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CASE REPORT

Adenoid Cystic Carcinoma: A Case Report

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ABSTRACT

Introduction: Adenoid cystic carcinoma (ACC) is a rare cancer originating from the salivary glands, displaying an indolent growth pattern but aggressive progression. Early diagnosis is crucial due to the high metastatic potential and recurrence rate associated with ACC. This case report aims to contribute to the understanding and timely identification of ACC and its impact on patient management.

Case: A 42-year-old male patient presented with bleeding and swelling of the gingiva in the right upper molar region. Radiological examination revealed a radiolucent lesion causing bone resorption. An excisional biopsy confirmed the diagnosis of ACC.

Discussion: ACC poses diagnostic challenges due to its diverse clinical features and histological types, often leading to delayed detection. A prompt diagnosis is essential for effective management. This case report emphasizes the importance of early detection and highlights the clinical and pathological aspects of ACC, aiding in its recognition and appropriate treatment.

Key words: adenoid cystic carcinoma, head and neck cancer, salivary gland tumors

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a very rare malignant neoplasm of the salivary glands[1]. ACC is most commonly observed in the salivary glands in the body. ACC is a rare tumor type, accounting for only 1% of all malignant tumors of the head and neck and 10% of total salivary gland neoplasms. ACC is more prevalent in the minor salivary glands than in the major salivary glands[2]. Additional regions of localization in the head and neck also include the tongue, palate, lacrimal glands, paranasal sinuses, and nasopharynx, as well as the external auditory canal. ACC can also arise in secretory glands in other tissues such as the tracheobronchial tree, oesophagus, breast, lung, prostate, cervix, Bartholin's glands, and vulva[2]. ACC is a slow-growing but aggressive neoplasm with a high recurrence rate[3]. It is characterized by a prolonged clinical course and the late onset of distant metastases. Distant metastases and regional lymph node involvement usually occur late in the course of the disease, many years after diagnosis. The organ involved in distant metastases is usually the lung, but bone, liver, and brain metastases have also been reported. Invasion of adjacent structures is quite aggressive, and hematogenous spread is more common than lymphatic spread. Perineural invasion is characteristic and occurs in more than 60% of cases[4].

The aim of this report is to present a case with a histopathological diagnosis of ACC causing bone destruction in the posterior molar region of the right maxilla and to contribute to the early diagnosis of oral cancer.

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CASE

A 42-year-old male patient was admitted to Karamanoğlu Mehmetbey University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery with bleeding and swelling of the gingiva in the region of the right upper molar. The patient's medical history revealed that he had a tooth extracted in the same area approximately 1.5 years ago. Clinical examination revealed a painless, hyperemic lesion on the alveolar mucosa (Figure 1). Radiological examination revealed a radiolucent lesion with indistinct margins in the right maxillary molar region, causing resorption of the alveolar bone on the panoramic radiograph (Figure 2). An excisional biopsy was taken under local anesthesia and sent for histopathological examination.

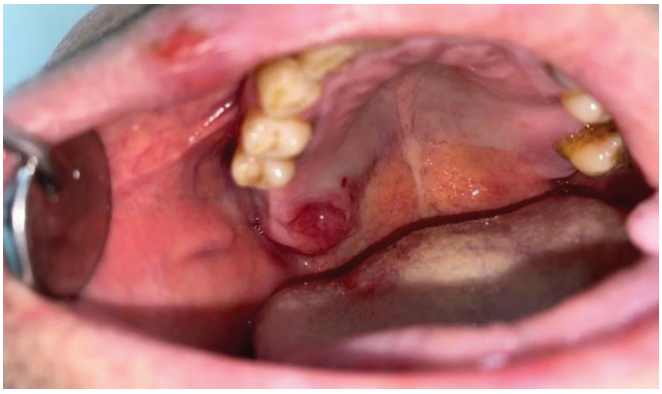

Figure 1: The intraoral appearance of the lesion as observed during a clinical examination.

Histopathological examination revealed a tumor structure consisting of myoepithelial and ductal cells with round pseudocystic areas. These cystic areas were filled with amorphous material. Basophilic glucosaminoglycans were histochemically positive for Alcian blue. Myoepithelial cells were immunohistochemically positive for p63 staining, and ductal cells were immunohistochemically positive for CK7.

Resection was performed under general anesthesia using an extraoral approach to include hard and soft tissues, and then a pedicled flap was transferred from the cheek area and closed primarily. The patient has been under control for 1 year, and no recurrence has been observed (Figure 3).

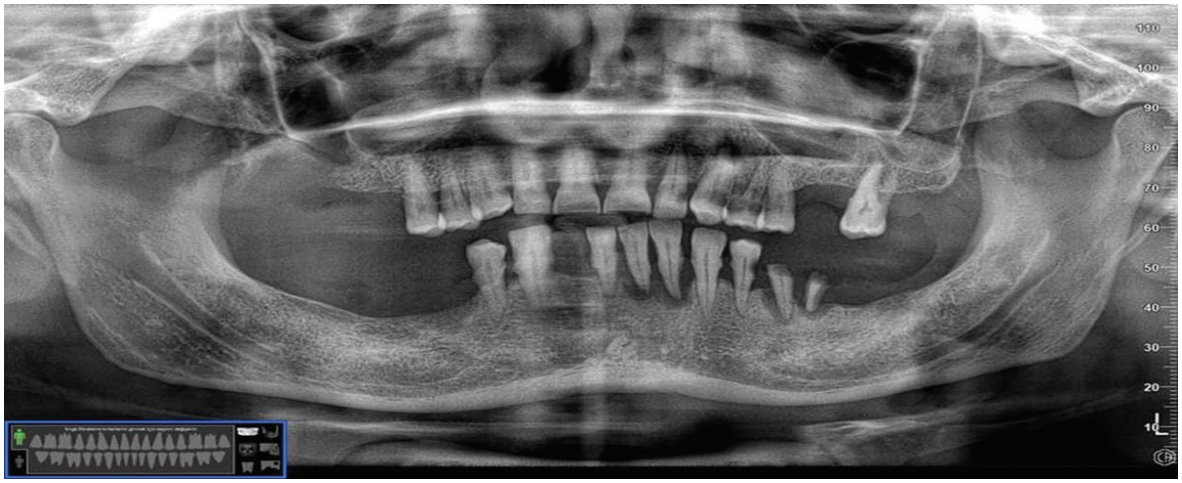

Figure 2: A panoramic image was obtained prior to the procedure.

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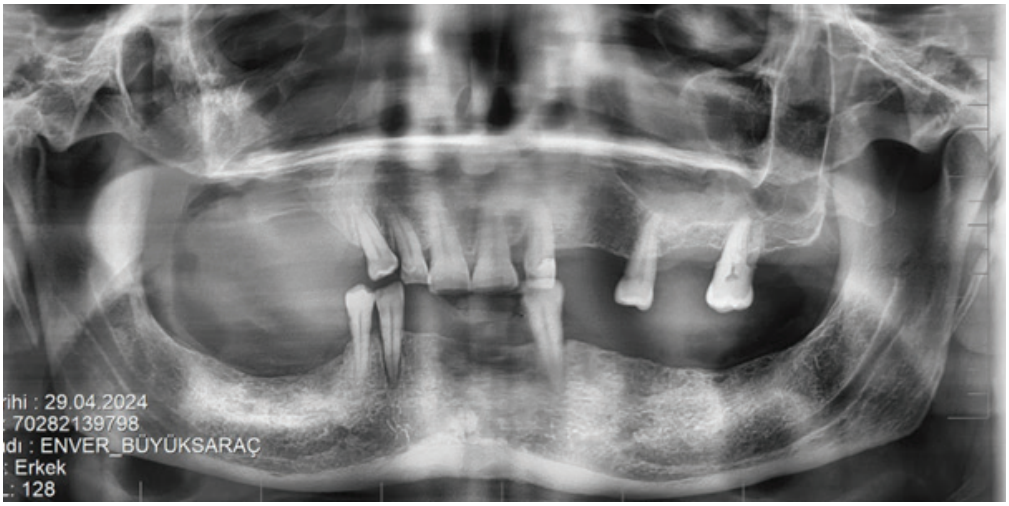

Figure 3: A panoramic view of the first year postoperatively.

DISCUSSION

ACC was first described in 1856 and named 'Cylindroma' due to its histological character of interweaving epithelial and connective tissue elements. The term 'adenoid cystic carcinoma' was defined by Ewing in 1954[5,6]. The lesion occurs predominantly in females and in the 5th and 6th decades of life. This case report presents a case of ACC in a 42-year-old male patient. Due to the slow growth of the tumor, it is characterized by the presence of the lesion for several years when patients present for treatment. In addition, pain, facial paralysis, and perineural invasion may be observed[6]. However, our patient did not have nerve involvement, as it was detected at an early stage.

The best way to treat local ACC is with surgery, resulting in total resection and negative surgical margins with no significant loss of organ function[7]. In spite of proper surgical technique, 5- to 10-year recurrence ratios have been reported to range from 30% to 75%[8]. Especially in the case of metastases, the survival rates drop significantly, with equally increased recurrence rates of up to 60%[9]. One strategy proposed in the literature to reduce local recurrence is postoperative radiotherapy. Although data from randomized trials is lacking, some studies suggest that this treatment is beneficial[10]. The majority of ACC types have slowly progressing growth dynamics and do not benefit from systemic chemotherapy. However, various chemotherapy studies have been conducted over the years. Previous studies have demonstrated that response rates to cytotoxic chemotherapy for ACC are consistently low. Consequently, there is no established standard of care for the systemic chemotherapy of patients with ACC tumors[2].

Particularly in the case of malignant lesions, early detection and treatment planning are crucial to prolong survival and prevent metastasis to distant sites. After diagnosis and treatment, ACC cases should be followed for many years because of the risk of recurrence.

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