



Case Report

Canalicular Adenoma- A case report of a rare benign tumor of minor salivary gland of upper lip

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ABSTRACT

Among all salivary gland tumors, Canalicular adenoma constitutes only 1% of all neoplasms. Oral cavity is the most common location for this rare benign neoplasm of minor salivary glands. Within oral cavity upper lip is the most common site for this tumor followed by buccal mucosa and sometimes it is rarely seen in parotid gland. Previously CA was considered as a type of monomorphic adenoma, however in latest World Health Organization (WHO) classification, CA is classified as a separate entity under benign neoplasms. Here we report a case of CA in a 65 year old female patient who presented with swelling upper lip since 10 months. The swelling was painless submucosal nodule, firm in consistency measuring approximately 2 cm in diameter. Diagnosis of mucous retention cyst was made clinically. Cyst was excised and on histopathological examination we made diagnosis of Canalicular adenoma. We report this case because of rarity of this lesion.

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1. Introduction

Canalicular adenoma accounts for 1% of all salivary gland tumors.¹ Oral cavity is the most common location for this rare benign neoplasm of minor salivary glands. Within oral cavity upper lip is the most common site for this tumor followed by buccal mucosa and sometimes it is rarely seen in parotid gland. Previous literature found that around 80% cases of CAs is present in upper lip. However rarely these have been also reported in the palate, parotid, and mandible. These tumors are commonly seen in older adults with female predominance.²

CA was previously classified as a type of monomorphic adenoma along with basal cell adenoma. Now days, CA is classified as a separate entity in the recent classification by World Health Organization(WHO). Although it is believed that CAs originates from terminal duct, however origin and separation of these neoplasms is still controversial.

Clinically, it presents as painless slow growing mucosal swellings ranging in size from 0.5 to 3cm. Clinically CA usually presents as solitary nodule or as multiple nodules.³

Here, we discuss a rare case of CA presented with upper lip swelling which was diagnosed clinically as mucus retention cyst.

2. Case Report

A 65 years old female presented with history of painless swelling in upper lip since 8 months. Swelling was smooth surfaced, firm in consistency and measuring approximately 2 cm in diameter with no prior history of trauma. No other significant medical history was found. Clinically, diagnosis of mucous retention cyst was made. The cyst was excised and further sent for histopathological examination. Grossly, we received a single encapsulated globular tissue piece measuring 1.2x0.8x0.3cm. External surface was smooth. Cut section was solid and grey white in color. No cyst identified grossly (Figure 1). Microscopy revealed a well demarcated lesion composed of canalicular structures

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with a central lumen. These structures show branching and anastomosing pattern and lined by double layer of cuboidal to columnar epithelial cells with vesicular nuclei and moderate amount of cytoplasm (Figures 2, 3 and 4). The intervening stroma showed fibrocollagenous tissue, numerous capillaries, focal hemorrhage, chronic inflammatory infiltrate and hemosiderin pigment. No cytological atypia or mitotic figures seen. The diagnosis of canaliculal adenoma was made finally on histopathology.



Fig. 1: Gross Examination: Cut section of globular tissue piece is solid and grey white in color

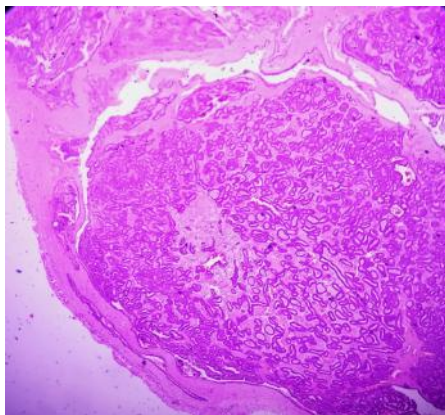


Fig. 2: Photomicrograph showing well demarcated lesion comprising of canaliculal structures with a central lumen. (H&E x 400)

3. Discussion

Benign salivary gland neoplasms were classified into two broad categories, monomorphic and pleomorphic adenoma by Rauch et al.⁴ CA was previously classified as a type of monomorphic adenoma along with basal cell adenoma. In 1972, the histological classification by WHO classified CA with other types of monomorphic adenoma. Now in recent classification by WHO, CA is classified as a separate entity under benign tumors.⁵

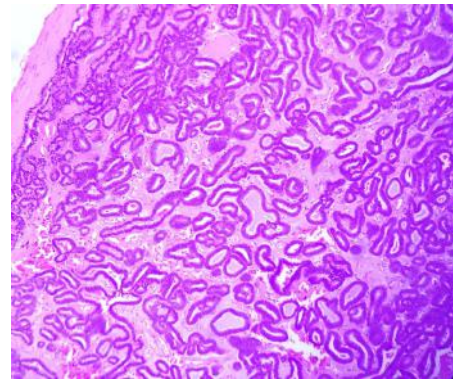


Fig. 3: Photomicrograph showing surrounding fibrocollagenous tissue stroma and blood vessels around canaliculal structures. (H&E x 100)

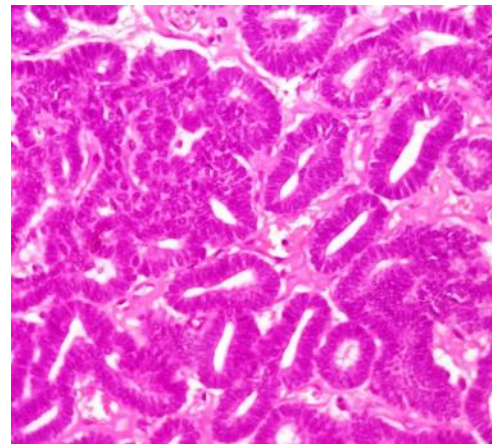


Fig. 4: Photomicrograph showing double layered cuboidal to columnar epithelial cells with vesicular nuclei and moderate amount of cytoplasm. (H&E x 400)

Canaliculal adenoma is rare benign tumor of minor salivary glands and it is rarely found in major salivary glands. After pleomorphic adenoma and mucoepidermoid carcinoma, it is the third common salivary gland tumor. Canaliculal adenoma constitutes 4% of the minor salivary gland neoplasms and only 1% all salivary gland tumors.³

These tumors commonly occur during seventh decade of life with 65 years mean age of presentation. It has female predominance.⁶ Most common location of CA is upper lip as in our case report (80% CA cases). Rarely it involves major salivary glands and buccal mucosa.⁷

It usually presents as a well-demarcated asymptomatic lesion measuring approximately 0.5 to 3 cm. The clinical details in our case report correspond to previous studies in the literature.³

Histologically, the tumor reveals a well demarcated lesion depicting double row of cuboidal to columnar cells forming cords in a loose well vascularized stroma. These

double rows appears to be tight or separate forming canaliculi. Minimal mitotic figures seen. In agreement with Sivoiella et al., the histological features are typical of Canalicular adenoma.⁶

Differential diagnosis include mucoid cyst, vascular tumor, lipoma, or another salivary gland tumor, including basal cell adenoma, low-grade adenocarcinoma, and adenoid cystic carcinoma. Distinction between these salivary gland tumors and multifocal CA can be difficult. Hence, immunohistochemistry is useful for the diagnosis.

Because of slow growth pattern, bland appearance, and circumscription of tumor, malignant change was not found in previous studies. Biopsy, enucleation and excision of the tumor, are proposed treatment options.⁸

In conclusion, we describe peculiar clinical and histopathological features that help in diagnosis. CA has an excellent prognosis and no malignant transformation has ever been described in the literature. Recurrences are rare but can be seen in multifocal CA.

4. Source of Funding

None.

5. Conflict of Interest

None.

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