Primary cutaneous adenoid carcinoma of the scalp

Primäres kutanes adenoidzystisches Karzinom der Kopfhaut

Abstract

Primary adenoid carcinoma are rare skin tumors. We present a 75-year-old female with this primary cutaneous tumor of the scalp with additional bone involvement. Wide scalp excision with bone enclosure, latissimus-dorsi-free-flap defect overage, and subsequent radiation slowed down the disease but could not prevent further skull infiltration.

Keywords: adenoid carcinoma, skin cancer, tumor of the scalp

Zusammenfassung

Primäre adenoidzystische Karzinome sind seltene Hauttumoren. Wir stellen eine 75-jährige Patientin mit einem derartigen Tumor der Kopfhaut vor. Weite Exzision unter Einschluss von Teilen der knöchernen Kalotte, Defektdeckung mit einem freien Latissimus-dorsi-Muskellappen und nachfolgende Bestrahlung führten zu einer Verzögerung des Tumorwachstums, konnten jedoch letztlich die weitere Schädelinfiltration nicht verhindern.

Schlüsselwörter: adenoidzystisches Karzinom, Hauttumor, Tumor der Kopfhaut

Meike Keck[1]
Klaus Ueberreiter[1]
Ursula Tanzella[1]
Dietrich Doll[2]
Björn Dirk Krapohl[2]

1 Plastic Surgery Center, Birkenwerder, Germany

2 German Armed Forces Hospital of Berlin, Plastic and Hand Surgery, Department of Surgery, Berlin, Germany

Case description

A 75-year-old female presented with two nodules of 1.5 cm and 1.0 cm on the scalp in an area of previous excision. A year before the patient's physician had excised a mass without taking samples for histology (initial situation: Figure 1; preoperative state one year after first excision: Figure 2). The tumour was indolent and accompanied by focal alopecia. After tumour excision (intraoperative situs: Figure 3) histopathological findings showed a firm tumour involving the dermis and subcutaneous fat, consisting of basaloid epithelial cells arranged in cribriform and tubular pattern as well as some focal solid pattern. No periodic acid-Schiff-positive material was found. The resection margins were infiltrated. Perineural invasion was not seen. It was discussed with the pathologist whether the tumour was most likely a distant metastasis or a rare primary cutaneous tumour.

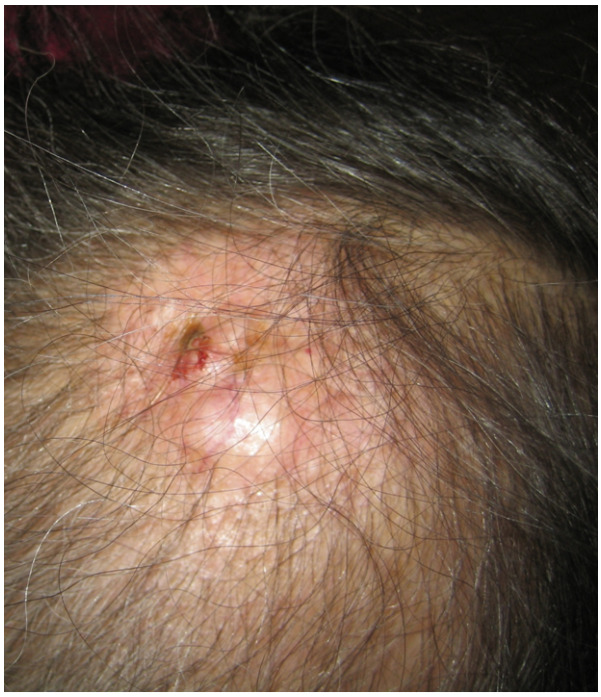

Figure 1: Preoperative finding

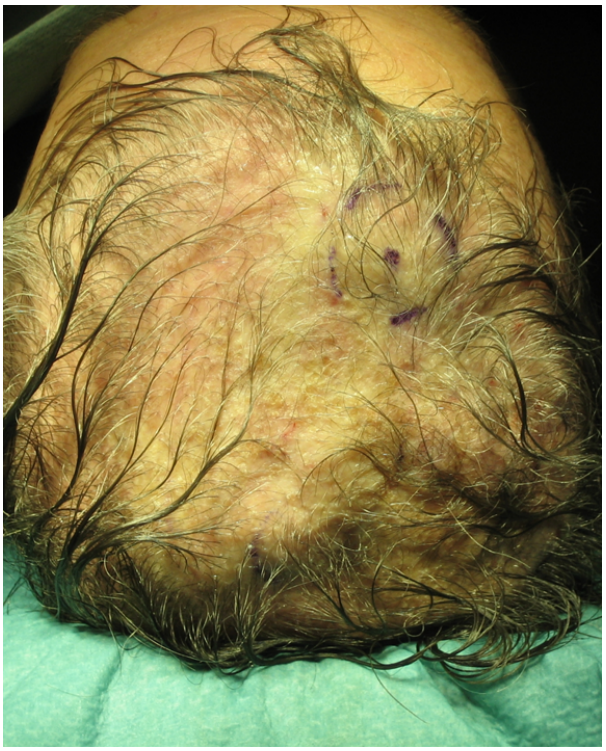

Figure 2: Local recurrence

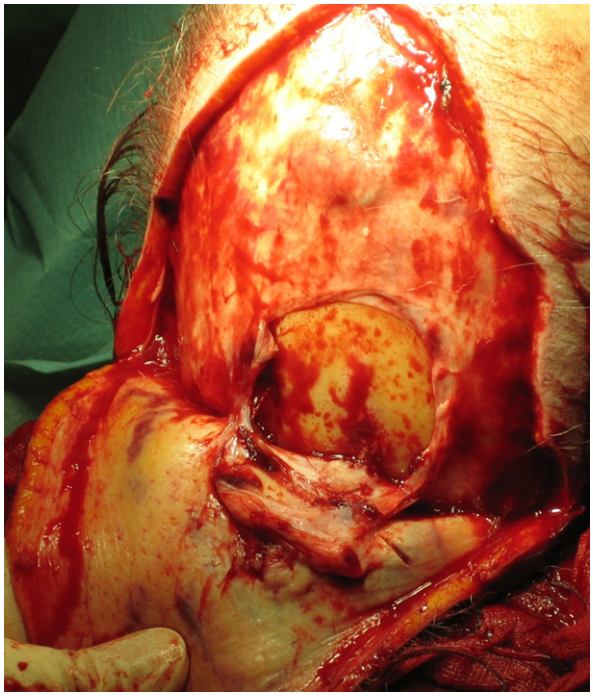

Figure 3: Intraoperativ findings: involvement of the skull

A primary salivary gland origin as well as any other primary origin like breast, thyroid gland, lungs, lacrimal, and mucosal glands was excluded by detailed clinical and radiological investigation (magnet resonance imaging, computer tomography). The remaining tumour was excised with 3 cm margins in width. Considering the clinical background, histological investigations were compatible with a primary cutaneous adenoid cystic carcinoma. Intraoperatively there were signs of cranial bone involvement. Histological investigations showed tumour free margins except for the involvement of the external cortical wall. Another postoperative CT scan showed no further infiltration of the skull. The external cortical wall of the cranial bone was removed (Figure 4). A lateral neck dissection was performed and the surgical defect (10x14 cm) was covered with a free latissimus dorsi flap (Figure 5) with a split-thickness skin graft (postoperative result: Figure 6).

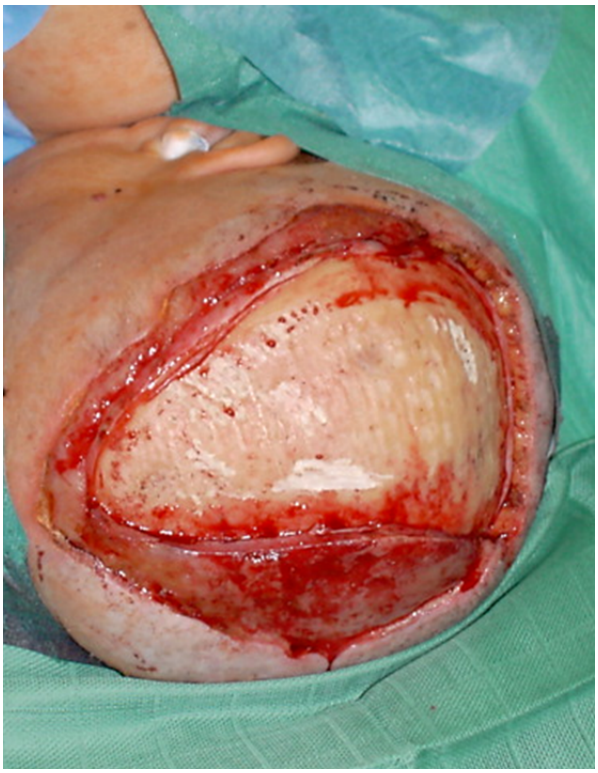

Figure 4: After abrasion of the external cortical wall

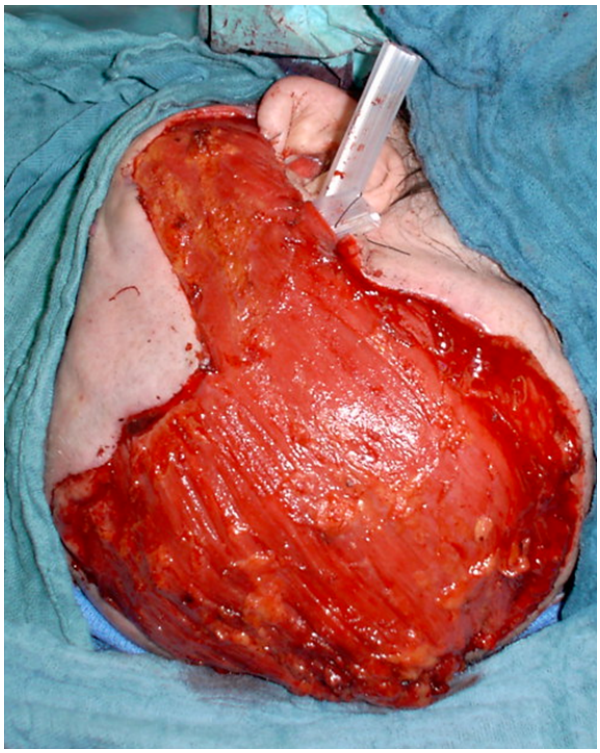

Figure 5: Coverage with M. latissimus dorsi free flap

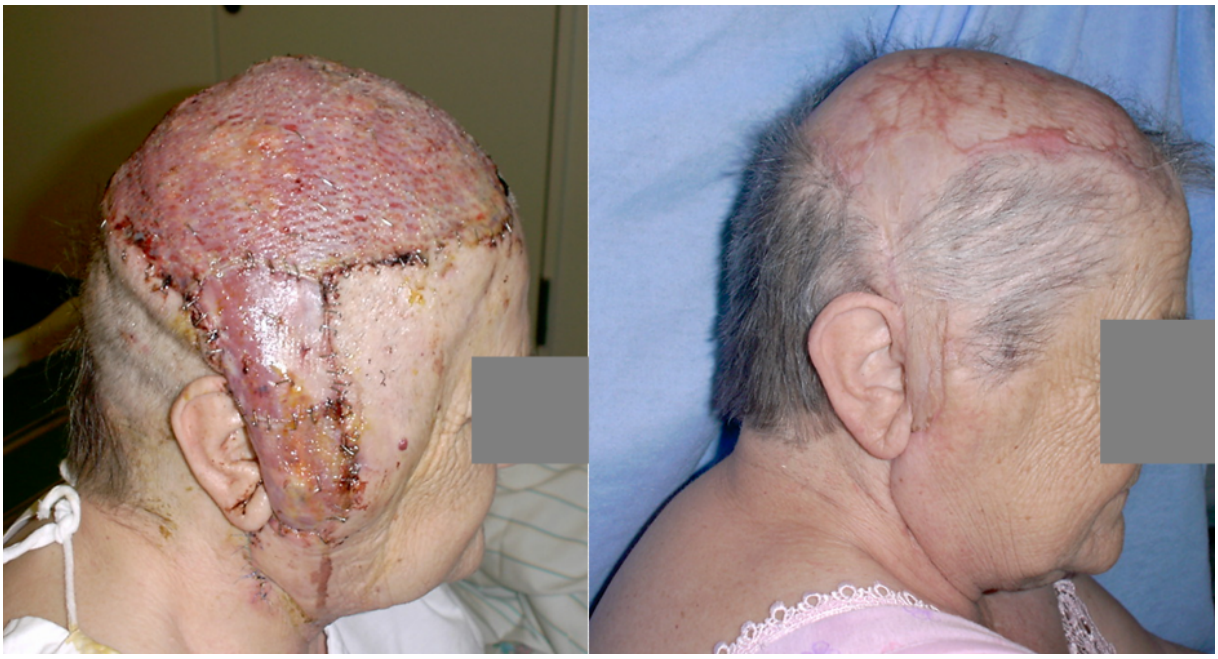

Figure 6: One week and six months after surgery

Discussion

Adenoid cystic carcinoma mainly occurs as a neoplasm of the salivary glands. Other less-frequent primary locations described have been the lacrimal glands, mucosal glands of the upper respiratory tract, external auditory canal, breast, Bartholini's glands of the vulva, uterine cervix, prostate gland, and the esophagus [1]. Primary cutaneous adenoid cystic carcinoma (PCACC) is a rare entity with less than 50 cases reported in the literature [2]. In approximately 40% primary cutaneous adenoid cystic carcinoma arises on the scalp, approximately 18% are found on the skin of the breast [3], [4], [5]. The male to female ratio is 1:1.2 [6]. The natural history of PCACC is a long, indolent course characterized by progressive local recurrence. PCACC is characterized by an aggressive infiltrative growth into the reticular dermis and subcutis with frequent perineural invasion. This leads to local recurrence after tumour excision in more than half of the patients [6]. Adenoid cystic carcinoma of all sites can invade lymph nodes by direct extension. Embolic metastases are rare [1]. In PCACC only 3 cases with distant metastasis have been reported. Metastatic focus was the lungs. Histolopathologically, PCACC resembles adenoid castic carcinoma (ACC) of the salivary glands and consists of basaloid neoplastic cells that have inconspicuous cytoplasm and round hyperchromatic nuclei without nuclear atypia [7], [8]. It exhibits cribiform, tubular, cystic, and solid patterns and mainly occupies mid and reticular dermis. The lumina of tubular structures and the surrounding stroma may contain mucin or eosinophilic necrotic cells. A hyaline membrane, which probably represents redublicated basal lamina, may surround the cell islands and tubules and is periodic acid-Schiff-positive [8]. In our case, no periodic acid-Schiff-positive material was found. In our patient perineural invasion was not seen although described in more than 50% of the previously published cases [3], [6], [8], [9]. Golden standard for PCACC treatment is still wide local excision with tumour-free margins established by permanent section. It has also been reported that Moh's surgery is a good opportunity for the excision of PCACC. Lateral neck dissection is recommended [1]. However, perineural extension may be discontinuous and may lead to false-negative margins with a higher recurrence rate [3], [6], [9], [10]. Therefore radiation therapy is recommended in addition to the surgical treatment. If subsequent radiation is considered, coverage of the defect with stabile and well vascularised tissue is obligatory. The best way to achieve this is the coverage with a muscle flap. Defects of the scalp can be successfully covered by a free M. latissimus dorsi flap as presented in our case. The pedicle may be anastomosed with temporal vessels or in case of insufficient vessel diameter, as in our case, bigger branches of the external carotoid artery and the jugular veins may be used for flap vascularisation. The muscle flap will then be covered by a split thickness skin graft. After approximately one month when wound healing is completed radiation therapy can be commenced.

Notes

Competing interests

The authors declare that they have no competing interests.

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Corresponding author:

Prof. Dr. Dr. Björn Dirk Krapohl, MD
German Armed Forces Hospital of Berlin, Plastic and Hand Surgery, Department of Surgery, Scharnhorststraße 13, D-10115 Berlin, Germany, Phone: +49(0)30/2841-1205, Fax: +49(0)30/2841-1299
PlastChirBWBerlin@web.de

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