Case Report

Intra-Mandibular Canalicular Adenoma: Report of a rare case

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Abstract

Canalicular adenomas are uncommon benign salivary gland neoplasms of the oral cavity. They are typically located on the upper lip, buccal mucosa and infrequently found on the palate and derived from minor salivary glands. Intra-mandibular localization of canalicular adenoma is extremely rare. Due to benign character of the tumour, canalicular adenomas rarely present with bone erosion. Histologically, trabecular type of basal cell adenoma, pleomorphic adenoma and polymorphous low-grade adenocarcinoma should be discriminated from canalicular adenomas. A-56- year old female patient with asymptomatic intra-mandibular canalicular adenoma was presented. The lesion was managed surgically under local anesthesia and 2 year's follow up was uneventful.

Only two other intra-mandibular canalicular adenoma cases have been reported up till now. This case report describes the third intra-mandibular canalicular adenoma, and reviews the literature.

Keywords: Salivary gland tumour, Pathology, Immunohistochemistry.

Introduction

Canalicular adenomas (CAs) are uncommon benign tumours that derive from minor intraoral salivary glands. The upper lip is the most common affected site about 90% of the cases, almost exclusively occurring after 4th decade with a slight female predilection.[1-3] Buccal mucosa, palate and parotid gland are the other sites.[7] Intra- mandibular localization of the CA is extremely rare. Clinically CA presents as painless, slow growing, submucosal mass either firm or fluctuant on palpation.[3] Owing to slow growing pattern CAs usually remain asymptomatic until the size of the tumour becomes visible. Histologically CAs are composed of columns of columnar and cuboidal cells with small cystic spaces with or without connective tissue capsule.[4-6] Color of the overlying mucosa is generally normal but it may have a bluish hue and mimics a mucocele.[7] Rarely epithelial surface of the lesion may exhibit focal ulceration.[1,3] Local surgical excision of the lesion is the appropriate treatment for CAs.[6] Trabecular type of basal cell adenoma, polymorphous low grade adenocarcinoma and the other rare salivary gland tumours

are differential diagnosis for CAs, thus immunohistochemical observations are required.[4,5]

Case Report

A 56 years old female patient referred to the Karadeniz Technical University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery for diagnosis and treatment of an expanding mass on the left posterior mandible. Her medical history was unremarkable. Clinical examination revealed a well-defined painless mass at the left posterior mandible since 4 years. First and second premolar teeth were extracted 6 years ago due to periodontal problems. Patient had no complaint after the extraction. Overlying mucosa was completely intact, without erythema, the lesion was slightly fluctuant on palpation. There was no cervical lymphadenopathy. In a panoramic radiograph a mass of 2x1cm, poorly circumscribed, intra-mandibular radiolucent lesion with bone erosion was observed, which was 5mm above the left mental foramen of mandible (Figure-1). No additional abnormalities were noted upon intra- and extra oral examination. The patient was operated under local anaesthesia via intraoral approach for local excision of the lesion, and during surgery all dark-tan and soft tissues were removed from the bone cavity. Histopathological examination revealed many tubular formations lined with cuboidal epithelium in hyalinized stroma (Figure-2). The lining epithelium of the tubules was flattened focally, no evidence of atypia or mitotic figures. The epithelial cells had a positive reaction with pan-cytokeratin and cytokeratin 7, they had no reactivity for vimentin immunhistochemically (Figure-2).

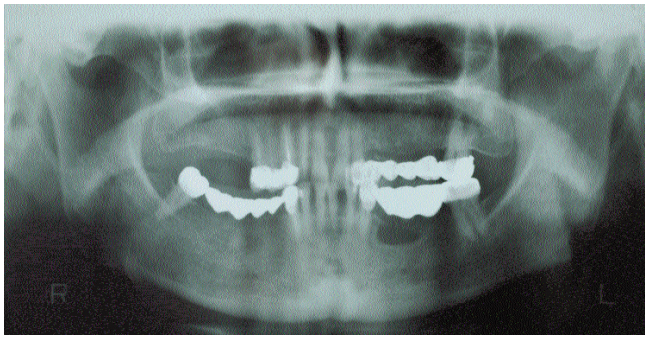

Figure-1: 2x1cm diameter, poorly circumscribed, radiolucent lesion with bone erosion 5mm above the left mental foramen of mandible. Note the superior border of the mandible is intact.

Patient is still coming for follow ups, 2 years follow-up was uneventful.

Discussion

The CA has been traditionally categorized with other several uncommon benign salivary gland tumours for many years.[1] Thus, CA and basal cell adenoma (BCA) were called together as 'monomorphic adenomas.'[3,6,7] Since the CA and BCA have been determined to have distinct clinical and histopathological features,[5,6,8-10] the term of monomorphic adenoma is revised. Latest World Health Organization (WHO) salivary gland tumours classification scheme is now separated these tumours into BCA, myoepithelioma, oncocytoma and CA.[8,9]

The CA is a rare neoplasm of the minor intraoral salivary glands.[4] Tumour has a predilection to occur in the upper lip, 70% or more of the cases were observed in this region.[1-4,7,11] Buccal mucosa, palate and parotid gland the other common sites respectively.[1,4] Intra-mandibular CA is extremely rare. Only two other intra-mandibular canalicular adenoma cases have been reported up till now.[11,12]

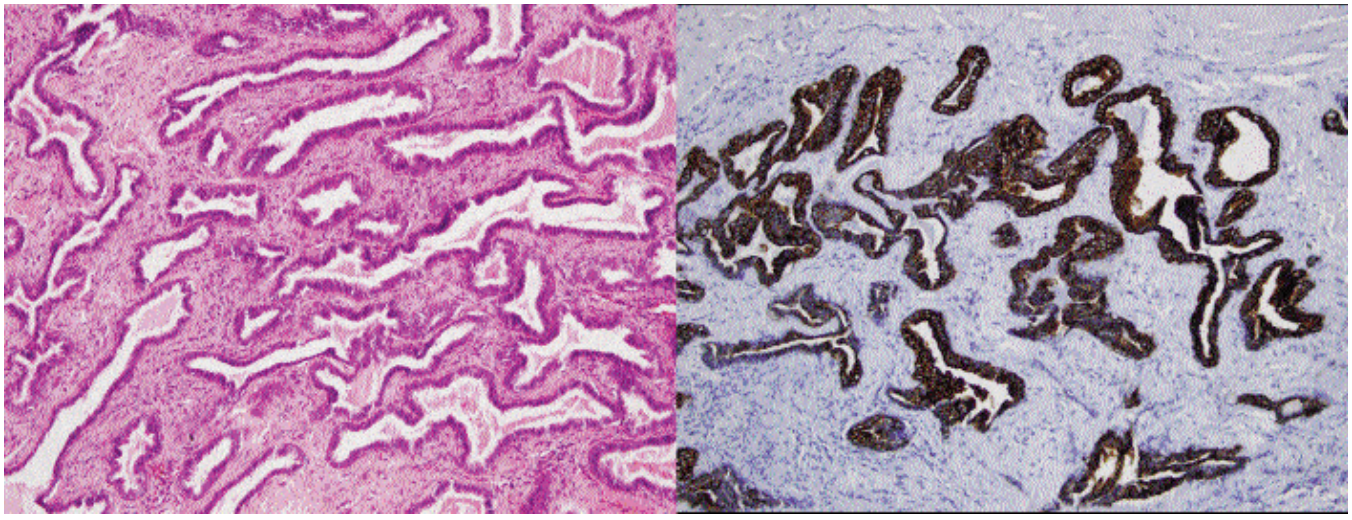

Figure-2: Canalicular adenoma showing characteristic rows of columnar epithelial cells in a fibromuscular connective tissue stroma (H-E staining 400X) (Right Side) Pancytokeratin 7 immunohistochemical staining of the lining epithelium of the ducts shows a positive reaction. (PANCK7 X 200) (Left Side).

The most common benign salivary gland neoplasm is pleomorphic adenoma, which constitutes varying proportions of ductal and myoepithelial cells.[1] Tumours with homogenous pattern are composed of either pure myoepithelial cells or ducts. Tubular variants of basal cell adenoma (TVBCA) still contains a layer of basal or myoepithelial cells. The CA is the only identified tumour with pure luminal cell differentiation.[8] Histologically CAs are composed of isomorphic columns of columnar and cuboidal cells with small cystic spaces with or without connective tissue capsule.[4-7] The BCA consisted of cords, neoplastic cells which were demarcated from surrounding stroma by a peripheral row of cells showing prominent nuclear palisading.[5] Immunohistochemical analysis is an important point to differentiate the CA from other salivary gland neoplasms such as polymorphous low-grade adenocarcinoma (PLGA) and BCA.[3,6] Immunohistochemical profiles of CA and BCA's are also different. While BCA mimics the development of salivary gland tissues, CA exhibits excretory duct origin.[5,8] Vimentin is the best indicator for differential diagnosis between these tumours; CA is not immunoreactive, while BCA and PLGA have positive immunoreactive profile.[6,13] In our case the tumour cells were also not immune-reactive for vimentin. The CA typically occurs in older patients as compared to BCA. Nearly all CA occurs in patients over 50 years of age with a female predominance.[1,4,7,13,14] Our patient was also 56-year-old female.

Management of CA is the surgical excision of tumour with a narrow margin of apparently healthy tissue.[1] Recurrence rate is also extremely rare.[1-3] and there has been only 1 case reported of recurrence in the lip, which occurred 11.2 years after excision.[15] Also CA lacks aggressive features (infiltration, mitotic activity, necrosis, atypia).[8] There have been no reported cases of malignant transformation of CA. Multifocal CA cases with poorly developed or absent capsule could be misdiagnosed as an PLGA.[2,3,7,16] Our case is also still under follow up procedure, and no recurrence was observed till 2 years follow-up.

As a conclusion awareness of the benign character of this neoplasm is important to differentiate from PLGA and the other several salivary gland tumours because the risk of aggressive treatment due to malignancy.

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