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CASE REPORT

Gerhard Seifert · Karl Donath · Günter Jautzke

Unusual choristoma of the parotid gland in a girl

A possible trichoadenoma

Abstract An 8-year-old girl had a painless circumscribed nodule in the right parotid gland for 4 weeks. A tumour (1.3 cm diameter) within the salivary gland parenchyma showed small cystic spaces with horn-like material macroscopically. The tumour tissue contained solid squamous cell formations and cystic spaces limited by multilayered squamous epithelium and covered by layers of ortho- and parakeratotic cells. The cystic spaces contained keratotic lamellae. In some areas pin-like epithelial proliferations were seen. All epithelial cells were characterized by uniform nuclei, and no atypical mitoses were seen. Keratinizated masses with partial calcification were occasionally located in the interstitial tissue and replaced by multinucleated giant cells. The tumour was classified as a choristoma and resembled a trichoadenoma. The ectodermally derived oral and salivary gland epithelium may be the source of skin-like or adnexal tumours.

Key words Choristoma (trichoadenoma) of parotid gland · Skin-like or adnexal hair follicle tumours · Ectodermally derived tissue

Introduction

Congenital salivary gland tumours [24] are extremely rare, and except for haemangiomas and lymphangiomas, salivary gland tumours in infancy are also rare [23]. In contrast, some types of sarcoma are more frequent in infancy, such as rhabdomyosarcoma [3]. Epithelial salivary gland tumours, especially pleomorphic adenomas and mucoepidermoid carcinomas, become more frequent during the second decade.

The revised WHO classification of salivary gland tumours [22] includes rare, but histologically well-defined tumours. In addition to the WHO classification, reports of some other rare tumour entities have been published in recent years, including congenital tumours [24], lipoadenoma [30], hidradenoma-like tumour [12] and salivary gland anlage tumour [8].

Ectodermally derived oral and salivary gland epithelium may be the source of skin-like or adnexal tumours in the salivary glands and of the occurrence of sebaceous cell metaplasia, sebaceous cell tumours or squamous cell metaplasia [10]. Our observation concerns a parotid tumour in an 8-year-old girl, which resembles a trichoadenoma, a special type of hair follicle tumour, in structure.

Dedicated to Professor Dr. U. Helmchen, Hamburg on the occasion of his 60th birthday

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Clinical history

An 8-year-old girl had a painless circumscribed nodule in the right parotid gland for some months. A tumour 1.3 cm in diameter was removed. No recurrence was observed.

Materials and methods

The biopsy specimen was routinely fixed in formaldehyde and embedded in paraffin wax. Several stains were used, including haematoxylin and eosin, PAS reaction, Astra blue, and Masson Goldner. In addition, the case was studied immunohistochemically by staining for total cytokeratin (CKMNF), cytokeratins Kl-1, low-molecular-weight CK and high-molecular-weight CK, CK 1/2, CK 3/6, CK 4, CK 7, CK 13, CK 14, CK 18–20, S-100 protein, vimentin, type IV collagen and the proliferative marker MiB1. A standardized procedure was followed, using the immuno-peroxidase method (PAP method) and the alkaline phosphatase method (APAAP method) with application of the following primary antibodies: Dako Diagnostika Hamburg (CKMNF, Kl-1, CK low, CK high, CK 1/2, CK 3/6, CK 18–20, S-100, type IV collagen), Progen Biotechnik, Heidelberg (CK 13, CK 18, vimentin), Sigma Deisenhagen (CK 4, CK 7),Biogenex, San Ramon (CK 14), Dianova, Hamburg (MiB1, type IV collagen clone 8C 55 A5).

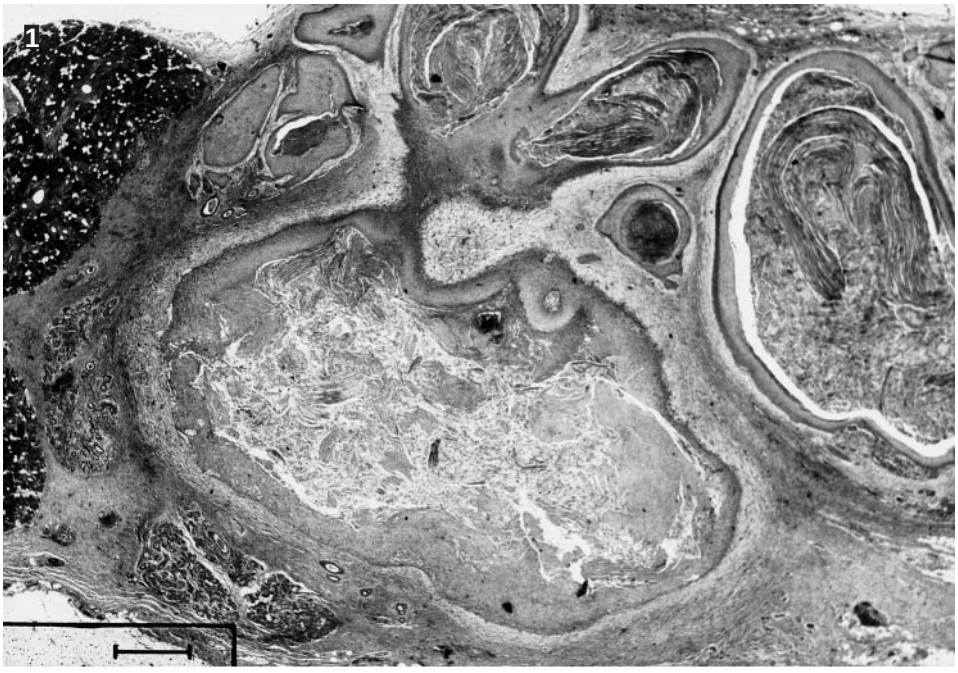

Fig. 1 Multiple cystic spaces limited by squamous epithelium and filled with horn-like material: intact parotid gland parenchyma nearby. H&E, ×3

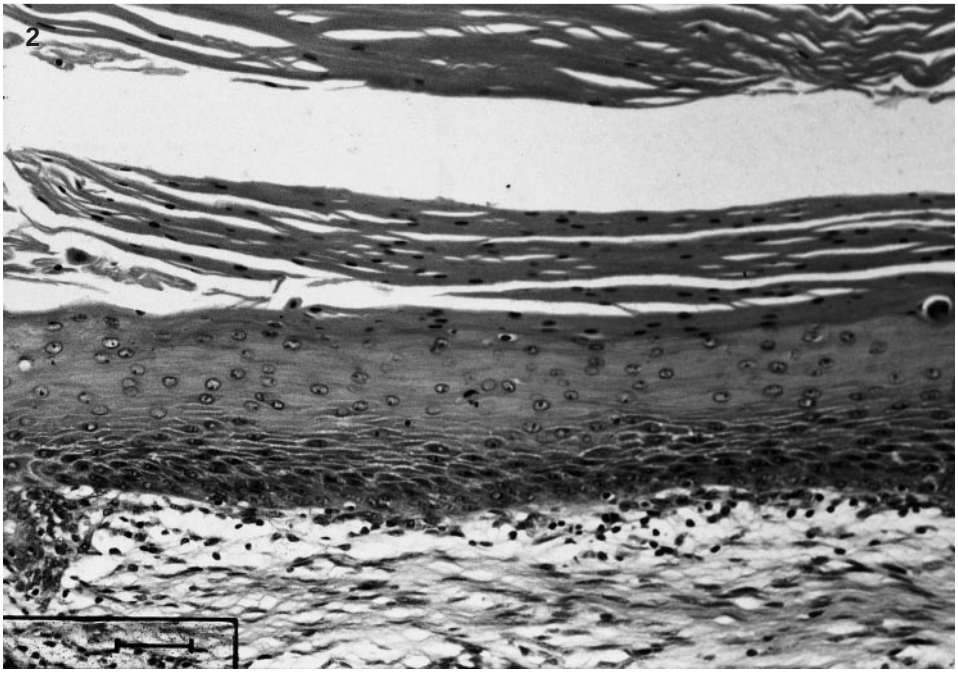

Fig. 2 Wall of a cystic space covered by distinct layers of squamous cells; keratotic lamellae in the lumen of the cyst; layers of ortho- and parakeratotic cells without a granular layer. H&E, ×50

Pathological findings

On the cut surface the tumour showed multiple small cystic spaces with horn-like material. Histologically, the lesion contained multiple cystic spaces (Fig. 1), which were limited by a multilayered squamous epithelium (Fig. 2). The epithelium of the cystic spaces was covered by layers of ortho- and parakeratotic cells without a granular layer. The cystic spaces contained keratotic lamellae. In other areas solid squamous cell formations were seen (Fig. 3), but with distinct demarcation from the surrounding parenchyma. The outer layer of the solid and cystic squamous cell formations showed a pin-like proliferation in some areas, similar to radicular cysts. This proliferation has the characteristic features of expansive growth rather than infiltrating growth. In the outer basal layer of proliferative areas focal distinct staining with the marker MiB1 was seen. All the epithelial cells had uniform nuclei without atypical mitoses. In addition, all proliferative areas were distinct from the surrounding tissue. The interstitial tissue of the salivary gland parenchyma was characterized by a moderate inflammatory reaction and an increase of collagen tissue. Solid or cystic squamous areas were present in the sur-

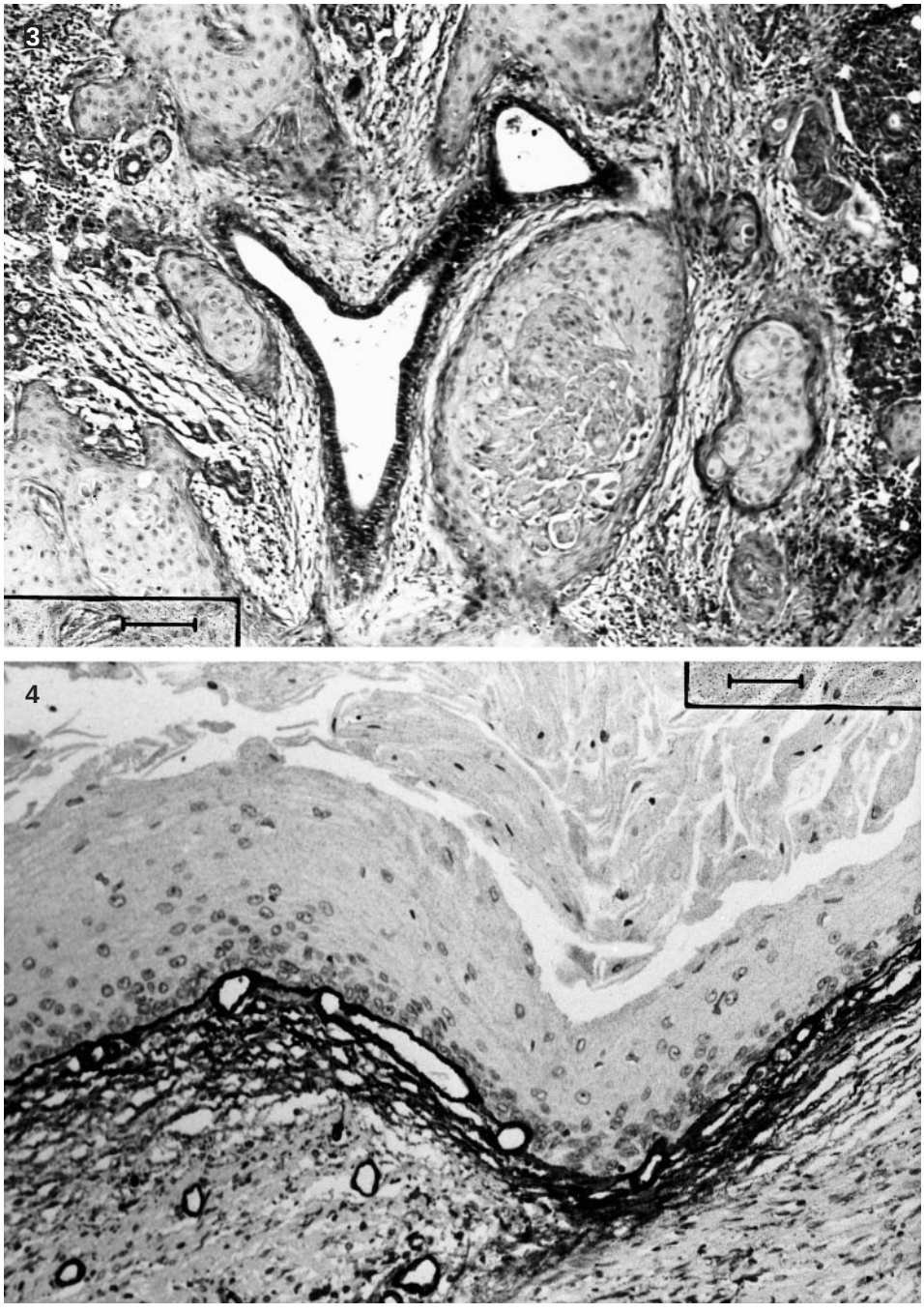

Fig. 3 Solid areas of squamous cell formations partly localized within the parotid gland parenchyma in the vicinity of an excretory duct, but with distinct demarcation. PAS, ×25

Fig. 4 Limitation of the outer layer of the cystic space by distinct expression of type IV collagen. APAAP, ×50

rounding parenchyma; these might be interpreted as invasive growth.

A further feature of the tumour was the presence of keratinizated masses outside the cysts, which were replaced by multinucleated giant cells. In some areas calcification of the keratin elements was observed. Immunohistochemically the solid squamous cell formations and the walls of the cystic spaces expressed distinct cytokeratins, especially CKMNF, CK Kl-1 and high-molecular-weight CK but also CK 3/6, 5/6 and 7, and moderate amounts only of low-molecular-weight CK and the other cytokeratins. In the walls of the cystic spaces type IV collagen was expressed (Fig. 4). Vimentin and S-100-protein were negative. Sebaceous cell differentiation was not observed, the formation of hair follicles was not seen. The tumour is similar to a group of hair follicle tumours, and specifically to trichoadenoma.

Discussion

We examined previously published cases of unusual salivary gland tumours or other unusual conditions of the major and minor salivary glands [19], but could not find

any other case with a similar structure. A comparison with adnexal tumours of the skin indicates the analogous development of the salivary gland tissue as a derivative of ectodermal tissue. Other examples of this origin are inclusions of sebaceous cells in the oral mucosa (Fordyce disease) or the group of sebaceous gland tumours of the salivary glands.

In the revised WHO classification of skin tumours, adnexal tumours of the skin are classified in four main groups: eccrine tumours, apocrine tumours, sebaceous gland tumours and hair follicle tumours [15]. Hair follicle tumours include many benign and malignant tumour entities and also follicular cysts [16, 18]. In comparison with our observations, the entity of trichoadenoma as a special type of trichoepithelioma is perhaps the most relevant [20]. In trichoadenoma multiple keratinous cysts predominate, with relatively little basaloid epithelial component or none at all. Keratinized masses outside the cysts with multinucleated giant cells and focal calcification are also observed. The general architecture of trichoadenoma suggests the development of immature hair structures. Some investigators emphasize the possibility of mistaking this tumour for well-differentiated squamous cell carcinoma.

Another point is the observation of squamous cell metaplasia in many salivary gland diseases [10]. Aetiological factors include radiation, duct obstruction and ischaemia [7, 29]. Extensive squamous cell metaplasia at the border of necrosis may make diagnosis difficult, but the metaplasia often shows a reactive epithelial proliferation with pseudo-invasion in the surrounding tissue, so that a squamous cell carcinoma is simulated. Squamous cell metaplasia at the border of tumour necrosis has some differences from necrotizing siaolometaplasia [1, 9], an ischaemic lesion of the salivary glands. Other types of squamous cell metaplasia in salivary gland tumours are metaplastic Warthin tumours with replacement of oncocytic cells by squamous cell metaplasia [13, 14, 25] and pleomorphic adenoma, 25% of the cases of which show squamous cell metaplasia. Focally circumscribed areas of squamous cell metaplasia can also be observed in oncocytic hyperplasia and oncocytic tumours [27] and also in basal cell adenoma or basal cell adenocarcinoma.

Sclerosing polycystic sialadenopathy [11] (sclerosing polycystic adenosis of major salivary glands [26]) has been observed in younger patients and is characterized by nodular, incompletely encapsulated tumour-like masses mainly of the parotid gland. This lesion is comparable to fibrocystic mastopathy.

In contrast to hamartoma [4, 28] and teratoma [21], choristoma [17] is defined as tumour-like formation of normal, but heterotopic, nonresident tissue that has developed in an abnormal location. Intraoral tumour-like masses of normal, but heterotopic, cells have been reported under a variety of other terms [6].

Heterotopic salivary gland tissue are observed relatively frequently in the lymph nodes of the parotid gland and in the upper or middle cervical area – mostly at the front rim of the sternocleidomastoid muscle but also in the gingiva, within the lower jaw and more rarely in the middle ear and other areas [23]. Heterotopic salivary gland tissue with tumour-like swelling of the gingiva is described as “gingival choristoma” [5].

Our report concerns an unusual choristoma of the parotid gland with the structure of a trichoadenoma. This finding is compatible with the occurrence of sebaceous cells and sebaceous gland tumours in the salivary glands [23], especially in the parotid. The exceptional feature is the development of a tumour structure comparable to skin adnexal tissue of the type of hair follicles. The ectodermally derived salivary gland epithelium may be the source of a skin-like or adnexal-like differentiation.

We have found only one remotely comparable case report, classified as “follicular choristoma” of the gingiva with inclusion of hair follicles [2]. In a 9-year old girl a tumour-like nodule developed in the gingiva lingual to the lower permanent incisors. The specimen contained “a large, cystic aggregate of disintegrated sebaceous cells associated with a foreign-body giant cell reaction”. Other findings were hairs within the follicles, sebaceous cells with acini-like organization and excretory ducts and a medium-sized cyst containing lamellar keratin. This lesion was classified as “follicular choristoma” or “folliculo-choristoma”. Differences between this peculiar lesion and our case are that our patient’s tumour contained no sebaceous cells or hairs, but only solid and cystic formations of squamous epithelium with inclusion of keratotic lamelles, focal calcification and foreign-body giant cells. Therefore, we have classified this choristoma as a possible trichoadenoma. We could not find an analogous case report in the literature with such a tumour structure in the parotid gland.

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