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ORIGINAL ARTICLE

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Sclerosing polycystic adenosis of parotid gland with dysplasia and ductal carcinoma in situ

Report of three cases with immunohistochemical and ultrastructural examination

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Abstract We describe three cases of sclerosing polycystic adenosis (SPA) of the parotid gland, a salivary condition analogous to fibrocystic disease of the breast. For the first time, immunoreactivity for oestrogen and progesterone receptors was demonstrated, suggesting a possible participation of hormone stimulation in its pathogenesis. In addition, all our cases showed foci of dysplasia of the ductal epithelium, which in one case was severe enough to amount to carcinoma in situ. This feature that has not previously been reported in SPA.

Keywords Salivary gland · Sclerosing polycystic adenosis · Sialadenopathy · Dysplasia · Ductal carcinoma in situ

Introduction

Several neoplasms of the major salivary glands bear a strong histopathological resemblance to breast tumours, but, although similar, they are not quite identical. Most notably, salivary duct carcinoma is morphologically identical to mammary ductal carcinoma with invasive and in situ components, the latter often taking the form of comedocarcinoma [15]. Biologically, however, it is closer to prostatic adenocarcinoma as it is usually negative for oestrogen receptors and positive for androgen receptors [10]. Salivary epithelial–myoepithelial carcinoma [8, 18] is morphologically identical to adenomyoepithelioma of the breast [14], although the former is a true malignancy often with invasion and with a low but definite metastatic potential, whereas the breast lesion is benign with a tendency to recur; evolution of a high-grade malignancy has been described in both [1, 16]. Adenoid cystic carcinoma and pleomorphic adenoma are found in both organs, and collagenous spherulosis too has been described in the breast [3], and salivary glands, particularly in some types of myoepithelial cell-derived salivary tumours [11, 17]. Acinic cell carcinoma of the breast represents another example of this interesting phenomenon [5, 13].

Strangely, the most common breast condition, benign fibrocystic disease, was not thought to have a salivary counterpart, but recently sclerosing polycystic adenosis (SPA) was described as a distinctive pseudoneoplastic lesion of the major glands [7, 21] and it has many histological similarities to its mammary counterpart. Here, we describe three cases of SPA with focal epithelial dysplasia that ranged from mild atypia to low-grade ductal carcinoma in situ. This is a feature that has not previously been reported in SPA.

Materials and methods

The specimens were fixed in 10% formalin, embedded in paraffin and routinely stained with haematoxylin and eosin. Additional histochemical studies included periodic acid-Schiff (PAS), with and without prior diastase digestion, and mucicarmine stains. For electron microscopy, formalin-fixed wet tumour tissue samples of case 1 were washed in phosphate buffer, fixed briefly in 4% paraformaldehyde, postfixed in 1% osmium tetraoxide, and embedded in epoxy resin (Durcupan-Epon). Sections 1-µm thick were stained with uranyl acetate and lead citrate, and examined under an electron microscope.

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Table 1 Antibodies used for immunohistochemical studies. *GCDFP-15* gross cystic fluid disease protein, *EMA* epithelial membrane antigen, *CEA* carcinoembryonal antigen, *MIA* anti-mitochondrial antigen

Antibodies	Clone	Dilution	Source
AE1/AE3		1:500	Boehringer Manheim
CAM 5.2		1:50	Becton-Dickinson, Mountain View
Muscle-specific actin	HHF-35	1:3000	Dako A/S, Glostrup, Denmark
Smooth muscle actin	1A4	1:1000	Dako A/S
Calponin		1:200	Dako A/S
MIA	113–1	1:100	Biogenex, San Ramon, Calif.
GCDFP-15 (BRST2)	D6 clone	1:2000	Signet Laboratories, Mass.
CD68	PGM1	1:100	Dako A/S
CD68	KP1	1:40	Dako A/S
CEA		1:800	Dako A/S
EMA		1:1000	Dako A/S
Oestrogen receptor		1:150	Novocastra, Newcastle upon Tyne, UK
Progesterone receptor		1:150	Novocastra
Ki67	MIB1	1:100	Immunotech, Marseille
Collagen type IV	CIV22	1:50	Dako A/S
S-100 protein	Polyclonal	1:100	Dako A/S
Anti-HER-2/neu Ab-17	Clone e2–4001and 3B5	1:2000	NeoMarkers, Fremont, Calif.

Immunohistochemical studies were performed on deparaffinized, formalin-fixed sections. The antibodies used are listed in Table 1. Specifically for the MIB1 antibody, and for oestrogen and progesterone receptor analyses, sections 4-µm thick were cut from the specimens and placed on slides coated with 3-aminopropyltriethoxy-silane (Sigma, St. Louis, Mo.). An antigen-unmasking technique, with which the sections were microwaved in an oven twice, each for 5 min at 700 W in citrate buffer (pH 6.0) was employed prior to incubation with the primary antibodies. The bound antibodies were visualised using the supersensitive streptavidin–biotin–peroxidase complex (Novocastra, Newcastle upon Tyne, UK), with 3,3-diaminobenzidine (Sigma) as chromogen.

Case reports

Clinical findings

Case 1

A 31-year-old woman was admitted to hospital for a slowly growing tender mass in her right parotid gland region, which she had noticed over the preceding 2 years. Grossly, the lesion measured 3×2.5×2 cm; it was hard in consistency, partly cystic, and freely mobile. A malignant parotid tumour was suspected clinically and a lateral superficial conservative parotidectomy was performed, sparing the facial nerve. The postoperative course was uneventful. No recurrence or other complaints have been experienced during a 3-year follow-up period, and the patient remains alive and well.

Case 2

A 27-year-old woman presented with a slowly growing painless retro-mandibular mass approximately 4.5×3×1.5 cm in size. The lesion was surgically removed from the right parotid gland. It was well circumscribed, firm in consistency, and measured 1.5 cm in maximum diameter. No abnormality was found in six adjacent cervical lymph nodes. The postoperative course was uneventful, and 4 years after surgery there are no signs of recurrence. This lesion was previously reported as *Case 3* in a series of sclerosing polycystic sialadenopathy by Donath et al. [7].

Case 3

A 9-year-old girl first presented to hospital with a mass at the angle of the left mandible, which had been present for 18 months. This was sometimes painful, particularly at night when lying on it in bed. On examination, there was a left parotid mass, 5 cm in diameter. It was spherical and slightly mobile but was not adherent to the skin or bone. Microscopic examination showed "calcular atrophy with sialadenitis". Six years later, a nodule was noted under the surgical scar. It was excised and reported as a pleomorphic adenoma. Thirteen years after that, she presented again with a "massive recurrence", although this has not yet been biopsied. Case 3 was taken from the consultation files, and sections were seen from both the original biopsy and the recurrence. Further tissue was not available for immunohistochemistry.

Microscopic findings

Histological examination in each of the three cases showed a well-circumscribed, partly encapsulated tumour with a peripheral rim of normal salivary gland tissue. Microscopically, the lesions were very similar; they demonstrated an abundant sclerotic collagenous stroma within which were ductal and acinar lobules composed of ectatic ducts, as well as smaller tubular and acinar structures arranged in a vaguely nodular pattern. The collagenous stroma was mostly hypocellular with focal lymphocytic infiltration and occasional lymphocytes encroaching upon the duct epithelium (Fig. 1). Many ducts were cystically dilated and the epithelium comprised flattened cuboidal cells (Fig. 1), but other ducts were lined by hyperplastic epithelium with intraluminal papillary projections. At higher power, smaller ducts and acini were seen to be composed mostly of large cells with abundant eosinophilic, amphophilic or clear cytoplasm. Many ducts contained large eosinophilic cells with abundant cytoplasm with rounded intraluminal projections suggestive of apocrine metaplasia (Fig. 2). The epithelial cells with apocrine morphology merged in places with vacuolated foam cells suggesting sebaceous differentiation (Fig. 2). A few ducts contained aggregates of foam cells within the lumina, and the same vacuolated cells were also observed focally within the collagenous stroma. A spectrum of vacuolated, foam, apocrine, mucous and acinic granular cells was often seen within the same duct or lobule (Fig. 3). Squamous metaplasia of the duc-

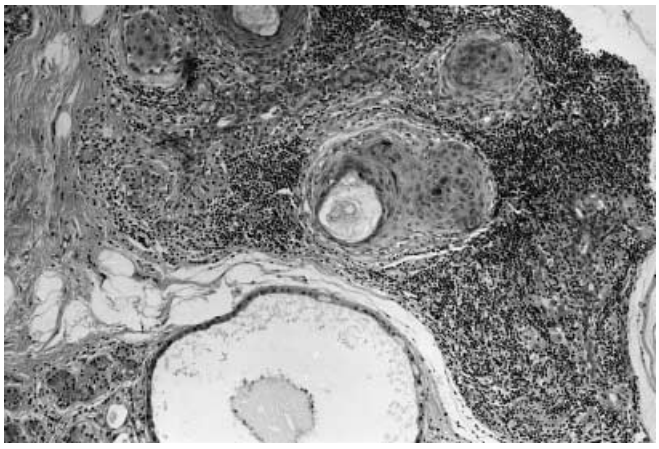


Fig. 1 The lesion is composed of tubular and acinar structures embedded in abundant sclerotic collagenous stroma with focal lymphocytic infiltration. Many ducts are cystically dilated and lined by flattened cuboidal epithelium. Haematoxylin-eosin, ×36

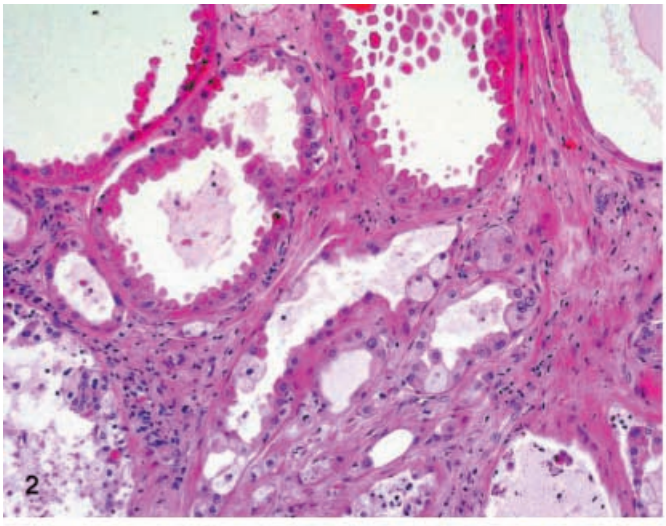
tal epithelium was identified in places. A very typical finding was partial preservation of some lobules, and these were composed of abnormal salivary serous acini consisting of large granular acinar cells with cytoplasm filled with numerous eosinophilic granules and globules. These were of various sizes, but often large, and they stained strongly with PAS (Fig. 4), as well as being faintly positive for mucicarmine. Among these foci of acinar differentiation, there were also cells with water-clear and vacuolated cytoplasm (Fig. 4).

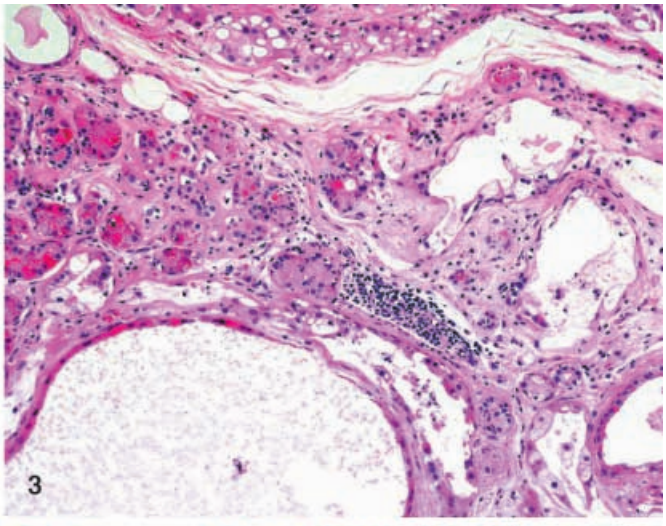
Fig. 2 Dilated ducts are lined by apocrine cells. Epithelial cells with apocrine morphology merge in places with foamy and vacuolated cells resembling sebaceous differentiation. Haematoxylin-eosin, ×180

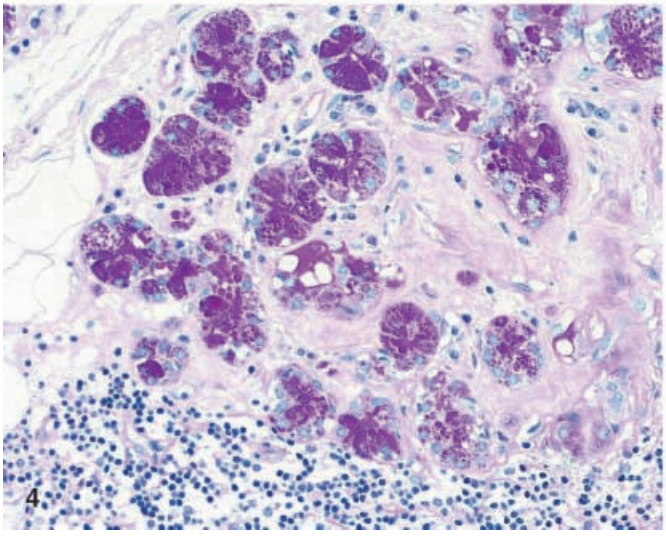
Fig. 3 A spectrum of vacuolated, foam, apocrine, mucous and acinic granular cells is seen within the same lobule. Heamatoxylin-eosin, ×360

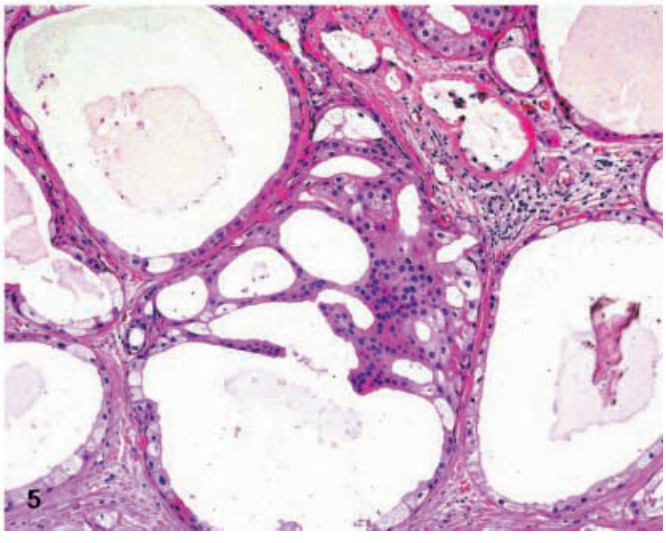
Fig. 4 Irregular serous acini consisting of large acinic cells with abundant granular cytoplasm. Haematoxylin-eosin, ×180

Fig. 5 Focally, the ducts contain – in addition to foamy cells – hyperplastic intraluminal epithelium forming interconnecting bridges in a cribriform pattern. Haematoxylin-eosin, ×180









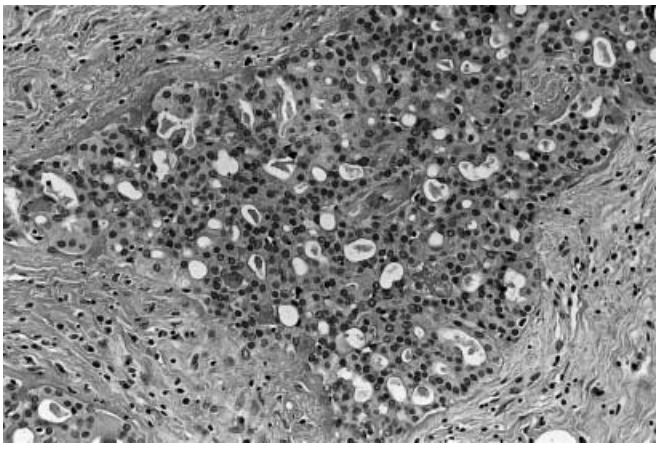


Fig. 6 Focal ductal in situ carcinoma pattern within the solid nest. Haematoxylin-eosin, ×180

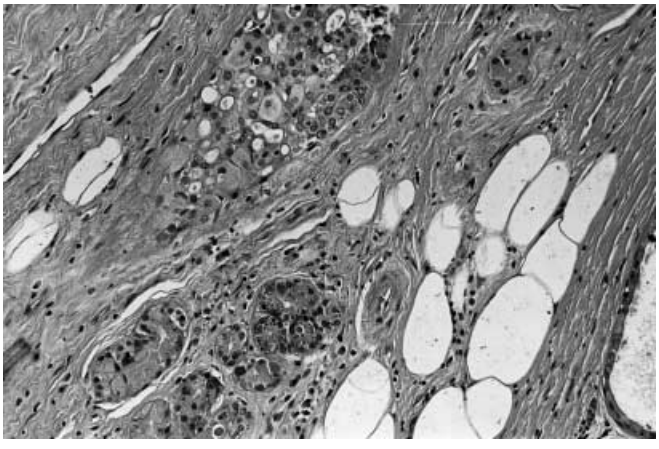


Fig. 7 Dysplastic cells in tiny aggregates and duct are embedded in collagenous stroma, thus suggesting invasive growth pattern. Haematoxylin-eosin, ×180

In all three cases, there were dilated ducts composed of hyperplastic intraluminal epithelium that formed interconnecting bridges in a cribriform pattern (Fig. 5). Within these solid and cribriform foci, the epithelial cells had eosinophilic or vacuolated cytoplasm and nuclei of different shapes and sizes. The nuclear atypia ranged from mild (cases 2 and 3) to severe (case 1) with changes focally amounting to ductal carcinoma in situ of low grade (Fig. 6). Tiny cell aggregates and small ducts composed of dysplastic cells were embedded within the collagenous stroma thus resembling an invasive growth pattern (Fig. 7). Furthermore, there was compression of the stroma to produce bundles of spindle cells mimicking nerves and perineural infiltration.

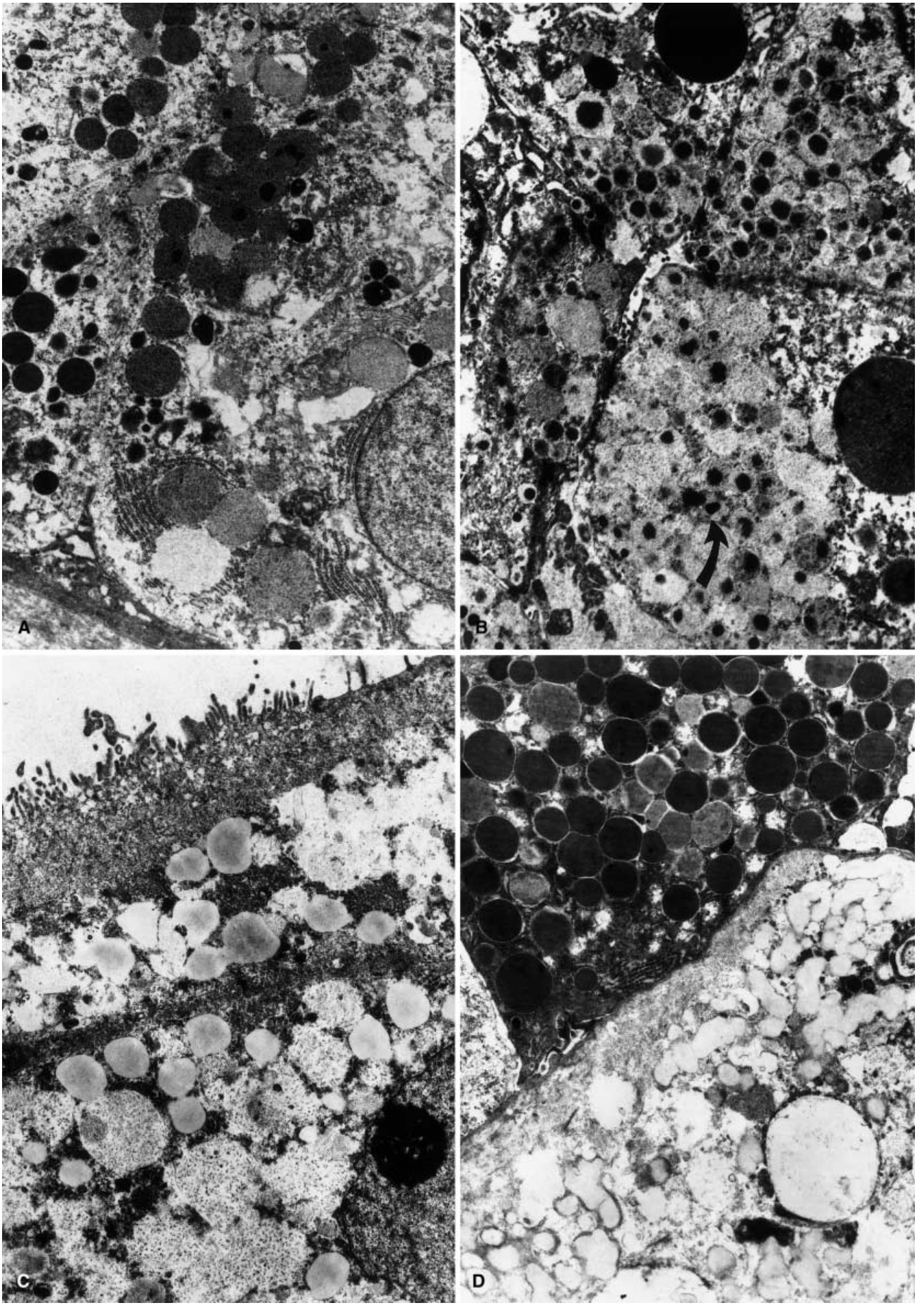
Immunohistochemical staining

Sections from cases 1 and 2 only were available for study. They showed that the ductal and acinar structures reacted uniformly for cytokeratins, as did the vacuolated and apocrine cells of the ductal lining epithelium. The foam cells that filled some lumina and formed stromal aggregates reacted with both anti-CD68 antibodies, but not cytokeratins. Epithelial membrane antigen (EMA) was positive in ductal and apocrine epithelium, while acinar cells were negative. Foam cells that were in contact with lining ductal epithelium were strongly positive for EMA, while intraluminal and stromal foam cells were negative. S-100 protein decorated most proliferating ductal cells and foci of dysplastic epithelium, but apocrine cells, the epithelial lining of ectatic ducts and foam cells did not react. Antimitochondrial antigen 113-1 was expressed in many eosinophilic ductal cells, including a few foam cells lining the ducts. GCDFP-15 was detected in the acinar cells with coarse eosinophilic granules, and also in the epithelium with apocrine-like "decapitation secretion" and in intraductal foam cells; other cells of the lesion were negative. Staining for carcinoembryonal antigen (CEA), and for oncoprotein HER2/neu/cerbB-2 was negative. Most epithelial cells of the cystic ducts, some foam cells lining the ducts, and the cells within the dysplastic foci expressed strong nuclear immunoreactivity for progesterone receptors (about 80%) while staining for oestrogen receptors was detected in about 20% of ductal cells in dysplastic foci only. Most ectatic ducts and serous acini were surrounded by continuous layers of myoepithelial cells that were decorated with S-100 protein, actin and calponin antibodies. Ducts filled with hyperplastic and dysplastic epithelium were mostly surrounded by intact layers of myoepithelial cells. Collagen type IV stained continuous basement membranes subjacent to ectatic and hyperplastic ductal structures, while no staining for collagen type IV was seen close to serous acini. Proliferative activity was low with the MIB1 index about 1%.

Electron microscopic examination

The ultrastructural appearance was variable. Most epithelial cells demonstrated features of secretory cells, and desmosomes were visible between adjacent epithelial cells (Fig. 8A, B). The cells displayed abundant round to oval secretory granules of various size and electron density. The cytoplasm of some cells was packed with large numbers of electron-dense secretory granules consistent

Fig. 8 **A** The secretory cell displays abundant structures of rough endoplasmic reticulum and many round to oval secretory granules of various size and electron density (×4000). **B** The cell shows mucous granules of varying morphology. Note large pale granules with a reticular or particulate content, dense granules, and an assortment of "bull's eye" granules with a central or eccentric core (*arrow*) set in a light or dense matrix (×4000). **C** The apocrine cell has the free surface covered by short widely spaced microvilli and the apical pole of the cytoplasm is filled with numerous electron-lucent secretory granules and empty vesicles (×4000). **D** Ballooned cells exhibit extensive vacuolisation of the cytoplasm (×4000) ▶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with zymogen granules (Fig. 8A), whilst others contained both electron-dense and lucent granules (Fig. 8B). The latter granules often demonstrated indefinite margins and irregular shapes, and many displayed a "bull's-eye" morphology (Fig. 8B). Rough endoplasmic reticulum was frequently prominent within the space between secretory granules (Fig. 8A). The apocrine cells had their free surfaces covered by short widely spaced microvilli and the apical pole of the cytoplasm was filled with numerous electron lucent secretory granules and empty vesicles (Fig. 8C). Ballooned cells exhibited extensive vacuolisation of the cytoplasm (Fig. 8D).

Discussion

Several non-neoplastic diseases and reactive lesions of the salivary glands can mimic malignant neoplasms, both clinically and histopathologically. For example, extensive squamous metaplasia in Warthin's tumour may be misinterpreted as squamous or mucoepidermoid carcinoma [6], and reactive changes in oncocytomas caused by fine needle aspiration injury can resemble acinic cell carcinoma [20]. SPA was first described by Smith et al. in 1996 as a slowly growing mass in the parotid gland suggestive of a neoplasm [21]. Most cases of SPA were initially misdiagnosed as tumours, such as mucoepidermoid and acinic cell carcinomas, cystadenocarcinoma, and pleomorphic adenoma. Similarly, the clinical impression in all our cases of SPA was a salivary gland neoplasm. The initial histopathological interpretation in case 1 was a low-grade cystadenocarcinoma with apocrine differentiation and in case 2 low-grade carcinoma most likely of mucoepidermoid type. Case 3 was diagnosed as pleomorphic adenoma, but, when it recurred, mucoepidermoid carcinoma was suspected.

SPA is quite rare; 21 cases have been published so far [2, 7, 21] and most histopathologists are not familiar with this lesion. Therefore, the lesion can represent a distinctive differential diagnostic problem. Major microscopic clues to a correct diagnosis include maintenance of the lobular architecture of the gland, ductal ectasia, scar-like hyalinised fibrous sclerosis, and a spectrum of foam, apocrine, granular, and mucous cells, in addition to the presence of tubuloacinar structures composed of large acinar cells with prominent brightly eosinophilic granules. In contrast, intraductal hyperplasia, particularly if associated with dysplasia as in our cases, may lead one to suspect a neoplastic process, but clues to the benign nature of SPA are that it is well-circumscribed, lacks an invasive growth pattern, and that mitotic/proliferative activity is low.

A particularly interesting finding in both our cases and previous studies of SPA is an overlapping spectrum of apocrine, vacuolated, foamy and sebaceous-like cells. The foam cells were observed floating free within the cystic lumina, clustered in the periductal stroma or in continuity with the epithelial lining of the glandular structures. An identical spectrum of cells showing apocrine morphology and abundant finely vacuolated cytoplasm (foam cells) is frequently found in most benign lesions of the breast, particularly in those associated with apocrine cystic changes and duct ectasia [4]. The origin of mammary foam cells has long been debated, and many morphological and immunohistochemical studies have supported either a histiocytic or an epithelial nature. Recently, Damiani et al. [4] used immunohistochemistry and in situ hybridisation to demonstrate a spectrum of phenotypes in mammary foam cells, from epithelial-apocrine cells to macrophage-derived phagocytic cells. This phenomenon of overlapping between epithelial-apocrine, epithelial-oncocytic, and macrophagic phenotype may be more common than is now realised. In the salivary glands, we have demonstrated a similar combination of oncocytic, apocrine and foam cells in a series of oncocytic myoepitheliomas [19]. The most probable explanation is related to the well-known propensity of myoepithelial and ductal cells in mammary and salivary gland tissues to undergo metaplastic changes.

Electron microscopic examination confirmed secretory activity in most epithelial cells of the lesion. Neoplastic cells had abundant cytoplasm filled by electron-dense granules of various sizes consistent with zymogen granules. Moreover, the serous acini of SPA did not show histologically architectural and cytological features of normal salivary gland acini. They resembled acinic cell carcinoma differentiation with histological features strongly reminiscent of those seen in cases of acinic cell carcinoma of the breast [5, 13]. The electron-lucent granules with indefinite margins and irregular shapes are most probably identical to the empty vesicles, observed in studies of normal and neoplastic apocrine epithelium [9, 12].

The pathogenesis of SPA is uncertain. Most histological features, such as duct ectasia, sclerotic fibrosis, intraluminal epithelial proliferation, acinic cell hyperplasia, and metaplastic changes of the epithelium, point to a reactive postinflammatory process, and a pathogenetic relationship with juvenile recurrent chronic parotitis has been suggested [2]. However, microscopic resemblance to fibrocystic disease of the breast, and expression of oestrogen and progesterone receptors suggest that SPA of the salivary glands and fibrocystic disease of the breast may share a similar aetiological basis.

In our cases, we found a variable degree of dysplastic changes within the epithelium that ranged between mild atypia (cases 2 and 3) to focally severe dysplasia (case 1): the latter conformed to the established criteria of ductal carcinoma in situ in the breast. The presence of hyperplastic intraluminal ductal epithelium with minimal nuclear atypia was briefly mentioned by Smith et al. [21], but dysplasia such as in our cases has not so far been described in SPA. We have seen foci of dysplasia in all our cases, and the lesions illustrated in Fig. 2 and Fig. 5 by Smith et al. [21] are at least suspicious of cribriform pattern of carcinoma in situ very similar to our lesions. Two of our patients remained well without any

signs of disease within limited follow-up periods of 4 years and 3 years after surgery, whilst the third still has recurrent disease. Also there were two recurring cases in the report of Smith et al. [21] and two recurrences in that of Batsakis et al. [2]. It is noteworthy that recurrences occurred after 4.5 years and 9 years after excision in the series by Smith et al. [21] and after 6 years in our patient, while the mean follow-up period in recurrence-free patients from both previous series [2, 21] is only 30 months. We believe that the recurrence rate and occurrence of malignancy in the lesions called SPA is more frequent than expected before and we are convinced that SPA is a neoplastic lesion with a low-grade malignancy potential.

In conclusion, regardless of the causative mechanism, sclerosing polycystic adenosis of major salivary glands is a distinctive histopathological lesion that may simulate malignant tumours, both clinically and microscopically, and, despite possibly alarming features, such as epithelial hyperplasia and dysplasia, SPA seems to have a favourable outcome. However, reports of cases of SPA are still few, and more are needed to evaluate the biological nature of the lesion.

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