A large vascular leiomyoma or an infected sebaceous cyst of the chest?
A case report

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INTRODUCTION: *Vascular leiomyoma is a rare tumour of smooth muscle origin. It has been reported in many anatomical sites, but it usually affects limbs. It's recurrence rate is slow but it's quite difficult to make diagnosis.*
CASE REPORT: *The study describes the case of a 53-year-old man, with a subcutaneous, large vascular leiomyoma of the right clavicular region. The patient had a painful, slow-growing mass, measuring more than 20 mm. The mass had a relatively homogeneous, hypoechoic-echo texture, with a small amount of posterior acoustic enhancement. No diagnosis was made and the patient underwent surgery for a suspected leyomioma. After surgery a hard, round-shaped tumor, measuring approximately 43X32 mm of diameter was found with histological features of leyomioma.*
DISCUSSION: *The case has an unusual clinical presentation, for the uncommon localization and size of the mass. US imaging and histopathologic features are reported. The rarity of vascular leiomyoma makes diagnosis difficult and frequently delayed. Differential diagnosis includes infected sebaceous cists, glomus tumors, hemangiomas, angiolipomas, ganglions, and traumatic neuromas.*
CONCLUSIONS: *This report highlights that leiomyoma, even of large dimension, must be included in the differential diagnosis of painful, subcutaneous masses.*

KEY WORDS: Angioleiomyoma, Benign Tumor, Sebaceous cyst, Subcutaneous Tumor, Vascular leiomyoma.

Introduction

Vascular leiomyoma or angioleiomyomas are benign, solitary tumors of smooth muscle cells originating from the muscular layer of vessel walls. This type of tumor can occur on any part of the body, and can be found in the dermis, the subcutaneous fat, or the fascia areas and are mostly apparent on the extremities, such as the lower limb (50-70%)[1-5]. Clinically, this tumor appears as a small (<20 mm), freely movable, subcutaneous nodule, causing pain in approximately 60% of patients [2,5-9]. This type of lesion occurs more frequently in females than in males, and pregnancy may increase the severity of the pain [1-4,10]. Diagnosis of leiomyoma is frequently delayed because of its rare occurrence, and for the lack of awareness by the treating clinicians [11].
Sonography is often used in the initial evaluation of soft tissue masses, and the treatment for these lesions usually consists of their surgical excision. Because of its biological features recurrence rate is low [2,7-8,11,12].
The study describes the case of a patient who had an angioleiomyoma of the right clavicular region of an uncommon size (> 20mm). Due to the large size of the mass the differential diagnosis was difficult.

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Case report

In April 2010, a 53-year-old man was brought to our attention in the Department of plastic surgery of Policlinico Umberto I of Rome, affected by a painful, slow-growing (seven years), right clavicular mass. During these 7 years he began having episodic pain in the affected area, increased by movements of elevation and rotation of the arm. He reported a progressive increase of the mass's size. He didn't complain fever, lymphadenopathy, involuntary weight loss, or loss of energy. The skin covering the mass was erythematous, hot and itchy and it presented peripheral telangiectasias and around grey central area (Fig. 1). Palpation revealed a firm, elastic, mobile mass, located close to the right clavicular region. The patient underwent an ultrasound examination, which confirmed the presence of a round nodule, located in the subcutaneous fat tissue. The mass had a

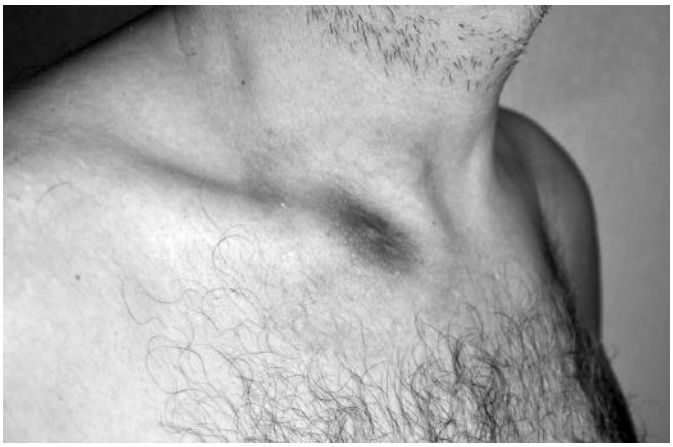

Fig. 1: At physical examination an oval lesion measuring approximately 6 cm with soft-elastic consistency at palpation of the clavicular region was observed. The skin covering the mass was erythematous and it presented peripheral telangiectasias and a round grey central area.

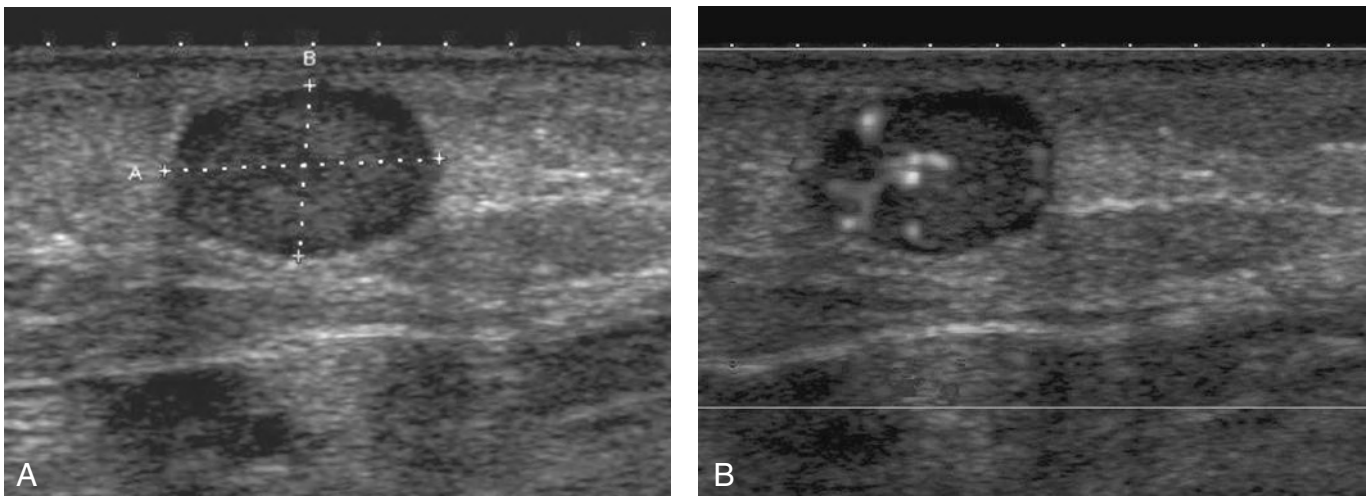

Fig. 2: (A) US examination showed homogeneous, hypoechoic-echo texture lesion, to the subcutaneous soft tissue with a small amount of posterior acoustic enhancement. (B) Power Doppler sonogram revealed diffuse arterial hypervascularity.

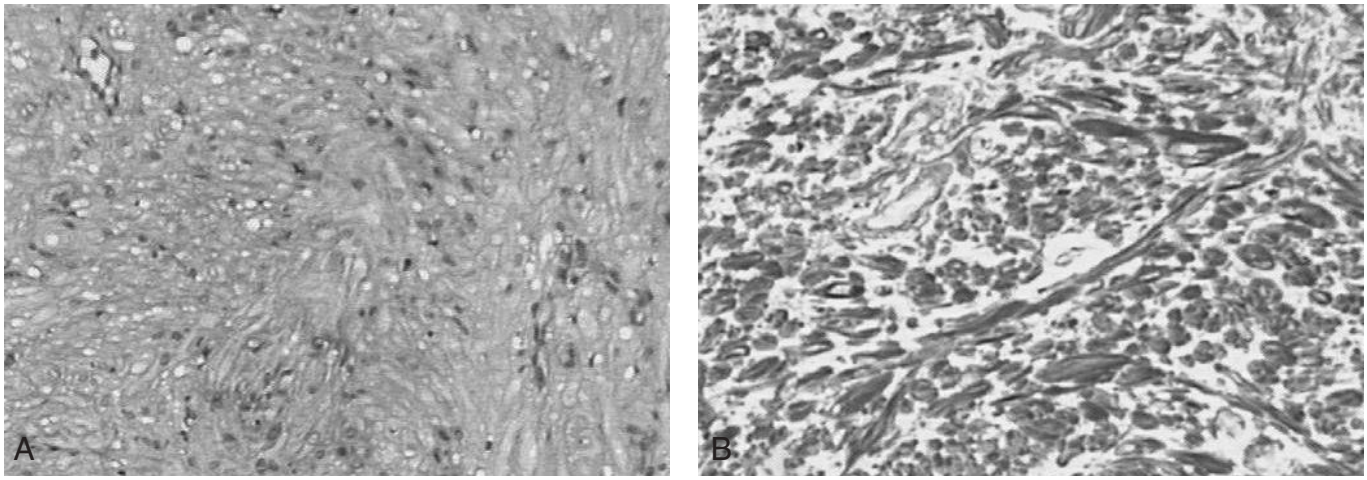

Fig. 3: Photomicrographs showing the histopathology of vascular leiomyoma. (A) Closely compacted, intersecting smooth muscle bundles surrounding slit-like vascular channels. No evidence of necrosis, pleomorphism, mitotes or nuclear atypia. (Hematoxylin-Eosin 100 X).
(B) Positive immunohistochemistry staining for smooth muscle actin (100 X).

relatively homogeneous, hypoechoic-echo texture, with a small amount of posterior acoustic enhancement. No internal calcification was present, and an evaluation by a color Doppler, revealed diffuse arterial hypervascularity (Fig. 2 A, B). The patient, with an uncertain preoperative diagnosis, underwent the surgical excision under local anesthesia. The patient had a complete recovery of his symptoms immediately after the operation. A incision of approximately 6 cm was made in the right clavicular region. In the subcutaneous tissue, a capsulated mass, was found. The mass was easily removed, showing a hard, round-shaped tumor, measuring approximately 43X32 mm of diameter, not adherent to the deep nor superficial tissues.

The hystopathological analysis described a well-circumscribed, not capsulated-nodular tumor made of smooth muscle bundles closely compacted, intersecting with each another, and surrounded by thick fibrous tissue. The smooth muscle fascicles had uniform spindle cells, with eosinophilic cytoplasm and cylindrical nuclei with blunted ends. The proliferation was surrounded by blood vessels and slit-like vascular channels (Fig. 3A). Immunohistochemical smooth-muscle-actin staining, highlighted the presence of muscular fibers, therefore confirming the diagnosis of a vascular leiomyoma (Fig. 3B). Postoperatively, he had immediate and complete relief of his symptoms.

Discussion

A vascular leiomyoma or an angioleiomyoma, is a rare, benign tumor that originates from smooth muscle cells of arterial or venous walls [6,10]. Stout13 published the first comprehensive review of this rare lesion in 1937, and it has been well characterized in literature since that time [1,2-14].

The condition is more common in females than in males (female to male ratio of 2.7:1) [2]. These tumors can affect any age, but are more common in people between 30 and 60 years of age [2]. Ninety per cent of these lesions occurs in limbs, with the head and trunk accounting for remaining cases. Lower limbs is involved in 50 to 70% of the extremity lesions, and is twice as common in females. Conversely, head and neck, and upper limbs lesions are more frequent in males. Pain and/or tenderness are the most characteristic symptoms reported in the majority of patients (60/75%) [2,10].

These days, three different histological subgroups of angioleiomyomas have been identified: solid, cavernous and venous14. Solid tumors are found more commonly in the lower limbs,1 and pain is the most common presenting symptom. Therefore, our patient's presentation was not typical for an angioleiomyoma, in fact we reported a case of a male patient with a painful solid large leiomyoma of the chest. Macroscopically, vascular leiomyomas are sharply demarcated, spherical tumors, usually within 10 mm of size. The reported case showed that angioleiomyomas may be very large reaching 20 mm.

Differential diagnosis for painful subcutaneous lesions includes infected sebaceous cists, glomus tumors, hemangiomas, angiolipomas, ganglions, and traumatic neuromas, among others [15].

Sebaceous cysts are usually painless, slow-growing, small bumps or lumps that move freely under the skin. Occasionally, infections may occur. Signs or symptoms that may indicate infection of sebaceous cysts include: redness, tenderness, increased temperature of the skin over the bumps or lumps and greyish white, cheesy, foul-smelling material draining from the bump or lump[16].

In this case report, clinical examination was compatible with an infected sebaceous cyst.

The diagnosis of vascular leiomyoma depends upon the histopathological identification using staining techniques, such as Masson's trichrome, hematoxylin-eosin, alcian blue, van Gieson, and PAS (Periodic acid-Schiff). Immunohistochemical stains for vimentin, desmin, and smooth muscle actin can be of great value when available [17]. In this case, we used a hematoxylineosin stain, and an immunohistochemical stain for smooth muscle actin.

There are no specific imaging techniques, capable of characterizing the vascular leiomyoma, anyway sonography represents an excellent modality for initial evaluation of this kind of lesion. In this case, sonography was essential to describe location and size of the tumor.

The homogeneity of the lesion, lack of calcification, and lack of compressibility excluded a possible hemangioma, and the solid nature of the mass, as evidenced by the hyperemia, basically excluded a cyst, and other fluid collections. Furthermore, the location of the mass and the lack of compressibility were unusual for a lipoma.

The vascular flow of these tumors showed a high resistance, thus suggesting the presence of muscular arteries. Sonographic findings suggestive of angioleiomyomas, include well-defined margins, hypervascularity, and of small dimensions. When a slow-growing, lower-limbs mass has these features, diagnosis of an angioleiomyoma should be consider even if mass's diameter is more than 10 mm.

Conclusions

In conclusion, although angioleiomyoma is an infrequent, soft-tissue tumor, it does have a typical, but non-specific, presentation. The vascular leiomyoma has to be included in the differential diagnoses of painful subcutaneous masses even in the case of tumors more than 20 mm. Excision of the lesion enables a histopathologic diagnosis, and offers a complete resolution of symptoms to the patient. In this case, the rarity of the vascular leiomyoma of the trunk and the clinical aspect of the

lesion make diagnosis difficult.The use of sonography and the histopathological examination helped to solve the clinical question.

Riassunto

Il leiomioma è un raro tumore del tessuto muscolare liscio, solitamente localizzato a livello degli arti inferiori. Il trattamento è sempre di tipo chirurgico. Il caso da noi presentato è quello di un uomo di 53 anni che lamentava una massa dolente a livello della regione claveare destra da circa sette anni. Gli esami ultrasuonografici sono stati effettuati ed hanno evidenziato una lesione ipoecogena senza calcificazioni al suo interno. Dall'aspetto ecografico non si poneva diagnosi. Il paziente è stato sottoposto ad intervento per sospetto leiomioma sebbene la localizzazione e le dimensioni (43x32 cm) fossero atipiche per questo tipo di neoplasia. L'esame istologico ha poi, però, confermato l'iniziale sospetto. La lesione in questione è quindi degna di nota sia per le ingenti dimensioni sia per l'atipica localizzazione.

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