TUMOUR AND SEPSIS

A rare case of angioleiomyoma around the ankle: case report and review of literature

Nakale NT[1], Saragas NP[2], Ferrao PNF[3], Strydom A[4]

[1] BSc(UNAM), MBChB(UKZN), Registrar, Orthopaedic Surgery Department, University of Witwatersrand, Johannesburg, South Africa
[2] MBBCh(Wits), FCS(SA)Ortho, MMed(Ortho Surg)(Wits), The Orthopaedic Foot and Ankle Unit, University of the Witwatersrand and Netcare Linksfield Hospital, Johannesburg, South Africa
[3] MBChB(Pret), FCS(SA)Ortho, The Orthopaedic Foot and Ankle Unit, University of the Witwatersrand and Netcare Linksfield Hospital, Johannesburg, South Africa
[4] MBBCh(Wits), FC Ortho (SA), MMed(Ortho Surg)(Wits), The Orthopaedic Foot and Ankle Unit, University of the Witwatersrand and Netcare Linksfield Hospital, Johannesburg, South Africa

Corresponding author: Dr NT Nakale, Division of Orthopaedic Surgery, Wits Medical School, 7 York Road, Parktown, 2193; Email: tn.nakale@gmail.com, Cell: +27728676140

Abstract

Angioleiomyoma is a slow-growing benign tumour that originates from the tunica media layer of vessel walls. It represents 4.4–5% of all benign soft tissue tumours and 0.2% of all tumours in the foot and ankle. Excisional biopsy of the tumour is both diagnostic and curative, with a low recurrence rate reported in the literature. Malignant transformation has been described in 1% of cases. We present a case of a 67-year-old female diagnosed with angioleiomyoma at the lateral malleolus.

Level of evidence: Level 5

Key words: angioleiomyoma, benign ankle, soft tissue tumour, vascular tumour

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Introduction

Angioleiomyoma of the foot and ankle is a rare benign soft tissue tumour of unknown aetiology originating from blood vessel walls typically affecting middle-aged women between the fourth and sixth decade. Angioleiomyoma should be considered as part of the differential diagnosis of painful masses around the foot and ankle as it has the potential of malignant transformation.

Case report

A 67-year-old female presented with acute pain, of three-month duration, over the right lateral aspect of the ankle. She described the pain as an extreme burning sensation, especially when touching the small nodule on the side of her leg. She described no prior history of injury. Clinical findings were those of a small hard nodule on the posterior border of the fibula approximately

5 cm proximal to the tip of the lateral malleolus *(Figure 1)*. This area was extremely sensitive to light touch, with a positive Tinel sign. The rest of the examination was unremarkable.

Ultrasound *(Figure 2)* reported a well-circumscribed 4.8×4.4×2.2 mm hypoechoic mass in the subcutaneous tissue overlying the right distal fibula with moderate to marked vascular flow *(Figure 3)*. A nerve entering the tumour was identified, measuring 1.7 mm and appeared to be thickened on exiting the tumour, with a diameter of 4.3 mm.

The patient was advised to have an excisional biopsy of the mass, to which she agreed. The tumour was excised in its entirety through a direct approach *(Figure 4).* The mass was found to be in the subcutaneous tissue with no obvious nerves or vessels leading from it.

The tumour measured 9×5×1 mm macroscopically *(Figure 5).* Microscopic histopathological evaluation revealed a circumscribed but encapsulated proliferation of smooth muscle bundles arranged in intersecting fascicles *(Figure 6).* These were centred around and spanned thick-walled blood vessels within the lesion *(Figure 7).* With no cytologic atypia or malignancy present, these features represented a benign angioleiomyoma.

The wound healed at two weeks and the patient returned to normal activities at four weeks. At one-year follow-up, the patient's symptoms resolved completely with no post-operative complications reported.

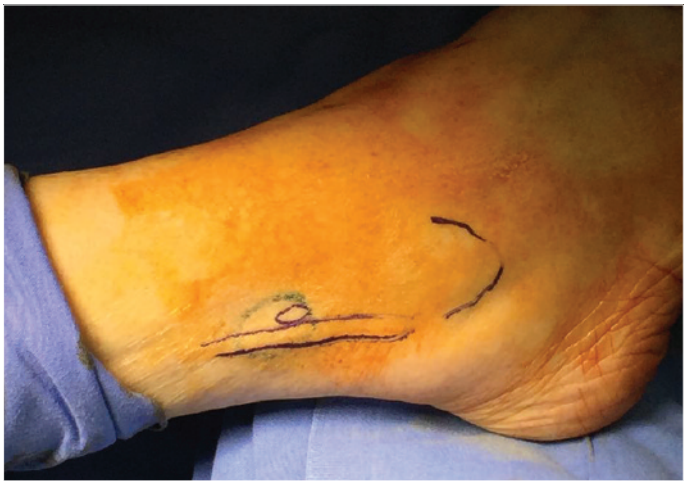

Figure 1. Clinical picture showing the location of the mass in relation to the lateral malleolus

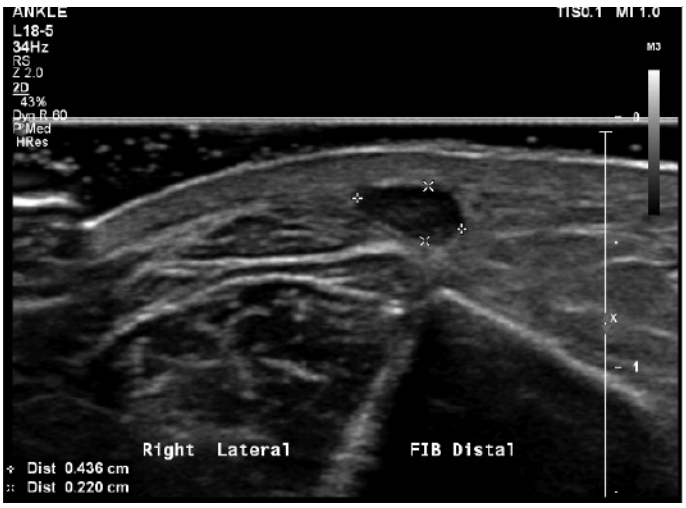

Figure 2. Sonar image demonstrating a hypoechoic mass measuring 4.8×4.4×2.2 mm

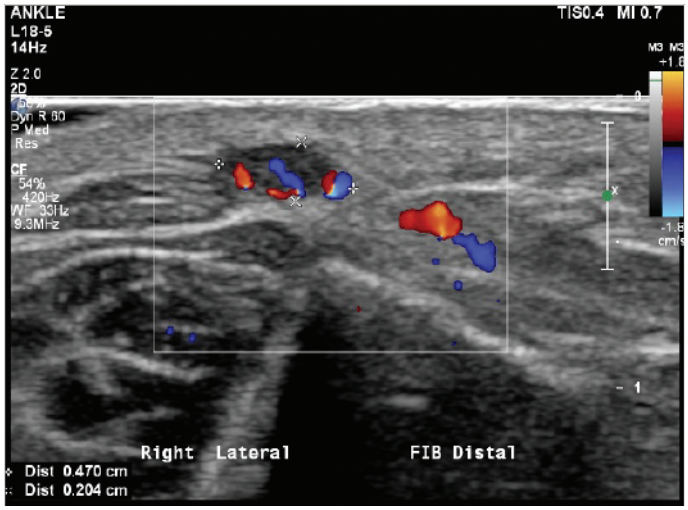

Figure 3. Duplex sonar image highlighting vascular flow to and from the mass

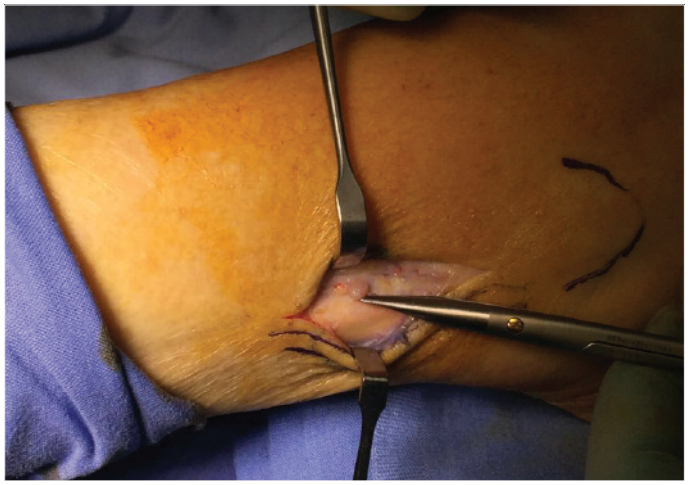

Figure 4. Excision of mass via lateral incision

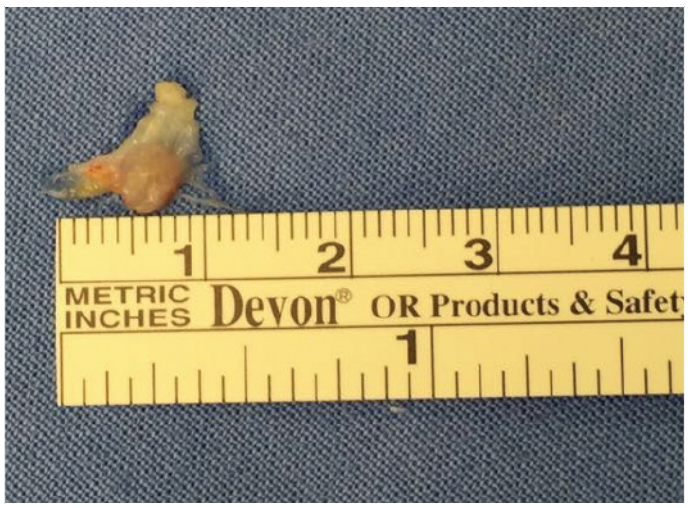

Figure 5. Macroscopic appearance of the mass post excision

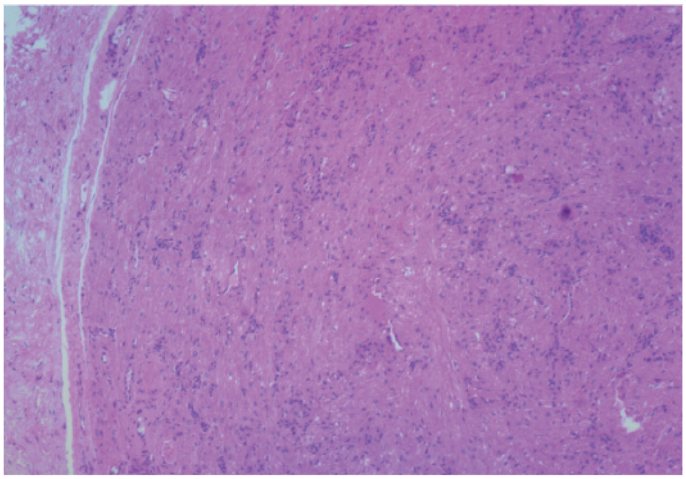

Figure 6. Histopathology slide: Low power (10×) view demonstrating a circumscribed lesion composed of spindled cells with eosinophilic cytoplasm

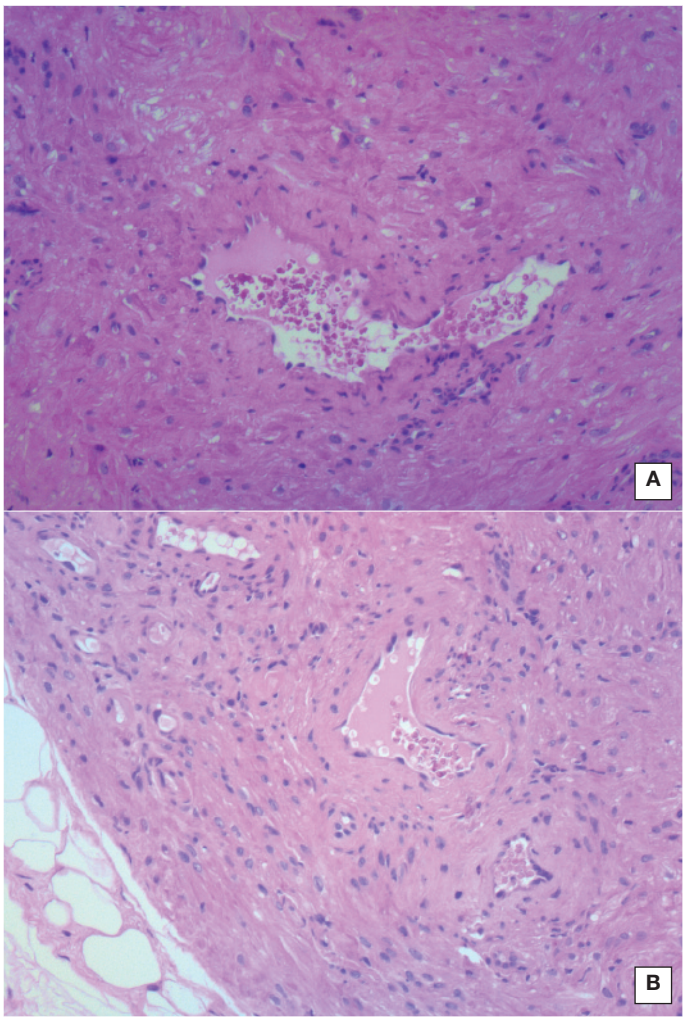

Figure 7. A and B Histopathology slides: High power (20×) demonstrating thick-walled blood vessels from which the spindle cells of the lesion originate

Discussion

Angioleiomyoma is a benign slow-growing tumour that originates from the tunica media layer of smooth non-striated muscle in blood vessel walls. Angioleiomyomas are quite rare, representing 4.4–5% of all benign soft tissue tumours.[1,2]

Angioleiomyomas have a predilection for the lower limb and are commonly found in women between the fourth and sixth decade.[1,3,4] The majority of cases of angioleiomyoma reported in the English literature are in the form of case reports and case series, some of which are summarised in *Table I.*

The reported incidence of angioleiomyoma around the foot and ankle is 0.2% of all tumours of the foot and ankle.[5,6] Craigen and Anderson reported three cases of angioleiomyoma out of the 161 cases of smooth muscle tumours in the foot and ankle over a 10-year period.[7] Macdonald *et al.* reported a 2% incidence of angioleiomyomas in 101 cases of foot masses during a four-year period.[8] Azevado *et al.* reviewed 72 cases of foot and ankle tumours treated at a single institution over a ten-year period and found that 5.5% of these tumours were angioleiomyomas.[9]

The aetiology of angioleiomyoma remains unknown, and multiple factors have been proposed ranging from minor trauma, venous stasis, pregnancy, oestrogen therapy and hormonal alteration.[2,3,10-12] A genetic predisposition has also been proposed in individuals with an autosomal dominant inheritance.[13]

Vascular malformations from arteriovenous anastomosis or haematoma formation following local minor trauma may also result in an angioleiomyoma. Up to 60% of cases will present with a painful, solitary subcutaneous nodule which may be associated with discomfort with shoe wear.[1] The tumour may be asymptomatic for years prior to making the diagnosis.[1] Presence of pain may be due to irritation of an involved nerve, smooth muscle contraction or blood vessel spasm secondary to ischaemia.[3] The tumour rarely exceeds 2 cm in size.[3,4,14,15]

Clinical findings and imaging are supportive, with the diagnosis being confirmed on histopathological analysis of the excisional biopsy specimen. However, it is still important to consider other possible causes for a painful foot mass including glomus tumour, fibroma, haemangioma, lipoma, schwannoma and giant cell tumour *(Table II).* A few cases of a calcified angioleiomyoma, seen on plain radiographs as a calcified mass located at the site of symptoms, have been reported.[4,14,16] The tumour may exert a compressive effect on bone which is visible on X-ray, otherwise plain X-rays are usually normal.[17]

Ultrasound features only show a homogenous structure with well-defined margins. MRI shows a smooth muscle mass, with numerous vessels within it, that is hyperintense on the T2-weighted sequence. There is strong enhancement after contrast media, consistent with abundant vascularity of the tumour.[3,4]

The classic morphological appearance is that of a sharply circumscribed but encapsulated mass usually measuring less than 2 cm in diameter.[16] Microscopically there are bundles of mature smooth muscle oriented around blood vessels showing solid growth of compact spindle cells with ovoid nuclei. Mitotic activity is usually sparse to absent.

Hachisuga *et al.* described three histological types, which was modified by Katenkamp *et al.* who added a fourth type *(Table III).*[1,18] Secondary changes may also be present including myxoid change, hyalinisation or calcification.[4,14,16] Excision biopsy is both diagnostic and curative, as recurrence is very rare. Malignant transformation has been reported in 1% of cases, into angioleiomyosarcoma.[1,19-22]

Conclusion

Angioleiomyoma is rarely encountered in the foot and ankle. However, it remains an important differential diagnosis for a painful foot and ankle soft tissue tumour, due to the risk of malignant transformation. Excisional biopsy is both diagnostic and curative.

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Table I: Summary of some cases of angioleiomyomas of the foot and ankle reported in literature

Author	Age (years)	Sex	Location	Symptom	Duration
Craigen (1991)[7]	62	F	Foot plantar	Pain with weight bearing	18 months
	47	F	Foot lateral	Painful mass	6 years
Santucci (2000)[15]	52	F	Foot dorsum	Occasional painful mass	3 years
Yates (2001)[17]	49	M	Great toe	Painless mass	14 years
	78	F	Forefoot plantar	Painless mass	12 years
	41	F	Third toe	Nerve compression symptoms	5 years
Murata (2007)[14]	58	F	Heel	Painless mass	5 years
	63	F	Heel	Painless mass	1 year
Maheshwari (2008)[4]	35	M	Dorsum foot	Painful mass	3 months
	75	F	Third toe	Painful mass	30 years
	73	F	First web space	Shoe wear difficulty	6 years
	73	M	Foot	Painful mass	6 months
Hamoui (2010)[3]	64	F	Tarsal tunnel	Pain	Several years
Gajanthobi (2013)[23]	44	M	Foot plantar	Painless mass	3 months
Blalock (2015)[16]	71	F	Lateral foot	Painless mass	15 years
Lopez (2014)[24]	39	F	Lateral ankle	Painful mass	3 years

Table II: Differential diagnosis of angioleiomyoma

Glomus tumour
Fibroma
Haemangioma
Lipoma
Schwannoma
Giant cell tumour
Ganglion

Table III: Hachisaga histological classification[1,18]

Type	Characteristics
Solid (capillary) 66%	Closely compacted smooth muscle and many slit-like vascular channels (66%)
Venous 23%	Thick, vascular walls
Cavernous 11%	Vascular channels dilated with less smooth muscles
Mixed* (solid/venous) 43%	Mixed

*Katenkamp modification

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