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CASE REPORT

Pleomorphic adenoma of a molar salivary gland

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KEYWORDS
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Summary Pleomorphic adenoma of a buccal or molar minor salivary gland, which lies on the external aspect of buccinator, has not been reported previously. We report a case of a pleomorphic adenoma apparently arising from such a gland. Histologically there was marked cystic degeneration producing an apparently empty lumen surrounded by an encapsulated cellular mass. While the final diagnosis was pleomorphic adenoma, there were a number of features of myoepithelioma and the differences between these entities are discussed.

Case report

A 49 year old Caucasian man was referred by his medical practitioner for management of a painless lump in his right cheek. He had become aware of the mass three months previously and over this period he felt that it had doubled in size. There was no facial asymmetry nor other symptoms. Intra-oral examination revealed a 3 cm firm, well-circumscribed, mobile mass with some fluctuance located posterior to the right parotid duct (Fig. 1). The overlying mucosa and skin were normal in appearance. Clear saliva was expressed from the right parotid duct. Insertion of a lacrimal probe into the parotid duct indicated that the lesion was not in continuity with the duct. CT scan showed a well-demarcated lesion lying on the lateral aspect of buccinator (Fig. 2).

Fine needle aspiration biopsy produced 2 ml of straw-coloured fluid that showed clusters of plasmacytoid cells, uniform bland spindled mesenchymal cells, occasional uniform cuboidal epithelial cells in a background of a thin proteinaceous material, suggestive of pleomorphic adenoma.

The tumour was excised via an intra-oral approach through the buccinator muscle. It was encapsulated, not involving the parotid gland or duct, and the wound was closed primarily.

Histological examination showed an encapsulated lesion with a solid periphery surrounding a central cyst-like space (Fig. 3). The solid regions were composed of sheets and strands of uniform cells with plump oval to spindle-shaped nuclei and poorly defined eosinophilic cytoplasm (Fig. 4). Some cells had a clear cytoplasm. Few plasmacytoid cells were found. There was no significant mitotic activity. There were areas where these cells were densely packed with only a small amount of mature fibrous connective tissue between them. Elsewhere, hyalinised connective tissue separated them. Occasional ducts containing acellular, eosinophilic material and lined by a single row of flat to cuboidal cells were present. A number of thick walled blood vessels could

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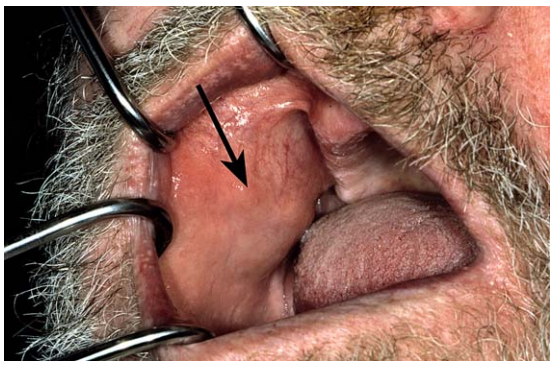

Figure 1 Clinical photograph showing the lesion lying posterior to the right parotid duct orifice (arrow).

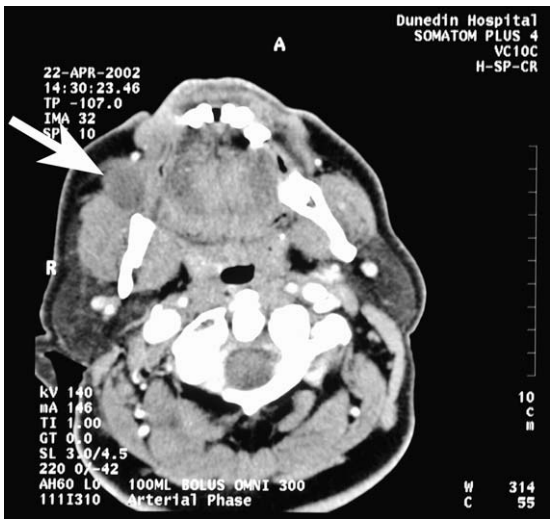

Figure 2 CT scan showing the well-demarcated lesion lying on the lateral aspect of the right buccinator muscle.

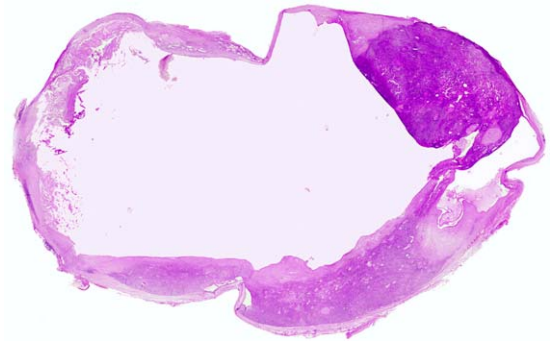

Figure 3 Photomicrograph showing the pleomorphic adenoma with extensive cystic degeneration—original × 4.

be seen. Also present were large zones of dense, paucicellular fibrous tissue. Amorphous eosinophilic material was present in the cyst space. There was no myxoid or cartilaginous stroma.

The tumour cells were positive for cytokeratin, vimentin and S-100, but did not react with smooth muscle actin (SMA).

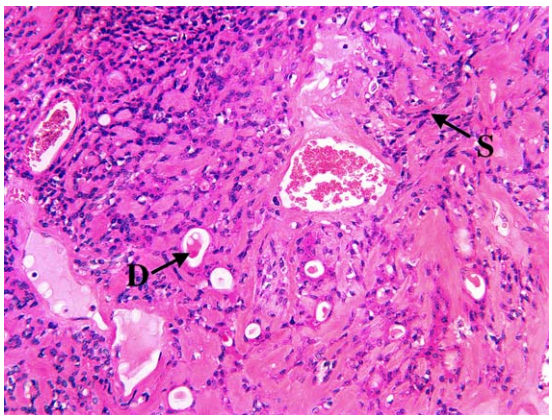

Figure 4 Photomicrograph showing oval to spindle-shaped myoepithelial cell nuclei (S) separated by fibrous connective tissue. Duct-like structures (D) containing eosinophilic material are present—original × 200.

Discussion

Most intra-oral pleomorphic adenomas develop in the minor salivary glands of the palate, followed by the glands in the upper lip and buccal mucosa.[1] Pleomorphic adenomas that have been described in the buccal mucosa are considered to have arisen in the *submucosal* buccal minor salivary glands.[1] We are unaware of any reports in the literature to date that have described salivary gland tumours arising in the minor salivary glands in the buccal space, external to the buccinator. These glands are referred to as the *buccal* or *molar* glands and their ducts pierce the buccinator to drain opposite the last maxillary molar teeth.[2]

Most minor salivary gland tumours present as a smooth, submucosal mass or nodule and the neoplasm's rate of growth is usually indolent. Rapid enlargement, as in the current case, is unusual in benign tumours, although may be seen in high-grade malignant salivary tumours.[3] In a study of 50 patients with minor salivary gland tumours of the lip and buccal mucosa, the median duration of symptoms before diagnosis was five months for buccal tumours.[3]

The histological features of the specimen were interesting in that most of the solid part of the tumour had a uniform architecture rather than the variable pattern frequently seen in pleomorphic adenomas. The predominant cell type was a spindle-shaped myoepithelial cell which was cytokeratin, vimentin, and S-100 positive, raising the possibility that this lesion was a myoepithelioma rather than a pleomorphic adenoma. Using the criteria of Dardick et al.[4] and Gnepp et al.[5] the current lesion would have fitted the diagnosis of myoepithelioma, since they accept this diagnosis as long as no more than 5% of the lesion is composed of ductal epithelium. In contrast, Seifert et al.[6] and Ellis and Auclair[7] comment that a myoepithelioma should not show ductal differentiation and, in examples with duct formation, the most appropriate diagnosis is pleomorphic adenoma with myoepithelial predominance. Whilst it is likely that myoepithelioma is at one end of the histological spectrum of pleomorphic adenoma, distinction between the two may be clinically significant in that myoepitheliomas have been reported to behave in a more aggressive manner and have a greater likelihood of

undergoing malignant transformation than pleomorphic adenomas.[6,8] In contrast, Ellis and Auclair[7] state that they are less likely to recur than pleomorphic adenomas.

Many pleomorphic adenomas and myoepitheliomas arising in minor salivary glands have a capsule that is often thin or incomplete,[5,7] but this lesion was enclosed in a well-defined fibrous capsule. There was no evidence of extension into or through the capsule.

Cystic degeneration in large pleomorphic adenomas is mentioned[5] and an example is illustrated in the Armed Forces Institute of Pathology fascicle.[7] De las Casas et al.[9] reported a cystic myoepithelioma in the minor salivary glands of the tongue.

Treatment of pleomorphic adenoma has included simple excision, excision with a margin and the use of adjuvant radiotherapy.[3,10,11] The tendency for pleomorphic adenoma to recur is well recognised and a five year follow-up period may be inadequate for these tumours as recurrences have presented up to 18 years after initial treatment.[11] A 10–20 year follow-up period may be more suitable for both pleomorphic adenomas and myoepitheliomas.

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