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Subcutaneous Myoepithelioma of the Foot: A Case Report

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Abstract | **Case Report**

Background: Myoepitheliomas of soft tissues are extremely rare neoplasms of uncertain histogenesis, accounting for less than 1% of all soft-tissue tumors. They belong to the "mixed tumor/parachordoma" family in the WHO classification. Their occurrence in the foot is exceptional, with fewer than ten cases reported in the literature. ***Case Presentation:*** We report the case of a 54-year-old male presenting with a progressively enlarging mass located in the first interdigital space of the right foot. Magnetic resonance imaging (MRI) revealed a well-circumscribed, lobulated soft-tissue lesion measuring 95 × 55 × 35 mm, displaying hypointense signal on T1 and hyperintense signal on T2 with heterogeneous enhancement after gadolinium administration. Histopathological examination of a biopsy specimen demonstrated an epithelioid proliferation composed of polygonal and plasmacytoid cells arranged in nests and lobules, separated by fibrous septa. Immunohistochemistry showed positivity for AE1/AE3, EMA, PS100, and INI-1, with negativity for CD34, Desmin, AML, and ACE, and a Ki-67 index of 5%. Complete surgical excision was performed with satisfactory functional outcome and no recurrence after six months of follow-up. ***Conclusion:*** Myoepithelioma of the foot is a rare benign tumor. The diagnosis relies on histopathological and immunohistochemical analysis. Surgical excision remains the treatment of choice, with regular follow-up recommended due to the risk of local recurrence

Keywords: Myoepithelioma; Foot; Benign soft-tissue tumor; Immunohistochemistry; Surgical excision.

INTRODUCTION

Myoepitheliomas are uncommon tumors arising from myoepithelial cells, which possess both epithelial and smooth-muscle characteristics. Initially described in salivary glands as pleomorphic adenomas, similar lesions have been recognized in extra-salivary sites, including skin, soft tissues, and viscera. The WHO classifies soft-tissue myoepitheliomas within the spectrum of "mixed tumors" or "parachordomas."

These tumors usually occur in the subcutaneous or subfascial planes of the extremities, predominantly in the thigh, arm, or forearm, and rarely in the foot. They affect both sexes but are slightly more frequent in middle-aged men. Their biological behavior is variable, ranging from benign to low-grade malignant forms.

Due to their rarity and non-specific clinical presentation, myoepitheliomas of the foot are frequently misdiagnosed as sarcomas or vascular lesions. We report a rare case of subcutaneous myoepithelioma of the right foot and discuss its clinical, radiological, and pathological features, along with therapeutic management and literature review.

CASE PRESENTATION

A 54-year-old male, with a history of chronic tobacco use (30 pack-years), presented to our orthopedic outpatient clinic with a painless swelling in the right foot, progressively increasing in size over several months. The patient mainly complained of aesthetic discomfort and mild difficulty while walking.

On physical examination, there was a firm, well-circumscribed, mobile mass in the first interdigital space between the great toe and the second toe, extending from the dorsal to the plantar aspect. The overlying skin was normal, with no inflammatory signs or tenderness. No regional lymphadenopathy was noted.

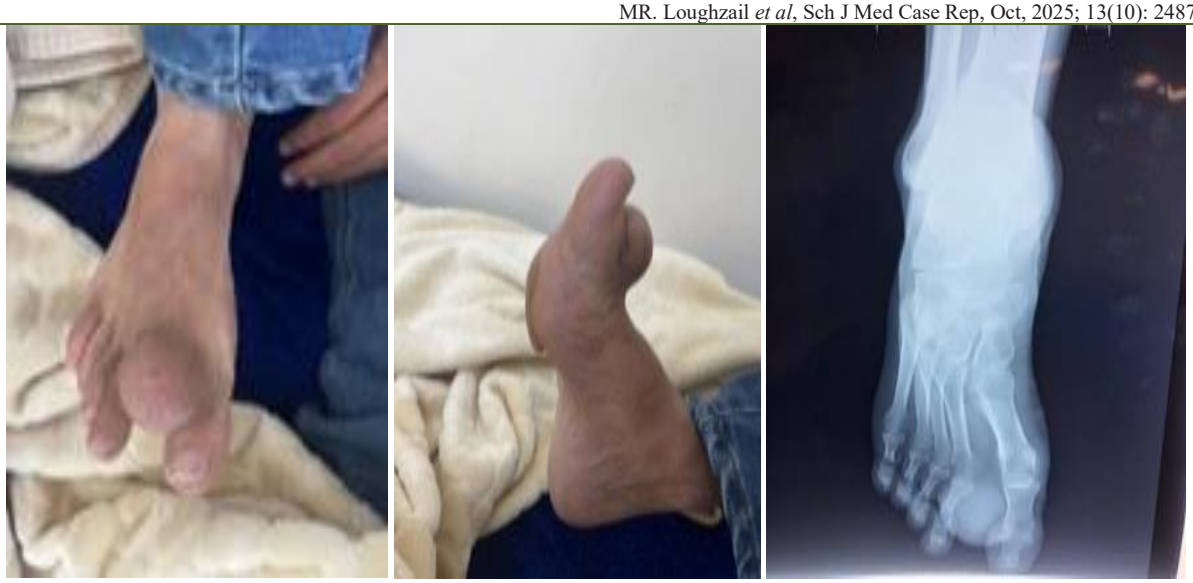

Figure 1: Clinical and radiological images showing the tumor

Radiological Findings

Plain radiography of the foot was unremarkable, showing no bony involvement. MRI with gadolinium contrast revealed a multilobulated soft-tissue lesion measuring 95 × 55 × 35 mm, located between the first and second rays. The lesion appeared hypointense on T1-weighted images and hyperintense on T2-weighted images, containing hypointense fibrous septa that enhanced intensely and heterogeneously after contrast injection.

The mass extended toward the plantar soft tissues, abutting but not infiltrating the flexor tendons of the second ray. The surrounding fat planes were preserved, and there was no cortical erosion or marrow signal abnormality of adjacent bones. The overall radiologic aspect was suggestive of a sarcomatous lesion, prompting histological evaluation.

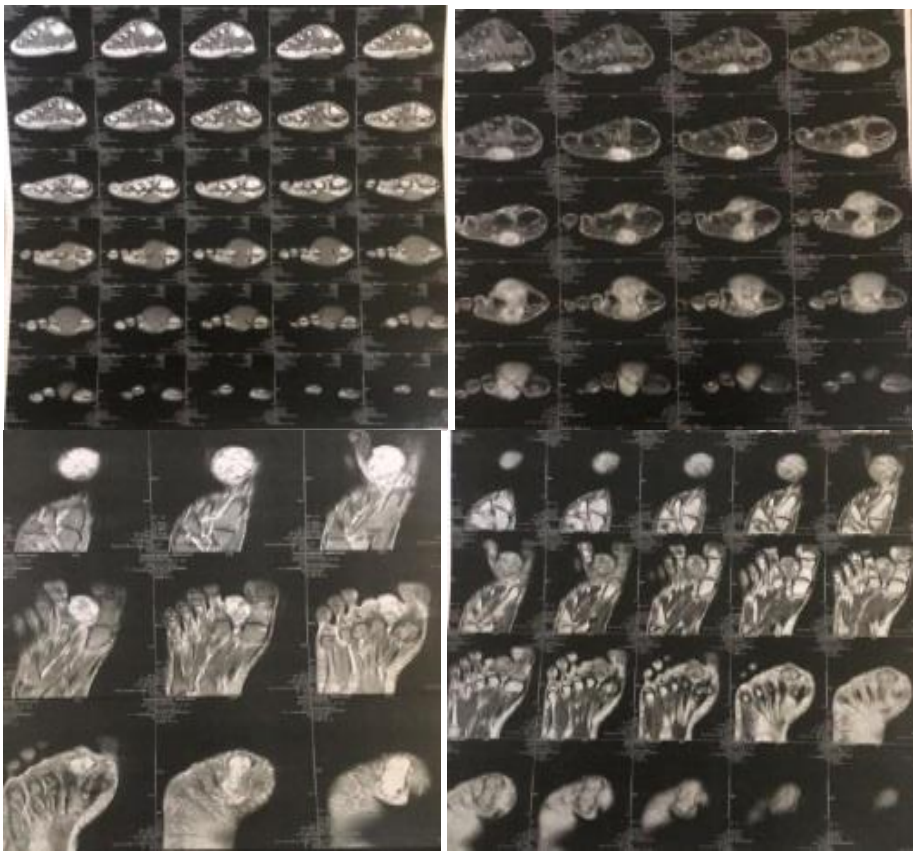

Figure 2: MRI images of the foot showing a soft tissue tumor process initially suggesting a sarcomatous origin

Histopathological and Immunohistochemical Findings

A needle biopsy was performed on December 30, 2022. Microscopic examination revealed a well-circumscribed proliferation of epithelioid and plasmacytoid cells arranged in sheets and lobules, separated by thin fibrous septa. The tumor cells had eosinophilic cytoplasm, hyperchromatic nuclei, and rare mitotic figures, without necrosis.

Immunohistochemical staining showed positivity for AE1/AE3, EMA, PS100, and INI-1, confirming myoepithelial differentiation. CD34, Desmin, AML, and ACE were negative. The Ki-67 proliferative index was estimated at 5%. These findings supported the diagnosis of a benign myoepithelioma of soft tissue.

Surgical Management

After multidisciplinary discussion and patient consent, a complete surgical excision was scheduled. Under spinal anesthesia, the patient was placed in the supine position with a pneumatic tourniquet applied to the thigh. An incision was made along the previous biopsy scar, and careful dissection was performed to isolate the tumor circumferentially. The lesion was excised en bloc, with coagulation of feeding vessels and meticulous hemostasis. The wound was irrigated and closed in layers with a sterile dressing applied.

The postoperative course was uneventful. The patient received prophylactic antibiotics and analgesia and was discharged after 48 hours. The excised specimen was sent for histopathological confirmation, which corroborated the diagnosis of a benign myoepithelioma with free surgical margins.

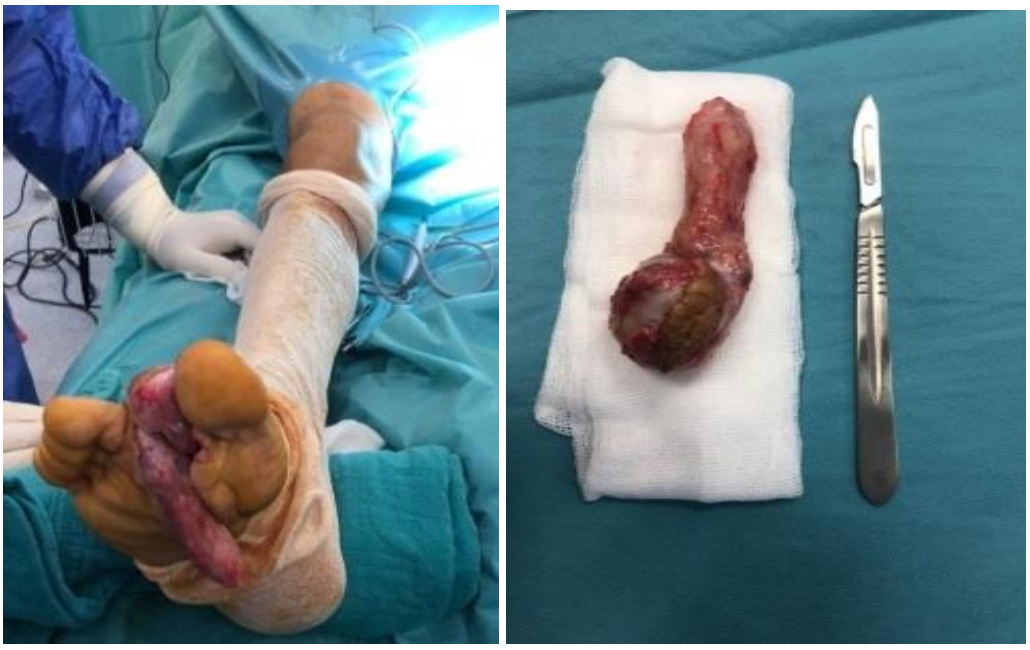

Figure 3: Photographs clinically showing the tumor sent to the pathology laboratory before and after its resection

Figure 4: Photographies du pied en post-opératoire immédiat

Follow-up and Outcome

At six-month follow-up, the patient remained asymptomatic with good wound healing and no evidence of recurrence. Mild dorsal paresthesia of the foot was reported but resolved spontaneously. Range of motion of the toes was preserved, and gait was normal. Regular clinical and MRI surveillance was recommended every six months due to the risk of local recurrence.

DISCUSSION

Soft-tissue myoepitheliomas are exceedingly rare, representing less than 1% of all soft-tissue neoplasms. They can occur at any age, with a predilection for adults between the second and fifth decades of life. The lower and upper limbs are the most frequent locations, whereas involvement of the foot is exceptional.

Clinical and radiological features:

Clinically, these tumors are usually slow-growing, painless, and well-circumscribed. Imaging typically shows a lobulated, well-defined mass with heterogeneous T2 hyperintensity and contrast enhancement. However, these findings are non-specific, often leading to confusion with sarcomas, hemangiomas, or synovial lesions.

Histopathological aspects:

Histologically, myoepitheliomas consist of epithelioid, spindle, plasmacytoid, or clear cells embedded in a myxoid, hyaline, or fibrous stroma. The key diagnostic feature is the dual epithelial and myogenic differentiation, confirmed by immunohistochemistry. Co-expression of cytokeratins (AE1/AE3), EMA, and S-100 protein, along with INI-1 positivity, is characteristic. CD34 and Desmin negativity help to exclude other soft-tissue tumors such as sarcomas and leiomyomas.

Differential diagnosis:

The main differential diagnoses include epithelioid sarcoma, metastatic carcinoma, synovial sarcoma, and parachordoma. Immunohistochemistry remains crucial to distinguish these entities.

Management and prognosis:

Complete surgical excision with tumor-free margins is the cornerstone of treatment. Incomplete resection is associated with a higher risk of local recurrence. Malignant transformation is rare but has been described, particularly in lesions showing high mitotic activity or cellular atypia.

In the largest published series of 401 soft-tissue tumors of the foot, Kaposi and synovial sarcomas were the most common malignant entities. However, benign myoepitheliomas can mimic these tumors clinically and radiologically, delaying diagnosis for several months. Hence, histopathological confirmation is mandatory.

Our case aligns with reports by Boldig *et al.,* (2023), Rekha *et al.,* (2020), Dix *et al.,* (2012), and Patrizi *et al.,* (2008), who described similar benign subcutaneous myoepitheliomas of the foot with favorable postoperative outcomes and no recurrence after complete excision.

CONCLUSION

Soft-tissue myoepithelioma of the foot is an extremely rare benign neoplasm. Due to its non-specific clinical and imaging presentation, histopathological and immunohistochemical analyses are essential for accurate diagnosis. Surgical excision with clear margins remains the treatment of choice and is usually curative. Long-term follow-up is recommended to detect possible recurrence.

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