

Case report

Multifocal dermal-type basal cell adenomas of parotid glands with co-existing dermal cylindromas*

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Summary. A basal cell adenoma of the parotid gland accompanied by multiple developing microadenomas of the same type and three dermal cylindromas, all observed in a 54-year-old woman, were examined by light microscopy. The histological structure of the basal cell adenomas shows striking similarities to co-existing dermal cylindromas, suggesting a common histogenesis of both tumors. We suggest that the term “basal cell adenoma of dermal type” should be used for these lesions. Multifocal ductal proliferations with nascent adenomas in the glandular parenchyma allow us to trace the development of these tumors. Studies of the literature reveal four additional cases in which there are co-existing basal cell adenomas of salivary glands and dermal adnexal tumors.

Key words: Multifocal basal cell adenoma – Parotid gland – Dermal cylindroma

Basal cell adenomas of salivary glands are a special group of monomorphic adenomas comprising 1–2% of all primary salivary gland neoplasms (Kleinsasser and Klein 1967; Seifert and Schulz 1979). The main features distinguishing them from other types of adenomas are (1) proliferation of basaloid epithelial cells with a peripheral layer of cells arranged in a palisade fashion and (2) almost total absence of myoepithelial elements and lack of myxomatous or chondroid material (Kleinsasser and Klein 1967; Hübner et al. 1971; Batsakis 1972; Thackray and Lucas 1974; Jao et al. 1976; Seifert and Schulz 1979).

Recently, a subgroup of basal cell adenomas has been recognized, exhibiting close histological similarities to dermal adnexal tumors, in particular to dermal cylindromas (Headington et al. 1977; Reingold et al. 1977; Bat-

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sakis and Brannon 1981). The peculiarity of this entity of dermal analogue tumor is based mainly on its coincidence with dermal cylindromas and trichoepitheliomas suggesting a common histogenesis of both salivary gland and skin tumors. So far, four cases of dermal-type basal cell adenomas of salivary glands and co-existing dermal adnexal tumors have been described (Headington et al. 1977; Reingold et al. 1977; Batsakis and Brannon 1981). In the present communication we report on a further observation with recurrent multifocal dermal-type basal cell adenomas of the parotid glands and dermal cylindromas of the scalp.

Case report

A 54 year old patient had been admitted to hospital with a well circumscribed tumor located in the dorso-caudal lobe of her right parotid gland. She also showed three small, lobulated dermal tumors of the scalp with a diameter of 7–25 mm. The parotid tumor was removed by lateral parotidectomy, with preservation of the facial nerve. The three dermal tumors of the scalp were excised at the same time.

Only one year later she returned with a diffuse, indolent swelling of her left parotid gland. The painless tumor was more diffuse, non-fluctuant and restricted to the dorsal lobe of the parotid gland. Lateral parotidectomy was carried out, the facial nerve remained undisturbed. Up to this time the postoperative follow-up has been uneventful.

The patient's family history revealed that two sisters, her mother and her mother's father showed similar dermal tumors of the scalp of varying sizes. Hitherto tumors of the parotid glands had not been observed in the family.

Histological examination

The tumor from the right parotid gland (Fig. 1) exhibits characteristic features of a basal cell adenoma. It is composed of densely arranged epithelial islands (Fig. 2a). Small dark-staining isomorphic cells are grouped in palisade fashion at the periphery of the islands. The central cells are larger containing pale staining nuclei. Occasionally, they form foci of squamous metaplasia. Abundant extracellular hyaline deposits, staining periodic acid-Schiff (PAS)-positive, diastase-resistant, are frequently seen in central areas (Fig. 2b). Small foci of tubular structures are present, lined by mucus producing columnar epithelial cells. Some epithelial nests show centrally located duct-like spaces with PAS-positive material. Prominent PAS-positive and diastase resistant hyaline bands surround the individual cell islands. Myoepithelial cells are not identified. Dense lymphocytic infiltration of the stroma and occasional formations of lymphoid follicles are seen in some areas of the tumor.

The glandular parenchyma adjacent to the adenoma (Fig. 1) and the segment of the contralateral parotid gland removed one year later both present similar and characteristic structures. Multiple minute focal proliferations of the intercalated ducts and incipient adenomas are conspicuous, allowing us to trace the development of basal cell adenomas from their origin (Fig. 3a–c). Earliest changes are characterized by hyperplasia of ductal cells frequently associated with widening of the lumen (Fig. 3a–b). Continuous tubular proliferations lead to the formation of microadenomas. Some of these microadenomas already exhibit typical features of basal cell adenomas, as described above, with broadened basement membranes and solid epithelial nests with peripheral palisading cells (Fig. 3c). Most of the ductal proliferations are associated with lymphocyte infiltrations of the surrounding stroma. The remaining areas of the glandular parenchyma contain small groups of acini separated by increased amounts of adipose tissue.

Light microscopic examination of the tumors of the scalp reveals characteristic dermal cylindromas closely associated with the adjacent cutis. Solid masses of epithelial cells consist of smaller dark-staining cells in the periphery suggesting a palisade formation and centrally located larger cells (Fig. 4a). There are occasional cystic and tubular formations, some of them resembling sweat ducts. Tumor lobules contain large areas of hyaline deposits that stain

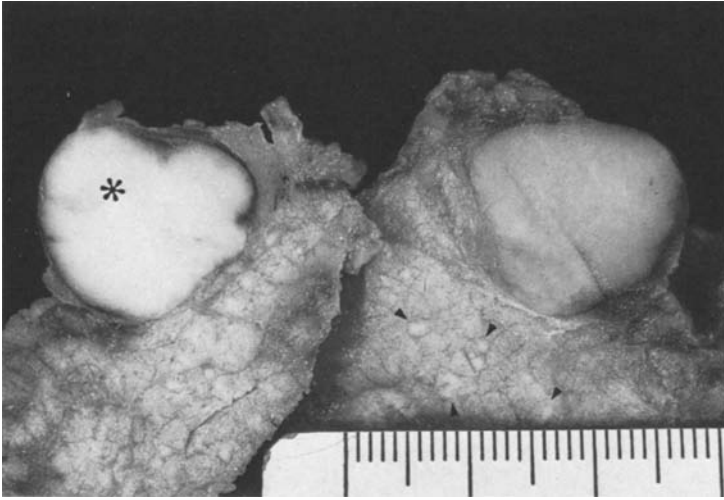


Fig. 1. Bisected portion of the right parotid gland showing the clearly demarcated basal cell adenoma measuring 18 mm in diameter (*). The small foci in the glandular parenchyma indicated by arrows represent microadenomas. Smallest unit of measure on the scale 1 mm

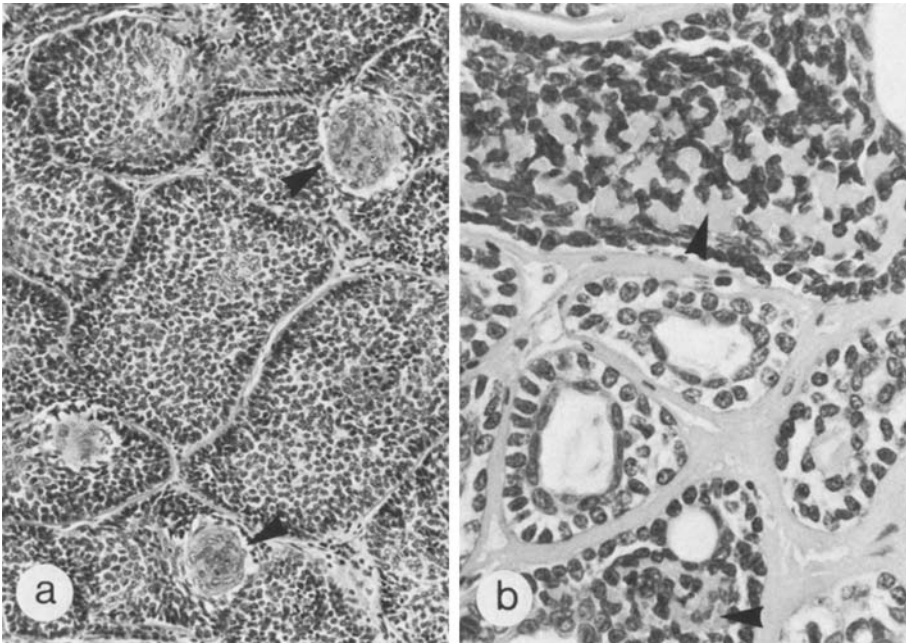


Fig. 2a, b. Dermal-type basal cell adenoma of parotid gland: **a** The epithelial islands are densely arranged like pieces of a jigsaw puzzle, the peripheral cells lying in a palisade fashion. Occasional foci of squamous metaplasia occur (*arrows*). HE $\times 140$. **b** Epithelial islands with tubular structures. Intercellular hyaline deposits (*arrows*) and prominent basement membranes are present. HE $\times 370$

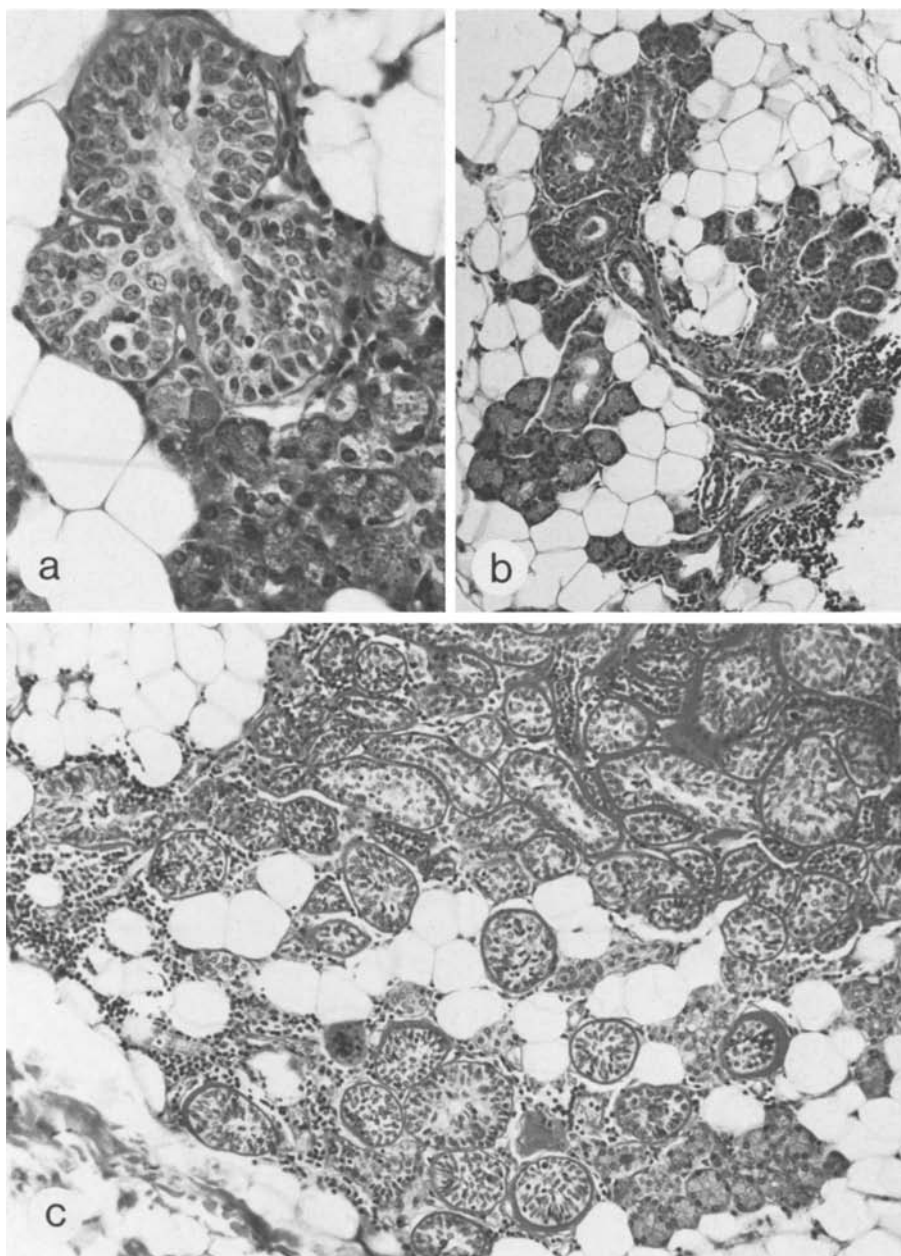


Fig. 3a–c. Different stages of ductal proliferations of the glandular parenchyma: **a** Hyperplasia of ductal reserve cells in close vicinity to acinar structures. PAS $\times 280$. **b** Small nodules of ductal proliferations some of them with widened tubular structures. PAS $\times 112$. **c** Microadenoma with predominance of solid epithelial islands. PAS $\times 136$

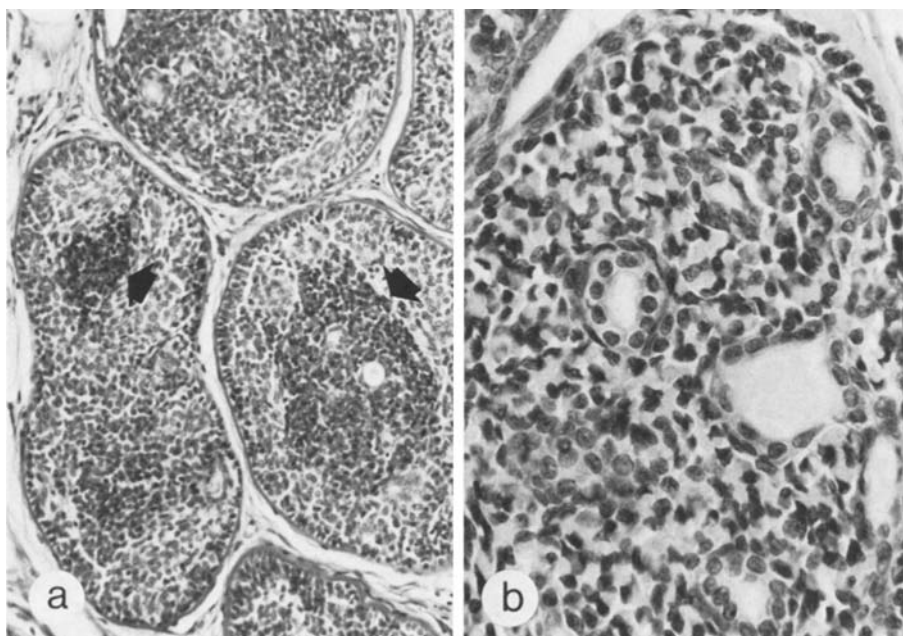


Fig. 4a, b. Dermal cylindroma of the scalp: **a** Tumor lobules exhibiting secretory activity with intercellular PAS-positive deposits (*arrows*); surrounding, PAS-positive basement membranes. PAS $\times 136$. **b** Area with tubular differentiation and intercellular hyaline deposits. PAS $\times 340$

strongly PAS-positive, diastase resistant, indicating secretory activity of tumor cells (Fig. 4b). Hyaline material, exhibiting the same staining reaction, is also present as a broad membrane surrounding the cell islands. Most of the tumor lobules are separated from each other by loose connective tissue.

Discussion

The present report describes the coincidence of basal cell adenomas of the parotid glands and dermal cylindromas of the scalp. Such association is rare, but characteristic as shown by the existence of four cases from the literature with similar features. Table 1 summarizes the observations made in our own case and in the four cases known from the literature (Headington et al. 1977; Reingold et al. 1977; Batsakis and Brannon 1981). There is no doubt that the adenomas of all five cases share common traits with those basal cell adenomas of salivary glands originally described by Kleinsasser and Klein (1967). The tumors of Kleinsasser and Klein (1967) represent a relatively heterogeneous group of adenomas with occasional tubular differentiation and a prominent PAS-positive basement membrane. Some basal cell adenomas exhibit secretory activities with small amounts of hyaline, PAS-positive material lying either intratubular or between the cells of solid trabecular proliferations. However, the adenomas included in Table 1 are characterized by their coincidence with dermal appendage tumors

Table 1. Survey of cases with co-existent basal cell adenomas of salivary glands and dermal adnexal tumors

Case	Sex/ age ^a (years)	Salivary gland tumor(s)	Recur- rence of salivary gland tumor	Ductal prolifera- tions of glandular parenchyma	Dermal tumors	Authors
1	M/70	Membranous basal cell adenoma of parotid gland	No	Yes	Cylindromas and trichoepitheliomas	Headington et al. (1977)
2	M/43	Multicentric dermal-type cylindromas of parotid gland (basal cell adenomas) and epidermoid carcinoma	Yes; 1, 15 and 17 years ^b	Yes	Cylindromas, trichoepitheliomas, eccrine spiradenomas, Bowen's disease, squamous cell carcinoma ^c	Reingold et al. (1977)
3	M/68	Multifocal dermal analogue tumors of submandibular gland (basal cell adenomas)	Yes; 2 ¹ / ₂ years ^b	Not reported	Cylindromas	Batsakis and Brannon (1981)
4	M/74	Dermal analogue tumor of parotid gland (basal cell adenoma)	No	Not reported	Cylindromas	Batsakis and Brannon (1981)
5	F/54	Multifocal dermal-type basal cell adenomas of parotid gland	Yes; 1 year ^b	Yes	Cylindromas ^c	Own case

^a Age at removal of the first salivary gland tumor^b Time period after removal of the first salivary gland tumor^c Familial occurrence of dermal tumors is reported

and a greater secretory activity. It is obvious that these adenomas possess traits identical with co-existent dermal cylindromas, suggesting a common histogenesis for both tumors.

The morphology of these particular basal cell adenomas indicates that they belong to a special category or subgroup of this tumor entity. They are described as "membranous basal cell adenoma" by Headington et al. (1977), as "dermal-type cylindroma" by Reingold et al. (1977) or as "dermal analogue tumor" by Batsakis and Brannon (1981). In order to underline the characteristic basal cell arrangement of the tumor cells and the close relationship to dermal appendage tumors, we prefer the term "basal cell adenoma of dermal-type". There is no doubt that dermal-type basal cell adenomas of the salivary glands represent a separate nosologic entity (Batsakis and Brannon 1981). Increased familial occurrence has not been reported for this type of adenoma.

The dermal-type basal cell adenomas of the salivary glands are benign tumors without infiltrative growth although there is no clear fibrous encap-

sulation (Headington et al. 1977; Batsakis and Brannon 1981). They may be multicentric (3 cases) and associated with multiple ductal proliferations of the surrounding glandular parenchyma (Table 1). These multiple ductal proliferations seem to be a characteristic trait of the diathesis. They obviously represent precursors of clinically recognizable adenomas and indicate a high propensity for relapse. Recurrence of salivary gland tumors was noted, indeed, after 1, 15 and 17 years in case No. 2 (Reingold et al. 1977) and after $2\frac{1}{2}$ years in case No. 3 (Batsakis and Brannon 1981) of Table 1. In our case (No. 5 of Table 1) ductal proliferations and microadenomas of the contralateral parotid gland were discovered.

Ductal proliferations and developing microadenomas as previously described (Headington et al. 1977; Reingold et al. 1977) and found in our case allow us to trace the development of the adenomas. Proliferating reserve cells of the intercalated ducts (Fig. 3a) apparently participate in the origin of these adenomatous neoplasms. These observations are in agreement with the concept of Eversole (1971), suggesting that the genesis of monomorphic adenomas (including basal cell adenomas) of salivary glands is attributed to transformed progenitor cells of the intercalated ducts. Early adenomatous proliferations generally show ductal structures (Fig. 3b). Larger adenomatous foci and microadenomas exhibit increasing amounts of solid cellular islands (Fig. 3c), indistinguishable from those of fully developed dermal-type basal cell adenomas.

The scalp tumors of our case and of those from the literature (Table 1) represent typical dermal cylindromas (Crain and Helwig 1961; Pinkus and Mehregan 1981). Dermal cylindromas may be inherited as a dominant disease (Becker 1964; Rasmussen 1975). This is true in the patient of Reingold et al. (1977) and in our patient. In two cases additional trichoepitheliomas were reported (Headington et al. 1977; Reingold et al. 1977). The patient of Reingold et al. (1977) developed further neoplastic changes, i.e. eccrine spiradenomas, Bowen's disease and dermal squamous cell carcinoma. Furthermore, an epidermoid carcinoma of his parotid gland was found. The observation periods of the other patients including our own are too short to exclude a predisposition to additional types of tumors.

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