



# Inguinal ulcerated sebaceous carcinoma: an unusual presentation\*

Carcinoma sebáceo inguinal ulcerado: apresentação incomum

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**Abstract:** Sebaceous carcinoma is a rare and aggressive skin tumor. It can be located in any area of the body, the most commonly involved area being the periorbital region. It does not entail a typical clinical presentation, which explains the often late diagnosis. The aim of this report is to outline the rarity of the disease and its atypical clinical description, since to this day, inguinal ulcers with clinical manifestation have not been reported. We present and discuss a case of sebaceous carcinoma with an unusual clinical presentation, in an elderly male patient. The precise approach to genital ulcers, as shown in this case, is a diagnostic challenge that requires a great deal of effort on the part of the clinician.

**Keywords:** Sebaceous gland diseases; Sebaceous gland neoplasms; Sebaceous glands; Ulcer

**Resumo:** O carcinoma sebáceo é um tumor cutâneo raro e agressivo. Pode localizar-se em qualquer área do corpo sendo a região periorbital a mais comumente envolvida. Ele não tem uma apresentação clínica típica, o que explica o diagnóstico frequentemente tardio. O objetivo deste relato é apresentar a raridade da doença e uma descrição clínica atípica, uma vez que, até a presente data, não foi relatada úlcera inguinal como manifestação clínica. Apresentamos e discutimos um caso de carcinoma sebáceo, com uma apresentação clínica incomum em um paciente idoso do sexo masculino. A abordagem de úlceras genitais, como mostrado no presente caso, é um desafio diagnóstico que requer uma grande quantidade de esforço por parte do clínico.

**Palavras-chave:** Doenças das glândulas sebáceas; Glândulas sebáceas; Neoplasias das glândulas sebáceas; Úlcera

## INTRODUCTION

Genital and perigenital ulcers are a common reason for appointments with dermatologists in daily practice. How to approach them represents an important diagnostic challenge and, in most cases, they are initially regarded as a sexually transmitted disease (STD). Currently, this is the main cause mentioned in reports on infectious diseases, particularly among people aged between 15 and 50.<sup>1</sup> In many countries, algorithms have been developed to treat urethral syn-

dromes and genital ulcers, based on the local reality. For ulcers that do not have a history of genital herpes and have endured for more than 4 weeks, the recommendation is to carry out treatment for chancroid and syphilis, early treatment for donovanosis, as well as a biopsy. The rationale is to cover all etiological possibilities, including cancer, taking into consideration the limited resources.<sup>2</sup>

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Skin cancer should always be considered a possibility, especially for ulcers that endure, grow rapidly and/or behave aggressively.<sup>3</sup> The possible types of cancer include, essentially: squamous cell carcinoma, basal cell carcinoma and melanoma.<sup>4</sup>

The following is a case report that was forwarded to our department due to the possibility of it involving an STD. However, during the case study, the biopsy of the lesion revealed the presence of moderately differentiated sebaceous carcinoma (SC). A recent search, drawing on sources such as the Cochrane Library and publications containing primary information, Medline and Embase, made no reference to inguinal ulcers as a manifestation of sebaceous carcinoma, rendering the documented case original.

### CASE REPORT

A 63-year-old male patient, with a two-year history of painful inguinal mass, progressively increasing in volume. Subsequently, the inguinal mass to the left fistulized and the ulcerated lesion grew, reaching 12cm. The pathological background included reports of pulmonary tuberculosis treated 15 years before. The physical examination revealed an ulcer in the left inguinal region with malodorous and purulent exudate containing live larvae; hard right inguinal lymph node, adhered to deep plans, along with multiple yellowish papules in the posterior region of the penis (Figures 1 and 2). Serology tests were carried out for syphilis, HIV and *Chlamydia trachomatis*, as well as screening for acid-alcohol-resistant bacilli and a PPD exam, and all the tests were negative. A histology of the ulcer's border revealed atypical cells with prominent nucleoli, eosinophilic cytoplasm with multiple micro-vacuolizations and atypical mitoses (Figures 3 and 4). A Sudan III test showed vacuoles containing fat inside the tumoral cells (Figure 5). The immunohistochemical exam came back positive for the epithelial membrane antigen (EMA) and CK-7. The EMA revealed transmembrane positivity in sebaceous cells (Figure 6). The biopsies of the papules located on the penis and the right lymph node, revealed the presence of atypical cells with similar characteristics. Thus, our diagnostic conclusion was moderately differentiated sebaceous carcinoma with lymphatic and skin of the penis metastases as well as secondary myiasis associated with the ulceration brought about by sebaceous carcinoma. An investigation was conducted to check for Muir-Torre syndrome. A complete colonoscopy, thorax radiograph and urine analysis did not show any pathological alterations. The possibility of neoplasias in other organs was explored and then discarded. However, the abdominal and pelvic computerized axial tomography showed extensive damage to

retroperitoneal lymphatic chains and multiple contralateral inguinal adenopathies and the tumor inside the deep muscular plan.



FIGURE 1: Ulcer in the left inguinal region, dirty bottom, 12cm in diameter, with live larvae, and purulent and malodorous exudate. Hard, irregular and unclear defined borders



FIGURE 2: Left inguinal ulcer after treatment of larvae

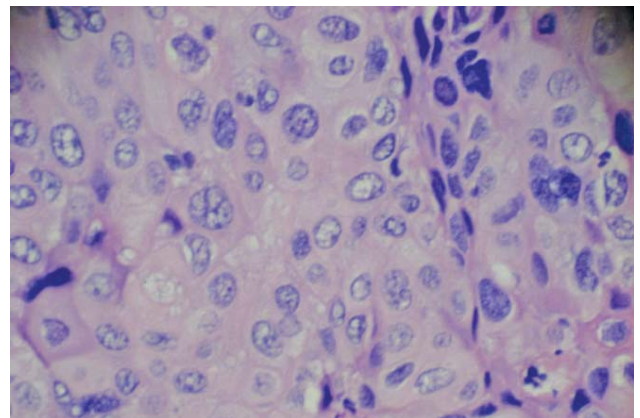


FIGURE 3: Atypical round and polygonal cells with pleomorphic nuclei, prominent nucleoli, and eosinophilic cytoplasm with micro-vacuolizations. Presence of atypical mitoses. (100x)



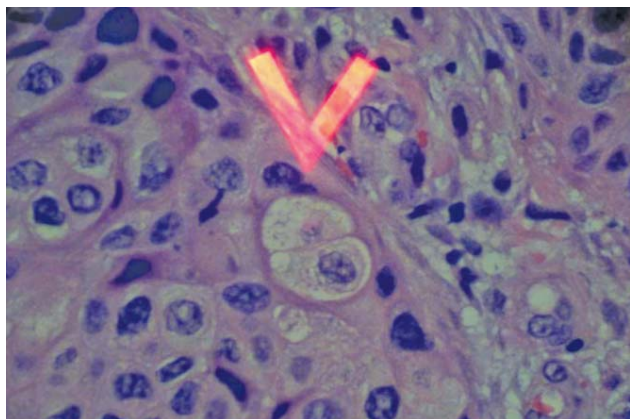


FIGURE 4: Detail of atypical cells with several intracytoplasmic microvacuolizations

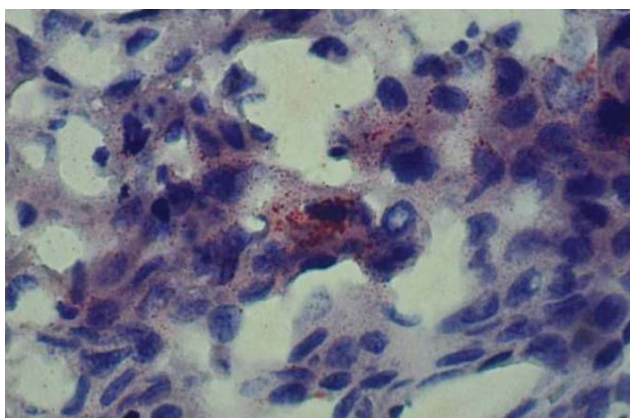


FIGURE 5: Sudan III - vacuoles with lipid content inside the tumoral cells

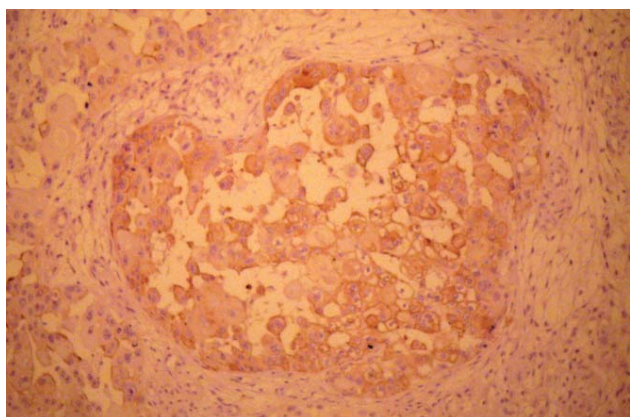


FIGURE 6: Epithelial membrane antigen (EMA), showing transmembrane positivity of the sebaceous cells

## DISCUSSION

SC is a malignant tumor, derived from the aneural epithelium of sebaceous glands. It can emerge in any area of the scalp but most tumors are found on the head and neck, especially the eyelids.<sup>5,6</sup> Wick *et al* reported that 25% of sebaceous carcinoma cases are extraocular.<sup>7</sup>

Clinical care is not specific, thus delaying diagnosis and affecting prognosis significantly. Extraocular lesions are described as firm, yellow-pink nodules that grow slowly and a third of cases present a hemorrhagic surface.

To date, it has been described in the following areas: the external auditory canal, oral mucosa, scalp, vulva, ovarian cysts, parotid, cervix, breasts, lungs, larynx, pharynx, palmoplantar region, nose, anal margin, penis and saliva glands.<sup>8,9</sup>

Its pathogenesis is still unclear. SC has been associated with Muir-Torre syndrome, infections from the human papillomavirus (HPV) and previous use of diuretics and radiotherapy.

The gold standard for diagnosis is a biopsy and histological examination of the lesion.<sup>5</sup> Some studies initially recommend a fine needle aspiration biopsy, achieving good results. However, if these results come back negative, a biopsy should be performed. The traditional coloring by hematoxylin-eosin makes diagnosis possible in most cases, though special colorings and immunohistochemistry might also be necessary. Coloring using Sudan is recommended, which identifies the lipid content of the interior part of the well-differentiated tumoral cells.<sup>4</sup> Immunohistochemistry is recommended to identify primarily the epithelial membrane antigen.<sup>10</sup>

Surgery remains the most efficient form of treatment, with margins of 5-6mm, histopathology and a detailed series of borders of the surgical piece, seeking pagetoid involvement in relation to its limits.<sup>5</sup> Radiotherapy and chemotherapy should be considered as palliative treatments in cases where the disease is advanced or has metastasized.<sup>5</sup>

As regards prognosis, Rao *et al* were able to establish the histopathological characteristics associated with worse results: vascular, lymphatic and orbital, invasion; involvement of both eyelids; low differentiation, multicentric origin; duration of over 6 months, tumor diameter of over 10mm; pagetoid invasion and infiltrative pattern.

Garrido *et al* have shown that extraocular locations seem to have a better prognosis,<sup>6</sup> based on the lower rate of metastases observed. However, extraocular case reports are on the increase, questioning previous observations, as is the case with our patient.

This case was described in order to provide a report on the first patient with this type of clinical manifestation. Ulcers in this area represent a constant diagnostic challenge, demanding great efforts to arrive at the correct diagnosis. Our report aims to draw attention to the presence of neoplastic lesions in this region. □

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