

Canalicular adenoma of the minor salivary gland in the upper lip: case report

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ABSTRACT:

Canalicular adenoma is a rare benign tumor of the minor salivary glands that is the most prevalent in older adults (mean age, 60 years). The upper lip is the most commonly affected site, followed by the buccal mucosa and palate. It presents as a nodular lesion without a tendency for recurrence. Here, we describe a case of canalicular adenoma presenting as a nodular, painful mass in the upper lip of a 62-year-old man with a history of trauma. Fine needle aspiration biopsy yielded inconclusive results but excluded malignancy. The lesion was enucleated, and a definitive diagnosis of canalicular adenoma was established by histological and immunohistochemical analyses. No signs of recurrence were noted at the 8-month follow-up examination. The case emphasizes the importance of differential diagnosis of nodular lesions in the oral mucosa for appropriate treatment and histological analysis for definitive diagnosis.

Keywords: adenoma, lip, salivary gland neoplasm.

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INTRODUCTION

Canalicular adenoma (CA) is an unusual benign tumor of the minor salivary glands¹⁻³ that arises from ductal tissue^{4,5}. It appears as a flat mobile nodule that is slow growing and asymptomatic^{6,7}. It occurs more often in adults, and the upper lip is the most affected site (70% of the cases), followed by the buccal mucosa and palate^{3-5,8}. The occurrence of multifocal tumors of minor salivary glands is rare; however, this phenomenon has been reported in the literature, especially in the upper lip⁸⁻¹⁰. The treatment of choice is enucleation, and recurrence is rare. Here, we describe a case of canalicular adenoma in the upper lip.

CASE REPORT

A 62-year-old man attended the Service of Oral and Maxillofacial Surgery at Erasto Gaertner Hospital complaining of a “ball in his mouth.” He reported trauma to the area at the age of 6 years, and the lesion progressed to a mass in his upper lip. Although it was asymptomatic, it was painful on palpation. Physical examination revealed a circumscribed, mobile, and hard submucosal nodular mass with normal color on the left side of the upper lip (Figure 1). Cervical lymph nodes were not palpable. Therefore, pleomorphic adenoma and mucocele were considered in the differential diagnosis.



Figure 1. Clinical examination showed a nodular mass in the upper lip.

The patient underwent fine needle aspiration biopsy (FNAB), which verified the presence of a yellowish liquid of viscous consistency, compatible with glandular lesions (Figure 2). Although the biopsy was inconclusive, it excluded malignancy, and enucleation was performed in an outpatient setting. The lesion was well defined and encapsulated (Figure 3). Histopatho-

logy showed a lesion characterized by proliferation of columnar and cuboidal epithelial cells forming ductal structures and sometimes anastomosis in loosely arranged connective tissue. The characteristics of the lesion and the fibrous capsule suggested a long period of evolution. Immunohistochemical analysis with antibodies to cytokeratin (CK) AE1/AE3 and vimentin confirmed the diagnosis of CA (Figure 4). During the 8-month follow-up examination, the patient showed no signs of recurrence, and had a normal lip and facial appearance.

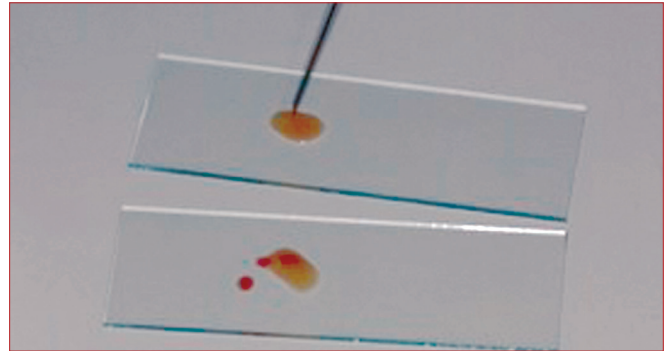


Figure 2. FNAB revealed a yellowish liquid of viscous consistency.

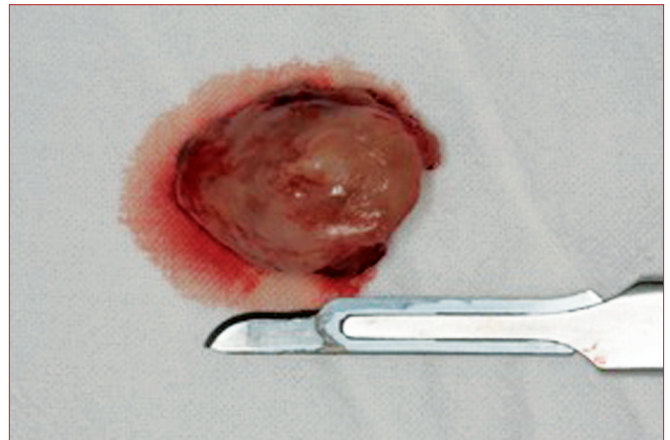


Figure 3. Enucleation revealed an encapsulated lesion.

DISCUSSION

CA and basal cell adenoma (BCA) are often described as monomorphic adenoma to distinguish them from pleomorphic adenoma. However, recently, the World Health Organization has classified CA and BCA as separate entities, and the term “monomorphic adenoma” is no longer used. The use of immunohistochemical techniques has provided increasing evidence that CA is derived from ductal luminal cells, whereas BCA is most likely derived from salivary gland parenchyma³. Sousa et al.⁴, in a comparative analysis of CA and BCA, reported that BCA arises from the intercalated duct, whereas CA originates from the excretory duct of minor salivary glands.

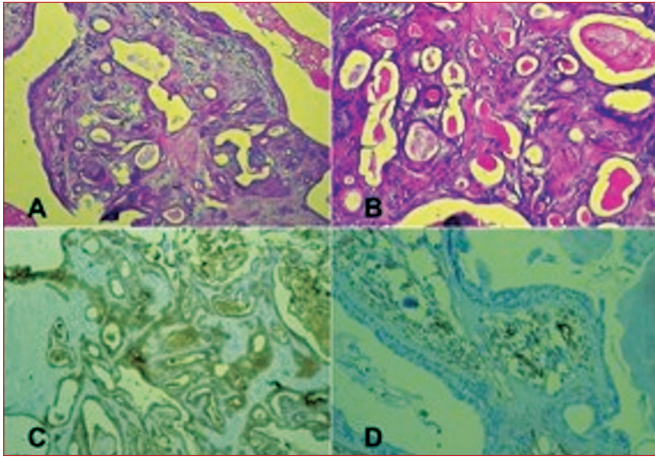


Figure 4. Histological features. A and B. Rows of columnar epithelial cells with beaded appearance and forming duct-like structures in fibrous stroma. C and D. Immunohistochemical staining with CK AE1/AE3 and vimentin, respectively.

Our case presented as an encapsulated lesion with proliferation of columnar epithelial cells featuring islands and cords as well as ductal structures. Immunohistochemically, CA strongly expresses CK7 and CK13, and most cells are negative for vimentin⁴. We observed a similar immunohistochemical pattern, showing CK AE1/AE3 positivity. The expression of Ki67, a marker of cell proliferation, was observed in a few tumor cells, suggesting the lesion had a long evolution time, consistent with the history of injury.

CA is often characterized clinically by a slow-expanding, well-circumscribed, and painless mass that can be firm, soft, or fluctuant on palpation¹¹. The tumor can present ulceration, hemorrhage, mucus extravasation, inflammation, and pigmentation. The clinical presentation and patient age in our case are in agreement with previous reports^{6,10}: the patient was over 60 years of age and the lesion was present on the upper lip. However, the patient reported a long period of evolution and limited growth.

The most common presentation of epithelial tumors of minor salivary glands is a single nodule. CA, however, can occur as multifocal lesions in the upper lip^{8,10}. According to some authors^{1,8}, the treatment of choice for both presentations is complete enucleation. Multifocal lesions have higher rates of recurrence. There are no reports of malignant transformation, and benign behavior is the usual presentation of CA^{8,9}. After 8 months, our patient had no clinical signs of recurrence.

Salivary gland tumors have varied microscopic features. CA is excluded from the classification of mixed tumors. Investigations of this lesion revealed the complete absence of changes in mesenchymal or chondromyxoid stroma and presentation as an encapsulated tumor⁶. Nelson et al.⁶ reported the importance of establishing the initial diagnosis of benign tumors of the

minor salivary glands, thus excluding malignant lesions such as adenoid basal cell carcinoma, papillary adenocarcinoma, and papillary cystadenocarcinoma. The provisional diagnosis should be established on the basis of the clinical features of the lesion, time course, presence of lymphadenopathy, and absence of malignancy by FNAB. After confirming these characteristics, the treatment of choice should be enucleation.

CONCLUSION

The present case emphasizes the importance of differential diagnosis of nodular lesions in the oral mucosa, through clinical features, time of evolution, and FNAB, which excludes a malignant tumor of salivary glands. Enucleation is the most appropriate treatment for CA. The definitive diagnosis of CA is established by microscopic and immunohistochemical analyses. A second surgical intervention is usually not required, because recurrence is rare.

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