Trichilemmal carcinoma - Case report *

Carcinoma triquilemal - Relato de caso

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Abstract: The trichilemmal carcinoma is a rare tumor that usually occurs on sun-exposed skin, especially on the face, scalp, neck and back of hands, mainly in elderly subjects but commonly between the 4th and 9th decades of life. It is not a gender-based illness. This study shows a difficult to treat case of recurrent trichilemmal carcinoma on the same location of a basal-cell carcinoma previously treated with surgery and radiotherapy.
Keywords: Neoplasms, adnexal and skin appendage; Neoplasms, radiation-induced; Skin neoplasms

Resumo: O carcinoma triquilemal é um tumor raro, que ocorre, geralmente, na pele exposta ao sol, principalmente face, couro cabeludo, pescoço e dorso das mãos, em indivíduos idosos, entre a 4ª e 9ª décadas de vida, sem predilação por sexo. O presente estudo mostra um caso de carcinoma triquilemal, recidivado, de difícil tratamento, em mesma topografia de um carcinoma basocelular tratado previamente com cirurgia e radioterapia.
Palavras-chave: Neoplasias cutâneas; Neoplasias induzidas por radiação; Neoplasias de anexos e de apêndices cutâneos

INTRODUCTION

The term trichilemmal carcinoma (TLCA) was originally described as a clinical entity by Headington in 1976, although it was not adopted by pathologists[1] for many years. Recently, the publication of some series has contributed to distinguish the TLCA from other groups of follicular tumors. [1]

It is considered a carcinoma of low malignancy for presenting low frequency and rare metastases[1]. As a rule, it appears as a solitary lesion after the fifth decade of life. [2]

The diagnosis is established by means of histopathological examination using hematoxylin-eosin staining which, when necessary, is complemented by immunohistochemistry of the lesion. The treatment may be Mohs' technique or simple lesion excision.

We report the case of a patient with recurrent trichilemmal carcinoma on the same location of a previous basal-cell carcinoma, treated at the Erasto Gaertner Hospital with radiotherapy.

CASE REPORT

The patient, NK, a 35-year old male, white, married, carpenter from Palmital, in the state of Paraná came to Erasto Gaertner Hospital presenting an ulcerous, crusty lesion with high irregular borders, measuring 3.5 x 1.5 cm, located on the anterior thoracic wall by the superior third of sternum. Lesion exeresis with free margins was performed. The diagnosis was basal-cell carcinoma (BCC).

One year later, the patient presented a nodular lesion on the left infraclavicular region with a 3 x 2 cm diameter. Lesion exeresis was performed. The anatomopathological examination revealed presence of

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basal-cell carcinoma with compromised deep margin and adjuvant radiotherapy with a 6000 cGy dose was recommended.

The patient missed follow-up and returned to the hospital 5 years later with crusty and ulcerous lesions on the sternum scar region. Biopsy revealed BCC. An ample lesion resection was performed, followed by reconstruction with a skin flap. The anatomopathological examination revealed presence of sclerodermiform BCC with free lateral margins and compromised deep margin. The patient was subjected to adjuvant radiotherapy with a total dose of 5600cGy.

One year later, he presented a cervical node in the left sternocleidomastoid muscle. The cervical lymph nodes at the left of levels II, III and IV were dissected and the supraclavicular space was explored. The anatomopathological examination revealed an ulcerated neoplasm composed of solid blocks with a lobular growth pattern, characterized by proliferation of epithelial cells with clear cytoplasm, at times presenting peripheral palisading, areas of central necrosis, keratinization, cytologic atypia and mitotic figures compatible with the trichilemmal carcinoma diagnosis and compromised margin bordering the clavicle. Lymph node metastasis was absent.

One month after surgical resection, the patient presented recurrence of the trichilemmal carcinoma, with bone exposure at the superior third of sternum. He was subjected to surgery again and 4 cycles of chemotherapy were prescribed.

(1000g/m² of 5FU (5 fluouracil) associated with 75mg/m² of CDDP (cisplatin)).

The patient had a pathological fracture of the left clavicle and bone exposure 4 months after the chemotherapy treatment. Upon examination, an extensive tumor on the anterior thoracic wall was detected, with internal jugular vein and left subclavian vein infiltration, extensive post-radiotherapy fibrosis invading the manubrium and the right sternoclavicular joint and infiltration of the anterior mediastinum, without an apparent cleavage plane for dissection (Figure 1). Lesion exeresis was performed, resection R2 (persistence of local macroscopic disease) (Figure 2). The anatomopathological examination revealed a trichilemmal carcinoma with compromised margins (Figures 3, 4 and 5) .

Once more adjuvant radiotherapy was prescribed, with a total dose of 4320cGy.

Two years after the radiotherapic treatment a vegetative bleeding lesion could be observed, with its wider diameter measuring 4 cm on the surgical scar. The biopsy revealed a trichilemmal carcinoma. There was no other therapeutic, surgical or radiotherapic possibility and the patient was referred to clinical follow-up. After this date, he abandoned follow-up.

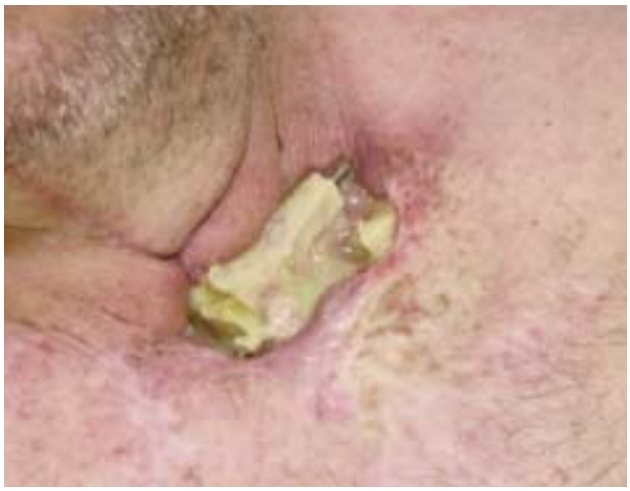

Figure 1: Lesion in the left clavicular region

DISCUSSION

The trichilemmal carcinoma is a rare tumor that usually occurs on sun-exposed skin, mainly on the face, scalp, neck and back of hands. It affects mainly elderly subjects but is more common between the 4th and 9th decades of life and is not a gender-based illness. [1,3-8] It has also been reported in patients exposed to radiotherapy treatment of benign head and neck diseases with long latency periods, related to the radiation dose to which the patient was subjected. During the 1920 to 1960 decades thymus (a gland that increases in size during childhood and regresses spontaneously) and tonsils radiotherapy was common, for acne treatment and other indications. At that time physicians believed this form of treatment was safe. [7]

Lesions may present clinically as papules, nodules or plaques, frequently ulcerated or with crusts. [1,2,3,5-10] A differential diagnosis is required and squa-

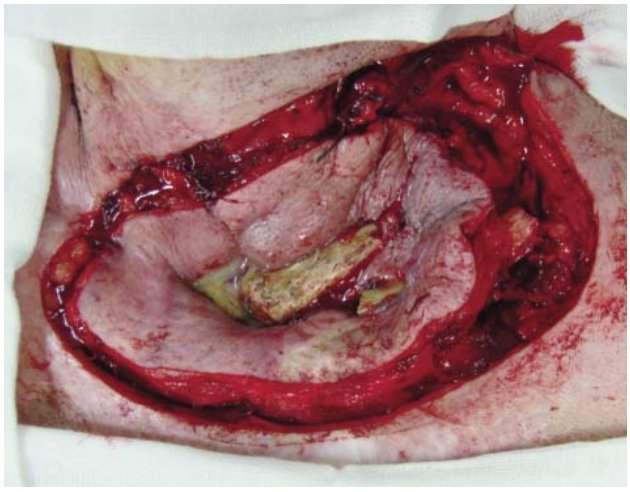

Figure 2: Lesion in the left clavicular region being resected

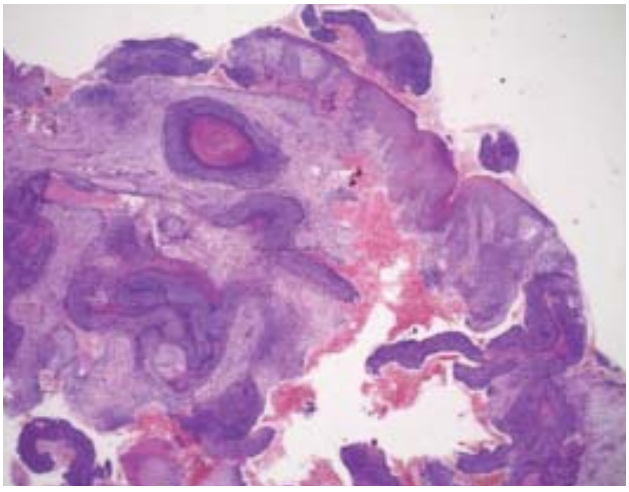

Figure 3: Histopathology (increased 25x) – An ulcerated neoplasm composed of solid blocks with a lobular growth pattern, reaching as far as the deep dermis

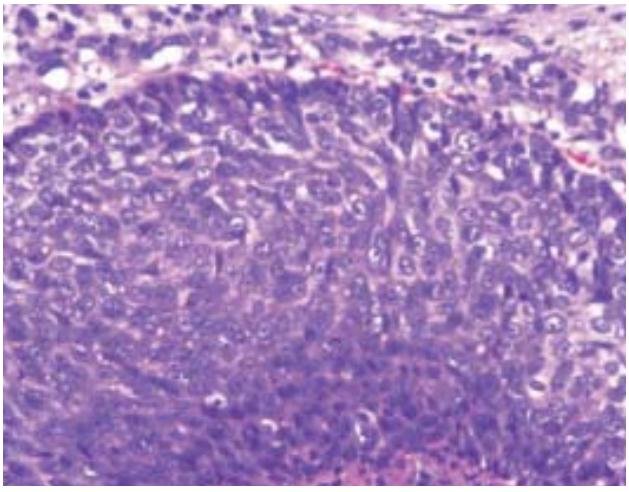

Figure 5: Histopathology (increased 400x) - Neoplastic blocks with keratinization, cytologic atypia and mitotic figures

mous cell carcinoma, basal-cell carcinoma, keratoacanthoma and malignant nodular melanoma should be considered. [1,7] However, such lesions are papulonodular, usually reach the reticular dermis, are prone to local recurrence and frequently metastasize. In contrast, the trichilemmal carcinoma presents a slow course and rarely recurs after its excision or metastasizes in other organs. [1]

Histologically, the tumor is purely intraepithelial or is more commonly associated with an invasive component centered around the pilosebaceous unit, which may reach from the epidermis to subcutaneous fat. [2] They are frequently continuous with the epidermis and the follicular epithelium. [4] When large, they have hemorrhage and/or necrosis focuses. Immunohistochemistry of the trichilemmal carcinoma is usually negative for CEA (carcinoembryonic antigen) and EMA (epithelial membrane antigen), although late positive results have occasionally been documented. [1,11]

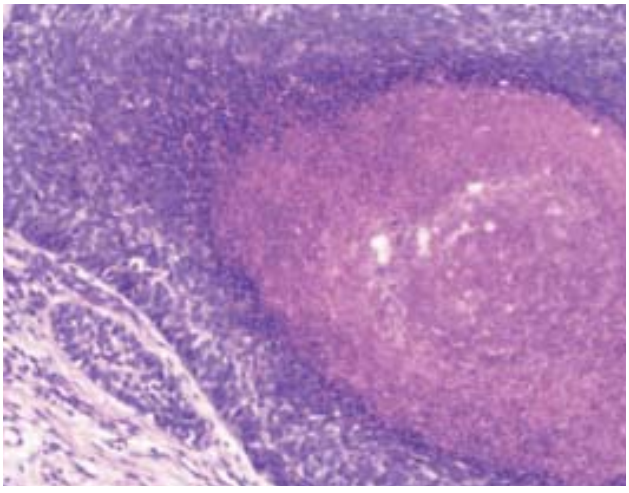

Figure 4: Histopathology (increased 200x)- Neoplastic blocks with proliferation of epithelial cells, at times presenting clear cytoplasm with peripheral palisading and areas of central necrosis

The treatment is exclusively surgical. [1,11] Simple excision with adequate margins is safe, inexpensive and effective for the treatment of trichilemmal carcinoma. [5,10]

The treatment recommended in the literature is micrographic Mohs surgery, as it does not show recurrence signs even after many years of treatment. [2,3,4,6]

The trichilemmal carcinoma generally has good prognosis and reports of deep invasion and local recurrence cases are uncommon. [4,5]

Ko T *et al.* report the case of 2 patients with trichilemmal carcinoma developed in burn scars. Histologically, such tumors present as proliferation of lobular cells continuous with the epidermis, composed of large atypical cells with clear cytoplasm and PAS-positive. [12]

The present study shows a recurrent trichilemmal carcinoma case of difficult treatment, which presented on the same location where a basal-cell carcinoma had been previously treated with surgery and radiotherapy. ❑

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