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Case Report
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Adenoid Cystic Carcinoma: Report of Two Cases with Review of the Literature

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Abstract

Adenoid Cystic Carcinoma (ACC) is an indolent, slow-growing malignant tumor which occurs frequently in hard palate associated with minor salivary glands. It is an aggressive neoplasm characterized by early neural invasion and a high incidence of multiple recurrence and distant metastases, which may develop years after the initial resection. Surgery followed by radiotherapy seems to be the best treatment. ACC patients have a limited prognosis, even after radical surgery and radiotherapy. Present article describes two cases involving adenoid cystic carcinoma of the maxilla with different treatment modalities.

Keywords: Carcinoma; Adenoid cystic; Maxilla; Computed tomography; Magnetic resonance imaging

Introduction

Adenoid Cystic Carcinoma (ACC) is the most frequent malignant tumor that originates from salivary glands and accounts for 1% to 2% of all head and neck malignancies and approximately 10% of all salivary gland neoplasm's [1,2]. ACC can include the nasopharynx, lacrimal glands, upper respiratory tract, lungs, mammary glands, skin, mastoid air cells, and genital tract [3-5]. Also intracranial invasion between the rates of 4% and 22% was reported [3-6]. ACC is characterized by a high rate of late local failure and distant metastasis [7]. The 60% to 70% of ACCs arise from the minor salivary glands, which mainly localize in the palate and sinonasal tract, although they may also occur in the parotid or submandibular glands [8]. The clinical behavior of ACC is a contradiction: First, tumor growth is slow, but its clinical course is relentless and progressive. Second, surgical intervention is usually feasible, but multiple local recurrences are not rare. Third, metastatic spread to regional lymph nodes is uncommon, but distant spread to the lungs and bones is frequent. And fourth, 5 year survival rates are optimistically high, but 10 to 20 year survival rates are dismally low [9].

Three histological patterns of growth have been described: solid, cribriform, and tubuloductal. Tubular and cribriform subtypes have better prognosis than solid subtypes [10].

Signs and symptoms of ACC depend largely on the site of origin of the tumor. Early lesions of the salivary glands present as painless masses of the mouth or face usually show slow growing character. Advanced tumors may present with pain and/or nerve paralysis, for this neoplasm has a propensity to invade peripheral nerves as a characteristic histological finding [11].

The purpose of this paper is to report two cases of ACC along with an analysis of the literature in order to make a contribution to the diagnosis, pathogenesis, treatment and differential diagnosis of the ACC.

Case Presentation

Patient 1

A 49-year old female patient was referred to the department of oral and maxillofacial radiology (Faculty of Dentistry, Erciyes University) with an expansion in her left maxilla. Extra-oral examination revealed a swelling in the left quadrant of the maxilla (Figure 1a) and intra-oral examination revealed a swelling with a significant expansion in the left maxilla bucco-palatinally (Figure 1b,1c).

According to the patient's anamnesis; she had first realized a swelling in her left maxilla seven months ago. She stated that the mass had been growing slowly over seven months and she complained about occasional pain. However, she neglected this condition and the swelling had been

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Figure 1: Preoperative Images of Case: a) Extra oral view of swelling in the left quadrant of the maxilla. b,c) Intraoral view of significant expansion in the left maxilla bucco-palatinally, d) Decrease in trabecular structure of the left maxilla on panoramic graphy.

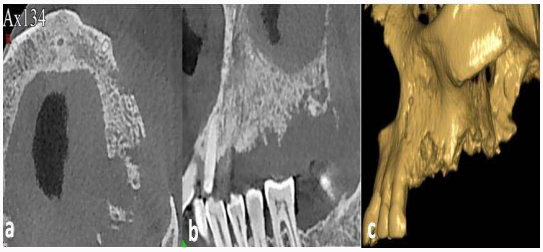

Figure 2: CBCT sections, a) Axial section b) Sagittal sections c) 3D images.

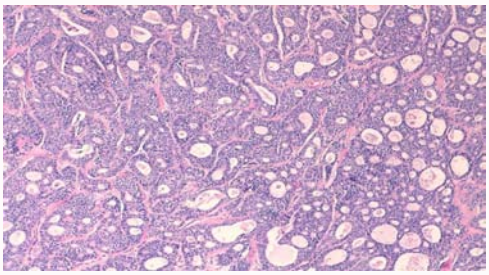

Figure 3: Histopathological section of case one, a) Adenoid cystic carcinoma consisting of solid nests and gland-like (pseudo cystic) areas. The lumens of the pseudocystic areas are seen filled with homogeneous eosinophilic material (H&E x100).

growing slowly ever since. In addition, we determined that a lesion had been diagnosed as an abscess and some medical treatments had been performed repeatedly in other clinics during the period of seven months.

Her panoramic radiograph showed a little decrease in trabecular structure of the left maxilla (Figure 1d). So, Cone-Beam Computed Tomography (CBCT) scans and Three-Dimensional (3D) reconstruction images were performed (Figure 2a-2c). 3D CT imagination showed that the large mass involving the left maxilla and the maxillary sinus. Clinical appearance and the radiological findings were consistent with Central Giant Cell Granuloma (CGCG).

An incisional biopsy including healthy and cystic tissue was performed through the oral cavity under local anesthesia. The histological examination of hematoxylin and eosin stained sections revealed solid type adenoid cystic carcinoma (Figure 3).

Patient was referred to the faculty of medicine, Department of Otorhinolaryngology for the treatment of the lesion and there

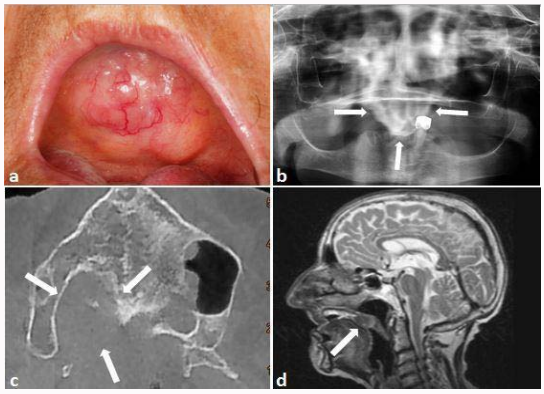

Figure 4: Preoperative images of case two, a: swelling in soft palate, b: Panoramic radiography, c: axial section on CBDT images, d: a well limited lesion in the middle of the soft palate with a radiolucent border in MRI.

underwent an inferior maxillectomy.

Patient 2

A 78 years old female patient was referred to Erciyes University dentistry faculty department of oral and maxillofacial surgery due to complain of swelling in the soft palate (Figure 4a). Her history revealed a tooth removal 2 months before the admission during which the swelling was noticed and the patient was referred.

Patient complained about pain during swallowing which was involving from soft palate to larynx. Clinical examination showed a lobulated and well limited lesion in soft palate. Panoramic (Figure 4b) an CBDT (Figure 4c) MR imaging were performed and a well limited lesion in the middle of the soft palate with a radiolucent border was revealed in MRI (Figure 4d).

A transoral incisional biopsy was carried out under local anesthesia. The histological examination of hematoxylin and eosin stained sections were compatible with cribriform type adenoid cystic carcinoma (Figure 5).

Patient was referred to the Faculty of Medicine, Department of Otorhinolaryngology for the treatment or the lesion and underwent total maxillectomy.

Discussion

ACC is a relatively rare tumor that forms about 1% of all

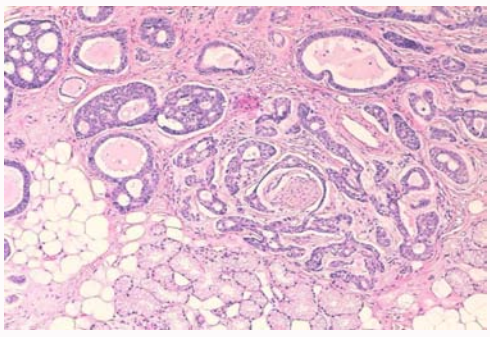

Figure 5: Histopathological Section of Case 2: Tumor is not surrounded by a prominent capsule but is infiltrated into the surrounding salivary gland. In addition, perineural invasion, which is the characteristic feature of the tumor is observed (H&E x100). B-5680: Typical cribriform pattern of adenoid cystic carcinoma. The tumor is composed of solid nests and gland-like structures of columnar cells that do not contain significant pleomorphism and atypia (H&E x100).

malignant tumors of the oral and maxillofacial region and about 10% of all malignant tumors of the salivary glands [9,12]. Malign lesions of the salivary glands can mess with other pathologies that arise in same locations; in the salivary glands these include benign mixed tumor, mucoepidermoid carcinoma and Polymorphous Low-Grade Adenocarcinoma (PLGA) [13]. The differential diagnosis is important for the selection of the main treatment modality.

ACC has a potential of recurrence and distance metastasis. Kokemueller et al. [12] literature review showed that overall survival and recurrence free survival rates decrease by the years and mean survival times of patients were 4, 7 years. In a recent study of a cohort of 160 ACC patients, disease specific survival was 89% at 5 years but only 40% at 15 years.

There are three major variant histologic growth patterns of ACC: Cribriform, tubular and solid. Solid types of the tumor have poor response to the treatment and survival rates are lower than cribriform type. Also, solid type has a potential of metastasis and recurrence [14].

ACC can develop in a variety of anatomical sites, such as major and minor salivary glands, the lacrimal gland, the mucous glands of the upper aerodigestive tract, the skin, and the breast [15]. In one of our cases, the tumor was localized in soft palate where the minor salivary glands exist, and in the other, in the left maxilla.

The diagnosis of the lesion can be late due to its slow growing potential and lack of symptoms may cause a delay applying to a healthcare professional. In the second case presented here, the mass was noticed by her dentist. The first case waited for 7 months to apply to a hospital with an expectation of self regression of the tumor.

Bernardes et al. [16] concluded that ACC occurs in fifth decade of the life and usually in female patients. On the other hand Spiro et al. [8] reported that ACC occurs commonly in male patients, (AA) but Kokemueller et al. [12] reported that ACC is not a certain tumor of a certain age however most of their patients occur in 5th or 6th decades of life. In our study one of our patients is in 7th decade of her life and the other is in the 5th decade and both were females.

Definitive diagnosis must be done with the histological analysis of biopsy or specimen of the lesion. It is important that clinical appearance of the lesion may mix with other lesions which occur in same area of oral and maxillofacial region. Definitive diagnosis will guide the clinician to apply the right treatment modality. As an example, in case 1, we first thought that the lesion was CGCG due to clinical appearance and the radiological findings but incisional biopsy and the histological examination showed that the lesion was an ACC.

In a recent study of the authors which about the biopsy results of the oral lesions during the period of the years 2006 and 2012 showed that ACC is not a common lesion of the oral region. In this unpublished study only 15 of the 476 biopsies were malign however none of them were ACC.

Conclusion

As a conclusion early diagnosis of ACC is very important because of the high potential of recurrence and distance metastasis. Incisional biopsy must be performed if the lesion occurs in soft palate and radiological findings show non-cystic appearance.

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