murphey MD, Carroll JF, Flemming DJ, Pope TL, Gannon FH, Kransdorf MJ. From the archives of the AFIP: benign musculoskeletal lipomatous lesions. *Radiographics*. 2004;24:1433–66.
- Campbell RS, Grainger AJ, Mangham DC, Beggs I, Teh J, Davies AM. Intraosseous lipoma: report of 35 new cases and a review of the literature. *Skelet Radiol*. 2003;32:209–22.
- Gonzalez JV, Stuck RM, Streit N. Intraosseous lipoma of the calcaneus: a clinicopathologic study of three cases. *J Foot Ankle Surg*. 1997;36:306–10.
- Greenspan A, Raiszadeh K, Riley GM, Matthews D. Intraosseous lipoma of the calcaneus. *Foot Ankle Int*. 1997;18:53–6.
- Bertram C, Popken F, Rütt J. Intraosseous lipoma of the calcaneus. *Langenbeck's Arch Surg*. 2001;386:313–7.
- Lagier R. Case report 128: lipoma of the calcaneus with bone infarct. *Skelet Radiol*. 1980;5:267–9.
- Elias I, Zoga AC, Raikin SM, Schweitzer ME, Morrison WB. Incidence and morphologic characteristics of benign calcaneal cystic lesions on MRI. *Foot Ankle Int*. 2007;28:707–14.
- Symeonides PP, Economou CJ, Papadimitriou J. Solitary bone cyst of the calcaneus. *Int Surg*. 1977;62:24–6.
- Bagatur AE, Yalcinkaya M, Dogan A, Gur S, Mumcuoglu E, Albayrak M. Surgery is not always necessary in intraosseous lipoma. *Orthopedics*. 2010;33:306.
- Milgram JW. Intraosseous lipomas. A clinicopathologic study of 66 cases. *Clin Orthop Relat res*. 1988;231:277–302.
- Milgram JW. Intraosseous lipomas: radiologic and pathologic manifestations. *Radiology*. 1988;167:155–60.
- Pappas AJ, Haffner KE, Mendicino SS. An intraosseous lipoma of the calcaneus: a case report. *J Foot Ankle Surg*. 2014;53:638–42.
- Sirry A. The pseudo-cystic triangle in the normal os calcis. *Acta Radiol*. 1951;36:516–20.
- Freysschmidt J, Wiens J, Brossmann J, Sternberg A. Borderlands of normal and early pathological findings in skeletal radiography. New York: Thieme Stuttgart; 2003. p. 1020–40.
- Hatori M, Hosaka M, Ehara S, Kokubun S. Imaging features of intraosseous lipomas of the calcaneus. *Arch Orthop Trauma Surg*. 2001;121:429–32.
- Narang S. Gangopadhyay. M. Calcaneal intraosseous lipoma: a case report and review of the literature. *J Foot Ankle Surg*. 2011;50:216–20.
- Hart JA. Intraosseous lipoma. *J Bone Joint Surg Br*. 1973;55:624–32.

39. Richardson AA, Erdmann BB, Beier-Hanratty S, Lautz D, Jacobs PM, Julsrud ME, et al. Magnetic resonance imagery of a calcaneal lipoma. *J am Podiatr med Assoc.* 1995;85:493–6.
40. Malghem J, Vande Berg B, Noël H, Maldague B. Imagerie de la moelle et de ses variations. In: Sintzoff S, Laredo J-D, Caroit M, editors. *Imagerie de l'os et de la moelle osseuse, Getroa Opus XXII.* Montpellier: Sauramps Médical; 1995. p. 123–34.
41. Lien HH, Holte H. Fat replacement of Hodgkin disease of bone marrow after chemotherapy: report of three cases. *Skelet Radiol.* 1996;25:671–4.
42. Malghem J, Vande Berg B, Lecouvet F, Koutaissoff S, Maldague B. Principles of analysis for sacroiliac joints imaging. *JBR-BTR.* 2007;90:358–67.
43. Lecouvet FE, Larbi A, Pasoglou V, Omoumi P, Tombal B, Michoux N, et al. MRI for response assessment in metastatic bone disease. *Eur Radiol.* 2013;23:1986–97.