

Left Buttock Sebaceous Carcinoma: A Case Report and Review of the Literature

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ABSTRACT: Sebaceous carcinoma is a rare, aggressive, malignant tumour, characterized by a tendency for local recurrence and distant metastases. This case report presents a rare form of sebaceous carcinoma in an 86 years-old female. Clinical examination revealed a 12cm nodular tumour, yellow-brown in colour, located on the left buttock of the patient. Ultrasound examination indicated that no ganglions were affected; dermatological examination suggested that the tumour had an unpredictable evolution. Treatment was surgical with a 2cm wide excision, followed by reconstruction with a VY skin flap, from the left side of the buttock. Histological exam supported the diagnosis of sebaceous carcinoma; the tumour was removed with a safety margin of healthy tissue. Sebaceous carcinoma is a malignant tumour with unpredictable evolution. Diagnosis is based on histological examination. Treatment of choice consists in complete wide surgical excision.

KEYWORDS: Sebaceous carcinoma, histological examination, malignant tumor.

Introduction

Sebaceous carcinoma is a rare, aggressive tumor that usually develops in the upper sections of the body (head, neck, quite frequently at the eyelid level), being associated with the presence of Meibomian glands or Zeiss glands [1-4].

It is the third most common malignant tumor of the eyelids.

A less common location for sebaceous carcinoma is the torso; very rarely, it may develop on the limbs or in non-specific areas [1,3].

Some authors claim that the tumor develops from the epithelial cells of the sebaceous glands [5], others support the idea that it comes from the differentiation of a pluripotent cell [6].

Most lesions occur de novo [6], but there are cases in which the tumor evolves from a pre-existing sebaceous nevus [7,8].

Shields et al. and Vosoghi et al. [9,10] reported that sebaceous carcinoma is more common in women, while Dasgupta et al. [3] found it more common in men.

The average age at which sebaceous carcinoma occurs is 70 years [3] or less [10].

The etiology is unknown, but there seem to be a number of incriminating risk factors: UV radiation exposure [1], advanced age [3,11], female patients [9,10], Asian race [3,9,12],

previous irradiation of the head and neck [3,13], a genetic predisposition to Muir-Torre syndrome [14] or possible familial retinoblastoma [13].

We present a case of sebaceous carcinoma, a rare form characterized by an unusual size (12cm), located at the level of the supero-internal quadrant of the left buttock.

Case Report

Patient B.E., an 86 years-old female, was admitted at the Clinical Emergency Hospital of Craiova, presenting a tumor at the level of the left buttock.

This patient had a history of hypertension, without other significant pathological conditions.

Anamnesis revealed that the tumor appeared several years ago, and had a slow growth.

In the last 12 months, the growth has intensified, which is why the patient requested immediate medical treatment.

We specify that B.E. expressed her informed consent for treatment and data processing.

This report was approved by the Ethics Commission, U.M.F. of Craiova.

Clinical examination revealed a cutaneous tumor formation with a diameter of approximately 12cm, with nodular shape, not

very prominent, yellow-brown with well-defined edges, located on the left buttock.

The formation had a dense consistency and was mobile compared to the deep plans.

There was no pain or other associated symptoms.

No localized or generalized lymphadenopathy was detected by palpation.

Usual blood tests, coagulation tests, renal and hepatic parameters were performed, which were within normal limits.

Ultrasound of the tumor revealed an invasive formation in the subcutaneous fat, which did not reach the muscle plane.

The lymph nodes showed no lymphadenopathy.

Dermatological examination suggested a tumor with unpredictable evolution and recommended excision with oncological safety margins (Figure 1).



Figure 1. Macroscopic aspect of the tumor.

We informed the patient regarding her health status and we explained the need to perform a biopsy, in order to have a correct and complete diagnosis.

However, the patient requested surgical excision of the tumor, considering the advanced age and the difficulty to return to hospital during the present COVID-19 pandemics.

Therefore, we decided to perform surgery under spinal anesthesia.

The patient was placed in a prone position.

We made an elliptical incision that circumscribed the tumor with a macroscopic safety margin of 2cm, considering that the safety margin recommended in these cases is 1cm (Figure 2).

Block excision was performed up to the fascial plane.

The excision sample marked at 12 o'clock was sent for histopathological examination.



Figure 2. Image after excision and creation of VY Flap.

To close the defect, a „VY” skin flap was created and mobilized from the side of the buttock (Figure 3).

Two suction drain tubes were installed, and the donor area was closed by suturing.

The evolution was favorable postoperatively (Figure 4).



Figure 3. VY Flap.



Figure 4. Flap is sutured in place.

The drainage tubes were extracted 2 days after the operation, and the patient was discharged 5 days postoperatively, with the indication to return for the histopathological result and to check-up every six months for 3 years.

The histopathological examination revealed a moderate differentiated sebaceous carcinoma in which areas of sebaceous differentiation were observed at the periphery of the infiltrative neoplastic islands (Figure 5, Figure 6).

The tumor showed irregular lobules and cords of cells with varying degrees of sebaceous and pattern differentiation of infiltrative growth in the dermis.

In the center of the tumor islands there were areas of necrosis (comedonecrosis), marked nuclear atypia and increased mitotic activity.

A mixture was observed in different proportions of cells with sebaceous differentiation, and smaller undifferentiated cells with a primitive appearance.

Cells with sebaceous differentiation have notched nuclei, centrally located, with foamy and multivacuolated cytoplasm, slightly basophilic, secondary to the accumulation of intracytoplasmic lipids.

The less differentiated cells contain intensely basophilic cytoplasm and high nucleocytoplasmic ratio, hyperchromatic nuclei with prominent nucleoli and intense mitotic activity.

Histopathological examination also confirmed that the tumor margins had healthy tissue.

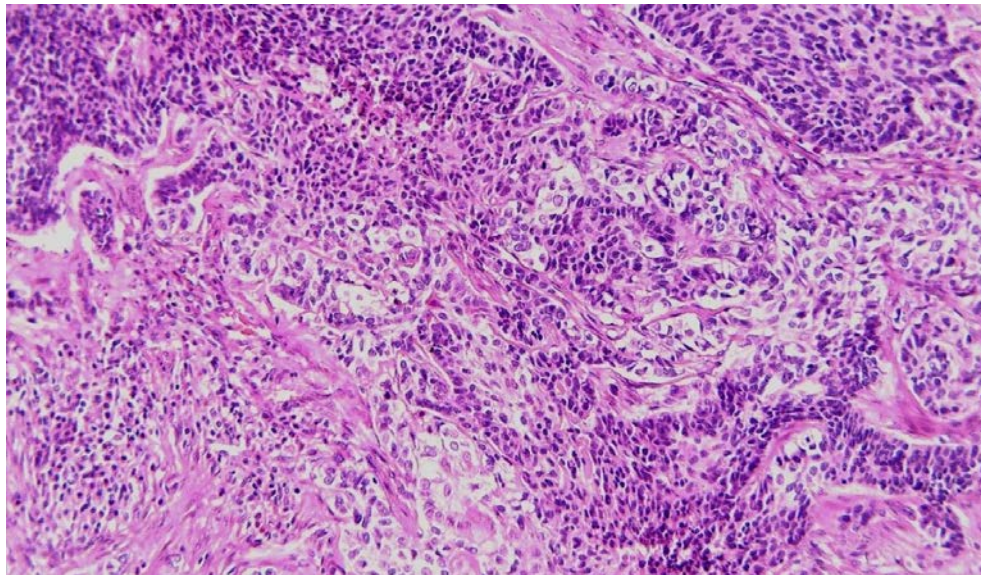


Figure 5. Sebaceous carcinoma moderately differentiated: areas of sebaceous differentiation in periphery of neoplastic islands, HE staining, x200.

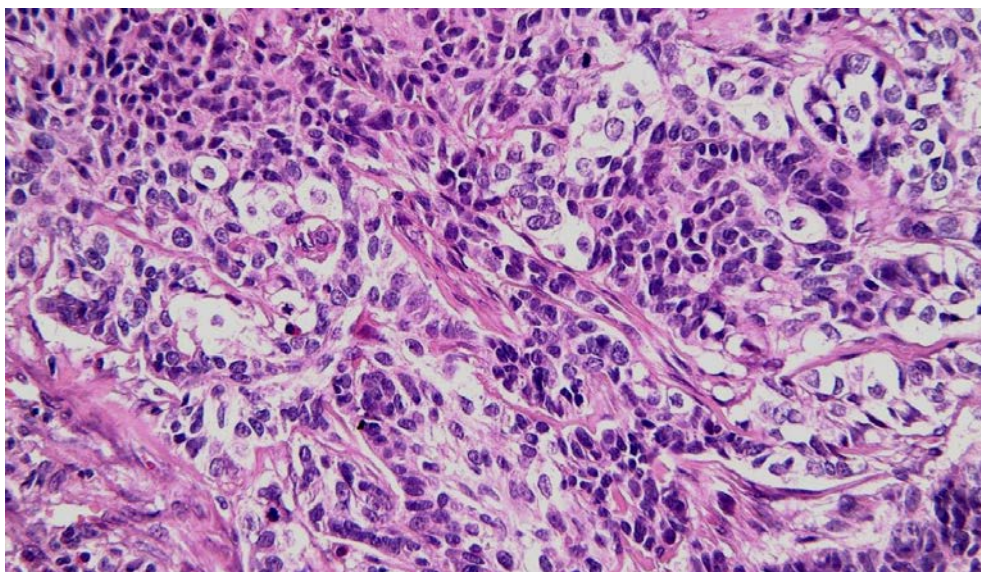


Figure 6. Moderately differentiated sebaceous carcinoma, HE staining, x400.

Discussions

Sebaceous carcinoma is a malignant skin tumor, extremely rare, aggressive, with a tendency for both local recurrence and distant metastases.

Being an extremely rare tumor, there are few studies that refer to its incidence; Dasgupta et al. [3] reported 2.03 cases per 1000000 for Caucasians, 1.07 cases per 1000000 for Asians; and 0.48 cases per 1000000 for Africans.

Tripathi et al. reported 0.32 cases per 100000 for males and 0.16 cases per 100000 for females [15].

Margo et Mulla reported 0.5 cases per 1000000 for eyelid sebaceous carcinomas, in patients over 20 years-old from Florida [16].

Although the case presented in this study concerns a woman, the literature shows that sebaceous carcinoma affects slightly more men 54% [3], 52% [17], 54.64% [15] with a mean age of over 70 years [3,15,17].

Regarding the location of this tumor, the vast majority of sebaceous carcinomas (75%) occur in the periocular region, usually in the eyelid area [1].

Extraocular sebaceous carcinomas account for only 25% of all reported cases of sebaceous carcinoma [5,18].

Few cases of sebaceous carcinoma with special localization are presented in the literature.

Thus, Mohamed et al. [1] described 5 cases of sebaceous carcinoma, of which 4 are in the cephalic extremity and one in the chest, Sen et al. [19] described a sebaceous carcinoma located in the cheek, Diaz et al. [18] described two cases, the first with the tumor located in the area of the armpit and the other with the tumor in the cheek, Panjwani et al. [20] described three cases with the tumor located on the armpit, chest, face, Natarajan et al. [21] described a sebaceous carcinoma of the scalp, Alzaraa et al. [22] described a case localized on the breast, Wick et al. [23] described an extremely rare localization of sebaceous carcinoma in the penis, and Manveen et al. described an intraoral sebaceous carcinoma [24].

For the patient presented in this case report, tumor started to develop many years before, in nodular form with a slow evolution, which intensified in the last year.

This form of onset was also described by Buitrago et Joseph [6], who stated that sebaceous carcinoma most often begins as a subcutaneous nodule, but the same author states

that it may also start as a nonspecific inflammation manifested by thickening of the skin, as a pedicle lesion or as an irregular mass with extensive tissue invasion.

Regarding the color of the tumor that in the case presented was yellow-brown, Mohamed et al, Diaz et al. [1,18] studies state that it varies between pink and red, sometimes yellowish or purple.

One third of cases has surface ulcers and occasionally bleeds [18].

Patients rarely report pain [5,6,25].

The size of the tumor can vary between 6 mm and 20cm [5,6,26].

Sometimes, clinical and even histological diagnosis is difficult, being similar to a benign or less aggressive lesion [25,27-29].

In these situations, the primary tumor does not appear to develop or has a benign appearance but produces metastases in the regional lymph nodes or other sites [25].

Shin et al. [30] estimated that regional or distant metastasis occurs in 8% to 28% of patients at some point during the disease [31], and Singh et al. [32] estimated that 22% of patients with sebaceous carcinoma die from visceral metastases.

Given that the clinical aspect is not conclusive for establishing the definite diagnosis, histological examination is compulsory in this sense.

However, we had to consider the current context of COVID-19 pandemics, patient's advanced age, and her agreement upon the proposed treatment.

Therefore, we opted to surgically remove the tumor and afterwards perform a histopathological examination, which would indicate the subsequent therapeutic actions.

Jawanda et al. [24] cited the WHO definition of sebaceous carcinoma: "a malignant tumor composed of sebaceous cells of varying maturity that are arranged in sheets and/or nests with varying degrees of pleomorphism, nuclear atypia and invasiveness" [32].

Shin-ichi et al. proposed the following classification of sebaceous tumors [33]:

- sebaceoma-benign neoplasm with well-defined architecture and without atypical neoplastic cells;
- LGSC (low-grade sebaceous carcinoma) or BSN (borderline sebaceous neoplasm)-tumor with intermediate malignancy, well-defined architecture, and nuclear atypia), and

- SC (sebaceous carcinoma)-malignant tumor with invasive growth and obvious nuclear atypia.

Sebaceous carcinoma is in turn classified according to the proportion of sebocytes on three levels [34]: well differentiated-sebocytes predominate; moderately differentiated-sebocytes are few but easily recognized, and poorly differentiated component-sebocytes are rare.

Several studies have shown variable proportions of these categories [35,36].

Commonly recognized histological patterns of infiltrative growth are lobular, papillary, and trabecular.

The lobes may vary in size within the same tumor, and some lobed tumors may display central necrosis (comedonecrosis).

This central necrosis is described as foci of exaggerated holocrine secretion, rather than true necrosis.

The main cell types found in sebaceous carcinoma are basaloid, basal squamous and epidermoid [37].

Basal squamous cells display intermediate cytoplasmic features of the basaloid and epidermoid types, and cytoplasm richer than the basaloid cell [37].

Ocular lesions often show a pagetoid pattern in the affected conjunctiva or eyelid epidermis [38], but this is rarely seen in extraocular sebaceous carcinoma [35].

In the present case, the appearance of moderate differentiated sebaceous carcinoma was present in the tumor tissue, but the edges of the excised mass showed no modifications.

The unanimously accepted treatment for sebaceous carcinoma is the surgical one and its main objective is the complete excision with free edges, maintaining the function and appearance.

The earlier this it is performed, the better the prognosis [1].

The proposed safety margin is 5-6mm [5,6,39].

In 2019, "Sebaceous Carcinoma: Evidence-Based Clinical Practice Guidelines" [40] was developed by a group of specialists in general dermatology, oncology (cutaneous, ocular, surgical, medical, radiological), plastic surgeons, statisticians, and other researchers, to determine the best conduct in establishing the diagnosis, risk assessment and management of extraocular sebaceous carcinoma and periocular in people without a genetic predisposition.

This guide proposed a management regimen for sebaceous carcinoma tumors.

Thus, recommendations were made for diagnosis, surgical treatment, radiotherapy and post-treatment care.

To establish the diagnosis, anamnesis and clinical examination are performed, but a biopsy is also required;

In addition, the differential diagnosis with other diseases is made by histological tests.

For our patient we performed anamnesis, clinical examination, dermatological examination which established that it was a tumor formation with unpredictable evolution, and ultrasound examination which established that there are no locoregional metastases.

Excision biopsy was performed to remove the tumor with 2cm safety margins.

The VY flap used to close the defect is an easy to create and reliable method [41].

The definite diagnosis was established after histological examination.

Overall, we consider that our actions comply with the recommendations given in the above-mentioned guide, since the removal was surgical, we assured a safety margin larger than the recommended one, and this treatment was in accordance with the patient's request.

Given the amount of cutaneous tissue in the buttock area, the reconstruction process assured the anatomic and functional restoration of that region.

Another variant of surgical treatment may be Mohs surgery, but it was not available for this case.

The same guide [40] states that following this surgical technique, there are the lowest chances of recurrence but at the same time the wide local excision cannot be compared with Mohs surgery due to the lack of studies.

Sentinel node biopsy is not usually performed in extraocular sebaceous carcinoma.

Radiation therapy is considered in cases of nerve or lymph node involvement and as primary treatment in patients who are not eligible for surgery.

It has an adjuvant role in combination with surgery and applies to advanced primary tumor formations, those with invaded resection margins or perineural invasion.

Further studies are needed to define the role of radiotherapy in the management of extraocular sebaceous carcinomas.

Post-treatment clinical examination should be performed at least every 6 months for 3 years.

In terms of prognosis, local recurrences occur in 32% of cases [6] after wide excision, and only 12% after MOHS surgery [42,43].

In the past, it was considered that the extraocular location of the tumor implied a worse prognosis (5), but recent studies show that the prognosis is not influenced by the location of the tumor [6,44-46].

Extraocular localization and smaller tumors are associated with a lower rate of metastasis, and therefore have a lower mortality [3].

Negative prognostic factors include lymph node involvement, multicenter localization, poorly differentiated tumors [5].

Mortality reported in the literature is 20%-32% [47].

In his study on 1349 cases of sebaceous carcinoma, Dasgupta et al. reported the 5-year survival of 91.9% of cases and 79.2% of cases after 10 years [3].

Conclusion

Sebaceous carcinoma is an aggressive, rare form of malignant tumor, mostly located on the eyelid.

We presented a rare case of a large sebaceous carcinoma (12cm in diameter), moderately differentiated, located on the left buttock area.

Even if the election treatment implies a histopathological examination prior to surgery, in the current context we decided that the best treatment for our patient was directly the surgical removal of the tumor, with a safety margin larger than the recommended one.

Time and continuous monitoring will define the efficiency of this method.

Conflict of interests

None to declare.

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