

Schwannoma-like pleomorphic adenoma of the parotid

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Abstract

Pleomorphic adenoma is the most common benign salivary gland tumour. It can occur in any salivary gland, but is most frequently found in the parotid gland. Chondroid metaplasia is a frequent finding in pleomorphic adenoma. Other forms of metaplasia have been described, but are encountered less frequently. We report a rare case of unusual pleomorphic adenoma of the parotid gland with schwannoma-like feature.

Introduction

Salivary gland tumors account for about 3% of all head and neck neoplasia. The parotid gland is the main site for these tumors and about 95% of them are of epithelial origin.¹ Benign tumors represent 54% to 79% and 21% to 46% are malignant. The proportion of benign versus malignant tumor varies greatly by site. In the major salivary glands, such as the parotid and the submandibular gland, the majority of the tumors are benign contrary to the minor salivary glands, such as the sublingual and the floor of the mouth, where most of them are malignant.

Case Report

A 47-year-old woman was referred to us for a pain localized in the right retromandibular area, since about two months. Careful head and neck palpation revealed only a slight swelling in the right retromandibular region, without any well delineated palpable tumor. Head and neck MRI revealed a well defined tumor of 3.2 cm of size in the deep lobe of the right parotid gland with parapharyngeal exten-

sion, consistent with a pleomorphic adenoma (Figure 1). A right total parotidectomy with identification and preservation of the facial nerve was performed to remove the tumor appropriately. Macroscopic examination showed an intra-glandular, well delimited, white and firm, homogeneous nodule of 3.5cm of size. Histopathologically the tumor was completely circumscribed by a thin fibrous capsule. The major part of the tumor consisted of spindle cells with nuclear palisading resembling Verocay bodies (Figure 2B). At the periphery, some foci were composed of myxoid stroma with epithelial and myoepithelial-lined tubules (Figure 2A). Immunohistochemistry demonstrated the expression of p63 (Figure 3A) and CD10 (Figure 3B) in both tumour components and cytokeratin was positive in epithelial and myoepithelial cells. Histopathology diagnosis was a schwannoma-like pleomorphic adenoma of the parotid gland.

Discussion

Benign mixed tumour, also referred to as pleomorphic adenoma, is the most common benign salivary gland tumour, of which approximately 80% occur in the parotid gland, 10% in the submandibular glands and 10% in the minor salivary gland of the oral cavity. This tumour is most often observed in 30 to 60 years old patients and is more frequent in women than in men.² It is usually a solitary slow growing painless mass.

Pleomorphic adenomas are known for their morphologic and architectural variability. They share common features of epithelial, myoepithelial and mesenchymal components. The proportion of each of these elements can vary widely.¹ The stromal component of these tumours is most often predominantly myxoid with focal chondroid or fibrous aspects. Modified myoepithelial cells are thought to play an important role in the histopathological changes of the stroma. Sometimes squamous or osseous metaplasia is found in pleomorphic adenoma but is encountered less frequently. On a retrospective study on 83 pleomorphic adenomas over a period of five years, we found an overall prevalence of 5% of metaplasia (*unpublished data*).

Squamous cell metaplasia in pleomorphic adenoma is an uncommon and most often accidental finding.³⁻⁶ Squamous cell metaplasia has been reported as a potential pitfall in fine-needle aspiration cytology (FNAC) of pleomorphic adenoma, which can be confused with a squamous cell or mucoepidermoid carcinoma. Different hypotheses have been advanced for its etiopathology. Firstly, it may be a repair process following FNAC, as reported by Li *et al.*⁷ In a series of 10 benign parotid lesions following

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Key words: salivary gland, parotid, metaplasia, pleomorphic adenoma, schwannoma-like.

Conflict of interest: the authors report no conflicts of interest.

Received for publication: 20 April 2011

Accepted for publication: 23 August 2011

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Rare Tumors 2011; 3:e40
doi:10.4081/rt.2011.e40

FNAC, they found 8 cases of squamous cell metaplasia. In our case, we did not have any proof of a previous FNAC. A second hypothesis could be intra-lesional ischemia as it can be found in chronic sialadenitis or necrotizing sialo-metaplasia. Experimental data support this hypothesis, squamous metaplasia can be induced in rat salivary glands by arterial ligation.⁸ FNAC can also induce necrosis and then ischemia, which may support this last explanation.

Osseous metaplasia can be found in all salivary gland locations such as the parotid, submandibular and minor salivary glands. In some cases an important chondroid matrix was present with the formation of enchondral ossification at the borders.⁹ Others described the formation of osteoid indicating a possible direct



Figure 1. Magnetic resonance imaging of the patient with a radio opac lesion in the right parotid gland.

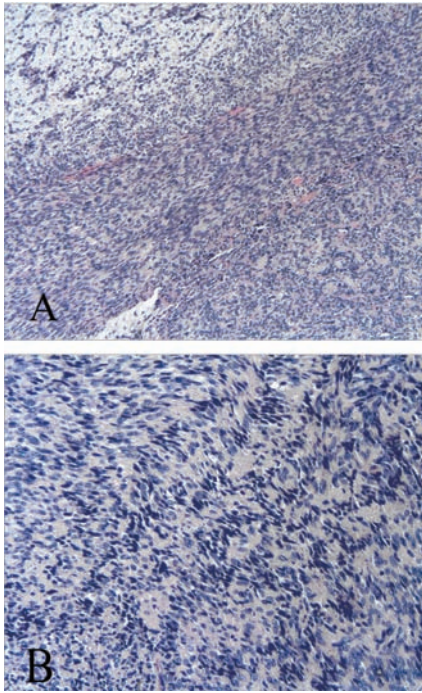


Figure 2. (A) Low magnification of schwannoma-like pleomorphic adenoma with both components (hematoxylin-eosin, magnification 100x); (B) Pathologic features of the schwannoma-like pleomorphic adenoma with palisading spindle-shaped cells (hematoxylin-eosin, magnification 200x).

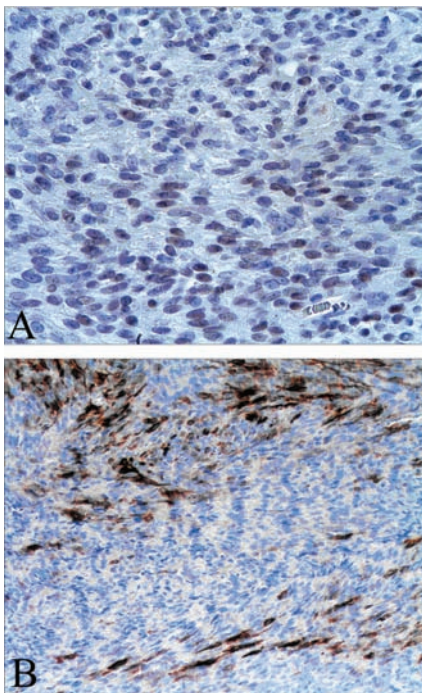


Figure 3. Immunohistochemical staining with (A) p63 and (B) CD10 (magnification for A: 400x, and B: 200x).

Table 1. Schwannoma-like features in pleomorphic adenoma. Review of the literature.

	Age, sex	Localisation
Takeda <i>et al.</i> 1999 ¹⁸	62-year-old man	Parotid
	48-year-old woman	Hard palate
Kajor <i>et al.</i> 2006 ¹⁶	75-year-old woman	Parotid
Merino <i>et al.</i> 1977 ¹⁷	74-years-old woman	Parotid
	39-years-old woman	Parotid
Present case	47-years-old woman	Parotid

differentiation from myoepithelial cells^{10,11} Osseous metaplasia can also be found in carcinoma ex pleomorphic adenoma^{12,13} and mixed tumour of the skin.^{14,15}

To our knowledge only five cases of schwannoma-like pleomorphic adenoma were reported in the English literature.¹⁶⁻¹⁸ Previous cases included four women and one man, aged from 39 to 75 years (Table 1). The majority of the lesions, as ours, were located in the parotid gland with one exception in the hard palate. All cases including this one had palisading areas of spindle-shaped cells in an otherwise classical pleomorphic adenoma.

Spindle cell tumours are rare in the salivary glands, representing from 1.9% to 5% of parotid neoplasms.^{1,19,20} Differential diagnoses of benign spindle cell tumours in salivary glands include neurogenic tumours: schwannoma, composed of areas with Verocay bodies and neurofibroma, as well as smooth muscle proliferations: leiomyoma.

Immunohistochemistry (IHC) may solve this dilemma. Leiomyomas are negative for cytokeratin positive for smooth muscle markers including alpha-smooth muscle actin, desmin and caldesmon, whereas benign neurogenic tumours, also negative for cytokeratine, express neurogenic marker (S100-Protein, CD57 and neurofilament).

Schwannoma-like pleomorphic adenomas are composed of modified myoepithelial cells expressing p63, CD10 and cytokeratin by IHC.¹⁶ Merino *et al.* demonstrated the presence of desmosomes in these spindle-shaped cells by electron microscopy confirming a myoepithelial origin.¹⁷ The pathogenesis of this phenomenon could be a result of plasticity of myoepithelial cells.

Fine needle aspiration cytology is an important diagnostic procedure used to evaluate salivary gland lesions and to help in their pre-operative management. Typically, FNAC of pleomorphic adenoma shows a combination of cohesive epithelial cells in a pale myxoid matrix. When the cellularity is abundant and no matrix is identified this diagnosis is challenging. In the presence of spindle-shaped cells immunohistochemical analysis may be used to identify the myoepithelial phenotype.

Pleomorphic adenoma with schwannoma-like feature is a rare variant that could be confused

with a schwannoma or a leiomyoma, but immunohistochemical study is helpful to differentiate these entities.

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