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Case Report

Calcifying aponeurotic fibroma of the sole of the foot in an elderly patient

Francisco Manuel Garcia Navas[a,*], Nuria Fernandez[b], Alberto Lopez[a], Pedro Montero[c]

[a] Trauma and Orthopaedics Service, General Universitary Hospital, Ciudad Real, Spain
[b] Geriatric Service General Universitary Hospital, Ciudad Real, Spain
[c] Pathological Service, General Universitary Hospital, Ciudad Real, Spain

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ABSTRACT

Calcifying aponeurotic fibroma (CAF) is a rare, locally aggressive fibroblastic lesion that occurs predominantly in the distal extremities of children and adolescents. In the present study, a case of pathologically proven CAF arising in the sole of the right foot of a 84-year-old man is presented. Physical examination revealed a firm, immobile, non-tender mass. Plain radiographs showed a faintly calcified soft tissue mass without bone involvement. Magnetic resonance imaging (MRI) revealed a well circumscribed subcutaneous mass. The patient underwent an excisional biopsy. Histologically, the tumor showed a biphasic pattern, composed of a moderately cellular fibromatosis-like component and irregular calcified areas with polygonal epithelioid cells. There has been no evidence of local recurrence six months following surgery. To the best of our knowledge, this case describes the oldest patient with this condition.

1. Introduction

Calcifying aponeurotic fibroma (CAF), also known as juvenile aponeurotic fibroma [1], is an unusual, benign soft tissue neoplasm that generally occurs in the palm of the hand and sole of the feet [2], although there are an increasing number of reports that place it in other anatomic locations [3]. It usually presents in children and adolescents, with a slight male predominance [4,5]. This tumor classically presents as a painless, slow-growing firm tumor, with the tendency to infiltrate the surrounding soft tissue, due to its CAF has a high rate of local recurrence following surgical excision with a rate of recurrence of 50% [6]. Malignant transformation is extremely rare [6,7]. It should be included in the differential diagnoses of any mass with calcification in the sole of the foot like plantar fibromatosis, nodular fasciitis, bizarre paraosteal osteochondromatous proliferation (BPOP), soft tissue chondroma, synovial sarcoma, and calcified epidermoid [1,3,6]. The age at presentation has been reported to range from birth to 69 years, with a median age of 12 years [6,8]. In the present study, a case of CAF arising in the sole of the foot of an elderly patient is presented, as well as a review of the literature.

2. Case report

A 84-year-old man presented with a 5-year history of a slow-growing, painless mass in plantar medial aspect of the sole of the right foot. The patient related that the mass had slowly increased in size during the past 2 years and it caused some discomfort with shoes. There was no history of antecedent trauma. Pain was aggravated by walking and unrelieved by accommodative orthotics and oral analgesics physical examination revealed a firm, immobile, non-tender palpable mass (Figs. 1 and 2). There were no neurologic or vascular deficits to the foot, and the results from laboratory tests were normal. The patient's past medical history was significant for hypertensión with left ventricular hypertrophy, dyslipidemia, chronic obstructive pulmonary disease, stroke, benign prostatic hyperplasia, paroxysmal atrial fibrillation and valvular heart disease.

Radiographs revealed a faint mass with dense calcifications plantar to the midfoot, with no cortical erosions (Fig. 3).

Ultrasound evaluation indicated a subcutaneous fibrous mass with diffuse hyperechoic areas representing calcification (Fig. 4).

Magnetic resonance imaging (MRI) revealed a relatively welldefined rounded subcutaneous soft tissue mass, 2 × 2.5 cm in size (Figs. 5 and 6). The mass showed low to intermediate signal intensity on T1-weighted images and heterogeneous high signal intensity with small foci of low signal intensity on T2-weighted spectral presaturation with inversion recovery images. Contrast-enhanced fat-suppressed T1-weighted images demonstrated intense heterogeneous enhancement throughout the mass. Based on these findings, the patient was diagnosed with a benign soft tissue tumor.

An excisional biopsy and surgical exploration was performed under

* Corresponding author.
E-mail address: fgarcianavas@gmail.com (F.M. Garcia Navas).

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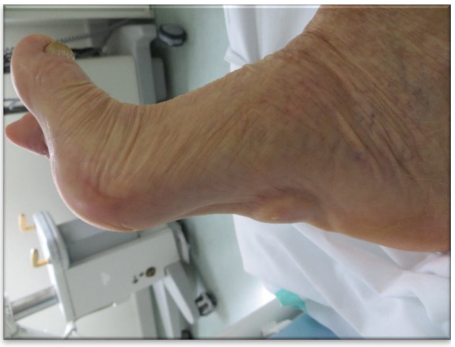

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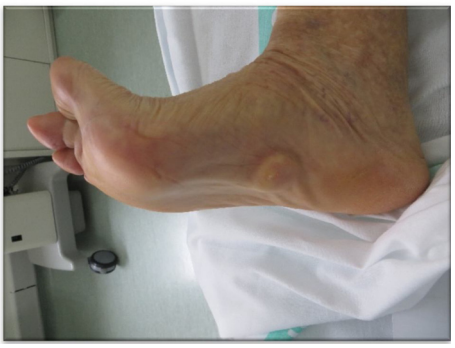

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Figs. 1 and 2. Physical examination showed a nontender, firm mass measuring 2 × 2,5 cm in the sole of the right foot.

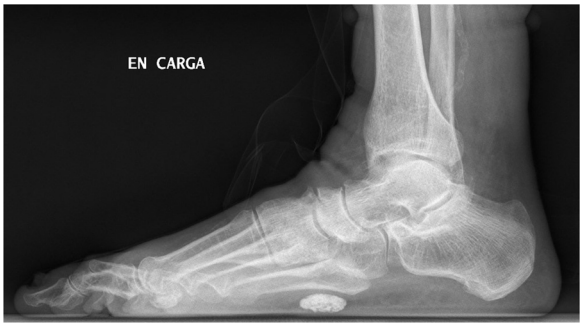


Fig. 3. Radiographs showed an oval-shaped calcified mass in the sole of the foot.

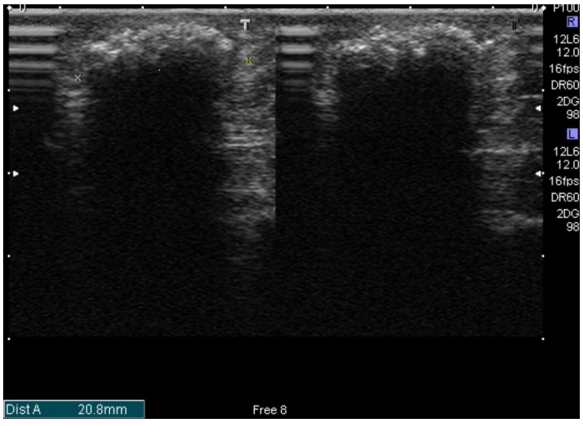
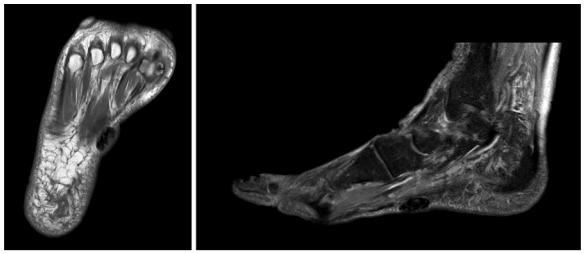


Fig. 4. Ultrasound evaluation indicated a subcutaneous hyperechoic mass representing calcification.

regional anesthesia with tourniquet control. A medial incision was made on the sole of the foot. The mass was noted to be well



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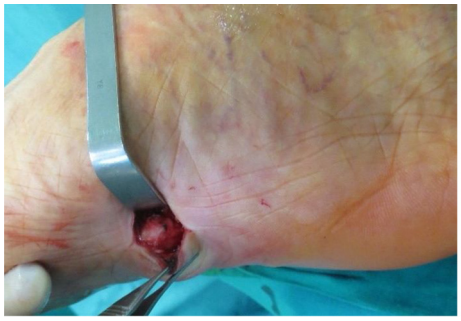
Figs. 5 and 6. MRI of our patient illustrates distinguishing characteristics of a calcified aponeurotic fibroma, including heterogeneous enhancement, speckled calcifications, and ill-defined margins on the plantar surface.



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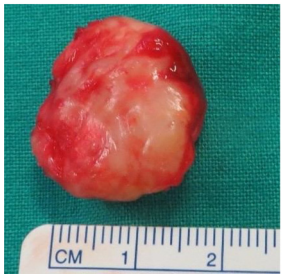

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Figs. 7 and 8. Clinical picture of the case, showing the tumor being resected from the sole of the foot. Intra-operative photographic views of the mass.

circumscribed and it was adherent to the plantar fascia (Figs. 7 and 8); however, the mass was fully excised.

Histologically, the tumor showed a biphasic pattern, composed of a moderately cellular fibromatosis-like component and irregular calcified areas with polygonal epithelioid cells (Figs. 9 and 10). The proliferating cells did not exhibit cellular atypia or evident mitotic figures. The results from the histopathological analysis were consistent with CAF.

Postoperative radiographs were consistent with removal of the mass and did not show any remaining areas of calcification. The

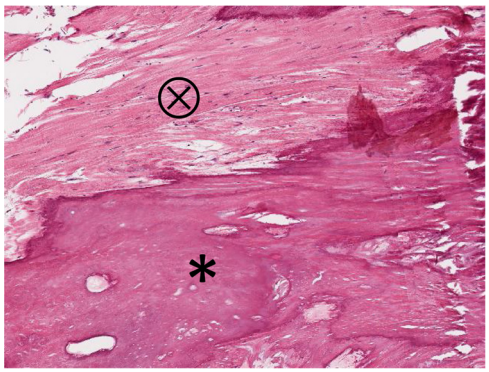

Fig. 9. Picture of the histological examination showing a multinodular pattern with large zones of calcification (*) and fibrosis (⊗). (4×, H&E).

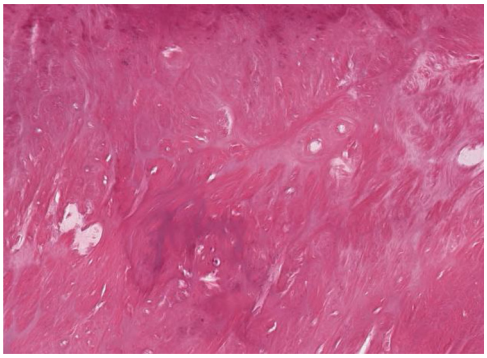

Fig. 10. dystrophic calcification (8×, H&E).

postoperative course was uneventful, and there was no evidence of local recurrence six months following surgery.

Written informed consent for publication was obtained from the patient and this study was approved by the Institutional Review Board.

3. Discussion

Calcifying aponeurotic fibroma (CAF) is a benign fibroblastic tumor localized in subcutaneous tissues. CAF was first characterized as a distinct clinicopathologic entity by Keasbey in 1953 [1]. Since her initial report, more than 150 cases have been documented in the literature, with the vast majority of examples occurring in the hands. Involvement of the plantar aspect of the feet is also well documented, but this event is less frequent. CAF occurs in patients over a wide age range; however, it is most common in children and adolescents [1]. The median ages for male and female patients are 11 and 12 years, respectively [3]. To the best of our knowledge, the present case is the oldest reported case of CAF [8].

CAF grows in a diffuse, poorly circumscribed manner and is often attached to the aponeurosis, tendons or fascia. Complete local excision is the treatment of choice. The histological examination of the lesion reveals two components: (i) fibromatosislike spindleshaped cell elements, and (ii) nodules of calcification, accompanied by more rounded, epithelioid cells [9]. Enzinger and Weiss [6] suggested the existence of biphasic development of the tumor, in the initial phase the tumor has an infiltrative growth and often lacks calcification, which is seen more often in the young, whilst in the later phase the tumor is more compact and nodular and exhibits a more prominent degree of calcification and cartilage formation, as seen in the present case. In cases of scant calcification, differentiating CAF of fibromatosis can be difficult. Advanced lesions exhibit increased soft-tissue density, poorly defined margins, and fine, narrow calcifications on radiographs [7]. These tumours are difficult to diagnose both for clinicians and for pathologists. Because of the fibrous tissue component, its histology is often mistaken for a fibroma or a dermatofibroma [10]. The term aponeurotic has been used because there is a subtle transition from fibrous connective tissue to fibrocartilage as tendons, aponeuroses, and ligaments insert into bone through Sharpey fibers, and the location of many of these tumors suggests an aponeurotic origin [10].

Imaging findings include subcutaneous neoplastic tumors with indistinct borders, showing a tendency of invasion into the surrounding tissues and edematous changes. The tumor is located next to the fascia and tendon sheath. Similar to most CAF lesions, imaging studies of the patient, showed an area of dense calcification with an intricate pattern, without any invasion of adjacent bone [11]. However X-ray imaging is not useful for the diagnosis because it's not specific. No calcification or only smudge-like radiopacities may appear initially. In the other hand, lesions that have been present for years may exhibit large calcified areas [12]. Ultrasound examination excludes the more likely diagnosis of a ganglion indicating a solid mass mainly fibrous with foci of calcification. CT scans are useful in order to determine the calcified areas of the lesion and its association with the adjacent bone. Magnetic resonance imaging (MRI) is the ideal modality for soft-tissue tumors and preoperative planning to evaluate the extent and complexity of the calcification and the margins of the lesion [7,13]. On MRI, heterogeneous combinations of high and low signal intensity on T2-weighted images and intermediate signal intensity on T1-weighted images are noted, with intense contrast enhancement [13,14]. The imaging results from the present case study were consistent with the aforementioned findings. On the other hand, Kransdorf et al. [15] reported that there are no reliable MR criteria for differentiating benign from malignant soft-tissue masses.

The pathogenesis of CAF remains uncertain; however, a afibroblastic/myofibroblastic origin has been suggested [3]. It has been previously demonstrated using immunohistochemistry that the tumor cells usually express vimentin and smooth muscle actin, but are negative for desmin [10], and these results are in accordance with this proposal.

The differential diagnosis should include both malignant and benign tumors, including glomus tumor, giant cell tumor of the tendon sheath, epidermoid cyst, chondromyxoid fibroma, osseous metaplasia of plantar fibromatosis, leiomyoma, calcified epidermoid, fibrosarcoma, fibrous subtype of synovial sarcoma, parosteal osteochondroma and extraskeletal chondroma [16–18]. In older patients, the distinction of CAF from soft tissue chondroma may be difficult. Like CAF, soft tissue chondroma tipically occurs in the finger or hand and feet, with no connection to the underlying bone [19]. However, unlike CAF, soft tissue chondroma is typically better marginated rather than having an infiltrative border. In addition, soft tissue chondroma is less likely than CAF to recur. On MRI, soft tissue chondroma usually appears as a welldefined mass with intermediate signal intensity on T1weighted sequences and high signal intensity on T2weighted sequences [20]. The most important histological finding distinguishing soft tissue chondroma from CAF is that the chondroma contain more abundant (and often lobulated) cartilage, and tend to have only a minor fibroblastic component and the presence of infiltrating fascicles of fibroblasts at the periphery. Metaplastic cartilage may be seen in both lesions but is rarely as well-developed in CAF as that seen in soft-tissue chondroma [21]. It is important not to confuse this lesion with a sarcoma or an aggressive fibromatosis. Despite its infiltrative attitude, the calcifying aponeurotic fibroma has in fact limited growth, which tends to exhaust with the end of body growth, and it does not cause important local disorders (such as joint limitation, vascular or nervous lesions) [6]. Panniculitis ossificans (fibro-osseous pseudotumor) exhibits a distinct mineralization pattern with a fibroblastic proliferation showing varying degrees of atypia, while parosteal (nodular) fasciitis can be readily distinguished by the presence of plump fibroblasts within a myxoid background [12]. Malignant tumors arising on the ankles are rare, but synovial sarcoma and clear cell sarcoma should be differentiated from CAF. They are sometimes misdiagnosed as a benign tumor, due to their slow clinical course and relatively well-defined margin. However, intralesional hemorrhage can be helpful for suspecting synovial sarcoma [22], and clear cell sarcoma may show high signal intensity on T1-weighted image, due to melanocytic differentiation [14].

Postoperatively, 50% of the calcifying aponeurotic fibromas recur locally (and occurs especially in infants and in cases of incomplete resection [1,4,6,7]. There are two reports of a malignant transformation of a calcifying aponeurotic fibroma in the literature [6,7].

In conclusion, in the present case report the imaging findings of CAF with pathologic correlation in an elderly patient are described. Although rare, CAF should be considered in the differential diagnosis of elderly patients with a calcified soft tissue mass, in particular in the sole of the foot.

Conflict of interest

The authors declare that they have no conflict of interest.

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