Ectopic Pleomorphic Adenoma: A Case Report

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The most common benign salivary gland tumor is pleomorphic adenoma. It affects major and minor salivary glands. Minor salivary gland tumors constitute <20% of all salivary gland neoplasms. Most minor salivary gland tumors occur on the palate. We present the case of a patient with pleomorphic adenoma that developed at a rare intraoral location. This patient presented to the Department of Oral and Maxillofacial Surgery of Seoul National University Dental Hospital.

Key words: Adenoma, Pleomorphic adenoma, Ectopic, Rare location

I. INTRODUCTION

1. Patient

A 70-year-old woman presented to Seoul National University Dental Hospital with a painless swelling in the left pterygomaxillary area. The patient had a history of medically controlled hypertension, with no other significant medical conditions.

On clinical examination, a 2.5 × 1.5 × 1-cm dome-shaped mass was noted in the left pterygomaxillary region. The covering mucosa was smooth; on palpation, the lesion was rubber-like and non-tender and had some mobility over the underlying tissues.

Magnetic resonance imaging (MRI) revealed a heterogeneous lesion with hyperintense T2 signals. The buccinator, lateral pterygoid, and temporalis muscles surrounded the lesion. The lesion was capsulated and had clear margins (Fig. 1). Based on the disease history and clinical findings, fibroma and a minor salivary gland tumor were in the top of differential diagnoses list.
2. Surgery

Under general anesthesia wide excision of the lesion with primary closure was performed. The excised specimen (size: 2.5 × 2.5 cm) was sent to histopathological lab for examination (Fig. 2, 3). Surgical removal of the entire mass was performed using the intraoral approach.

Fig. 1, MRI: A, coronal view (T1); B, axial view (T2)

Fig. 2, Wide excision of the lesion with primary closure was performed. The excised specimen (2.5 × 2.5cm) was sent for histopathological examination. The tumor lies in the left pterygomaxillary region.

Fig. 3, The mass after excision (size, 1.5 × 2.5 cm)
3. Histopathological examination

Histopathological examination revealed a well-circumscribed, non-encapsulated lesion with biphasic growth of epithelial and myoepithelial cells, forming ductal structures and pushing on the peripheral lymphoid structure. The abluminal myoepithelial cells showed plasmacytoid and spindle-cell architecture. Large cystic dilations surrounded by a proliferation of ductal epithelial cells were also noted. The proliferated myoepithelial cells merged into areas of densely hyalinized stroma at some places. The diagnosis was pleomorphic adenoma of the minor salivary glands (Fig. 4).

4. Follow-up

The patient was followed up for 9 months. Uneventful healing of the surgical site was observed and no evidence of recurrence.

5. Comments

In this case the tumor developed at a rare intraoral location. The tumor lies in the left pterygomaxillary region.

II. DISCUSSION

Minor salivary glands are mucous glands scattered across the upper respiratory tract. Most minor salivary glands are found in the palate, whereas others are distributed at the inner surface of the lips; around the opening of the parotid duct; in the submucosa of the uvula, mucous membrane of the cheek, floor of the mouth, palatoglossal folds, and superior pole of the tonsils (Weber's glands); on the inferior surface of the tongue; near the frenulum of the tongue; within the palatine tonsil; and in other uncommon places, such as the pterygomaxillary area.

Pleomorphic adenomas are benign heterogeneous (myoepithelial and epithelial) tumors of salivary gland origin and are considered a benign mixed tumor. Pleomorphic adenomas arising in the pterygomaxillary area are rare and the most common site is the palate. It usually appears as a firm, rubbery, submucosal mass. In the present patient, based on clinical examination alone, we placed pleomorphic adenoma at the top of our differential diagnosis list.

Pleomorphic adenoma is known to have some degree of recurrence either due to spillage, inadequate removal, or enucleation at the time of operation, but there are no reports of distant metastasis. In the literature, a recurrence rate of 2%-44% has been reported. Wide local excision is

Fig. 4. Hematoxylin and eosin staining Microscopic examination of the excised mass. A: Proliferation of plasmacytoid myoepithelial cells with intervening areas of hyalinized stroma. B: Well-circumscribed, non-encapsulated lesion with biphasic growth of epithelial and myoepithelial cells forming ductal structures. C: Large cystic dilation surrounded by a proliferation of ductal epithelial cells.
the treatment of choice for pleomorphic adenoma with the removal of the periosteum and/or bone if they are involved.2,4,7

Rarely, malignant lesions develop within this tumor (e.g.; carcinoma ex pleomorphic adenoma). The most common clinical features for identifying malignancy include an infiltrative growth pattern, vascular permeation, perineural invasion, and marked cytologic atypia with abnormal mitotic figures.12-14

REFERENCES