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-
differentiation from myoepithelial cells\textsuperscript{10,11} Osseous metaplasia can also be found in carcinoma ex pleomorphic adenoma\textsuperscript{12,13} and mixed tumour of the skin.\textsuperscript{14,15} To our knowledge only five cases of schwannoma-like pleomorphic adenoma were reported in the English literature.\textsuperscript{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.\textsuperscript{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).

Shwannoma-like pleomorphic adenomas are composed of modified myoepithelial cells expressing p63, CD10 and cytokeratin by IHC.\textsuperscript{16} Merino et al.\textsuperscript{17} demonstrated the presence of desmosomes in these spindle-shaped cells by electron microscopy confirming a myoepithelial origin.\textsuperscript{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 preoperative 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.

\begin{table}
\centering
\caption{Schwannoma-like features in pleomorphic adenoma. Review of the literature.}
\begin{tabular}{|c|c|c|}
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& Age, sex & Localisation \\
\hline
Takeda et al. 1999\textsuperscript{18} & 62-year-old man & Parotid \\
 & 48-year-old woman & Hard palate \\
Kajor et al. 2006\textsuperscript{14} & 75-year-old woman & Parotid \\
Merino et al. 1977\textsuperscript{17} & 74-years-old woman & Parotid \\
 & 39-years-old woman & Parotid \\
Present case & 47-years-old woman & Parotid \\
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References

11. Nakano K, Watanabe T, Shimizu T, Kawakami T. Immunohistochemical char-