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LETTER - CLINICAL

Amelanotic nodular melanoma in Marjolin ulcer on the plantar region☆

Dear Editor,

This case describes an 80-year-old female Caucasian patient, who sought dermatological evaluation for a lesion on the left foot after local trauma, with progressive growth over two years. Dermatological examination showed a nodular, vegetating, tumor-like lesion covered by hematic crusts on the left plantar region, overlapping an extensive area of deformities in the foot anatomy, caused by healing by secondary intention of burn lesions that occurred in childhood (Fig. 1). Histopathology showed an ulcer filled with a fusiform cell proliferation arranged in bundles in different directions, occupying the superficial and deep dermis, and moderate cell pleomorphism (Fig. 2). Immunohistochemistry was positive for S-100 protein, gp100 and Melan-A, confirming the diagnosis of melanoma (Fig. 3). The association of clinical and laboratory findings led to the diagnosis of acral amelanotic nodular melanoma on a burn scar. The authors found in the literature four case reports of amelanotic melanoma arising on chronic scars when searching Pubmed using the terms ''burn and amelanotic melanoma'' and ''Marjolin ulcer and amelanotic melanoma'', demonstrating the rarity of such a presentation.

The lesion was completely excised, as well as the ipsilateral lymph nodes, after confirmation of a positive sentinel lymph node for metastatic melanoma. Imaging exams showed alterations suggestive of metastasis in face and skull bones. The patient died after eight months.

Marjolin ulcer is a rare and aggressive tumor, occurs mainly in the fifth decade of life, and is prevalent in the male sex.[1] It arises over scar tissue, particularly after burns, occurring in 0.7% to 2% of these lesions.[1–3] Clinically, the lesions arise over previous scarring, as a non-healing, ulcerated or hardened, rapidly growing lesion that may become exophytic and bleeding. The most frequent histopathological type of neoplasia arising in Marjolin ulcers is squamous cell carcinoma (80%–90%), followed by basal cell carcinoma (9.6%) and, rarely, melanoma (2.4%).[1–3] The low incidence of melanoma in these scars can be explained by the small number of melanocytes in the scar tissue.[1]

In the epidemiology of melanomas, the nodular type represents the second most frequent one (approximately 15%

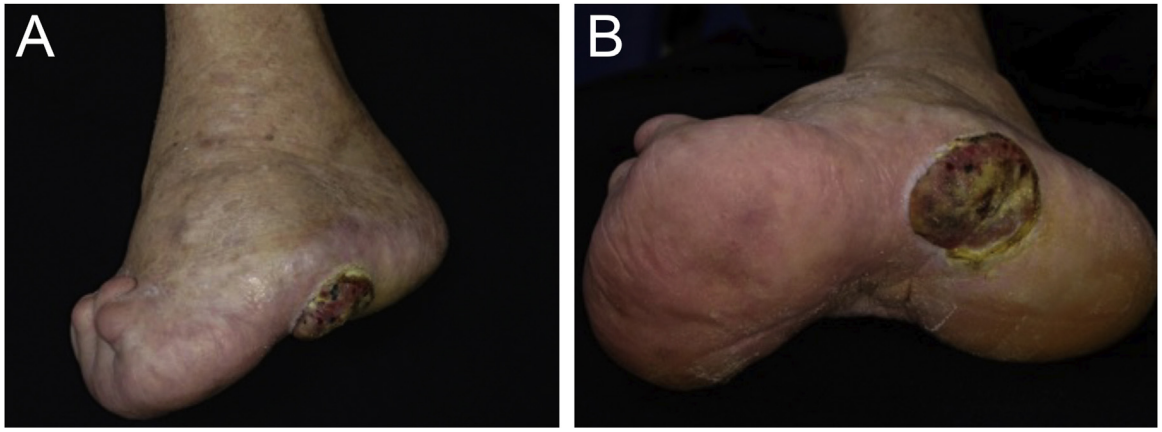

Figure 1 (a) Anatomical deformity in the left foot and absence of toes, secondary to a burning accident in childhood, that healed by second intention. (b) Vegetating nodular-tumor lesion with hematic crusts, well-defined borders, blackened spots, and overlying keratosis, affecting the plantar region and lateral face of the left foot.

☆ Study conducted at the Hospital Universitário Cassiano Antônio Moraes, Universidade Federal do Espírito Santo, Vitória, ES, Brazil.

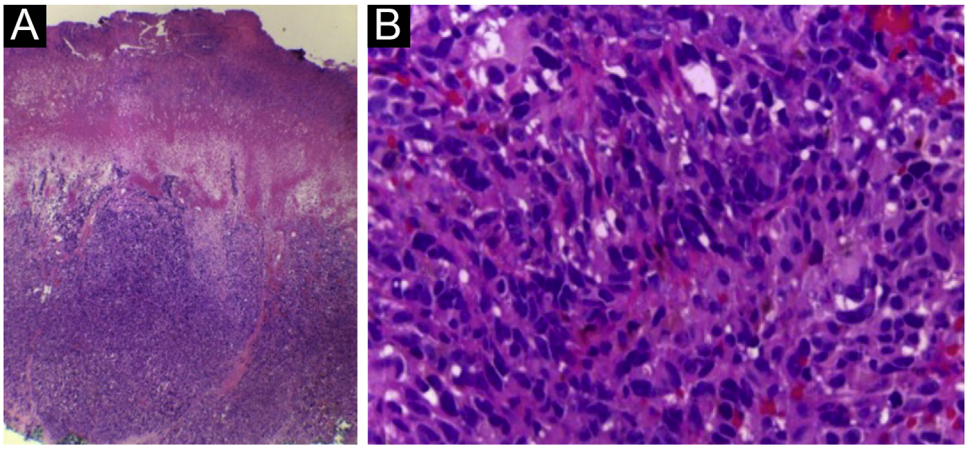

Figure 2 (a) Ulcerated lesion with cell proliferation occupying the superficial and deep dermis. (b) Moderately pleomorphic, fusiform and epithelioid cells, with hyperchromatic nuclei; nucleoli are difficult to distinguish.

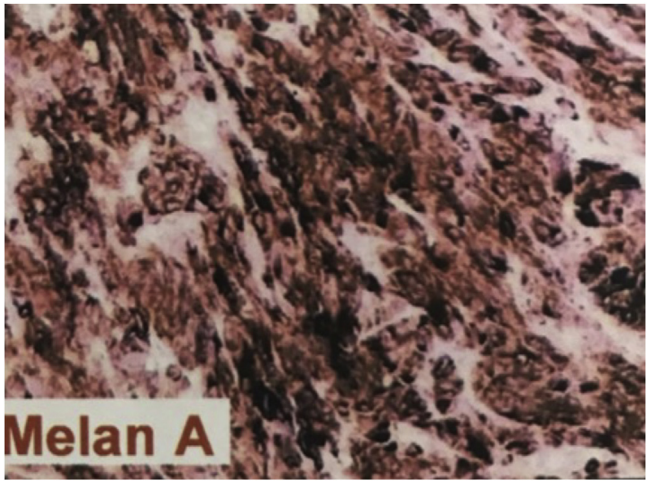

Figure 3 Immunohistochemistry was positive for Melan-A monoclonal antibody, which recognizes a specific melanocytic differentiation protein expressed in benign and malignant melanocytes.

of diagnosed melanomas), and 5% of nodular melanomas are amelanotic.[4]

Amelanotic acral melanoma has an incidence of 1.8% of melanoma cases,[4] and sometimes manifests as hyperkeratotic lesions.[2,5]

The diagnoses of Marjolin ulcer and melanoma are defined by clinical history, physical examination, and histopathology. The therapeutic options depend on TNM staging, including excision surgery with wide margins or limb amputation, in addition to chemotherapy and radiotherapy.[1]

Scars are susceptible to trauma due to a lack of collagen organization and vascular supply compromised by fibrosis, which obstructs vessels. Virchow's theory attributes the development of malignancy in scars to repeated trauma in the area, which leads to chronic irritation, repeated re-epithelialization, and local damage to the skin immune system, with a reduction in Langerhans cells and production of toxins in the affected area, in addition to genetic predisposition.[1–3]

A rare manifestation of Marjolin ulcer is presented in this case, which presented as a nodular acral amelanotic melanoma, reinforcing the need for advice to patients, care, and regular follow-up of burn scars, for early diagnosis and treatment, reducing morbidity and mortality.

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Authors' contributions

Valentina Lourenço Lacerda de Oliveira: Design and planning of the study; drafting and editing of the manuscript; collection, analysis, and interpretation of data; critical review of the literature; critical review of the manuscript.

Lucia Martins Diniz: Approval of the final version of the manuscript; effective participation in research orientation; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the manuscript.

Karla Spelta: Intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the manuscript.

Elton Almeida Lucas: Intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the manuscript.

Conflicts of interest

None declared.

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Case for diagnosis. Bilateral ulcerations on the distal phalanges of the second and third fingers - Ulcerative carpal tunnel syndrome☆

Dear Editor,

A 60-year-old female, hypertensive, type 2 diabetic patient with dyslipidemia, who worked in gastronomy, reported bilateral paresthesia in the thumb, index and middle fingers associated with nocturnal pain, which had started three years before. Two years before, she started to present ulcerated, bleeding, painless skin lesions on the second and third fingers, whereas on the third finger of the left hand, she had a healed lesion (Fig. 1A and B). The patient did not have any other skin lesions or other signs suggestive of leprosy, such as neural thickening, or loss of strength in other muscle groups. She also had hypoesthesia in these fingers, reporting no pain from professional injuries.

Neurological examination showed that Tinel's sign was present, as well as pain on wrist flexion. Electroneuromyography showed severe sensorimotor axonal neuropathy of the median nerve, which affected its distal segment, with an absence of potential in the nerve conduction test. X-rays of the hands showed osteolysis of the distal phalanx of the index fingers. A wrist magnetic resonance imaging showed bilateral thickening of the median nerve in the retinaculum (Fig. 2A and B), with normal thickness outside this region (Fig. 2C), whereas the ulnar nerves showed a normal appearance.

What's your diagnosis?

a) Leprosy
b) Diabetic neuropathy
c) Ulcerative carpal tunnel syndrome
d) Dermatitis artefacta

Discussion

Carpal tunnel syndrome (CTS) is a focal mononeuropathy caused by compression of the median nerve as it goes through the osteofibrous carpal tunnel. In the case reported herein, the nerve measured 18.7 mm^2 in the left retinaculum and 22.1 mm^2 in the right one, with normal size being up to 15 mm^2; measurements above 19 mm^2 are considered a severe form of the disease.[1]

The idiopathic form of CTS occurs more often in women aged between 40 and 60 years, and in half of the cases it is bilateral,[2] similar to the case described herein.

Secondary forms are caused by trauma, such as dislocation of the carpal bones, or by joint alterations in the wrist, such as osteoarthritis or inflammatory arthritis.[2]

Cutaneous involvement in CTS is rare, having been described as an ulcerative-mutilating form in 1979 by Bouvier,[3] with some reports in the dermatological literature,[4–6] which are generally described as ulcerative or ulcerative-mutilating forms.

There is a Portuguese report of nine cases in a retrospective study,[7] confirmed by electromyography. All of them started with paresthesia in the second and third fingers and one case had nocturnal pain. All radiographed cases (one-third) had osteolysis of the distal phalanges. Seven cases (77%) had ulcerations on the second and third fingers, also similar to the case described herein, and two cases had ulceration in only one finger.

The lesions observed in this patient are characteristic of peripheral neuropathy, with a clear background and callused edges and without pain, probably aggravated by the patient's professional activity. CTS should be suspected whenever there is involvement restricted to the second and third fingers, and the thumb is usually spared, as it has combined innervation of the radial nerve.[7]

It is relevant in dermatology to know about this disease, given the important differential diagnosis with leprosy (although it cannot be completely ruled out), so these cases can be referred for orthopedic evaluation and treatment, which can be conservative or surgical, with retinaculotomy, which gave good results in this patient..[8]

☆ Study conducted at the Liga de Dermatologia, Universidade Federal de Pelotas, Pelotas, RS, Brazil.