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Foot soft tissue myopericytoma: Case-report and review

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ABSTRACT

INTRODUCTION: Myopericytoma is a rare tumor of deep soft tissues, originating from pericytes and characterized by numerous thin walled blood vessels.
CASE REPORT: We report a case of myopericytoma found at the level of the second toe of the right foot.

A patient came to the Endocrinology Surgery Department of Catania Polyclinic because of a presence of a small swelling in the plantar region, between the 2nd and 3rd toe of the right foot. At the anatomopathological examination, the escalated lesion showed a neoformation of 0.6 cm in diameter, well circumscribed, capsulated, with myopericytoma diagnosis.
DISCUSSION: Its histopathological appearance is similar to myofibromatic lesions from glomic and angiomyoma tumors. It is a rare tumor that affects all ages with a peak after 50 years 3. The most frequent localization is at the lower extremities, particularly in soft subcutaneous tissues, but can rarely occur in other sites.
CONCLUSION: At the anatomopathological evaluation, the immunohistochemical examination for the correct formulation of the diagnosis is essential and an adequate surgical excision is important.
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1. Introduction

Myopericytoma is a rare tumor of deep soft tissues, originating from pericytes and characterized by numerous thin wall blood vessels [1].

Anatomically-pathologically is described as a perivascular proliferation of mesenchymal cells with pericytic differentiation, characterized by the radial and vertical growth of the cells, which take an oval form. Often the blood vessels take on a characteristic form of a "deer horn" [2–4].

The rarity and complexity of morphopathological aspects, during the years, have re-evaluated this pathology in order to correctly frame this cancer. Diagnosis is often based on the evaluation of immunohistochemical and structural characteristics [5–8]. We report a case of myopericytoma found at the level of the second toe of the right foot. The work has been reported in line with the SCARE criteria [9].

2. Case report

In December 2016, a patient, woman, 65 years old, came to our office for a small tumefying at the level of the plantar region, in the web second web space between the 2nd and 3rd toe of the right foot. The presence of this swelling has been reported for some months with a recent increase in size, associated with painful symptomatology, exacerbated by the use of footwear. Objective examination allowed the appreciation of a small nodule of the size of a lentil covered with normal, well-delimited and circumscribed skin, movable on the underlying planes.

Ultrasound showed a complex, hypoechoic, well-circumscribed, solid mass. The size of the mass was 0.6 × 0.5 cm.

There was nothing of relevance in his medical history and she had no trauma to her leg. The surgical approach included the total removal of neoformation, in local anesthesia, with cold scalpel, after section of a portion of a lozenge skin, including neoformation and preserving about 0.5 cm of indefinite margins. Once the clamping point was found, the piece was removed completely. After careful examination and hemostasis, the skin was sutured amd closed directly. The patient could walk after 1 day of surgical stay.

At the anatomopathological examination, the escalated lesion showed a neoformation of 0.6 cm in diameter, well circumscribed, capsulated and with free resection margins. Neoformation showed ovoid and soft cells with eosinophilic cytoplasm and perivascular proliferation of deer horns Fig. 1. The immunohistochemical profile was consistent for positive alpha-actin and negative desmine and allowed to formulate the diagnosis of myopericytoma.

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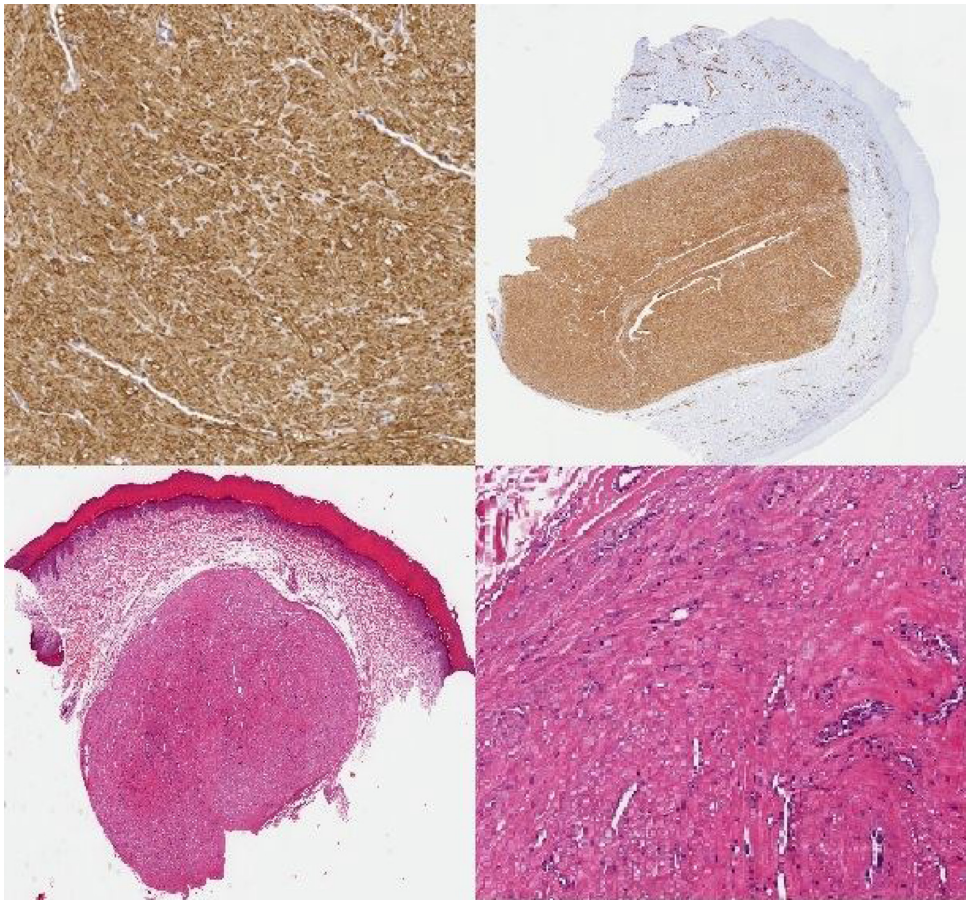

Fig. 1. Anatomopathological examination of neoformation.

3. Discussion

The anatomy-pathological entity of Miopericitoma is still a cause of scientific debate, although in the latest World Health Organization classification this tumor has been classified as a perivascular tumor of soft tissues and bones [2].

Its histopathological appearance is similar to myofibromatic lesions from glomic and angiomyoma tumors. It is a rare tumor that affects all ages with a peak after 50 years [3]. The most frequent localization is at the lower extremities, particularly in soft subcutaneous tissues, but can rarely occur in other sites. Usually, it is presented as nodular formations, with unspecific symptoms, often asymptomatic or paucidolent, with slow esophytic growth [10]. There is also a case of mypericytoma occurring in literature, approximately 2.1 cm at the right bladder trigonum in a 75-year-old patient treated with transurethral resection [11]. Although the use of instrumental examinations is not always indicated, myopericytoma in Computer Tomography appears to be iso-hypodensis, homogeneous or heterogeneous and occasionally calcified [12–14]. Other unusual cases reported: a myopericytoma of about 5cm × 5 cm at the right shoulder, previously mistakenly diagnosed as lipoma by RMN and subsequently surgically excised [15]; a myopericytoma of 9 cm × 8 cm diameter located in the occipital region, in a 16 year old girl [16]. Usually, the average size of these neoplasia is less than 2 cm in diameter. It is part of the same group of myopericytoma pathologies, Hemangiopericitoma in its lipomatosis and sinosal variants [5,6]. During clinical diagnosis, this neoplasia is easily confused by foreign body or fibrous reaction cause its macroscopic appearance [17,18]. There is also a rare malignant variant of this neoformation, characterized by infiltrative growth towards deep tissue layers, high atypia and increased mitotic index. In these patients, in addition to a radical and extensive excision, it is necessary to use chemotherapy and adjuvant radiotherapy [15,19–24]. In literature there are 11 cases of myoperaciotosis with diffuse subcutaneous involvement by microscopic myopericytomatous nodules [25].

Agusti et al. describe another case of myopericitoma on the foot plantar region and explains the likely origin from a cutaneous vascular malformation [26]. Another area where myopericitoma may develop is the heel [27]. Finally, Squillaci et al. described another myopericitoma of foot soft tissue concluding that clinicopathological features are similar to those previously reported in the literature [28] (Table 1).

4. Conclusion

In conclusion, myopericytoma is a rare benign tumor with a particular vascularization that characterizes it. In anatomopathological evaluation, the immunohistochemical examination for the correct formulation of the diagnosis is essential. Also, it is important to have a proper surgical technique during the excision of neoformation, in order to maintain the marginal benefit and to guarantee its radicality. Finally, despite the relative rarity of recurrences, it is evident that such patients need an adequate follow-up.

Conflicts of interest

No conflicts of interest.

Table 1
Cases of Myopericytomas in literature.

Study	Cases, n	Gender	Age, years	Site	Size, mm	Histopathologic features	Immunohistochemistry
Hemangiopericytoma: a vascular tumor featuring Zimmerman's pericytes [1]	9	5M 3F 1 unknown	45, 21, 31, 42, 37 3 infants, 1 unknown	Dorsal surface of the left ring finger. Outer margin of the breast. Parietal region. Shoulder. Right anterior chest wall. Righ infra-orbital region. Auricle. Left index finger. Lower part of the right thigh.	"Dressmaker's pin". "birthmark". 1 cm 17×10 mm 4 cm 5 mm $14 \times 7 \times 6$ mm.	The primary tumor is composed of capillary tubes and groups of endothelial cells. In some, the cells are spindle-shaped and suggest the appearance of smooth muscle.	N.D.
Myopericytoma of skin and soft tissues [3]	54	34M 18F 2 unknown	13–87 years (median, 52 years)	Lower extremities (26 cases); upper extremities (16 cases); head and neck (4 cases); trunk (2 cases); unknown (5 cases).	Confined to the dermis. Extension into the subcutis. Arose in subcutaneous. Deep soft tissue	Thin-walled vessels and a concentric, perivascular arrangement of ovoid, plump spindled to round myoid tumor cells.	Positive for Alpha-smooth muscle actin (ASMA) and H-caldesmon antobodies. Desmin is usually negative.
Myofibromatosis in adults, glomangiopericytoma, and myopericytoma: a spectrum of tumors showing perivascular myoid differentiation [5]	24	N.D.	23–67 years (median, 37 years)	N.D.	Subcutaneous tissue and the superficial soft tissue of the extremities.	Fascicles of spindle cells that resembled smooth muscle, in addition to a population of more primitive spindled cells associated with a hemangiopericytomalike vascular pattern. Subset of tumors characterized by concentric periluminal proliferation of bland, round to ovoid cells.	N.D.
Sinonasal-type hemangiopericytoma [7]	104	47M 57F	5–86 years (median, 62.6 years)	Nasal cavity Paranasal sinus	Average 3,1 cm	Spindle-shaped to round/oval nuclei with indistinct cell borders. Richly vascularized. Staghorn vessels.	Reactivity Vimentin, smooth muscle actin, muscle specific actin, factor XIIIa, laminin.
Lipomatous hemangiopericytoma: a rare variant of hemangiopericytoma [8]	16	12M 4F	54 years (range, 33–74 years)	Deep soft tissue	Average 10 cm	Oval to round cells surrounding a sinusoidal and staghorn vasculature often with perivascular hyalinization.	Factor XIIIa, type IV collagen, CD34 and smooth-muscle actins.
Myopericytoma: a unifying term for a spectrum of tumors that show overlapping features with myofribroma. A review of 14 cases. [10]	14	8M 6F	37 years old (F); 35.5 years old (M)	Distal extremities, head, neck region	10–30 mm 10–45 mm	Concentric perivascular arrangementof plump spindle shaped cells. Presence of a zonation/biphasic appaìearance.	Positive staining for SMA. Desmin was negative in five cases. Negative staining for S100 protein(4), HMB45(3), Cytokeratin(3) and CD34(3)
Myopericytoma in urinary bladder: a case report. [11]	1	1M	75 years old	Right trigone of bladder	2,1 cm	Spindle-shaped to oval-shaped cells; midly hyperchromatic nuclei arranged in perivascular whorls around often-hyalinized blood vessels.	Positive for alpha-smooth muscle actin (SMA) Desmin, CD34, h-caldesmon, CD99. Negative for S-100, CAM5.2, AE1/AE3, EMA, STAT6, MUC4, claudin-1.
Myopericytoma presenting as multiple pulmonary nodules [12]	1	1M	26 years old	Middle lobe of right lung	$1,7 \times 1,5 \times 1$ cm	Oval cells with a striking concentric arrangement of cells around variably sized lesional blood vessels.	Pattern CD31 and CD34 Positive for Vimentin and smooth muscle actin. Negative for desmin or S-100.

Table 1 (*Continued*)

Study	Cases, n	Gender	Age, years	Site	Size, mm	Histopathologic features	Immunohistochemistry
Renal Myopericytoma: a case report with a literature review [13]	1	1M	39 years old	Upper pole of the left kidney	9 × 10 × 18 cm	Spindle –shaped myoid cells with a concentric arrangement	Reactivity for Smooth muscle actin and CD10. Ki–67 <1%. Negative for CD34, desmin, S-100 protein, cytokeratin, HMB-45, Bcl-2, CD99
Incidental detection of subcutaneous myopericytoma of trunk [14]	1	1M	53 years old	Right lower back	4 cm	Myoid spindle cell proliferation with thin walled slit-like vessels	Immunoreactive for actin; positive for desmin. Ki67 index between 5% and 25%
Malignant myopericytoma of shoulder: a rare lesion [15]	1	1F	15 years old	Left shoulder	4,4 × 5,2 cm	Non-capsulated tumor composed of oval cells with eosinophilic cytoplasm	Positivity for smooth muscle actin vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34.
A giant myopericytoma involving the occipital region of the scalp [16]	1	1F	16 years old	Scalp in the occipital region	9 × 8 cm	Spindle-shaped cells forming characteristic rosettes around the blood vessels	Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34
Intravascular myopericytoma [17]	1	1M	54 years old	Subcutaneous tissue of the thigh	1,5 cm	Myoid spindle cells arranged in a striking concentric pattern around blood vessels	Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31
Malignant myopericytoma: expanding the spectrum of tumors with myopericytic differentiation [18]	5	2M 3F	19–81 years (67 median)	Neck, Arm, Thigh, foot	N.D.	Myoid to spindle cells with focally striking perivascular orientation	Positivity for Smooth muscle actin, one case for Demsin
Myopericytoma tumor of the glans penis [20]	1	1M	N.D.	Penile	N.D.	Perivascular proliferation of tumor cells with ovoid shaped nuclei abundant eosinophilic cytoplasm	Positivity for Smooth muscle actin negative for BRAF
Benign perivascular myoid cell tumor of the urinary tract [21]	2	2F	59 years old, 52 years old	Kidney, Urinary bladder	N.D.	Blood vessels surrounded by plump perivascular myoid cells.	Reactivity to SMA, caldesmon/calponin. Negative for CD34, cathepsin K, S100.
Myopericytoma of the kidkney [22]	1	1F	59 years old	Upper pole of the left kidney	3,5 cm	Richly vascularized, perivascular proliferation, oval to spindle cells cells were arranged in concentric fashion around vascular lumina	Reactivity for SMA, CD34, bcl-2, Collagen IV negative for Desmin, keratin, EMA.

Renal Myopericytoma: case report and review of literature. [23]	1	1F	40 years old	Kidney	3.8 × 3 × 3 cm	Characteristic pattern and additional glomus tumorlike pattern	Reactivity for Vimentin, SMA, smooth myosin heavy chain CD34. Ki-67<5%. Negative for desmin, S100, HMB-45, Mart 1.
Myopericytomatosis: clinicopathologic Analysis of 11 cases. [25]	11	3M 8F	9–63 years (37 median)	Lower extremities (foot/ankle, calf, knee, thigh, neck)	1,5 to 11 cm (median 6,0)	Diffuse infiltration by innumerable discrete myopericytoma/myofibroma-like nodules of bland spindled-to-ovoid cells (smooth muscle actin positive), in a mainly perivascular distribution.	PDGFRB alterations in all cases. No BRAF, NOTCH, or GLI1 alterations were detected. Strongly activating PDGFRB mutation N666 K.
Intravascular Myopericytoma of the plantar region: case report and Discussion. [26]	1	1F	63 years old	Left foot sole; subcutaneous nodule	1 cm	A nodular solid mass surrounded by vascular muscular wall. Solid areas of round to spindle cells with eosinophilic cytoplasm arranged in a multilayered and concentric perivascular pattern.	Diffusely positive for SMA and h-caldesmon; negative for desmin and CD34.
Intravascular Myopericytoma in the Heel: Case Report and Literature Review [27]	1	1M	48 years old	Heel of the right foot	1,5 × 0,4 cm	Lesion in the subcutaneous tissue; a venous-type vascular structure, was partially occupied by a proliferation of spindle-shaped cells	Positive for SMA and caldesmon, but not CD34 antobody, desmin, factor VIII, epithelial membrane antigen, or cytokeratins.
Myopericytoma-type perivascular myoma located in the soft tissue of the foot. [28]	1	1M	68 years old	Subcutaneous tissue of the right foot.	1,4 cm	Concentric perivascular spindle and ovoid cell proliferation and an extensive hemangiopericytomatous growth component.	Positive for vimentin, smooth muscle actin, desmin and calponin, negative for S-100 protein, CD34, CD31 and cytokeratins (AE1/AE3, Cam 5.2).

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Ethical approval

Department of "Scienze Mediche, Chirurgiche e tecnologie Avanzate – G. F. Ingrassia", University of Catania.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

I have obtained written consent from the patient and I can provide this should the Editor ask to see it.

Author contribution

Provenzano D: Write, translation.
Lo Bianco S: design, write, traslation.
Belfiore M: translation.
Buffone A: design.
Cannizzaro MA: design.

Guarantor

Cannizzaro Matteo Angelo.

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