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 myofibromatous lesions from glomic and angiomymomas. It is a rare tumor that affects all ages with a peak after 50 years. The most frequent localization is at the lower extremities, particularly in soft subcutaneous tissues, but can rarely occur in other sites. 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 scalp, 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 and 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. Neofomation 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.

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 scalp, 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 and 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. Neofomation 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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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 myofibromatous 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 myopericitoma 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, myopericitoma in Computer Tomography appears to be iso-hypodensis, homogeneous or heterogeneous and occasionally calcified [12–14]. Other unusual cases reported: a myopericitoma of about 5cm × 5 cm at the right shoulder, previously mistakenly diagnosed as lipoma by RMN and subsequently surgically excised [15]; a myopericitoma 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 myopericitoma pathologies, Hemangiopericitoma in its lipomatosis and sinusal 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 myopericitosis with diffuse subcutaneous involvement by microscopic myopericitomatous 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, myopericitoma 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 | 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. Right 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 × 7 × 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                                 | 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 spindle to round myoid tumor cells. Fascicles of spindle cells that resembled smooth muscle, in addition to a population of more primitive spindle cells associated with a hemangiopericytomalike vascular pattern. Subset of tumors characterized by concentric periluminal proliferation of bland, round to ovoid cells. Spindle-shaped to round/oval nuclei with indistinct cell borders. Richly vascularized. Staghorn vessels. | Positive for Alpha-smooth muscle actin (ASMA) and H-caldesmon antibodies. Desmin is usually negative. | N.D. |
| Myofibromatosis in adults, glomangiopericytoma, and myopericytoma: a spectrum of tumors showing perivascular myoid differentiation | 24       | N.D. | 23–67 years (median, 37 years) | Subcutaneous tissue and the superficial soft tissue of the extremities. | | | |
| Sinonasal-type hemangiopericytoma                                      | 104      | 47M 57F | 5–86 years (median, 62.6 years) | Nasal cavity Paranasal sinus | Average 3.1 cm | | Reactivity Vimentin, smooth muscle actin, muscle specific actin, factor XiiiA, laminin. |
| Lipomatous hemangiopericytoma: a rare variant of hemangiopericytoma     | 16       | 12M 4F | 54 years (range, 33–74 years) | Deep soft tissue | Average 10 cm | | Factor XiiiA, type IV collagen, CD34 and smooth-muscle actins. |
| Myopericytoma: a unifying term for a spectrum of tumors that show overlapping features with myofibroma. A review of 14 cases. | 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 arrangement of plump spindle shaped cells. Presence of a zonation/biphasic appearance. | 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.                      | 1        | 1M     | 75 years old | Right trigone of bladder | 2.1 cm | | 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                 | 1        | 1M     | 26 years old | Middle lobe of right lung | 1.7 × 1.5 × 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. |
| 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   | Upper pole of the left  | 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 Immunoreactive for actin; positive for desmin. Ki67 index between 5% and 25% |
| Incident detection of subcutaneous myopericytoma of trunk [14]        | 1        | 1M     | 53 years   | Right lower back        | 4 cm         | Myoid spindle cell proliferation with thin walled slit-like vessels                       | Positive reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31. |
| Malignant myopericytoma of shoulder: a rare lesion [15]               | 1        | 1F     | 15 years   | Left shoulder           | 4.4 × 5.2 cm | Non-capsulated tumor composed of oval cells with eosinophilic cytoplasm                 | Positivity for smooth muscle actin, desmin, and CD34. Negative reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31. |
| A giant myopericytoma involving the occipital region of the scalp [16] | 1        | 1F     | 16 years   | Scalp in the occipital region | 9 × 8 cm     | Spindle-shaped cells forming characteristic rosettes around the blood vessels          | Positive reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31. |
| Intravascular myopericytoma [17]                                      | 1        | 1M     | 54 years   | Subcutaneous tissue of the thigh | 1.5 cm       | Myoid spindle cells arranged in a striking concentric pattern around blood vessels     | Positive reactivity for smooth muscle actin, desmin, and CD34. Negative reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. 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 | Positive reactivity for smooth muscle actin, desmin, and CD34. Negative reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31. |
| 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 | Positive reactivity for smooth muscle actin, desmin, and CD34. Negative reactivity for smooth muscle actin, vimentin, CD99. Negative for cytokeratin, S100, desmin and CD34. Positive staining for smooth muscle actin (SMA). Negative for Desmin, Bcl2 and CD34. Positive for SMA, CD34. Negative for desmin, cytokeratin, S100, HMB-45 and CD31. |
| Benign perivascular myoid cell tumor of the urinary tract [21]        | 2        | 2F     | 59 years   | Kidney, Urinary bladder | N.D.         | Blood vessels surrounded by plump perivascular myoid cells.                           | Reactivity to SMA, caldesmon/calanpinon. Negative for CD34, cathepsin K, S100. |
| Myopericytoma of the kidney [22]                                     | 1        | 1F     | 59 years   | Upper pole of the left  | 3.5 cm       | Richly vascularized, perivascular proliferation, oval to spindle cells were arranged in concentric fashion around vascular lumina | Reactivity to SMA, CD34, bcl-2, Collagen IV negative for Desmin, keratin, EMA. |
Table 1 (Continued)

| Study                                                                 | Cases, n | Gender | Age, years | Site                  | Size, mm     | Histopathologic features                                                                 | Immunohistochemistry                                                                 |
|----------------------------------------------------------------------|----------|--------|------------|-----------------------|--------------|------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------|
| Renal Myopericytoma: case report and review of literature. [23]     | 1        | 1F     | 40 years   | 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. PDGFRB alterations in all cases. No BRAF, NOTCH, or GLI1 alterations were detected. Strongly activating PDGFRB mutation N666K. |
| Myopericytomatosis: clinicopathologic Analysis of 11 cases. [25]     | 11       | 3M 8F  | 9–63 years (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 N666K. |
| Intravascular Myopericytoma of the plantar region: case report and Discussion. [26] | 1        | 1F     | 63 years   | 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 b-caldesmon; negative for desmin and CD34. |
| Intravascular Myopericytoma in the Heel: Case Report and Literature Review [27] | 1        | 1M     | 48 years   | 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 antibody, 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   | 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, translation.
Belfiore M: translation.
Buffone A: design.
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