

Pathology in Focus

Recurrent canalicular adenoma of the minor salivary glands in the upper lip

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Abstract

Canalicular adenoma is a recently classified uncommon salivary gland neoplasm. This biologically benign growth tends to be multifocal and occurs most often in the upper lips of elderly people. Histologically and clinically it differs from the basal cell adenoma, for which it may be mistaken, in a number of ways. Its clinical importance lies in the fact that it may be confused with malignancy. Little information is available regarding the recurrence and long-term follow-up of these tumours, and where available it covers only relatively short periods.

We report the recurrence of a canalicular adenoma after an 11.2 year disease-free period.

Key words: Salivary gland neoplasms; Adenoma, canalicular; Lip

Case report

A 67-year-old lady presented with a one-year history of slowly growing swellings in her upper lip. These caused some discomfort, but were not painful. She was otherwise healthy.

She had a previous history of a single tumour of the upper lip in December, 1985. This had been removed and reported, at the time, as a basal cell adenoma of the minor salivary glands. She had not had any further related problems until the recent presentation.

On examination, five solid swellings of variable sizes (from 5 to 12 mm) were palpated across the upper lip. These were firm in consistency, but smooth, mobile and not tender (Figure 1). There were no other lesions in the oral cavity or salivary glands, and no palpable lymph nodes in the neck. Excision was planned and this was done under



FIG. 1

Diagrammatic illustration of the lesions in the upper lip.

general anaesthesia through two horizontal incisions placed on the inner lip surface. Five brownish-grey lesions were easily dissected from the surrounding tissues. The incisions were closed with absorbable sutures.

Histology of the lesions (Figure 2) showed the tumours to consist mostly of bilayered strands of columnar cells forming canalicular structures, with beading of the strands. The cells displayed moderate amounts of eosinophilic cytoplasm. The stroma consisted of loose connective tissue with rather myxoid areas and numerous small blood vessels. There was no evidence of malignancy.

The appearances were those of canalicular adenoma of the minor salivary glands. Review of the previous histology from 1985 (Figure 3), revealed similar features and would today be classified as a canalicular adenoma. The patient will be reviewed on a regular basis to exclude further recurrences.

Discussion

Canalicular adenoma is a recently classified, biologically benign, salivary gland neoplasm (Seifert and Sobin, 1991; Simpson, 1994). These tumours tend to be multifocal and usually occur on, or near, the upper lip of elderly patients (Daley *et al.*, 1984). Some authors regard it as an almost exclusively labial or oral tumour (Batsakis, 1991). Canalicular adenomas are thought to be derived from the cells of the excretory ducts of the minor salivary glands (McMillan *et al.*, 1993).

Clinically these tumours are uncommon, and as in our patient, usually present as painless submucosal swellings or nodules. Black people may have an increased incidence of occurrence, and most patients are over 50 years of age (Batsakis, 1991). Surgical removal is generally easy, but

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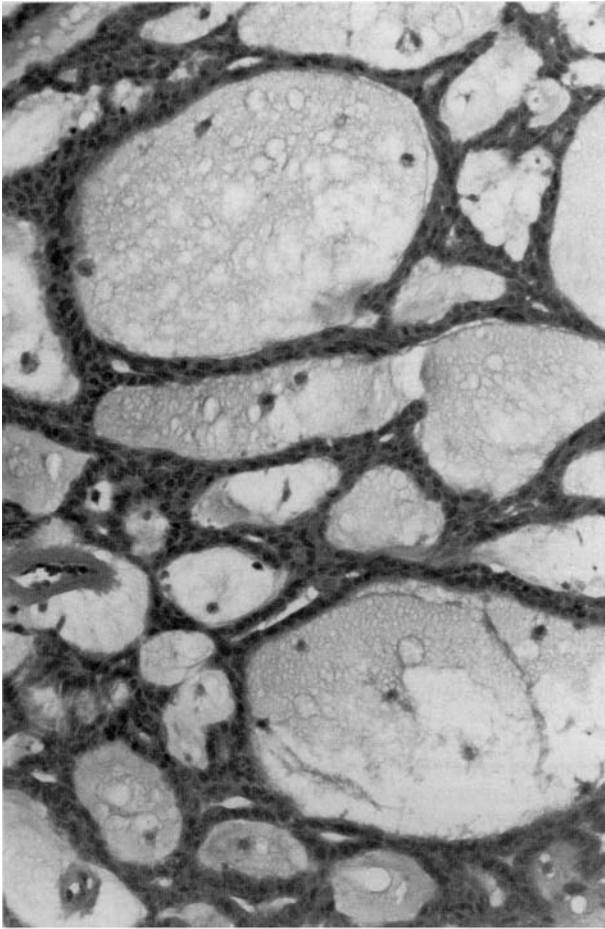


FIG. 2

A photomicrograph showing canalicular structures consisting of bilayered strands of epithelial cells with occasional 'beading'. (H & E; $\times 160$)

the multifocal nature of the lesion should be borne in mind.

The canalicular adenoma has previously been regarded as a variant of the basal cell adenoma, as was the case in our patient following the excision of the solitary tumour in 1985. In the revised WHO classification of salivary gland tumours, the canalicular adenoma is regarded as a separate entity (Simpson, 1994).

Morphologically canalicular adenomas differ from basal cell adenomas in a number of ways. Canalicular adenomas are largely composed of bilayered strands of cells displaying a moderate to abundant amount of eosinophilic cytoplasm and set in a loose, vascular stroma (Daley *et al.*, 1984; Simpson, 1994). Some canalicular adenomas also display so-called 'beading', which is separation of the bilayered epithelial strands (Daley *et al.*, 1984). In contrast, basal cell adenomas are composed of basaloid cells with scanty basophilic cytoplasm, set in a rather fibrous stroma with less conspicuous blood vessels (Daley *et al.*, 1984).

The most important differential diagnosis that must be excluded is a low grade adenocarcinoma (Batsakis, 1991). The delicate vasculature, a parvicellular and often watery stroma with a uniform cell population arranged in nests, cords and canaliculi surrounded by a well-defined basement membrane point to an adenoma rather than an adenocarcinoma (Nelson and Jacoway, 1973; Mintz *et al.*, 1982).

Although much work has been carried out regarding the morphology, ultra-structural characteristics (McMillan

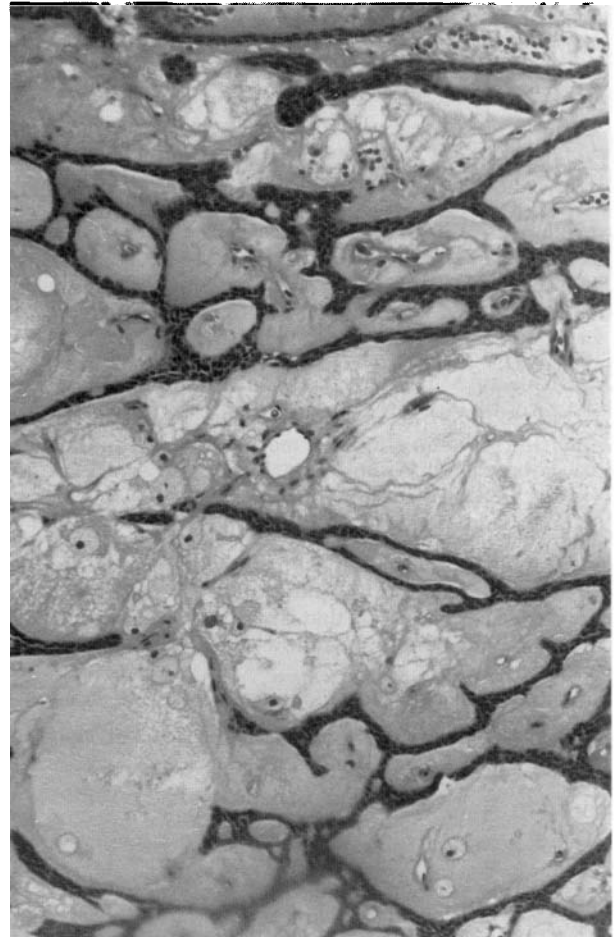


FIG. 3

A photomicrograph showing the features of canalicular adenoma in the original tumour. Note the loosely textured stroma containing thin walled blood vessels. (H & E; $\times 145$)

et al., 1993), and immunohistochemical analysis (Ferreiro, 1994) of canalicular adenomas, little information is available regarding recurrence and long-term follow-up. Available information often covers only short periods.

One study revealed no recurrence over a six to seven year period (Daley *et al.*, 1984). A review article by Batsakis, (1991) reported no recurrence during follow-up periods of up to 10.6 years. It should be noted that the multifocal nature of these tumours can set the stage for 'recurrence' if all the multifocal nodules are not excised at the first excision (Batsakis, 1991).

Conclusion

This case illustrates recurrence of a canalicular adenoma after a disease-free period of 11.2 years. In addition, where the initial disease presented as a single tumour, the recurrent disease is multifocal. Histological appearances in both instances are typical of canalicular adenoma.

This case provides, to the best of our knowledge, the longest follow-up information with a positive recurrence of canalicular adenoma in the English literature.

References

- Batsakis, J. G. (1991) Oral monomorphic adenomas. *Annals of Otolaryngology, Rhinology and Laryngology* **100**: 348–350.
- Daley, T. D., Gardner, D. G., Smout, M. S. (1984) Canalicular adenoma not a basal cell adenoma. *Oral Surgery, Oral Medicine and Oral Pathology* **57**: 181–188.

- Ferreiro, J. A. (1994) Immunohistochemical analysis of salivary gland canalicular adenoma. *Oral Surgery, Oral Medicine and Oral Pathology* **78**(6): 761–765.
- McMillan, M. D., Smith, C. J., Smillie, A. C. (1993) Canalicular adenoma: Report of five cases with ultrastructural observations. *Journal of Oral Pathology and Medicine* **22**: 368–370.
- Mintz, G. A., Abrams, A. M., Melrose, R. J. (1982) Monomorphic adenomas of the major and minor salivary glands. Report of 21 cases and review of the literature. *Oral Surgery, Oral Medicine and Oral Pathology* **53**: 375–386.
- Nelson, J. F., Jacoway, J. R. (1973) Monomorphic adenoma (canalicular type). Report of 29 cases. *Cancer* **31**: 1511–1513.
- Seifert, G., Sobin, L. H. (1991) Histological typing of salivary gland tumours, 2nd Edition, Springer-Verlag, Berlin.
- Simpson, R. H. W. (1994) Classification of tumours of the salivary glands. *Histopathology* **24**: 187–191.

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