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A minor salivary gland tumour presenting with dysphagia

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Abstract
A case is reviewed of a giant benign myoepithelioma of the soft palate presenting in an elderly female patient. Due to the large size of the lesion and its mass effect the patient developed dysphagia with subsequent significant weight loss. The clinico-pathological features of this rare tumour are described and the literature reviewed.

Key words: Myoepithelioma; Deglutition disorders; Salivary glands, minor

Introduction

Myoepithelioma is a rare tumour of the salivary glands composed of myoepithelial cells and was first described by Sheldon (Sheldon, 1943). It accounts for less than one per cent of all salivary gland tumours and the sites of predilection are the parotid gland and palate (Seifert and Sobin, 1991). These neoplasms vary considerably in both their architecture and cytology and may cause diagnostic confusion with the myoepithelial-cell-predominant variant of pleomorphic adenoma. This paper presents an unusually large myoepithelioma arising from the soft palate and discusses its clinical and pathological features and differentiation from pleomorphic adenoma.

Case report

A house bound 85-year-old female presented with a giant intra-oral tumour arising from the soft palate. The lesion had been present for five years but had grown more rapidly in the previous six months. The large tumour mass had resulted in dysphagia with the inability to swallow solid food. As a consequence the patient had lost 5 kilograms which represented more than 10 per cent of her total body weight.

Examination revealed a 6 × 6 cm tumour arising from the right side of the soft palate attached by a broad base (Figure 1). The patient was edentulous with no other intraoral abnormality. No cervical lymphadenopathy was detected. A provisional diagnosis of benign salivary tumour was made. In view of the medical history of hypertension and severe ischaemic heart disease, the patient underwent simple enucleation of the lesion under general anaesthesia. This involved preservation of the nasal mucosal layer, excision of expanded redundant oral mucosa and primary closure. The patient made an uneventful post-operative recovery. On review one year post-operatively she remains tumour free. Histopathological examination showed a well circumscribed partly encapsulated tumour composed of solid sheets of cells with areas of single cells, clumps of cells and trabeculation. No ductal differentiation was present. The neoplastic cells had large eccentric pleomorphic nuclei with abundant diffusely eosinophilic cytoplasm and stained positively with S–100 (Dako rabbit anti-cow S–100) and Cam 5.2 (Becton Dickinson murine monoclonal antibody) (Figures 2–4) reflecting myoepithelial differentiation. There was an abundant myxoid stroma. No malignant features were seen. In view of these appearances a definitive diagnosis of benign myoepithelioma of the plasmacytoid (hyaline) type was made rather than myoepithelial-cell-predominant pleomorphic adenoma.

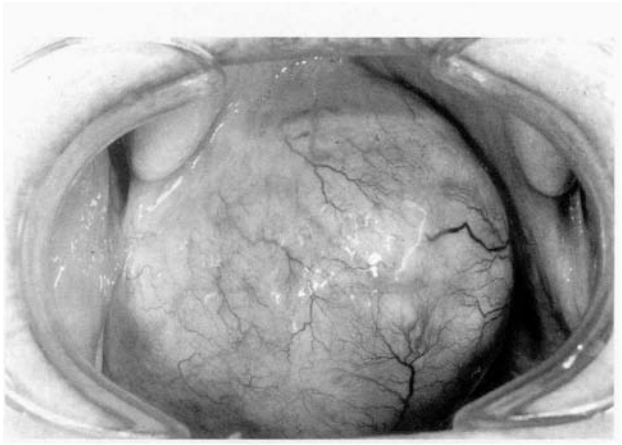

FIG. 1
Intraoral view of the tumour at presentation arising from the soft palate.

Discussion

The incidence of myoepithelioma is less than one per cent if only tumours composed solely of myoepithelial cells with no ductal differentiation are considered (Seifert and Sobin, 1991). However, if the definition is widened to include otherwise typical tumours with a limited amount of ductal differentiation the incidence increases (Simpson *et al.*, 1995). Myoepithelioma occurs over a wide age range with a median of 53 years and affects both sexes equally. It usually presents as a painless, slow-growing mass. The parotid gland is the most common site followed by the

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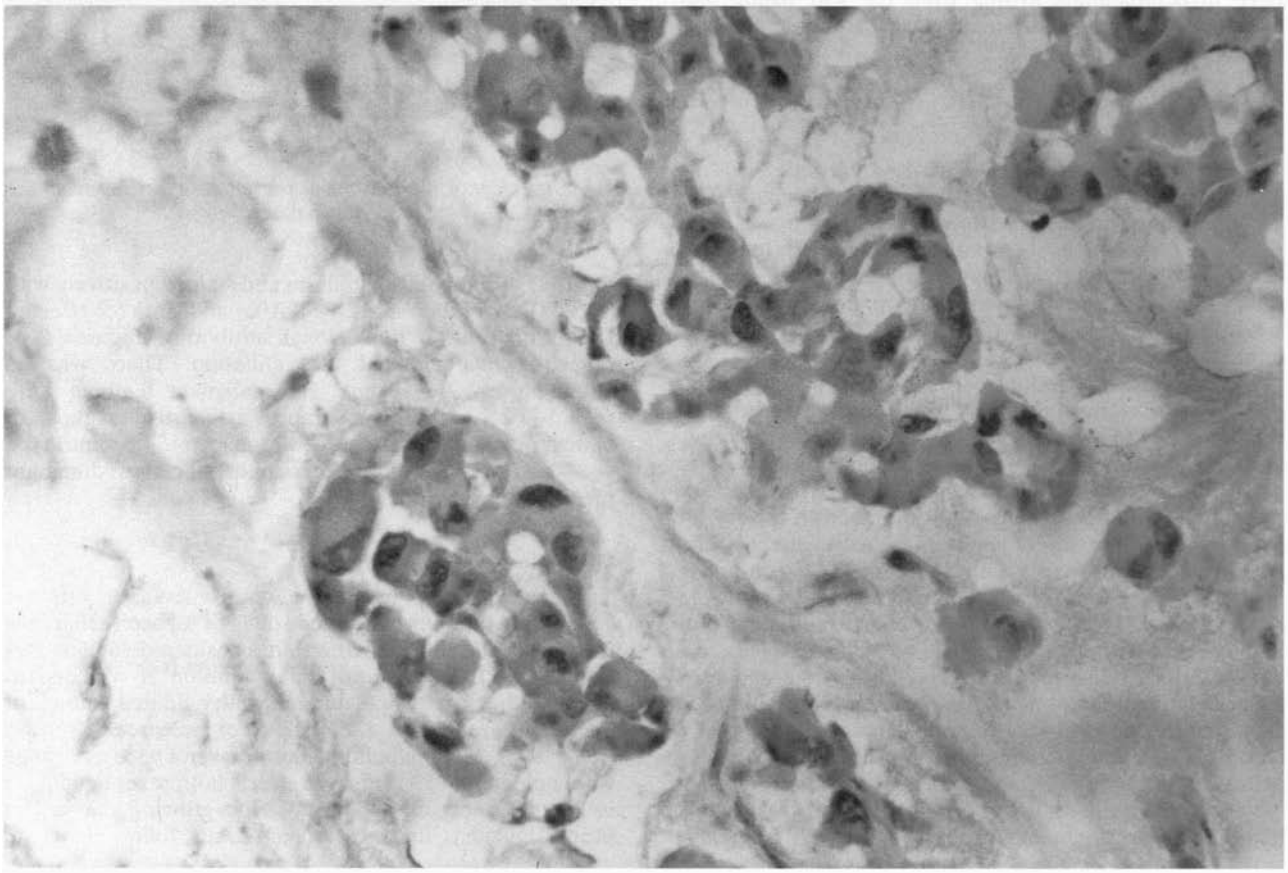

FIG. 2
Photomicrograph showing islands of epithelioid cells in a loose myxoid stroma. (H & E; ×200)

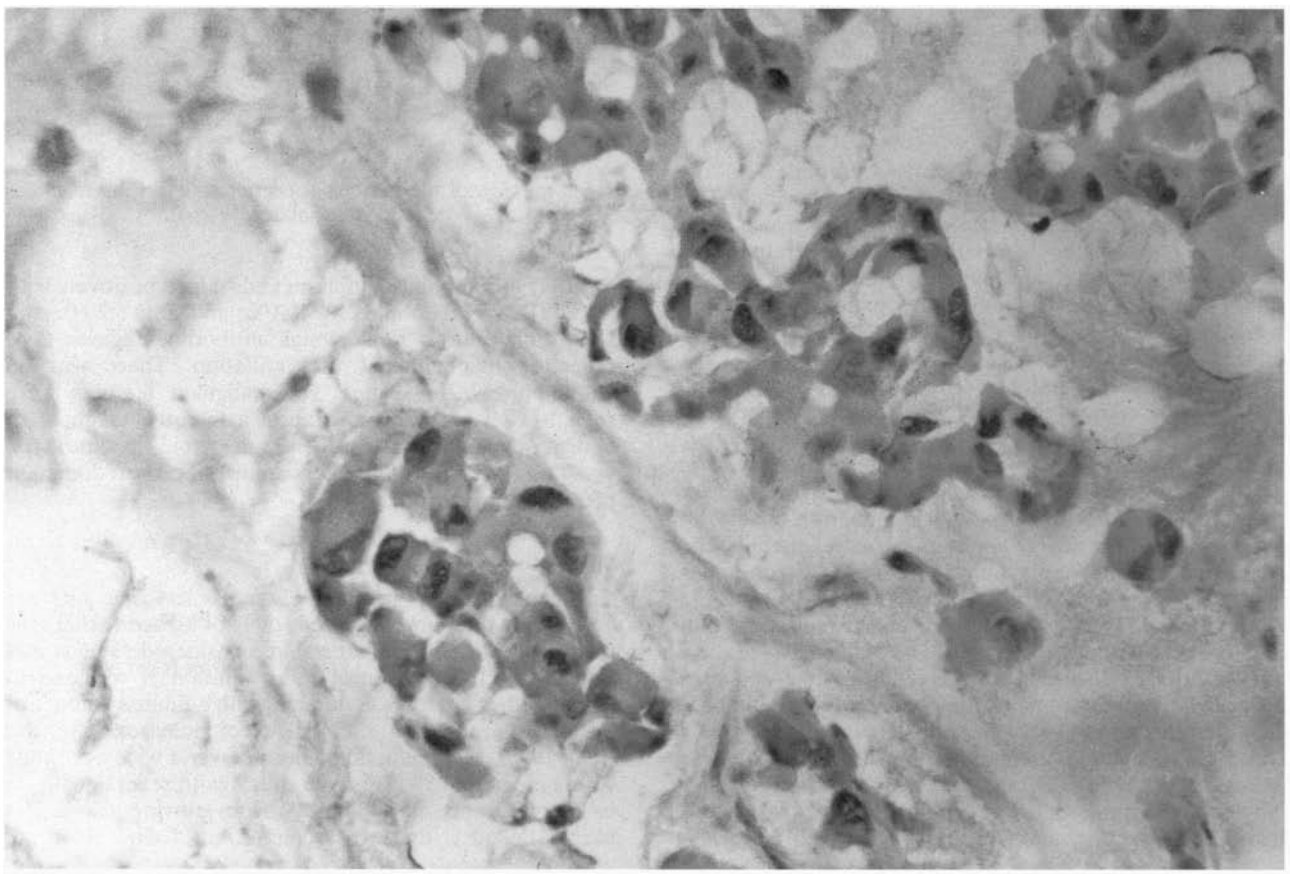

FIG. 3
Photomicrograph showing positive S100 staining (×400)

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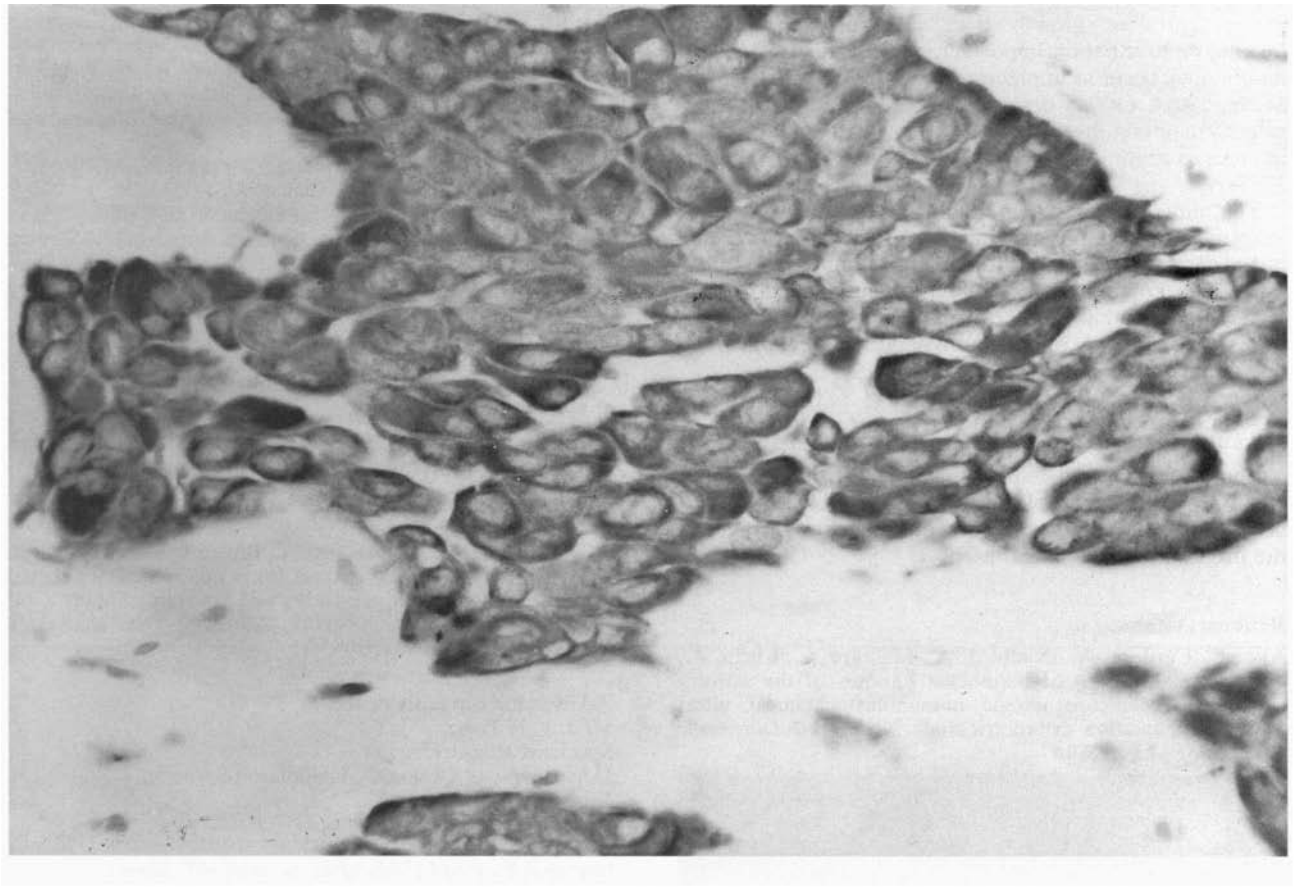

FIG. 4
Positive anti-cytokeratin (Cam 5.2) immunostaining of the neoplastic cells (×400)

minor glands of the palate (Sciubba and Brannon, 1982). Facial nerve palsy does not occur in parotid lesions and palatal lesions rarely ulcerate. Myoepithelial carcinoma is an uncommon locally invasive tumour which may metastasize (Barnes *et al.*, 1985).

Macroscopically, benign myoepitheliomas are well-circumscribed masses with a smooth, sometimes bosselated external appearance and a uniform white, tan or grey cut surface. They are surrounded by a thin fibrous capsule except when they arise in the palate, in which case the capsule may be partial as in the case reported or absent (Barnes *et al.*, 1985). The microscopic growth patterns encompass solid, myxoid and reticular forms or combinations of these. The cell types may be spindle-shaped, plasmacytoid or more rarely clear or epithelioid. In minor salivary gland tumours the plasmacytoid type predominates as in the case reported, whereas in parotid lesions the spindle cell variant is more common. However, neither architecture or cell type appears to carry prognostic significance.

Due to their infrequency and multiplicity of histopathology, myoepitheliomas present difficulties in diagnosis and classification. Accurate diagnosis is important to prevent misdiagnosis of cellular lesions as malignancy and confusion with other lesions, such as those of plasma cell, smooth muscle, or perineural origin, which it may superficially resemble (Sciubba and Brannon, 1982; Dardick, 1995).

The most useful immunohistochemical marker is S–100 protein and it would be difficult to make the diagnosis of myoepithelioma if this were absent. Although reaction with other antisera are more variable, cytokeratin antibodies such as AE1 and Cam 5.2 are useful in highlighting some cells throughout most myoepitheliomas, as in the case presented, and in distinguishing epithelial from non-epithelial neoplasms (Simpson *et al.*, 1995). The major differential diagnosis of myoepithelioma is from pleomorphic adenoma. Myoepitheliomas are composed completely, or almost completely, of myoepithelial cells, whereas in pleomorphic adenomas the numbers are variable, but may approach those in myoepithelioma. Pleomorphic adenomas contain plentiful ducts, whereas myoepitheliomas have few, if any. Indeed, many authors have widened the working definition of myoepitheliomas to neoplasms that have the histology and growth patterns of the myoepitheliomatous component of pleomorphic adenomas, but lack or have only limited (<5 to 10 per cent) differentiation of the ductal phase of this tumour (Dardick, 1995; Simpson *et al.*, 1995; Alos *et al.*, 1996). The range of stromal components is identical with pleomorphic adenomas, and myxoid and even chondroid areas can be seen in both. The only difference is quantitative in that the amount of stroma is likely to be much more in a pleomorphic adenoma. Considerable work has been done to distinguish between these tumours with immunohistochemical and ultrastructural techniques (Luna *et al.*, 1973; Dardick *et al.*, 1989; Takai *et al.*, 1995; Alos *et al.*, 1996). Nevertheless, the distinction is not generally considered to be a true biological one. A consensus is emerging that pleomorphic adenoma is in the middle of a spectrum of benign salivary tumours, including myoepitheliomas at one end and some (non-membranous) basal cell adenomas at the other (Simpson *et al.*, 1995).

The prognosis of myoepithelioma parallels that of pleomorphic adenoma and thus distinction between the two tumours can be said to have no clinical relevance (Barnes *et al.*, 1985; Simpson *et al.*, 1995). However, some authors consider myoepithelioma to be characterized by

more aggressive growth than pleomorphic adenoma making differentiation important, and malignant transformation may occur as in pleomorphic adenoma (Seifert and Sobin, 1991). Others deem that it is important academically to maintain the 'purity' of the myoepithelial category in order to acquire a more accurate database (Barnes *et al.*, 1985).

The aim of treatment should be complete surgical excision with a margin of normal uninvolved tissue being included within the surgical excision (Sciubba and Brannon, 1982; Pogrel, 1994). Although recurrence of benign myoepithelioma has been reported (Sciubba and Brannon, 1982), the prognosis of benign myoepithelioma appears to be good if surgical excision is complete.

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