Case Report

**Stromal Bone Formation in Pleomorphic Adenoma of
Minor Salivary Gland Origin**

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Abstract

We report an unusual case of pleomorphic adenoma with multifocal stromal bone formation, occurring in the upper lip of a 44-year-old Japanese woman. Islands of fibrous bone were in direct contact with stromal fibrocytes and parenchymal epithelial cells. A review of the literature revealed that stromal bone formation in pleomorphic adenoma of minor salivary gland origin is exceedingly rare.

Introduction

Pleomorphic adenoma is the most common tumor of both major and minor salivary glands, and histologically it is well known that pleomorphic adenoma consists of a capsule of varying thickness and completeness, and epithelial and modified myoepithelial elements arranged in a variety of configurations, intermingled with a stroma containing of fibrous, mucoid, hyalinized, myxoid, or myxochondroid tissues. In addition, there may be formation of true chondral tissue, but bone formation is rare. Bone formation has been seen almost exclusively in pleomorphic adenomas of parotid gland origin^[1-3], and is very rare in those of intra-oral minor salivary gland origin.

Here we report a case of pleomorphic adenoma of the upper lip and showing stromal bone formation.

Case Report

The patient was a 44-year-old Japanese woman. She had first noted an intra-oral, nodular swelling in her upper lip about 5 years previously, but was not concerned about it. She consulted a nearby dentist because the swelling had gradually increased in size, and was referred to our dental hospital. Clinical examination showed a well circumscribed, elastic-hard and mobile mass, measuring 12 mm in diameter, located in the left side of the upper lip. The overlying mucosa was normal in color. A clinical diagnosis of benign salivary gland tumor was made, and surgical resection of the lesion was performed. The postoperative course was uneventful.

Pathologic Findings

Histological sections revealed a well demarcated typical pleomorphic adenoma showing variously arranged neoplastic epithelial cells intermingled with a fibrous, myxoid, myxochondroid, and hyalinized stroma. In addition, several islands of calcified tissue were found (Fig.1), and these were regarded as fibrous bone, because they contained entrapped cells and Sharpey's fibers (Fig.2). Both stromal fibrous cells and parenchymal epithelial cells were closely associated with these islands (Fig.2). No true chondral tissue was formed around or near the fibrous bone.

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Discussion

Pleomorphic adenoma arising in the minor salivary glands tends to be more cellular with less prominent hyalinized, myxoid, myxochondroid and chondral components than that seen in parotis tumors^[4, 5]. In addition, stromal ossification in pleomorphic adenoma is a rare finding and, when present, is most commonly seen in parotid lesion^[1-3]. There have been very few descriptions of stromal ossification in pleomorphic adenoma arising in minor salivary glands, such as that presented here.

In previously described pleomorphic adenoma with bone formation^[1-3], the bone was formed within areas of myxochondroid or chondral tissue, that is, by a process of endochondral ossification. The bone tissue in the present case was surrounded by stromal fibrocytes and parenchymal epithelial cells, but no myxochondroid or chondral tissue was evident around or near the bone.

Recent pathological studies of pleomorphic adenoma have indicated that modified or neoplastic myoepithelial cells play an important role in the expression of morphologic diversity, especially in myxochondroid and chondral tissue formation. Yagn et al.^[6] examined the presence of BMP (bone morphogenetic protein) in pleomorphic adenoma, and considered that the simultaneous presence of glycosaminoglycans as a matrix substance and S-100 protein for calcium signalling are associated with BMP-mediated cellular activity of neoplastic myoepithelial cells during the formation of myxochondroid and chondral tissues in pleomorphic adenoma. However, the question arises as to why bone formation is rarely seen in pleomorphic adenoma, in contrast to frequent formation of myxochondroid and chondral tissues.

References

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Fig.1 In addition to the characteristic histologic features of pleomorphic adenoma, islands of calcified tissue are seen (HE, original magnification $\times 40$).

Fig.2 High-magnification view of two calcific islands shown in Fig.1 (a and b, HE, original magnification $\times 300$). These calcified tissues are regarded as fibrous bone, because they contain entrapped cells and Sharpey's fibers (a). Both stromal fibrous cells and parenchymal epithelial cells are closely associated with these islands of bone (a and b), and no myxochondroid or chondral tissue is evident around or near the islands.