Case report

Adenoid cystic carcinoma of the palate with squamous metaplasia or basaloid-squamous carcinoma? Report of a case


A case of an unusual tumor of the palate is presented, in which the differential diagnosis focuses on either adenoid cystic carcinoma with squamous metaplasia or a basaloid-squamous carcinoma.

Adenoid cystic carcinoma (ACC) is a malignant salivary gland tumor, most commonly arising in the minor salivary glands, especially those of the palate (1). Three different histological types are commonly described: glandular (cribriform), tubular and solid (basaloid) (2). Fusion of solid (basaloid) ACC with the overlying epithelium makes the differential diagnosis with a basaloid-squamous carcinoma (BCS), as described by WAIN et al. in 1986, very difficult (3), as is shown by the present case report.

Although the possibility of a salivary gland tumor, (i.e. an adenoid cystic carcinoma) could not be excluded.

Treatment consisted of a partial maxillectomy followed by postoperative radiotherapy. The tumor seemed to be completely removed. Half-way through the course of radiotherapy (3780 cGy of the planned 6300 cGy) the patient refused further treatment.

Nine years postoperatively he developed a primary lung carcinoma with regional metastasis. There are no signs of local recurrence or regional or distant metastases of the palatal tumor.

Nine and a half years after primary

Fig. 1. Clinical view of the lesion of the palate.
surgery he died because of his primary lung carcinoma.

**Histopathology**

The tumor of the surgical specimen from the maxilla was composed of lobules and cords of cells with scant cytoplasm and dark hyperchromatic nuclei intermingled with cribriform areas. Few mitotic figures and hardly any cellular or nuclear pleomorphism were present.

Small, cystic, gland-like spaces containing Alcian blue-positive material were seen. Between the cells there were thin septae of connective tissue, which were PAS- and Alcian blue-positive. At several sites the tumor cells appeared to fuse with the overlying surface epithelium. Furthermore, squamous metaplasia of tumor cells was observed in several areas. There was no evidence of perineural spread (Figs. 2–5).

Semi-thin plastic sections showed basal lamina-lined “pseudocysts” and small duct lumens among the more numerous and angular basaloid cells, which was considered to be suggestive of ACC.

Immunohistochemical studies were performed on paraffin sections (Table 1). Keratin markers (AE1/3-CAM 5.2) were strongly positive. Vimentin was focally positive in spindle-shaped cells. Amylase, CEA, GFAP, and actin were not detected. Foci of tumor cells were positive for EMA. Only some diffusely scattered cells surrounding “pseudocysts” were positive for S-100.

No final histologic diagnosis could be made. Instead, a differential diagnosis of BSC and ACC has been proposed. Although the histological slides have been sent to various (oral) pathologists, no final consensus could be reached.

**Discussion**

Adenoid cystic carcinomas are among the most common malignancies of the minor salivary glands, with the palate being the favourite location (4). According to the new WHO classification (1991) there are two distinctive histologic types, a glandular/tubular type and a solid type (2). Basaloid-squamous carcinoma is a variant of squamous cell carcinoma most often arising in the hypopharynx, base of tongue and larynx. Histology shows basaloid cells with foci of squamous differentiation, continuity with the overlying epithelium, squamous dysplasia and distinct nuclear pleomorphism (5).

Another diagnosis that has been considered in this case is adenosquamous carcinoma, although this entity is a de-
Table I. Antibodies used for immunohistochemical staining

<table>
<thead>
<tr>
<th>Reagent</th>
<th>Source</th>
<th>Dilution/Technique</th>
<th>Pretreatment</th>
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<tbody>
<tr>
<td>AE1/3 (M)</td>
<td>Boehringer</td>
<td>1:100 ABC</td>
<td>Pepsin</td>
</tr>
<tr>
<td>CAM 5.2 (M)</td>
<td>Beeton &amp; D.</td>
<td>1:10 ABC</td>
<td>Trypsin</td>
</tr>
<tr>
<td>EMA (M)</td>
<td>Dakopatts</td>
<td>1:50 ABC</td>
<td>-</td>
</tr>
<tr>
<td>CEA (M)</td>
<td>AVL Hospital</td>
<td>1:4000 IP</td>
<td>-</td>
</tr>
<tr>
<td>S-100 (P)</td>
<td>Dakopatts</td>
<td>1:400 PAP</td>
<td>Trypsin</td>
</tr>
<tr>
<td>GFAP (P)</td>
<td>Dakopatts</td>
<td>1:50 IP</td>
<td>-</td>
</tr>
<tr>
<td>Actin (M)</td>
<td>Organon</td>
<td>1:5 ABC</td>
<td>-</td>
</tr>
<tr>
<td>Vimentin (M)</td>
<td>Free University</td>
<td>1:4000 ABC</td>
<td>-</td>
</tr>
</tbody>
</table>

(M): Monoclonal; (P): Polyclonal; ABC: 3-step avidin-biotin-complex; IP: 2-step i

operoxidase method; PAP: 3-step peroxidase-antiperoxidase technique

buttable one (2). Since no intercellular bridges or keratin were demonstrable in the squamous component, the remote possibility of an adenocarcinoma with small foci of squamous differentiation has also been considered.

The histopathologic features of the solid type of ACC are similar, if not identical, to those of the basaloid component of BSC (3, 6). Cribriform and pseudoglandular patterns can be identified in both BSC and ACC (5). Since the BSC is a variably differentiated squamous cell carcinoma, that recognizable phenotype should be present (7). Despite the squamous metaplasia, epithelial dysplasia and the intimate relation with the overlying squamous epithelium in the present case, the ultrastructural findings of the basal lamina-lined “pseudo-cysts” and the small duct lumens among the angular basaloid cells seem to support the diagnosis of ACC.

The immunohistochemical findings have been compared with those of BSC and ACC, as presented in the literature (5, 8, 9). Because of positive staining for keratin, focal positivity for EMA, absence of CEA in the basaloid cells and absence of staining for actin and S-100, there is a slight preference for the diagnosis BSC (5, 8).

ACC with squamous cell carcinoma, either in situ or invasive, and showing a tendency to form solid or basaloid areas have been reported in the esophagus (10, 11). GNIEPP described that the majority of sinonasal tract adenomatous neoplasms originate from the mucosal lining of the sinonasal tract (12). The most acceptable concept for the genesis of coexistence of adenoid cystic and squamous elements is suggested to be a “field effect” wherein both mucosal and submucosal epithelial structures are similarly affected during the process of carcinogenesis (11).

Both BSC and solid ACC are found to be very aggressive and are associated with poor prognosis (3, 13). However, BSC shows a tendency for metastatic spread to cervical lymph nodes and short survival, and ACC shows a tendency for distant (lung) metastases which can occur even more than 20 years after primary surgery (5, 13). The patient in this report died nine and a half years after primary treatment without evidence of local, regional or distant metastatic disease, which is somewhat in favor of a diagnosis of ACC.

In conclusion, although the immunohistochemical findings are slightly in favor of a diagnosis of BSC, the clinical histopathological and ultrastructural findings lead to the diagnosis ACC with squamous metaplasia.

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References

9. CASELITZ J, SCHULZE I, SEIFERT G. Ade-