Pleomorphic Adenoma of the Nasolabial Region

Nazolabial Bölgenin Pleomorfik Adenomu

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Özet

Anahtar Kelimeler
Pleomorfik Adenom; Tükürük Bezi Tümörleri; Baş Boyun Tümörleri

Abstract
Pleomorphic adenoma often occurs in the major salivary glands, most commonly on the parotid gland. Although unusual cases of pleomorphic adenoma of the nasal cavity, nasopharynx, trachea, and oesophagus have been reported, to the best of our knowledge the present case is the first reported case in the nasolabial region. In this article, clinical presentation, surgical findings, histopathological features, and therapeutic challenges of this rare lesion are discussed.

Keywords
Pleomorphic Adenoma; Salivary Gland Tumors; Head And Neck Neoplasms
Introduction

Pleomorphic adenoma (PA), the most common type of benign salivary gland tumor, histopathologically consists of epithelial and stromal components. PAs usually appear as solitary, painless, asymptomatic, and slow-growing lesions. Though it is classified as a benign tumor, PA may undergo malignant transformation into carcinoma ex-pleomorphic adenoma, a risk that is believed to increase with time [1]. Also it can recur after resection; distant metastases have been reported after long time intervals [2, 3].

PA often occurs in the major salivary glands, most commonly on the parotid gland [4]. The palate is the most affected site among the extra-major salivary glands [3, 5]. The nasolabial region (NLR) is an unexpected involvement site for these tumors. Our study represents a case of a PA of the NLR excised by an intraoral (sublabial) approach. In this article, clinical presentation, surgical findings, histopathological features, and treatment of this rare lesion are discussed.

Case Report

A 21-year-old female presented with a complaint of a painless lump, having gradually increased in size, in the right NLR. She had first noted the lesion about nine months previously. The patient reported no trauma. On palpation the lesion was elastic, well-circumscribed, movable, painless, and 1.5 to 2 cm in diameter. Bimanual palpation of the upper lip was normal. The overlying skin and labial mucosa was not fixed. She was found to have a mild, barely noticeable asymmetry of the face. No other abnormality was noted on otolaryngologic examination. She had no significant previous medical history. The mass was totally excised with an intraoral (sublabial) approach. An incision was made in the oral cavity mucosa along the gingivolabial sulcus under local anesthesia. Dissection exposed a smooth, well-circumscribed, encapsulated solid mass that was approximately 2 cm in diameter (Figure 1). No adhesion to the underlying bone was encountered. The lesion was excised without difficulty from the surrounding tissue. The specimen obtained was a gray-white lesion, fully encapsulated with normal margin (Figure 2). The wound was closed by primary intention. The specimen was fixed in 10% buffered formalin and submitted for histopathologic diagnosis. The postoperative course was uneventful. No additional treatment was required. Subsequent follow-up after three years showed no signs of recurrence. Histologically, the material showed a well encapsulated epithelial salivary gland tumor. The lesion had a thin fibrous capsule and consisted of epithelial and fibromyxoid stromal components. A diagnosis of PA was made based on the characteristic histological pattern (Figure 3).

Discussion

Our study presents a PA of the NLR causing a painless mass as the primary symptom. Although unusual cases of PA of the nasopharynx, orbital area, trachea, nasal septum, and external auditory canal have been reported, to the best of our knowledge the present case is the first to be reported in the NLR [5]. Based on the clinical appearance, the preliminary diagnosis in our case was a nasolabial cyst, the most likely tumor found in this region. Nasolabial cysts are frequently-asymptomatic developmental masses occurring inferior to the nasal alar region.

A differential diagnosis of foreign body granuloma, lipoma, neurofibroma, myoepithelioma, or other benign mesenchymal tumors was also considered. A salivary gland tumor was not considered because of its deep location. A fine needle aspiration biopsy may be performed, but, due to the probable benign nature of the lesion, an excisional biopsy was planned. In our case, PA may have arisen from minor salivary glands of the nasal alar region and expanded to the NLR, possibly explaining the unusual location of the PA.
Nasolabial Pleomorphic Adenoma

Tumors arising from the minor salivary glands are uncommon clinical entities. Limited information is available in the literature on the management of PAs located in the extra-major salivary glands. In general, it is suggested that the lesion be removed together with the surrounding normal tissue for PA treatment [3-5]. However, wide excision of extra-major salivary gland PA may not always be possible due to aesthetic concerns, therefore resulting in a close surgical margin. In their study, Kuo et al. retrospectively investigated 37 patients who had undergone primary surgery for extra-major salivary gland pleomorphic adenoma of the head and neck region and concluded that although adequate surgical margins were not always achieved during operation, the rate of recurrence was relatively low in the 4.5-year average follow-up period [5]. In the present case, total excision by a narrow margin via a sublabial approach was performed. The lesion was easily released from the surrounding tissue with clinically normal margins and the mass appeared to be fully encapsulated. Follow-up three years after surgery showed smooth healing of the wound with no evidence of recurrence. But considering that the capsule may have been infiltrated by extension of the tumor, the patient will be followed up for at least 10 years due to the risk of recurrence.

In conclusion, PA should be considered in the differential diagnosis of tumors located in the NLR. Wide surgical excision may not be always possible to achieve with adequate surgical margins because of aesthetic concerns in this region. Special surgical attention must be paid not to rupture the capsule during the operation. Long-term follow-up of patients is necessary due to the risk of recurrence.

Competing interests
The authors declare that they have no competing interests.

References

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