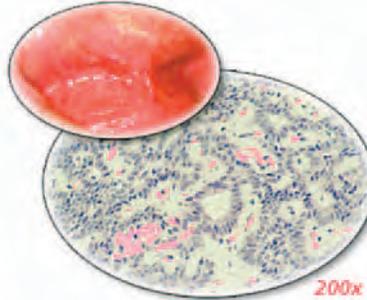


Canalicular Adenoma Presenting as an Asymptomatic Swelling of the Upper Lip: A Case Report

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Abstract

Aim: The purpose of this report is to present the clinical and histological features of a canalicular adenoma (CA) occurring in the upper lip and vestibular fornix of a 62-year-old woman.

Background: CA is an uncommon benign salivary gland tumor occurring almost exclusively in the intraoral glands. This tumor has often been referred to as a variant of the basal cell adenoma. However, the World Health Organization's latest histological classification of salivary gland tumors recognizes it as a separate entity under the broader heading of monomorphic adenoma, which is not related to any of the subtypes of basal cell adenomas.

Case Report: A 62-year-old woman with a chief complaint of an extraoral swelling in the upper lip. The evolution of the lesion was not known by the patient who recognized it when she touched her upper lip and found "an acne" on it. The patient wore complete dentures since age 17 and was not aware of any pain or tenderness in the area. Extra and intraoral examination revealed a mobile nodular lesion located in the right aspect of the upper lip near the nose and in the vestibular fornix between the lateral incisor and canine. Microscopic examination confirmed the final diagnosis of CA. The patient is currently free of disease 54 months after surgical excision of the tumor.

Summary: Local excision of symptomatic nodules seems to be sufficient to manage patients with multifocal CA.¹⁰ But, unlike other benign tumors, CA may need a longer follow-up due to its tendency towards multifocal occurrence and late recurrence.^{3,7}

Keywords: Canalicular adenoma, CA, salivary gland tumors, monomorphic adenoma

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Background

Canalicular adenoma (CA) is an uncommon benign salivary gland tumor occurring almost exclusively in intraoral glands and accounts for 1% of all salivary gland neoplasms.¹⁻⁵ In a series of 213 cases of intraoral minor salivary gland neoplasms, Yih⁶ found 25 cases of CA which was the second most common benign gland tumor found.

Historically, this tumor has often been referred to as a variant of the basal cell adenoma. However, the World Health Organization's latest histological classification of salivary gland tumors recognizes it as a separate entity under the broader heading of monomorphic adenoma which is not related to any of the subtypes of basal cell adenomas.^{3, 7-9}

CA usually arises in the minor salivary glands with the upper lip being the most common site accounting for 70-90% of all tumors followed by the buccal mucosa and palate.¹⁻¹³ Involvement of parotid glands is extremely rare.^{7,8,14} This tumor occurs mainly in elderly people after 60 years of age¹ and shows a slight female predilection.^{2-6,8,11-13}

CA is an older term than either basal cell adenoma or monomorphic adenoma, and the origin of the term is unclear.¹⁵ According to Triantafyllou et al.¹⁶ salivary parenchyma cells proliferate and activate dormant matrix genes. Proliferating cells fail to develop anatomical coupling with nerves and synthesize glycosaminoglycan, a primitive, or immature glycoprotein. Eventually a tumor with embryonal features is formed.

CA clinically is usually the color of the overlying mucosa and manifests as a painless, slowly growing encapsulated submucosal mass which may be firm or slightly fluctuant on palpation.^{1-4,7,13,15} It is typically smaller than 2 cm.^{2,4} The overlying mucosa is rarely ulcerated.^{4,11,14}

CA is not likely to be diagnosed preoperatively using fine needle aspiration or on clinical inspection.⁷ Clinically, it can be confused with

mucoceles, epidermal inclusion cyst, nasolabial cyst, lipoma, benign peripheral nerve lesion, benign fibrous lesions, mucous retention cyst, and pleomorphic adenoma.^{1,3,4} Adenoid cystic carcinoma and basal cell adenoma must be included in the differential diagnosis.^{2,13}

Case Report

Diagnosis

In August, 2001 a 62-year-old woman was referred to the Dental Clinic at Pontifícia Universidade Católica do Paraná in Curitiba, PR, Brazil with a chief complaint of an extraoral swelling in her upper lip. She did not know the exact evolution of the lesion and was only aware of it when she touched her upper lip and found "an acne" on it. The patient had been wearing complete dentures since she was 17 years old and was not aware of any pain or tenderness in the area. Extraoral and intraoral examination revealed a firm, mobile nodular lesion measuring 0.3 to 0.4 cm located near the nose in the right portion of the upper lip and extending to the vestibular fornix between lateral incisor and canine (Figure 1).

The overlying mucosa appeared slightly reddened. There was no cervical lymphadenopathy and no alteration of the nasolabial sulcus. The initial diagnostic hypothesis was mucocele, followed by CA and nasolabial



Figure 1. Discrete intraoral swelling in the upper lip of a 62-year-old edentulous woman.

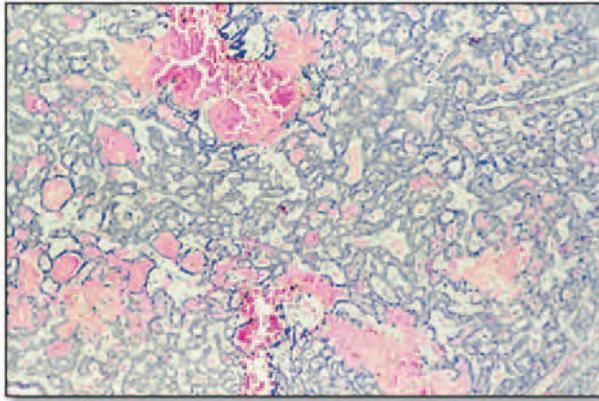


Figure 2. Typical histology of a CA composed of single layers of cubo-columnar cells in cords, forming “canal-like” structures (Hematoxylin-eosin stain; original magnification X 100).

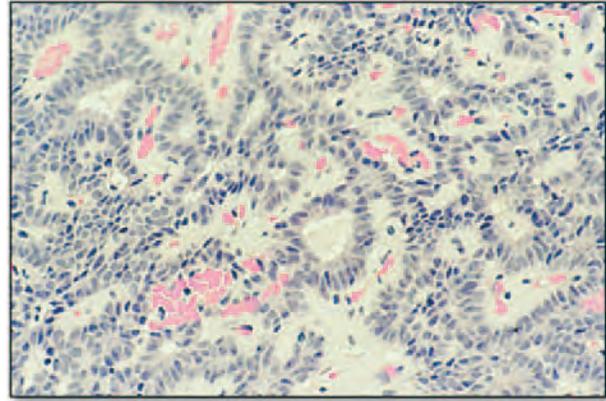


Figure 3. Photomicrography showing single and double layers of cubo-columnar cells in a fibrous stroma (Hematoxylin-eosin stain; original magnification X 200).

cyst. A mucocele was the first consideration due to its clinical characteristics and high frequency in the lips. CA was added to the list of possibilities since the most common site for this tumor is also in the upper lip. The nasolabial cyst was also considered because of the anatomic location and the age of the patient despite the apparent normality of nasolabial sulcus because it could have been an incipient lesion.

An excisional intraoral biopsy was performed under local anesthesia. Microscopic examination revealed a tumor encapsulated and well circumscribed. The epithelium was well-demarcated. The tumor was composed of single and double layers of cubo-columnar cells in cords, forming “canal-like” structures (Figures 2 and 3).

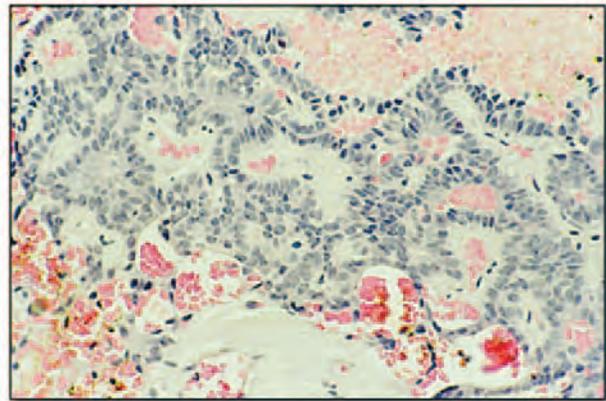


Figure 4. Glandular epithelial elements arranged in anastomosing monolayered duct-like pattern (Hematoxylin-eosin stain; original magnification X 200).

Uniform epithelial cells were arranged in an anastomosing monolayered duct-like pattern. The stroma consisted of loose fibrovascular tissue with dilated blood vessels (Figure 4). There was no evidence of satellite islands. The final diagnosis was CA.

Treatment

As a result of the surgical elimination of the tumor during the excisional biopsy, the patient is currently free of disease 54 months after surgery (Figure 5).



Figure 5. Upper lip and vestibular fornix showing characteristics of normality 54 months after surgery.

Discussion

It refers to a 62-year-old woman, showing an asymptomatic nodule in the upper lip that was initially noted extraorally. Mucocele was the first diagnostic hypothesis, followed by CA and

nasolabial cyst. This clinical case is partially in accordance with the findings of other studies presented previously.

Sometimes large and locally aggressive palatal CA may invade into the nasal cavity and cause

partial obliteration of maxillary sinus and even destruction of the maxillary walls and hard palate, which can be verified by panoramic radiographs and a maxillofacial CT scan.¹⁴

The diagnostic value of a preoperative CT and MRI imaging for CA is emphasized by Yamada et al.¹² The authors suggest CA should be included in the differential diagnosis when a nodule is located in the upper lip or buccal mucosa, the tumor is less than 2 cm in diameter, if the CT findings reveal a highly contrast-enhanced mass with a clear margin, and if the patient is over 50 years. MRI findings were not diagnostically specific.

Microscopically CAs are well circumscribed and generally composed of monomorphic epithelial cells that are frequently columnar in appearance and arranged in bilayered strands and ducts which may be associated with a focal collection of basaloid cells. The stroma is loose containing numerous thin-walled blood vessels with little collagen and few fibroblasts present.^{1-3,6,8,9,13-16} Scanning electron microscopic analysis revealed single-layered low columnar to cuboidal tumor cells lining the tubules that were uniform in size, shape, and structural appearance. Oval nuclei contain abundant euchromatin, sparse heterochromatin, and small nucleoli.^{5,7,8,17,18} The stroma of CA is primarily composed of mucoid material that is PAS-positive diastase resistant, mucicarmine-positive, alcian-blue-positive, and appears as a stellate reticulum.^{5,16,17} Cytologic features of CA are similar to those of tall columnar cells and small basal cells of the excretory ducts of salivary glands which supports a possible origin from excretory duct cells.⁵ CA shows positive staining with anti-keratin, anti-vimentin, and anti-S-100 protein, but immunohistochemical analysis is seldom needed in view of the characteristic histologic and clinical appearance.^{2,19}

A histological differential diagnosis between CA and basal cell adenoma (BCA) of the trabecular type seems necessary. BCA is composed of nests and multilayered cords of polygonal or cuboidal cells which sometimes exhibits peripheral palisading. The stroma of the latter is typically fibrous with less conspicuous blood vessels.⁸ These features serve as distinguishing features between BCA and CA.

A study comparing the extent of p63 immunoreactivity in adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma, basal cell adenoma, and CA found CA did not exhibit any p63 immunoreactivity.²⁰

Sometimes CA has a tendency to present as a multifocal lesion (approximately 22% of the cases)¹ especially in the lips.^{2,3,4,6} It is this multifocal nature that can lead to recurrence which may happen over ten years later.⁷ The multifocality of these tumors may be identified only microscopically rather than clinically.² It is highly likely the presence of tiny foci of tumor (microadenomas), developing in minor salivary glands at a site remote from the original lesion,^{1,4,10,16} are the cause of the multifocal nature of CA.⁹ However, it is important to emphasize these tumors are completely benign in behavior and no malignant transformation has been reported.³ Care must be taken to avoid interpreting the multifocal nature of these neoplasms as a “field of cancerization,” leading to a diagnosis of malignancy and unnecessary surgery.¹ In the present case there was no evidence of satellite islands in the microscopic examination. This negative finding excludes or avoids the misdiagnosis of malignancy.

Conversely, Khullar and Best¹⁰ suggest the multifocal occurrence and late recurrence of CA represents a field of neoplastic change although it appears to be benign. This indicates a high neoplastic potential because the recurrence may arise after a short time.

Adequate treatment consists of conservative surgical excision with a narrow margin of apparently normal tissue for minor salivary glands.^{1,2,3,12,14,16} Superficial or total parotidectomy, depending on extent and location of the tumor, is recommended for parotid lesions which yields excellent outcomes.^{2,14}

Summary

Local excision of symptomatic nodules seems to be sufficient to manage patients with multifocal CA.¹⁰ But, unlike other benign tumors, CA may need a longer follow-up due to its tendency towards multifocal occurrence and late recurrence.^{3,7}

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