

Angioleiomyomas Around the Tarsal Tunnel: A Case Report and Review of Literature

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Abstract

Angioleiomyoma is a benign, rare tumor. Our case speaks about a 41-year-old otherwise healthy man presented with a painful mass in his left medial retromalleolar region. It was described as an acute retromalleolar pain irradiated to the plantar aspect of the foot towards the big toe and the second and third toe. A positive Tinel's sign was found with regional numbness and paresthesia. During the surgery the lesion was easily cleavable and it was lying in the supra-fascial region just over the neurovascular bundle. Posterior tibial artery, vein and nerve were located below an intact fascial layer; no signs of compression were detectable. Our case is the second case in the literature describing an angioleiomyoma mimicking a tarsal tunnel syndrome.

Keywords: Angioleiomyoma; tumor; Tarsal tunnel

Introduction

Angioleiomyoma is a benign, rare tumor of smooth non-striated muscle origin that arises from the muscular layer of the small veins and arteries [1]. Angioleiomyomas are usually solitary subcutaneous lesions whose most common site of occurrence is the lower limb (67%), followed by the upper extremity (22%), the head and the trunk [1,2]. A predilection for the lower extremities of middle-aged women is reported, but they can be found throughout the body in male and female adults of all ages [3]. A painful swelling or a tender tumor mass growing slowly in the subcutaneous tissue is the most common complaint. Sub-fascial and intra-muscular occurrence is unusual and only rare cases are reported. Angioleiomyomas are rare in the foot and they account for less than 0.2% of all benign lesions in this region. To our knowledge, only two of angioleiomyomas occurring around the tarsal tunnel are reported in the literature. All these cases were characterized by complaints of tarsal tunnel syndrome, but just in one case symptoms were caused by true nerve compression [4,5]. We present our case of angioleiomyoma sited in the subcutaneous soft tissue above the tarsal tunnel; it was suprafascial and without direct involvement of the neurovascular structures. Our observations may contribute to the definition of the peculiar clinical manifestations of the angioleiomyomas occurring around this anatomical region [4-16].

Case Report

A 41-year-old otherwise healthy man presented with a painful mass in his left medial retromalleolar region. The mass had slowly grown over the previous years and pain was triggered by unintentional local compression. It was described as an acute retromalleolar pain irradiated to the plantar aspect of the foot towards the big toe and the second and third toe. Clinical examination revealed a swelling in the retromalleolar region at the tarsal tunnel (VAS score 8/10). The overlying skin was thinned and dyschromic (Figures 1 and 2). At manual examination it was possible to appreciate a mobile subcutaneous mass. No sensory or motor deficits were clinically detectable and no abnormalities of the hindfoot and forefoot were noticed. Gait pattern was not altered. A positive Tinel's sign was found with regional numbness and paresthesia. No muscle wasting was noticeable and ankle range of motion was normal. Laboratory examinations were normal. Electromyographic assessment did not reveal any abnormality.

Radiologic investigations

Standard X-rays showed no skeletal abnormalities or intra-lesion calcium deposits. Sonography revealed a relatively homogeneous,



Figure 1: Discromic aspect of the lesion.



Figure 2: Swelling at the tarsal tunnel.

solid, oval mass with hypoechoic echo texture and a small amount of posterior acoustic enhancement; located in the subcutaneous layer. No internal calcifications were seen, and color Doppler evaluation revealed considerable hypervascularity (Figures 3 and 4).

Magnetic Resonance Imaging (MRI) defined the diameters of the mass: measuring 22 mm × 12 mm × 20 mm along its main axes. The lesion had well defined margins and was confined in the subcutaneous tissue overlying the tarsal tunnel in close proximity to the neurovascular

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bundle with no signs of continuity or compression of the posterior tibialis nerve (Figures 5 and 6).

Surgical procedure

The patient was positioned supine after peripheral anesthesia by femoral and sciatic nerves block. Skin incision ran longitudinally in the retro-malleolar region over the palpable mass. The incision was slightly curved with its concavity towards the medial malleolus so as to lie over the neurovascular bundle. Through blunt dissection the mass was identified in the subcutaneous tissue. It appeared well demarcated by a defined capsule without adhesions to the surrounding tissues. Marginal excision was carried out and the surgical specimen was sent for histology. The lesion was easily cleavable and it was lying in the supra-fascial region just over the neurovascular bundle. Posterior tibial

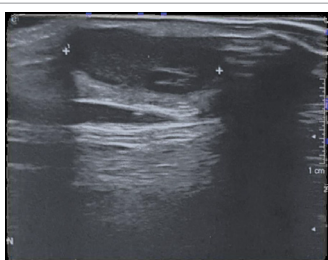


Figure 3: Pre-op ultrasound imaging showing the maximum diameter of the lesion.



Figure 4: Doppler showing a high vascularization of the swelling.



Figure 5: MR coronal cut of the lesion.

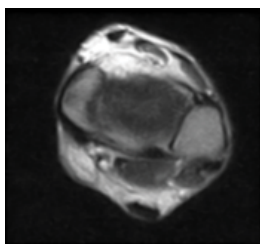


Figure 6: MR transverse cut of the lesion.

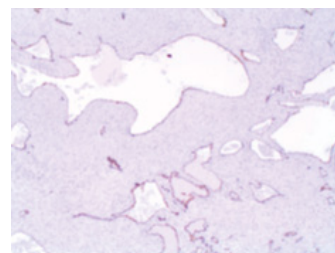


Figure 7: Immunohistochemistry for CD34.

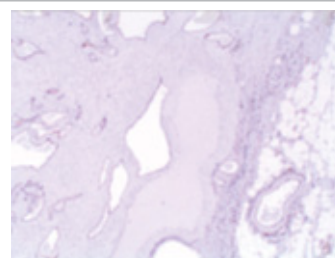


Figure 8: Immunohistochemistry for CD34.

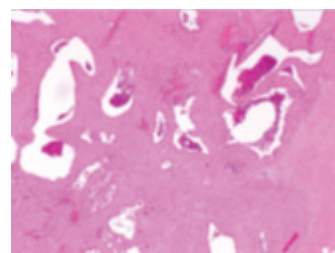


Figure 9: Hematoxylin and eosin stain.

artery, vein and nerve were located below an intact fascial layer; no signs of compression were detectable.

At 6 months follow-up there was complete relief in symptoms and no recurrence (VAS score 0/10); the full weight-bearing and return to normal ADL had been permitted by 5 months.

Histology interpretation

Macroscopic examination revealed a 2 cm well defined lesion. Histologically it was composed of slit-like and dilated vessels sometimes with clots. These structures were lined by a thin CD34 positive endothelial cells (Figures 7 and 8) and surrounded by a closely compacted spindle-shaped cells with eosinophilic cytoplasm arranged in bundle pattern (Figure 9). The latter component was positive for Smooth Muscle Actin and HHF35. There were no atypical cells and mitosis count was <1/10 HPF.

The morphological and immunohistochemical features were of a solid (capillary) angioleiomyoma without any sign of malignancy (Figure 10).

Discussion

Angioleiomyomas arising in the foot are generally described as solitary painful superficial masses wholly localized into the subcutaneous tissue. They are small, nodular and highly vascularized. Exceptionally they can develop in deeper regions and grow up to dimensions exceeding the usual limit of 2 centimeters.

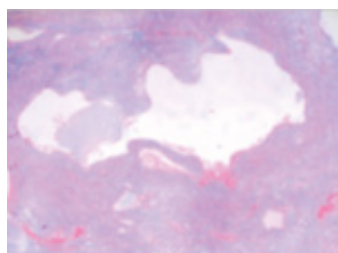


Figure 10: Immunohistochemical features were of a solid angioleiomyoma without any sign of malignancy.

In our review of the literature we found only another two cases of angioleiomyoma of the foot that presented symptoms such as those of a tarsal tunnel syndrome. The first one has been described from Cancilleri et al. [13] within a series of four unusual cases of the syndrome. In their case the lesion had a close relationship with the posterior tibial nerve inside the tarsal tunnel. Although they report some uncertainties in detecting and locating the tumor at the pre-operative MRI assessment, at surgery they found it in the nerve continuity, with signs of nerve compression. Pain was described as irradiating to the plantar aspect of the foot down to the first three toes and, although electromyography was negative, it is likely that it was a genuine tarsal tunnel syndrome caused by a mechanical nerve irritation due to the expanding mass. Hamoui et al. reported the other case. It concerns of an angioleiomyoma that occurred in the subcutaneous tissue overlying the tarsal tunnel. Symptoms were described as mimicking a tarsal tunnel syndrome. A positive Tinel's sign was reported with numbness and paresthesia on the plantar surface of the foot in the territory of the medial plantar nerve. The tumor was small (8 mm × 5 mm), located superficially, outside the tarsal tunnel, but from the description reported it is not clear how close to the posterior tibial nerve the mass was. Pain is a characteristic of angioleiomyomas and it occurs in 67% of the cases [17]. It is triggered by local pressure or rise of temperature [7,8,13]. It can increase with pregnancy and duration of the disease [7,8,13]. However, the size of the lesion does not correlate with symptoms [17]. Freedman and Meland [17] in 39 angioleiomyomas did not report a significant difference in the mean size of the tumor between the symptomatic and asymptomatic groups. Besides, it is a relatively common observation that also small angioleiomyomas can be extremely painful [5]. Pain is the main symptom and it is exceptionally accompanied by neurological findings. They have been described only in few cases of angioleiomyomas with intra-neural occurrence [4,5].

Some hypotheses have been suggested to give an explanation of the cause of pain in angioleiomyoma. proposed that pain was the result of irritation of nervous fibers contained in the capsule and in the interstice of the tumor. Claimed that the violent contraction of smooth muscle cells was the cause of pain. None of the above hypothesis however can explain the occurrence of numbness and paresthesia along the territory of a neighboring nerve in the absence of a mutual close anatomical relationship.

Conclusion

Our case is similar to that reported by Hamoui et al. [15], and would be the second case describing the peculiarity of an angioleiomyoma mimicking a tarsal tunnel syndrome. In both cases the lesion occurred in the soft tissue overlying the tarsal tunnel and in both cases pain was accompanied by neurological findings erroneously suggesting a nerve entrapment. It is difficult to give an explanation to the phenomenon observed and we simply record it. The only relevant discrepancies we noticed between our case and that Hamoui et al. regard the size of the mass and its position. In our case the mass was clearly placed straight over the neurovascular bundle and it was considerably bigger (22 mm as opposed to that of Hamoui et al. 8 mm) because of the patient delay in referring to medical care.

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