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(CASE REPORT)

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Temporo-frontal and parotidial hidradenocarcinoma: A case report

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Abstract

Introduction: Hidradenocarcinoma is a very rare malignant tumor of the sweat glands. Diagnosis is based essentially on histological and immunohistochemical studies. Standard treatment is surgery combined with radiotherapy if there is a prognostic factor for local recurrence. We report a case treated in our department.

Observation: A 24-year-old female patient with no specific pathological history consulted the Joliot Curie Institute of the Dantec Hospital for a forehead nodule that appeared in 2012 and was painless, evolving towards February 2021 into two right temporo-maxillary masses progressively increasing in volume. Examination revealed two tumors of stony consistency, with smooth, ulcerated surface, infiltrated center, raised borders, budding, not bleeding on contact, painless, the largest measuring approximately 16cm, located at the temporofrontal and right parotid level with extension to the right auricle associated with homolateral periorbital edema and right retroauricular and submental adenopathies, the largest measuring 3x2cm. Parotid skin biopsy revealed nodular tumour masses in the deep dermis, composed of a proliferation of medium- to large-sized epithelial cells, sometimes clear with central nuclei and numerous atypia and abnormal mitoses. Epidermoid differentiation can be seen in places. The stroma is fibrino-hyaline. The appearance was suggestive of a hidradenocarcinoma. The cervical CT scan revealed an inhomogeneous tissue process, strongly enhanced after well-limited contrast injection, in close contact with the right parotid gland. Similar homolateral parietofrontal lesions were noted, as well as a spiky homolateral frontoparietal appearance with cortical irregularity. The extension work-up was negative.

Conclusion: Hidradenocarcinoma is a very rare malignant tumour with a high risk of locoregional and distant recurrence, mainly in lymph nodes. Its prognosis is poor.

Keywords: Hidradenocarcinoma; Rare tumors; Sweat glands; Cancers; Young

1. Introduction

Hidradenocarcinoma is a rare malignant tumor of the sweat glands, accounting for less than 0.001% of all cancers [1,2]. It is an aggressive tumor with a very poor prognosis, occurring preferentially in the head and neck and rarely in the limbs [3]. Diagnosis is based on histological and immunohistochemical studies [4,5]. Standard treatment is surgery, with a recurrence rate of 50% [6]. And for good local control, external radiotherapy may be indicated in cases where local recurrence is a factor [7]. In metastatic forms, several lines of chemotherapy have been used in the literature, notably 5-Fluorouracil and capecitabine [8,9].

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We report the case of a 24-year-old female patient with temporofrontal and parotid hidradenocarcinoma and cervical adenopathy.

2. Observation

A 24-year-old patient with no specific pathological history reports the appearance of a painless forehead nodule in 2012, which evolved into two progressively enlarging right temporo-maxillary masses around February 2021. After a long period of misdiagnosis and worsening health, she finally consulted the dermatology department at Hôpital Dantec and then the Joliot Curie Institute, where physical examination revealed two tumours of stony consistency, with a smooth, ulcerated surface, with an infiltrated center, raised margins, budding, not bleeding on contact, painless, the largest measuring approximately 16cm, located in the right temporofrontal and parotid region, with extension to the right auricle (Fig. 1) associated with homolateral periorbital edema and right retroauricular and submental adenopathies measuring 3x2cm. Parotid skin biopsy revealed a malignant lesion deep in the dermis. The lesion consisted of nodular tumour masses composed of a proliferation of medium to large epithelial cells, sometimes of clear appearance with central nuclei showing numerous atypia and abnormal mitoses. Epidermoid differentiation can be seen in places. The stroma is fibrino-hyaline. The histological appearance was that of an eccrine sweat carcinoma developed at the expense of the deep secretory portion of the eccrine glands, in particular an eccrine hidradenocarcinoma (Fig.2). Cervical CT revealed an inhomogeneous tissue process, strongly enhanced after well-limited contrast injection, in close contact with the right parotid gland. Similar homolateral parietofrontal lesions were noted, as well as a spiky homolateral frontoparietal appearance with cortical irregularity. The extension work-up was unremarkable.

The case was presented at a multidisciplinary consultation meeting. The decision was taken to perform carboplatin-paclitaxel induction chemotherapy followed by concomitant radio chemotherapy with weekly cisplatin. The patient died before the first course of treatment.

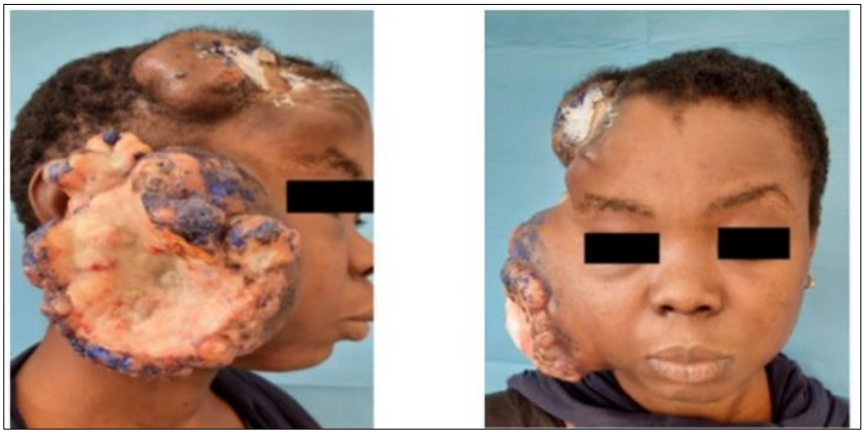

Figure 1 Macroscopic aspects of a hidradenocarcinoma in a 24-year-old woman

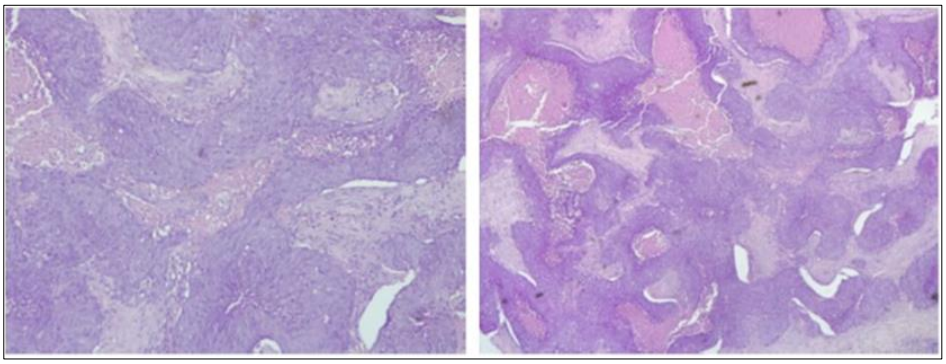

Figure 2 Hidradenocarcinoma in a 24-year-old woman

3. Discussion

Hidradenocarcinoma was first reported in 1954 by Keasby and Headley [10]. It accounts for around 6% of malignant eccrine tumours and less than 0.001% of all tumours [1,2]. It is also known as clear-cell hidradenocarcinoma, clear-cell eccrine hidradenocarcinoma, clear-cell eccrine carcinoma, apocrine hidradenocarcinoma, malignant nodular hidradenoma or malignant acrospiroma. The average age of onset is 50, with a slight female predominance and no racial prevalence [11,12]. It classically occurs de novo and rarely results from a benign hidradenoma [13,14]. It occurs preferentially in the head and neck and rarely in the extremities [3,15]. However, other localizations have been more rarely described in the literature, such as the trunk, abdomen or scalp [16]. In our case, the age of onset was 24, and the localization was temporofrontal and parotid.

The diagnosis of certainty is based on anatomopathological study, which shows a massive intradermal tumour proliferation, with sweat differentiation cut by a dense, hyalinized stroma with nuclear pleomorphism and mitosis figures [4,5]. This tumoral proliferation is made up of medium-sized, rounded cells with eosinophilic or clear cytoplasm. Smaller, basophilic cells are visible in the periphery. Mitoses are numerous and sometimes atypical. Sometimes, nuclear atypia and mitoses are rare, and the appearance is closer to that of a benign hidradenoma. Tumor patches are hollowed out by small glandular cavities. Foci of keratinization, papillary aspects and mucin deposits may be noted [17]. Unlike benign hidradenoma, which is well limited, hidradenocarcinoma shows an asymmetric configuration, less cystic architecture, areas of necrosis and invasion of neighbouring tissues [15,17]. Immunohistochemistry shows expression of androgen receptor (AR), estrogen receptor (ER), progesterone receptor (PR), epidermal growth factor receptor (EGFR) and human epidermal growth factor receptor 2 (HER-2) [18].

Clinically, hidradenocarcinoma can be confused with infundibular and pilar cysts, cutaneous tuberculosis or dermatofibrosarcoma protuberans [3, 15]. The differential diagnosis of hidradenocarcinoma includes benign tumors such as hemangiomas, lipomas and lymphangiomas, and malignant tumors such as basal cell carcinomas, squamous cell carcinomas and malignant melanomas, other malignant adnexal carcinomas such as eccrine adenocarcinomas, cystic adenoid eccrine carcinomas, mucinous eccrine carcinomas, aggressive digital papillary adenocarcinomas and metastatic skin tumours [19]. Hidradenocarcinoma can also resemble primary carcinomas of the breast, salivary glands and lungs [19]. However, the former two tumors are generally differentiated by their positivity to TTF-1 (thyroid transcription factor-1), and the latter by the presence of focal granular necrosis and hemorrhage within the lesion, as well as its prominent vascularization. Renal cell carcinoma also expresses CD10 and epithelial membrane antigen (EMA) [14]. Basaloid eccrine carcinoma, ductal eccrine carcinoma, clear-cell eccrine carcinoma and other unspecified sweat gland carcinomas are further differentiation criteria. These tumors have a prominent eccrine component/differentiation, no characteristic clinical picture and are generally very difficult to differentiate from metastatic carcinomas. They are generally devoid of epidermal connection and simulate carcinomas of other parts of the body, including breast, thyroid, salivary gland and renal cell carcinomas. Eccrine ductal carcinoma resembles mammary ductal carcinoma. The differential diagnosis of eccrine basaloid carcinoma is broad and includes Merkel cell carcinoma, Ewing's sarcoma, metastatic carcinoma, as well as small-cell melanoma and squamous cell carcinoma [14].

The course of the disease may be marked by the occurrence of visceral and lymph node metastases, which are less frequently described in the literature [20,21].

As hidradenocarcinoma is an aggressive and extremely rare tumour, there is no consensus treatment to date [15]. The treatment of choice remains surgery based on wide excision with healthy margins [7,22,23]. The role of the sentinel node technique is widely debated in the literature [24]. Lymph node dissection is indicated in cases of clinical adenopathy and histological criteria of malignancy [25]. Prognostic factors such as large tumour size, low resection margins, presence of lymphatic emboli, nerve sheath invasion or lymph node capsular effraction are correlated with a high risk of recurrence, indicating adjuvant radiotherapy at 50-70Gy [16,25,26]. In metastatic forms, several lines of chemotherapy have been used (5-fluorouracil, capecitabine, cisplatin, docetaxel, paclitaxel) with partial or transient responses [11]. Trastuzumab may also play a role in disease stabilization [15].

4. Conclusion

Hidradenocarcinoma is a very rare and aggressive malignancy. The standard treatment remains surgery with wide excision followed by radiotherapy in case of local recurrence factors. Our patient's case reinforces the need to insist on a proper diagnostic approach for early and appropriate management.

Compliance with ethical standards

Disclosure of conflict of interest

The authors have declared no conflicts of interest.

Statement of ethical approval

The present research work does not contain any studies performed on animals/humans subjects by any of the authors.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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