Primary Adenosquamous Carcinoma Of The Skin In The Occipital Region: A Case Report

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Citation

Abstract
Primary cutaneous adenosquamous carcinomas are very rare entities with only few published reports in literature. We describe a primary cutaneous adenosquamous carcinoma found in the occipital region of a 53 year old man with a pediculated tumor of 7 cm in diameter and an ulcerated, verrucous aspect with serous exudations. There was no clinical or radiologic evidence of invasion of the cranial vault or another primary tumor e.g. of the salivary glands.

The histologic picture consisted of two different components: masses of cords of a squamous cell carcinoma and a second element with goblet cells in the center of the epidermal areas. Histochemical studies and immunohistochemistry demonstrated the presence of the two components described.

The epidermis was dysplastic. The carcinoma was infiltrative with a multifocal origin from the basal layer of the epidermis.

INTRODUCTION
Adenosquamous carcinomas - sometimes called mucoepidermoid carcinoma - are quite common neoplasms in some localizations as the salivary glands [1,2], upper respiratory tract or nasal mucosa. Nonetheless they are very rare primary lesions of the skin [3,4,5,6]. They are malignant epithelial neoplasms composed of mucin-secreting cells and squamous cells in varying proportions [7].

We describe a case of primary cutaneous adenosquamous carcinoma of the occipital region.

CASE REPORT
A 53 years old man noted a occipital scalp lesion (diameter about 1 cm) eight month prior to consultation. When consulting in our hospital the tumor had reached 7 cm in diameter, was pediculated with an ulcerated verrucous aspect and serous exudations. A peripheral biopsy was taken and reported as squamous cell carcinoma of the acantholytic type. Radiologically there an osseous infiltration was suspected (Figure 1) what histologically could be discarded. There was no clinical or radiologic evidence for any kind of metastasis or any other primary tumor e.g. of the salivary glands. Thereafter, the patient received radiation therapy (5000 cGY) resulting in a 50 % diminution of tumor followed by surgical excision.

Figure 1
Figure 1: CT scan of the tumor region after the radiation

MATERIAL AND METHODS
After fixation stains with hematoxylin and eosin, periodic acid of Schiff (PAS) with and without diastase digestion alcian blue (pH 2.5) were prepared. Immunohistochemistry was performed with the Streptavidin–Biotin-peroxidase complex technique using monoclonal antibodies to cytokeratin 7 and 8 (DAKO), carcinoembryonic antigen (CEA, DAKO, 1:100) and CA 15-3 (DAKO, 1:50).
PATHOLOGIC FINDINGS

GROSS DESCRIPTION

The specimen consisted of a 7.5 x 7.5 x 1.2 cm ellipse of skin and a segment of the cranial aponeurosis with a centrally-ulcerated nodular lesion (2.3 x 2.3 x 1.2 cm).

MICROSCOPIC DESCRIPTION

A malignant epithelial neoplasm was found in an focally ulcerated, elevated hyperplastic epidermis. The 0.8 cm infiltrating neoplasm had the features of a non-conventional squamous cell carcinoma (SCC). In the dermis, the polyedric, squamous cells with moderately hyperchromatic and pleomorphic nuclei that sometimes displayed nucleoli and an acidophil cytoplasm, formed cords, nests and islands and presented individual keratinization or horn pearl formation. Underneath in the dermis, the neoplasm tended to form nests more than cords and randomly situated in the masses of epidermoid cells glandular differentiation with and without the presence of central lumina was observed. The gland cells were cuboidal, high or low columnar goblet cells, with a peripherally situated nucleus and one huge intracytoplasmatic vacuole each. When there was a lumen, it contained slightly acidophil material that seemed to be mucinous secretion of the gland cells. The same material was observed in the cytoplasm of some cells originated close to the lumen. Both cell types displayed a moderate number of mitosis (Figure 2).

Figure 2

Figure 2: Squamous cell tumor nest with keratinization, gland formation by goblet cells and stromal response (a - H&E, b – Alcian blue, 40 x)

Under or close to the superficial dermis close to the ulcerations a foreign body reaction could be observed and a desmoplastic reaction surrounded the tumor masses in the deeper dermis. No invasion of blood vessels, nerves or the aponeurosis was observed.

The formerly described goblet cells and glandular secretions stained positive with PAS with and without prior diastase digestion and Alcian Blue (pH 2.5) thereby proving the presence of diastase-fast acid mucins.

Immunohistochemical stains using antibodies against Cytokeratin 7 and 8, Ca 15-3 and Carcinoembryonic Antigen (CEA) were performed. The epidermoid cells stained positive for Cytokeratin 7 and 8 with a cytoplasmatic pattern (Figure 3). The Ca 15-3 antibody marked primarily the membranes and less the cytoplasm of epidermoid cells. Those epidermoid cells located next to goblet cells stained positive while goblet cells did not stain at all. The CEA antibody stained the membranes of goblet cells (not shown).
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Figure 4
Figure 3A: – Squamous cells with positive staining for Cytokeratin 7. Note lack of staining in goblet cells

Figure 5
Figure 3B: Positive staining of paraluminous goblet cells and squamous cells for cytokeratin 8. Squamous tumor cell nests negative for cytokeratin 8.

DISCUSSION

We presented a case of a primary cutaneous adenosquamous carcinoma (ASC) in a 53-year-old man. Due to incomplete mapping of the tumor mass in a first biopsy resulting in an inappropriate, not at all representative specimen, the first microscopic diagnosis was forcibly equivocated. After re-revising the biopsy sections later, we confirmed this diagnosis as there were only signs of the acantholytic subtype.

The described tumor arose in the occipital regions, was elevated and not relatively flat what is contrary to earlier observations. Furthermore, we observed a notable sensitivity of the carcinoma to radiotherapy (50 % diminution of volume) and favorable postsurgical evolution and wound healing. The rapid growth - though we could not observe invasion of blood vessels, nerves, the aponeurosis or bone, the cytologic signs of a “low-grade” carcinoma and the larger tumor size are contrary to earlier descriptions [1]. The final outcome is to be determined by a thorough follow-up of our patient.

As previous reports of ASC stated that the sensitivity to radiotherapy was low, it should be emphasized that thereby we observed an important diminution of volume. Clearly, the principal therapy is surgical, but the possibility of radiotherapy should be considered if huge lesions are present or if important anatomic structures are involved.

The surgically excised specimen of the herein reported ASC exhibited the described histologic features [8]. There was a predominant epidermoid carcinoma component in nests with the striking presence of scattered islands of mucin-secreting cells. Both components were undoubtedly identified by histochemical and immunohistochemical stains. Weidner and Foucar [4] did not identify mucin secretion in 50 conventional squamous carcinomas and Nappi and colleagues [10] found CEA positivity only in one focus of keratinization in one of 37 adenoid (acantholytic) squamous cell carcinomas. The multiple epidermal connections, a markedly dispersed growth pattern, a relative deep infiltration with rapid evolution and a prominent hyalinization fibroblastic stromal response set this tumor further apart from classic squamous cell carcinoma. Taking these findings, the diagnosis of ASC can be made with certainty.

As mucoepidermoid carcinomas are a well-described entity of adnexal tumors with sweat gland differentiation, it could be possible to interpret ASCs likewise. Many characteristic features of ASCs like the dermal localization, the glandular structures and their obvious mode of coalescent formation, the mucin production and the positivity for CEA are found in adnexal tumors, too. But there are differences like the number and extension of epidermal connections that are reminiscent of acrosyringical structures in adnexal tumors, the cytologic grade, more frequent mitotic figures and a markedly less favorable biologic course. In order to differentiate between sweat gland carcinomas and the presented lesion, we favor the term “Adenosquamous Carcinoma” as it is highly descriptive and names the dominant cell type. Besides, the term “mucoepidermoid” is established for a different type of neoplasms [11] and its use should be restricted to those. Riedlinger and colleagues [12] even state that the term “mucoepidermoid carcinoma of the
skin” should be reserved to a disease with a different histologic picture and is a different entity that should be distinguished from primary cutaneous adenosquamous carcinomas due to its different clinical behavior.

Weidner et al. (4) and Landman and colleagues (13) suggested that ASC might have their origin in pluripotent epithelial cells near or within the acrosyringial portions of sweat ducts. The secretion of diastase resistant mucins of sweat gland tumors (2) and the presence of CEA in eccrine and apocrine glands (14) make this view tenting yet such a cell has not been described till date. This remains therefore a theoretical possibility of explanation.

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References
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