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CASE REPORT

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Plantar forefoot pain due to vascular leiomyoma: a case report

Dor plantar no antepé devido a leiomioma vascular: relato de caso

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ABSTRACT

The diagnosis of plantar forefoot pain is very extensive. Plantar pain in the forefoot usually occurs due to mechanical aetiologies such as metatarsalgia, stress fractures and nerve damage. Tumours are a rare cause of plantar pain in the forefoot. We report the case of a patient with chronic pain in the plantar region of the forefoot due to a vascular leiomyoma.

Level of Evidence V; Diagnostic Studies; Expert Opinion.

Keywords: Leiomyoma; Angiomyoma; Forefoot, human; Pain.

RESUMO

O diagnóstico de dor plantar no antepé é muito extenso. A dor plantar no antepé ocorre usualmente devido a etiologias mecânicas como metatarsalgia, fraturas por estresse e lesões nervosas. Tumores são uma causa rara de dor plantar no antepé. Apresentamos um relato de caso de uma paciente com dor crônica na região plantar do antepé devido a um leiomioma vascular.

Nível de Evidência V; Estudos Diagnósticos; Opinião de Especialista.

Descritores: Leiomioma; Angiomioma; Antepé humano; Dor.

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INTRODUCTION

The differential diagnosis of plantar forefoot pain is extensive. Plantar pain in the forefoot region is usually due to mechanical aetiologies, including metatarsalgia, plantar fasciitis, stress fractures and Morton's neuroma. Tumour is a rare cause of pain in the plantar region of the forefoot. A vascular leiomyoma or angioleiomyoma is a rare benign smooth muscle tumour that originates from the medial tunica muscularis of the veins. It can occur anywhere in the body where smooth muscle is present and can be found in the dermis, subcutaneous fat or fascia. This tumour causes pain in approximately 60% of patients, and pain is usually more severe in tumours of the lower extremities (50% to 70%) than in tumours in the upper extremities, such as the head or neck[1-5].

We report a case of plantar forefoot pain with increased local volume and functional limitation caused by a vascular leiomyoma.

Work performed at the Santa Casa de Misericórdia de Porto Alegre, Porto Alegre, RS, Brazil.

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CASE REPORT

This study was approved by the Research Ethics Committee with registration in the Brazil Platform under CAAE number: 93547818.8.0000.5335.

An 81-year-old patient complained of pain and discomfort and noted the appearance of bulging in the plantar region of the right forefoot for 2 years. There was no history of previous local injury or any history of trauma in the area. The mass was increasing in size and making walking painful. The patient had difficulty wearing closed shoes due to the local volume between the second and third toes and had paraesthesia in the second and third toes. The patient presented hypertension as the only associated comorbidity and did not have renal or endocrinological disease.

Clinically, the patient presented a 2.5-cm mass in the plantar region of the forefoot between the second and third toes, with a hard consistency and discomfort on palpation (Figure 1), which caused apparent dorsal subluxation of the metatarsophalangeal joint of the second toe. The lesion was not evident on X-ray examination (Figure 2). Nuclear magnetic resonance imaging showed a nodular ovoid mass with signs of inflammatory changes in the plantar fat of the right foot (Figure 3).

The tumour (size 3.0 × 2.2 × 1.5cm) was excised in March 2017 via a plantar approach between the second and third toes with resection of a well-defined spherical

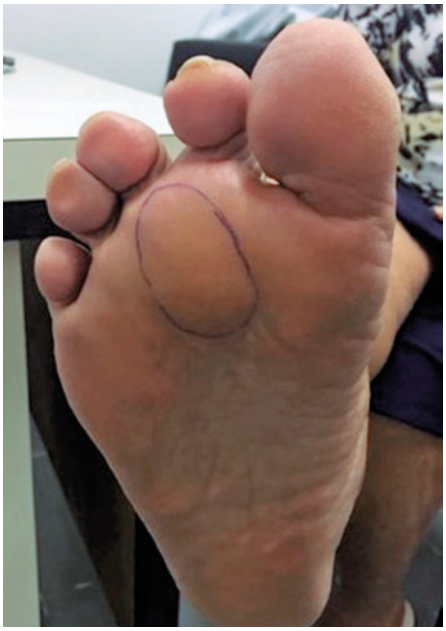

Figure 1. Plantar location of the lesion on the right foot.
Source: Author's personal archive.

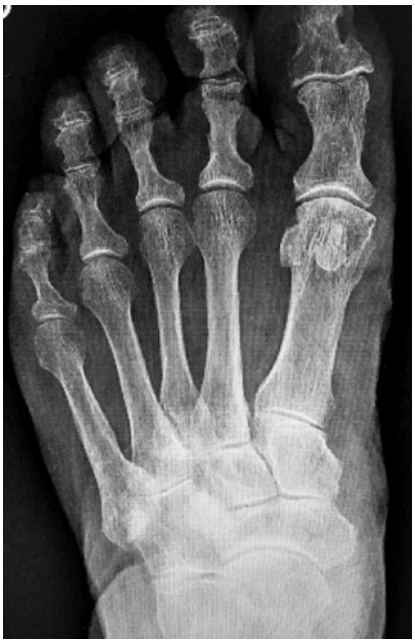

Figure 2. Anteroposterior radiograph of the right foot without signs of lesion calcification.
Source: Author's personal archive.

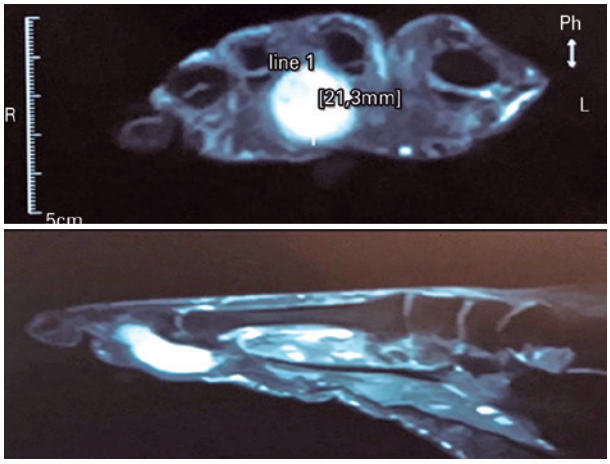

Figure 3. Sagittal and coronal views of the right foot on T2 magnetic resonance images showing hyperintense signal in the forefoot, with an increase in volume between the second and third toes.
Source: Author's personal archive.

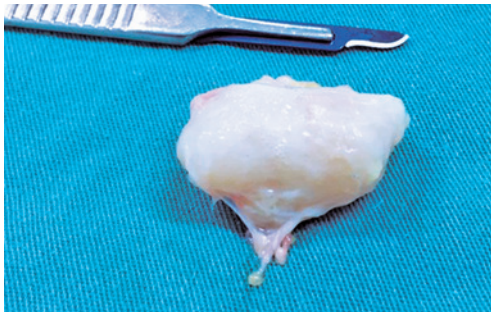

Figure 4. Aspect and size of the lesion.
Source: Author's personal archive.

mass. A pathological examination revealed a mass with nodular, yellowish-grey and elastic tissue (Figure 4). After surgery, the pain, paraesthesia and deformity in the forefoot improved fully, and the patient was able to walk again and wear closed shoes without pain.

DISCUSSION

Vascular leiomyoma usually develops late in life, typically between the fourth and sixth decades of life, and mainly affects women. Lower limb tumours occur twice as often in women as in men, and upper limb tumours occur more often in men than in women. The tumours are usually located in the subcutaneous tissue and, less frequently, can affect the dermis more deeply. Acral sites such as the fingers, hands and feet are rarely documented[5,6]. Vascular leiomyoma is characterised by a painful or painless subcutaneous nodule. Pain is present in approximately 50% of patients. The pain is described as acute and intermittent. It is stimulated even by subtle touch of the affected region, exposure to wind, temperature changes and other imperceptible stimuli. It may also be exacerbated during pregnancy and the menstrual period[4,5,7]. Sometimes symptoms may mimic local nerve compression when the tumour is located near peripheral nerves[2,8]. Even though an association exists between histological signs of myxomatous changes and a clinical history of pain, the aetiology of pain remains unknown. The pain could be the result of local tissue anoxia; however, it has been speculated that the pain results from the compression of cutaneous nerves by the tumour. Although the pain is often paroxysmal and precipitated by mild touch and temperature changes, another possible cause includes increased nerve pressure due to vasodilation resulting from increased venous stasis[7].

Clinically, the mass is spherical and well-defined, and most masses are less than 2cm in diameter. This tumour, however, is rarely diagnosed prior to surgery, even with these characteristic clinical manifestations[4]. Internal calcifications in leiomyomas are common. Generally, calcifications signify tumour degeneration[7]. Radiographically, 3 patterns of calcifications have been reported: small diffuse patches or standard fine grain calcifications, plate-like calcifications and large mulberry-like calcifications[5]. Nuclear magnetic resonance T2-weighted images supporting vascular leiomyoma show a mass with a mixture of areas that present both hyperintense and isointense signal relative to skeletal muscle and reveal a hypointense halo; these images correlate with the histopathological findings of smooth muscle, vessels, fibrous tissue, intravascular thrombi and the fibrous capsule[4].

Differential diagnoses should include benign and malignant tumours as well as non-neoplastic masses associated with calcification. Among benign tumours, haemangioma may have typical circular calcifications with lucent nuclei, whereas other tumours may be associated with collections of circumscribed acute peripheral calcifications (myxoma, xanthoma, hamartoma, lipoma) or small and large fine grain calcifications (pilomatrixoma). Malignant tumours such as sarcoma, synovial sarcoma, rhabdomyosarcoma, malignant fibrous histiocytoma or rabdomiosarcoma can lead to haemorrhage and necrosis with secondary calcifications with irregular appearance and weak amorphous areas of increased density on radiographs. Extra-skeletal chondrosarcomas or osteosarcomas may show irregularities, poor marginal deposits of calcification and ossifications that differ from other calcification patterns[9].

Non-tumoural causes include tumour calcification and other calcified lesions with tumour patterns located in superficial soft tissues, which may be associated with chronic renal disease with secondary hyperparathyroidism, Burnett's syndrome, hypervitaminosis D and patients with excessive osteolysis and calcium mobilisation due to destructive neoplasms and bone infections. Scleroderma, dermatomyositis, CREST syndrome, systemic lupus erythaematosus and overlapping syndromes can develop skin calcinosis known as calcinosis circumscripta, calcinosis universalis and dystrophic calcifications, which can result from tissue damage secondary to an injury, ischaemic necrosis or the necrotic infectious process[5]. Diagnosis is only possible by histological examination, with confirmation by immunohistochemical examination[8,10]. Three histological subtypes have been described according to their predominant component: solid (capillary), cavernous or venous. Each histological subtype presents different clinical characteristics, according to the location of the lesion and the gender of the patient, and the solid form is the most common subtype. The solid form is three times more common in women and often involves the lower extremities. It is usually accompanied by pain[7,9]. The cavernous subtype is four times more common in men and mainly affects the head and upper extremities[10].

Local recurrence or malignisation of the lesion after excision are extremely rare[3]. A simple marginal excision is generally more than adequate for the treatment of angioleiomyoma[7,10].

No article addressing this pathology has been found in the national literature.

CONCLUSION

We can conclude that, as presented in the case report, even though angioleiomyoma is a rare soft tissue tumour, it has a typically nonspecific presentation. Patients with chronic pain are a major challenge for surgeons when the cause of their pain is difficult to diagnose, and because angioleiomyoma is a rare and well circumscribed lesion, it is perfectly manageable with surgical treatment[1]. Although rare, vascular leiomyoma should be included in differential diagnoses of subcutaneous bulging, especially at the extremities, as it is a curable pathology through surgery. Excision of the lesion and histopathological examination provide a definitive diagnosis and may completely resolve the patient's symptoms.

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