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Pathology in Focus

Co-existing pleomorphic and tubular basal cell adenomas of the parotid gland

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Abstract
We report a hitherto undescribed case of co-existence of a pleomorphic adenoma and a tubular basal cell adenoma affecting the superficial lobe of the left parotid gland of a 53-year-old man. The histology of the pleomorphic adenoma is also of interest in that the prominent adipose metaplasia of its myxoid stroma yielded an appearance reminiscent of myxoid lipoma. The tubular basal cell adenoma showed gross cystic change, and its solid portion consisted of closely packed tubules lined by double layers of cuboidal cells with little intervening stroma. Unlike Warthin's tumour and membranous basal cell adenoma, both pleomorphic and tubular basal cell adenomas exhibit no propensity towards multicentricity or bilaterality. We, therefore, believe that their co-existence in the superficial lobe of the parotid gland of our patient is a mere concidence rather than association.

Key words: Parotid neoplasms; Adenoma, pleomorphic; Adenoma, tubular basal cell

Introduction

In spite of its relatively common occurrence in the major salivary glands, it is exceedingly rare to find more than one pleomorphic adenoma in the same salivary gland, or to detect this tumour in more than one salivary gland in the same individual (Gardner *et al.*, 1994; Ellis and Auclair, 1996). Warthin's tumours, on the other hand, are bilateral in five to 7.5 per cent of the cases, and may even be multiple, with more than one tumour developing in the same salivary gland (Patey and Thackray, 1970; Ellis and Auclair, 1996). Such multicentric property has also been described for membranous or dermal analogue basal cell adenoma but not in the other types of basal cell adenomas, including tubular basal cell adenoma (Batsakis *et al.*, 1991; Ellis and Auclair, 1996).

In this article, we report an extremely rare and hitherto undescribed case of co-existing pleomorphic and tubular basal cell adenomas occurring in the same parotid gland of a middle-aged man, who remained well with no evidence of disease more than three years after local resection of the tumours.

Case report

A 53-year-old previously healthy man noticed a slowly enlarging and painless mass in his left retromandibular region for one year, and was referred to our hospital for further management. Physical examination showed a firm mass in the parotid region, 3.5 × 3 cm across. Fine needle aspiration cytology revealed features consistent with those of a pleomorphic adenoma. During surgical operation, two discrete nodular masses were found in the superficial lobe of the left parotid gland. A left superficial parotidectomy, with removal of both masses, was performed.

The resected superficial lobe of the parotid gland measured 4.5 × 4 × 2 cm, and contained two distinct and well circumscribed tumour masses (Figure 1). The larger one was located anteriorly, measured 2.5 × 2 × 1.5 cm, and showed a uniform, pale yellow, firm cut surface (Figure 1). The smaller one was located in the posterior aspect, measured 1.5 × 1 × 1 cm and exhibited prominent

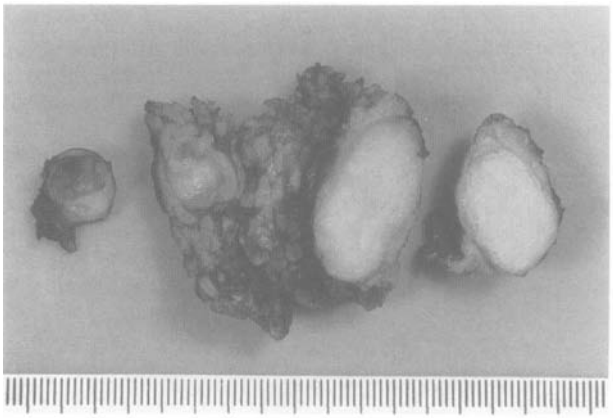

Fig. 1

Gross appearance of the resected superficial lobe of the parotid gland, showing the discrete nature of the two nodular masses. The pleomorphic adenoma on the right shows a uniform light yellow firm cut surface, whilst the tubular basal cell adenoma on the left demonstrates prominent cystic change. One smallest scale mark represents 1 mm.

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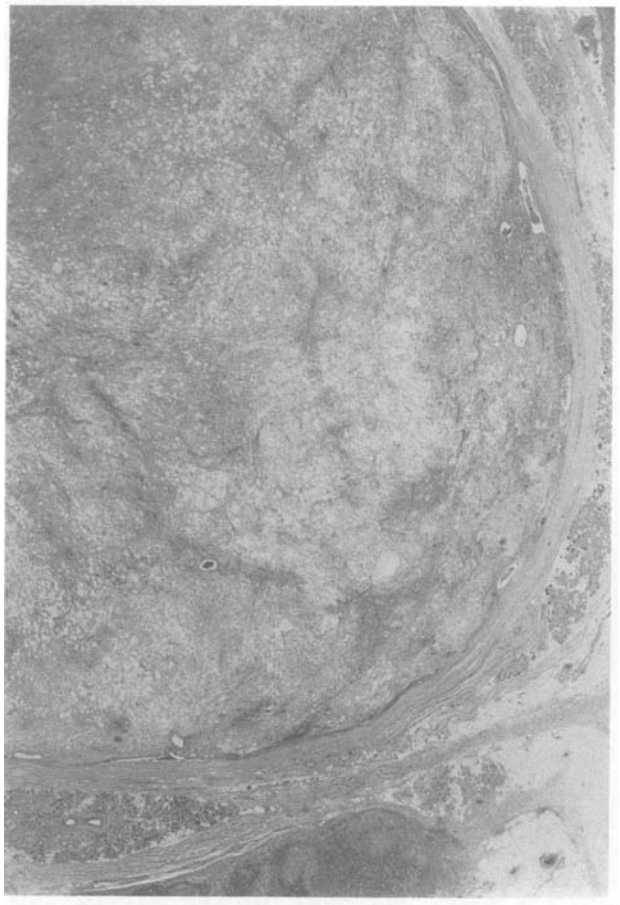

FIG. 2A
Low-power magnification of the pleomorphic adenoma, showing its encapsulation and extensive stromal adipose metaplasia (H & E; × 7.5).

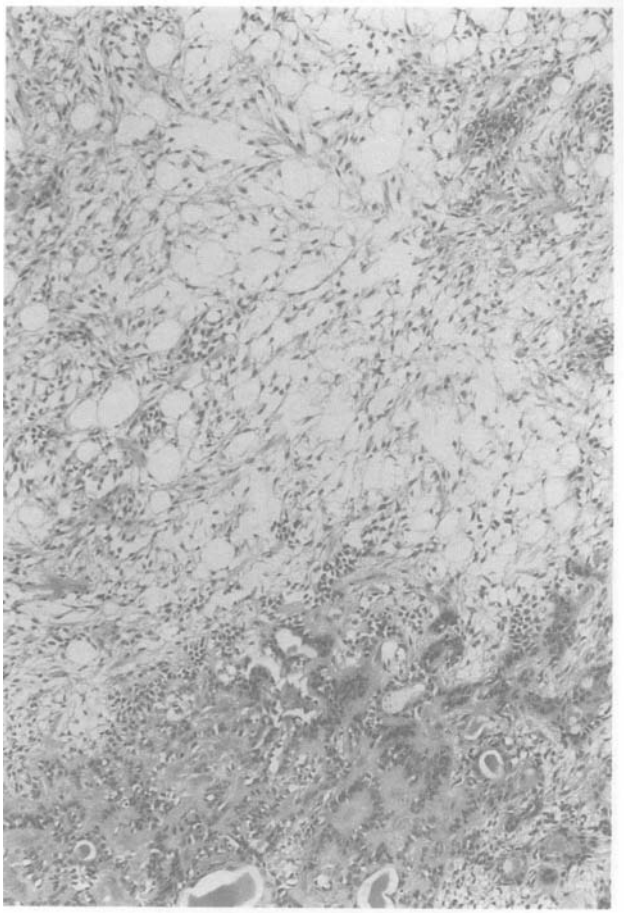

FIG. 2B
High-power magnification of the tumour, showing tubules lined by inner ductal and outer myoepithelial cells. The latter stream into the surrounding myxoid stroma where prominent adipose metaplasia is noted, reminiscent of myxoid lipoma. (H & E; × 60).

cystic change, with a white firm solid portion, 0.5 × 0.5 × 0.3 cm (Figure 1).

Representative blocks were taken and 4 μm thick paraffin sections cut and stained with haematoxylin and eosin, alcian blue (pH 2.5) and periodic acid-Schiff (PAS). Immunohistochemical study was performed on the paraffin sections using the avidin biotin-peroxidase technique, with appropriate positive and negative controls. The panel of antibodies included AE1/AE3 (Hybritech, San Diego, USA) and CAM 5.2 (Becton Dickinson, San Jose, USA) as epithelial markers, Muscle-specific actin (HHF-35) (Enzo Biochem, New York, USA) and S-100 protein (Dakopatts, Copenhagen, Denmark) as markers for myoepithelial differentiation. In addition, a tissue block from the larger solid tumour was taken from the formalin-fixed specimen, immersed in gum sucrose overnight, frozen, cryostat-sectioned, and stained with oil red O stain for fat.

Histologically, both tumours were well encapsulated (Figures 2 and 3). The larger one demonstrated typical features of pleomorphic adenoma, consisting of small aggregates of tubules lined by inner ductal and outer myoepithelial cells, supported in a myxoid stroma (Figure 2A). The myoepithelial cells surrounding the tubules streamed into the surrounding myxoid stroma where prominent adipose metaplasia was noted, reminiscent of myxoid lipoma (Figure 2B). The fat content of these stromal adipose cells was confirmed by oil red O stain, and the myxoid stroma showed positive staining with alcian blue at pH 2.5. The ductal cells stained positively for AE1/AE3 and CAM 5.2, whereas the myoepithelial cells showed positive staining for muscle-specific actin and S-100 protein.

The solid portion of the smaller tumour displayed a uniform histology, consisting of closely packed tubules, with little intervening stroma, bordered by double layers of cuboidal cells (Figure 3A). Many of these tubules contained eosinophilic material in their lumens (Figure 3B). PAS stain highlighted only a thin layer of basal lamina surrounding these tubules. In particular, thick bands of PAS-positive hyaline basal lamina surrounding nests of basaloid cells, characteristic of membranous or dermal analogue basal cell adenoma, was not seen. Many cuboidal cells showed positive staining for AE1/AE3 and CAM 5.2, whereas only focal positive staining for muscle-specific actin and S-100 protein was noted. The resection margins were clear of tumour.

The features were those of co-existing pleomorphic and tubular basal cell adenomas of the parotid gland, completely excised by superficial parotidectomy. The patient made an uneventful recovery and remained well with no evidence of disease more than three years after operation.

Discussion

Pleomorphic adenoma is the commonest tumour of the major salivary glands, accounting for 65 to 75 per cent of all tumours of the parotid gland (Eneroth 1971; Ellis and Auclair, 1996). Monomorphic adenomas other than

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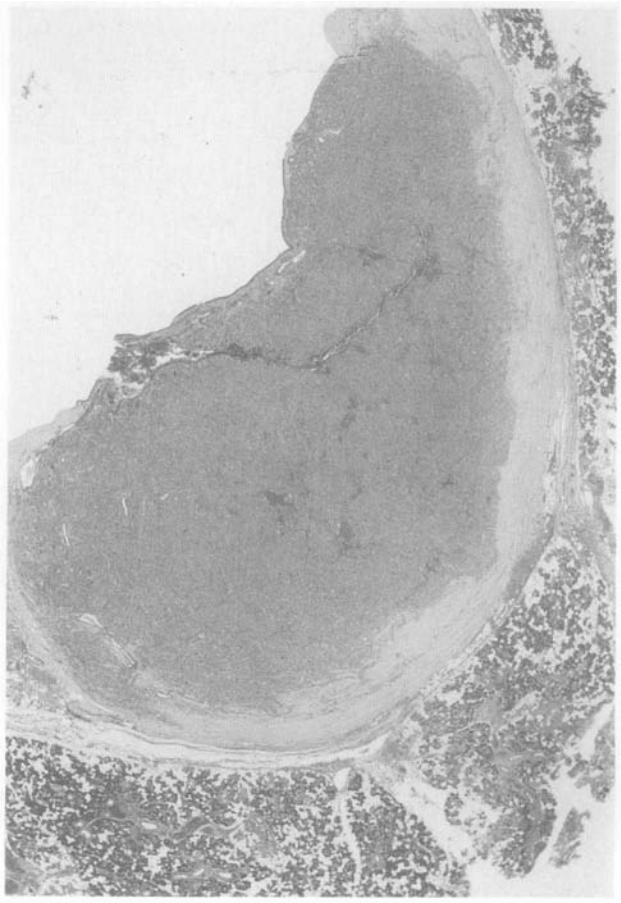

FIG. 3A

Low-power magnification of the tubular basal cell adenoma, showing prominent cystic change and uniform histology of the solid portion. (H & E; × 7.5).

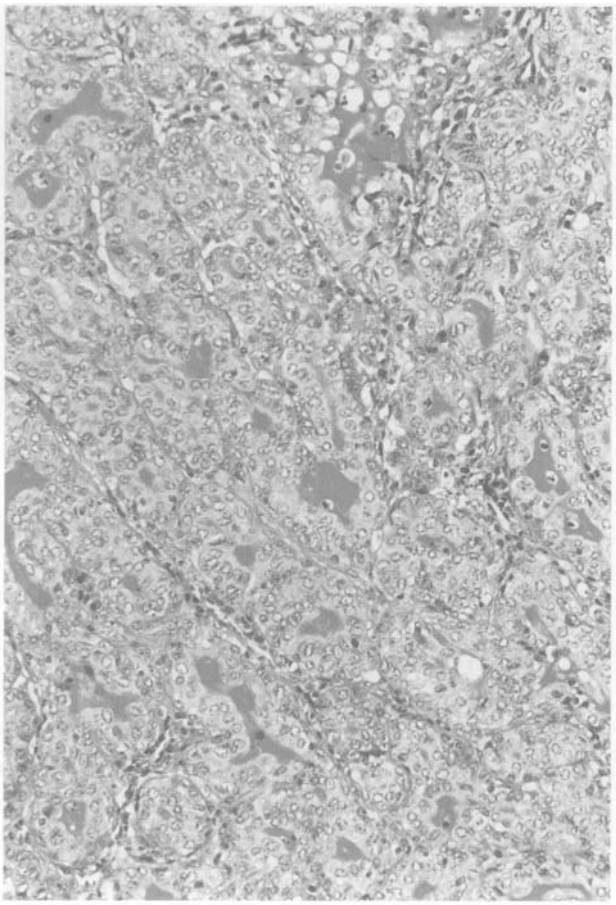

FIG. 3B

The tumour consists of tubules bordered by double layers of cuboidal cells. Eosinophilic material is seen in many of the tubular lumina (H & E; × 125).

Warthin's tumour, on the other hand, are rare, making up only two to four per cent of the parotid tumours (Maurizi *et al.*, 1990). Furthermore, although bilateral and multicentric occurrences are not uncommon for Warthin's tumours (Patey and Thackray, 1970; Ellis and Auclair, 1996), such phenomena are exceedingly rare for pleomorphic adenoma (Gardner *et al.*, 1964; Ellis and Auclair, 1996). With the exception of the membranous or dermal analogue type of basal cell adenoma, which is notorious for its frequent multicentricity and respectable rate of recurrence, all other basal cell adenomas, including the tubular type, are usually solitary (Batsakis *et al.*, 1991; Ellis and Auclair, 1996). The co-existence of a pleomorphic and a tubular basal cell adenoma in the same parotid gland in our patient is unique and, to our knowledge, has never been described in the English literature.

The histology of the pleomorphic adenoma of our patient is also of interest. Although aggregates of mature adipose cells may occasionally be seen in the stroma of pleomorphic adenomas (Ellis and Auclair, 1996), extensive and prominent adipose metaplasia in a myxoid stroma, reminiscent of myxoid lipoma (Figure 2), is distinctly uncommon and has only been recently described (Ng and Ma, 1995; Jin *et al.*, 1996).

Due to its small size, the tubular basal cell adenoma was not detected during clinical examination and, consequently, also escaped fine needle aspiration. Fortunately, it was superficially located and noted by the surgeon during operation. It was therefore resected together with the pleomorphic adenoma. The gross cystic change, together with the uniform histology of closely packed tubules bordered by double layers of cuboidal cells (Figure 3), are characteristic of tubular basal cell adenoma (Cho and Kim, 1989; Maurizi *et al.*, 1990; Ellis and Auclair, 1996). In contrast to the membranous or dermal analogue type, the recurrence rate of tubular basal cell adenoma is so low as to be almost nonexistent after adequate local excision (Bataskis *et al.*, 1991; Ellis and Auclair, 1996). The prognosis is thus excellent and indeed our patient remained well with no evidence of disease more than three years after operation.

Since both pleomorphic and tubular basal cell adenomas lack the propensity for bilaterality and multicentricity (Gardner *et al.*, 1964; Batsakis *et al.*, 1991; Ellis and Auclair, 1996), we believe that the co-existence of these tumours in the same parotid gland of our patient is a mere coincidence rather than association.

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