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A tumour derived from Ebner's glands: microcystic adnexal carcinoma of the tongue

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Abstract
Microcystic adnexal carcinoma (MAC) is known as an infiltrating but non-metastasizing tumour of the skin, that derives from sweat glands or follicular epithelium. We report on a rare case of MAC of the tongue. The patient had noticed the tumour for two years with slowly increasing dysphagia but no other symptoms of an oropharynx carcinoma. Histological and immunohistochemical analyses showed a similarity between the tumour derived from Ebner's glands of the tongue and MAC of the sweat glands.

Key words: Tongue neoplasms; Immunohistochemistry

Introduction

The term microcystic adnexal carcinoma (MAC) was introduced by Goldblatt *et al.* (1987). They described six cases of a locally aggressive tumour most frequently found in the upper lip of middle-aged females. Both follicular and sweat gland differentiation have been observed (Kato *et al.*, 1990). MAC of the tongue or a carcinoma of Ebner's glands (Assali *et al.*, 1989; Krstic, R. V., 1991) has not been reported so far. Histological similarity between sweat glands of the skin and Ebner's glands of the tongue, might explain these findings.

Case report

A 65-year-old man presented with induration, immotility, and reduction in tongue size; a nodular protrusion was observed (Figure 1). He complained of dysphagia and language disorders that progressed over a two-year period.

The patient was scanned by magnetic resonance imaging (MRI) at 1.5 T (Magnetom, Siemens), with a Helmholtz surface coil. T1- and T2-weighted multislice spin-echo images were obtained in the coronal and axial planes. MRI revealed involvement of the entire tongue, with local infiltration of the sublingual muscles, the right tonsillar pillar, and the parapharyngeal space to the hypopharynx (Figure 2). No enlargement of the cervical lymph nodes or other organs was found.

An incisional biopsy was performed. Formalin-fixed paraffin sections were stained with haematoxylin-eosin and periodic acid-Schiff (PAS). Immunohistochemical analyses were performed using the alkaline phosphate-antialkaline phosphatase (APPAP) method with CAM 5.2 (cytokeratin proteins 8 and 18) monoclonal mouse antibody (Becton Dickinson, USA), the CEA (carcinoembryonic antigen) monoclonal mouse antibody (Dako, Germany), and EMA (epithelial membrane antigen) monoclonal mouse antibody (Dako, Germany).

Histological analysis showed a poorly circumscribed, deeply infiltrating tumour (Figure 3) consisting of vertically oriented tubules lined by two layers of cuboidal or flat epithelium (Figure 4). These tubules open into the spaces between the papillae. In the upper region some tubules show microcystic dilatation with luminal PAS-positive diastase labile material interpreted as glycogen (Rongioletti *et al.*, 1986). Squamous epithelial differentiation occurred close to the epithelial surface without

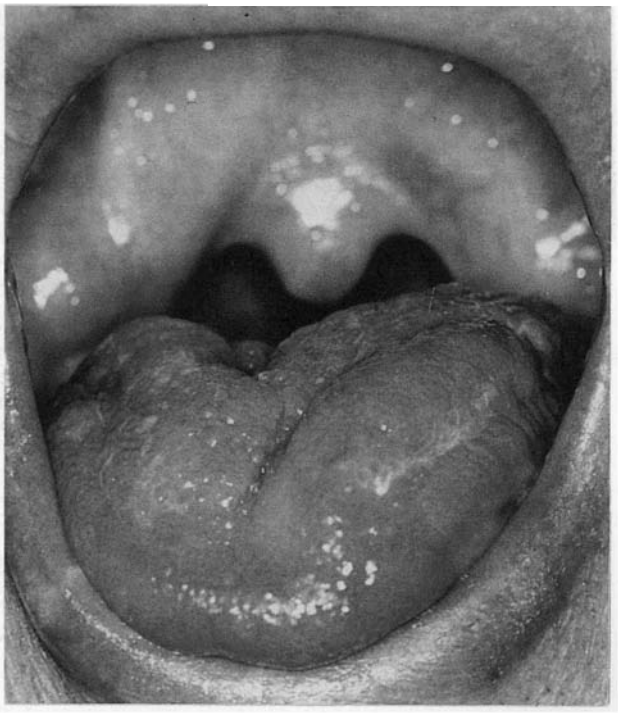

FIG. 1
Enoral photograph of the mouth. The tumour is on the right side, in front of the tonsillar pillar.

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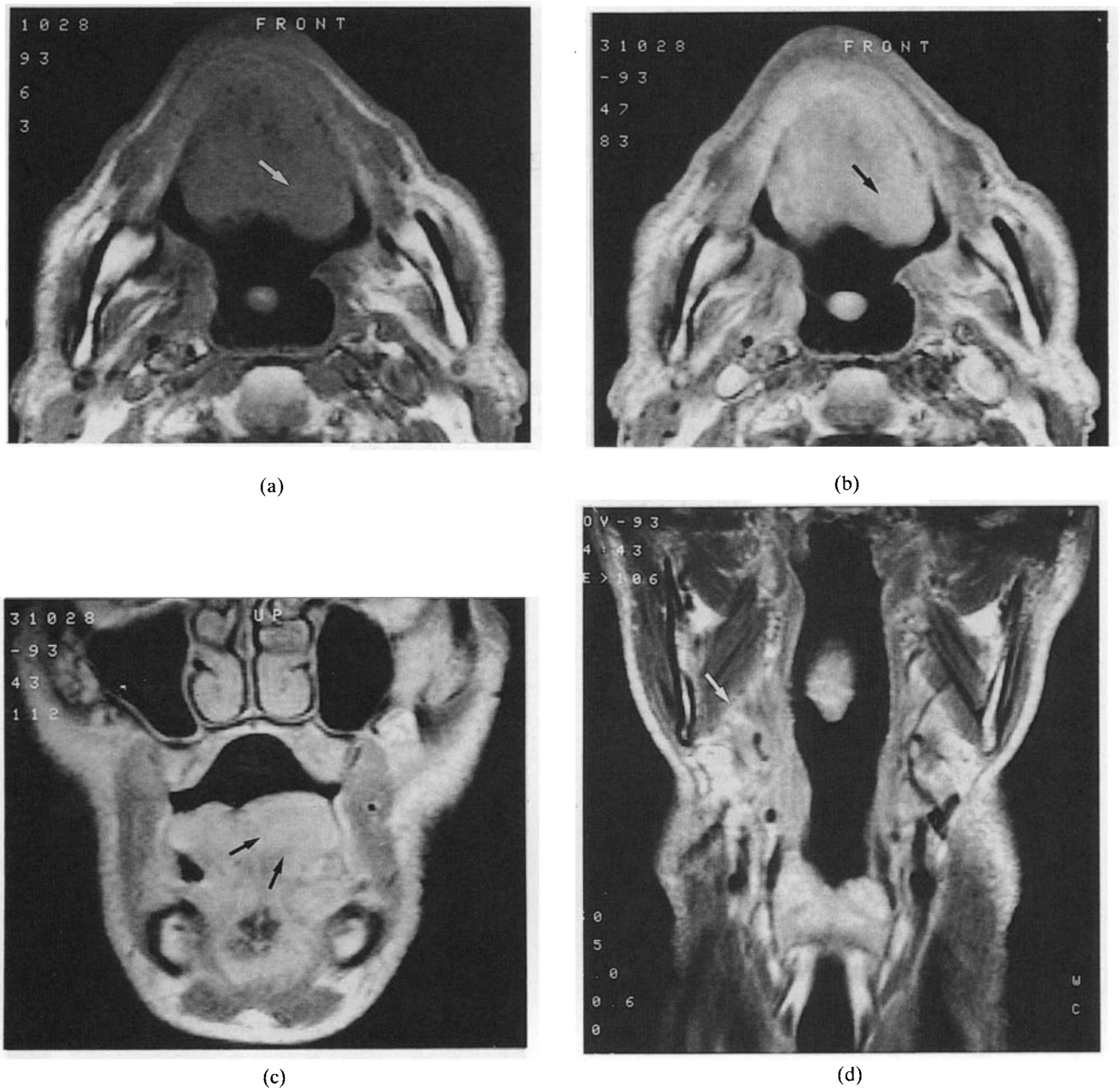

FIG. 2

MRI examination of the oropharynx. In the axial view of a T1-weighted sequence, the tumour was nearly indistinguishable from the normal musculature (arrow, a). After application of a contrast medium (gadolinium-DTPA), only slight enhancement was demonstrated on the right side of the tongue (arrows, b). In the coronal section there was again a slight enhancement reaching from the glossotonsillar sulcus to the midline of the tongue (arrows, c). In the more posterior position of the coronal cuts, the parapharyngeal space was thickened from the glossotonsillar sulcus down to the hypopharynx (arrow, d).

keratinization. The stroma between the glandular structures appeared sclerotic.

In the immunohistological analysis, strands of tumour cells showed a strong staining with CAM 5.2: CEA staining was evident in the inner layer of the microcysts, whilst EMA expression was slightly positive.

Discussion

There are three kinds of lingual glands: serous, mucous, and the seromucous glands. Ebner's glands are serous and are situated in the posterior part of the tongue beneath the circumvallate papillae. These serous glands resemble eccrine sweat glands of the skin (Kato *et al.*, 1990). Each circumvallate papilla is surrounded by a circular trench; these trenches are joined by the excretory ducts of Ebner's glands.

MACs of the skin typically contain cysts lined by two or more layers of a basaloid or flat epithelium that have keratin in their lumina or even show differentiation toward hair follicle structures. In the MAC of the tongue described here, microcysts contained only PAS positive substance in their lumina, and nests of basaloid cells did not differentiate into hair follicles (Figure 4). This is not surprising because in the tongue only the tips of papillae show keratinization; the epithelium between the papillae is not keratinized.

The immunohistological analysis corresponds to this histological finding. MAC of Ebner's glands of the tongue as well as MAC derived from sweat glands of the skin

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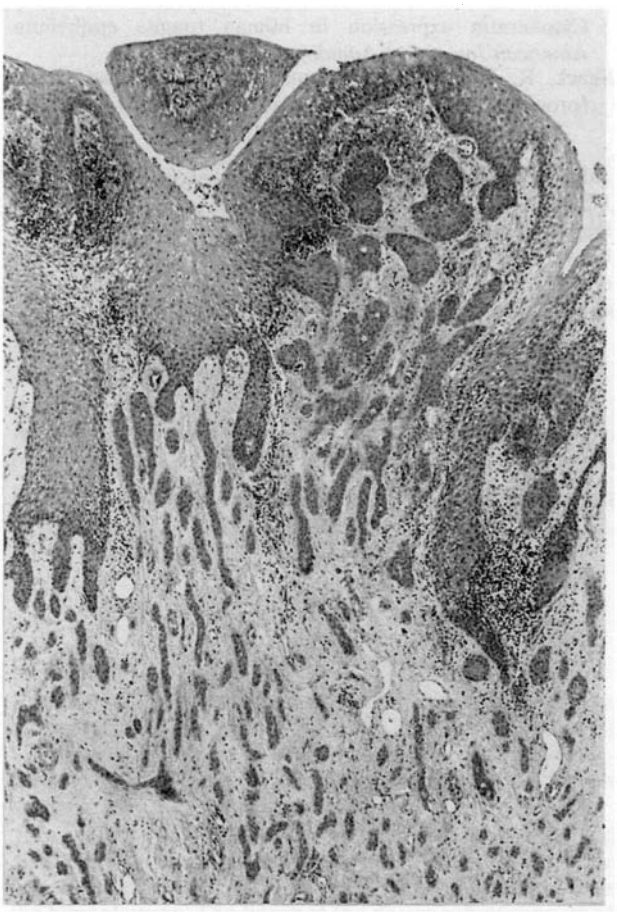

FIG. 3

A section of the papillae of the tongue with an infiltrating MAC, stained with the CAM 5.2 monoclonal antibody (keratin proteins 8 and 18) by the APPAP method and with haematoxylin-eosin.

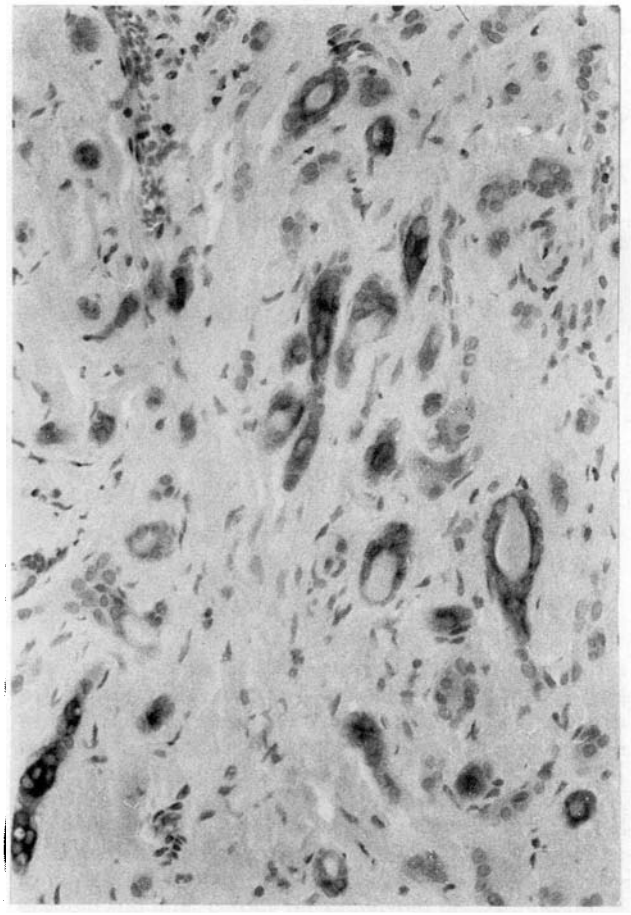

FIG. 4

Expression of the keratin proteins 8 and 18 by the APPAP method of a haematoxylin-eosin stained section of the MAC of the tongue.

express the keratin proteins 8 and 18, which characteristically react with serous glands. The expression of CEA was usually found in the inner surface of the microcysts of skin MAC. In the MAC of the tongue we could see the same distribution. The EMA expression was less significant, as previously described in the literature (Kato *et al.*, 1990).

Differential diagnoses to MAC in the skin include desmoplastic trichoepithelioma and syringoma. MAC may be distinguished histologically from desmoplastic trichoepithelioma by the presence of deep infiltrating ductal structures and from syringoma by keratin cysts, sometimes with calcification. In contrast to MAC, neither desmoplastic trichoepithelioma nor syringoma show subcutaneous and perineural involvement. The malignant acrospiroma also infiltrates subcutaneous fat and the perineurium, but it lacks the microcysts. Sweat gland carcinoma displays more pleomorphism and mitoses than MAC. Generally MAC of the skin would not metastasize but would infiltrate in the surrounding tissue structures. Other malignant salivary gland tumours of the tongue are the low-grade mucoepidermoid carcinoma (38 per cent), adenocarcinoma (20 per cent), high-grade mucoepidermoid carcinoma (14 per cent), adenoid cystic carcinoma (10 per cent), and clear cell carcinoma. The most common benign tumour is the myoepithelial variant of the benign mixed tumour (Sawaf *et al.*, 1990; Shack, R. B., 1986). Neoplasms of accessory salivary glands are much less common than those arising from major salivary glands. The tongue is an especially rare site for a salivary gland tumour. Salivary glands are at least partly of mucous diffentiation, so they differ from Ebner's glands, which are purely of serous differentiation. A purely serous tumour of the tongue, like the MAC described here, has not been reported before as far as we know. Therefore there was no therapeutic management described of such a tumour in the literature. In this case only a total glossectomy to resect the whole tumour, in combination with a laryngectomy to preserve the swallowing procedure, would be usful as an operative procedure. A neck dissection procedure would not be necessary, because similarly to MAC of the skin no metastases are expected. Primary radiation therapy was used because the patient refused operative treatment, although MAC of the skin seems to be insensitive to irradiation (Neumann, L.,1986). After radiation therapy the tumour has not changed in size. Follow up of this patient (21 months) showed no new tumour nor the appearance of metastases.

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