



Case Report

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LOCALIZED HARD PALATE ADENOID CYSTIC CARCINOMA

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Abstract

Adenoid cystic carcinoma (ACC) also called cylindroma in the oral cavity is a high grade malignancy tumor due to perineural spread tendency. It is the second most frequent malignant tumor of the salivary glands. It is characterized by slow expansion, local invasion, rare and generally occurred in minor salivary glands of the head and neck. Clinically local pain, facial nerve weakness or paralysis and superficial ulceration may occasionally be the initial presenting symptoms, but there are observed especially in late-stage. Bone invasion rarely occurs, at the beginning there are no radiographic changes. This case report aimed to present the oral and radiographic findings of ACC in a female patient. A 81-year-old female patient applied to our clinic complaining with hard swelling that appeared in the middle line of the hard palate 2 months ago and eating difficulty. Intraoral examination showed a fibrotic mass in the midline of the hard palate, extending from the insisive teeth to the molar teeth and the mucosa on the lesion was normal. Panoramic radiograph showed radiolucent lesion with well-defined borders extending from maxillary molar teeth to the nasal cavity and maxillary sinus. In our case, biopsy was performed and lesion diagnosed as ACC histopathological. After maxillary resection, postoperative radiotherapy was performed. Adenoid cystic carcinomas are rarely seen salivary gland tumors in the head and neck region. Dentists can play an important role in the diagnosis of intra-oral tumors.

Keywords: Adenoid cystic carcinoma, Salivary glands, Palatal neoplasms

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1. Introduction

Salivary gland tumors are a small part of the salivary glands diseases. They constitute 3% of all neoplasms in the body, approximately 5-10% of head and neck tumors (Khan et al., 2001). ACC is the most common malignant tumor in the major and minor salivary glands which constitutes 3% of all salivary gland cancers. It is mostly

seen in patients between 20-60 years old. Approximately 50-70% of reported cases in the head and neck region were seen in minor salivary glands, mainly in the palate region. The parotid gland is most affected among the major salivary glands (Regezi et al., 2016). It can be seen in other parts of the body, such as lacrimal gland, nasopharynx, breast, trachea, lung (Bradley, 2017). The

ACC, which has been typed histopathologically as cribriform, tubular and solid by the World Health Organization (WHO), has been generally described as a fetal type of basoloid tumor (El-Naggar and Huvos, 2005). It is a slow developing lesion, pain and fixation to the surrounding tissues are mostly seen in this tumor (Neville, 2015). This finding is associated with the tendency of the tumor to perform neural invasion. ACC is the most common tumor of the parotid gland (Bradley, 2017). Mucosal surface ulceration may occur in palatal tumors and bone invasion is common in lesions occurring in palatal and maxillary sinuses (Neville, 2015). Large resections are applied in the treatment as the tumor tends to local recurrences and later metastasizes. Postoperative radiotherapy can be used to reduce the likelihood of recurrence (Bradley, 2017).

The aim of this report is to highlight the diagnosis and importance of oral cancers in terms of dentists adding a new case of ACC.

2. Case Report

A 81-year-old Caucasian woman was admitted to Karadeniz Technical University, Faculty of Dentistry, Oral and Maxillofacial Radiology Department for a hard swelling of hard palate occurring 2 months ago and difficulty in eating. Anamnesis revealed that hypertension and history of myocardial infarction. Only operation history was the cardiac stent implantation four years ago. Patient was using antihypertensive and antiaggregant drugs. In the dental history it was learned that the last prosthodontic treatment was performed about 8 years ago. No abnormalities were found in the extraoral examination. The patient's intraoral examination revealed a fibrotic mass in the midline of the hard palate, extending from the insisive teeth to the molar teeth and the mucosa on the lesion was normal. The teeth in the region of the lesion were in normal position and there was no mobility (Figure 1).

On the panoramic radiograph obtained from the patient, there was a well-defined, homogeneous radiolucent lesion in the vicinity of the maxillary sinus and the nasal cavity, extending from the mesial of the tooth 21 to the tooth region 27 (Figure 2).

Contrast-enhanced CT (CECT) was used as a further imaging technique to evaluate the lesion's borders within the bone. CECT images revealed a heterogeneous contrast enhanced mass with size of 44x32 mm which originating from hard palate, expanding to the nasal cavity, left maxillary sinus and buccal and palatal bone. It destructed both lower nasal concha, nasal septum and buccal palatal bone (Figure 3).



Figure 1. Intraoral examination of the buccal and palatal bone that is observed to cause expansion of fibrotic swelling and normal mucosa on the image.



Figure 2. Radiolucent lesion extending between the teeth 21 and 27 in panoramic radiography.

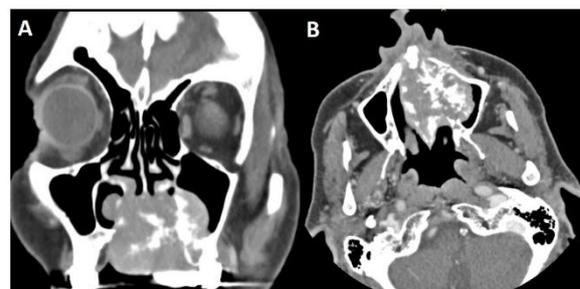


Figure 3. Contrast-enhanced CT coronal image (A) and axial image (B) of the hard palate origin, bilateral lower nasal concha and left maxillary sinus invasive mass lesion.

Patient's preliminary diagnosis was made as ACC and patient was referred to the Otorhinolaryngology Department. The histopathological examination confirmed our preliminary diagnosis. Our patient was operated and post-operative radiotherapy was applied. One year after the operation, the patient was admitted to our clinic with the desire to renew her obturator. In the radiographic examination, a defect area which was applied maxillectomy was observed (Figure 4).

Intraoral mucosal tissues was observed normally in clinical examination. The patient was referred to the relevant department to renew the obturator and the

patient was followed for a long time.



Figure 4. 1 year follow-up of the patient who was operated for adenoid cystic carcinoma.

3. Results and Discussion

ACC was first described by Billroth in 1856 and named as cylindroma due to its histological character (Hadfield et al., 1996). ACC is one of the rare tumors in the head and neck region. However, it is the most common tumor in the minor salivary glands and is usually seen in the palatal region. It, a slowly developing tumor, is characterized by being asymptomatic until the need for treatment, high local recurrence tendency, perineural invasion and distant metastasis (Kowalski and Paulino, 2002). It was observed more frequently especially in women in the 5th-6th decade (Waldron et al., 1988). In our case, the patient was 81-year-old female patient and the lesion was localized in the hard palate compatible with the literature. The mucosa on the ACC usually appears smooth, domed swelling without ulceration. Pain, facial paralysis and perineural invasion are among the characteristics of this tumor (Bradley, 2017). Our patient did not have these symptoms. There was asymptomatic swelling in our case and the mucosa on the lesion was observed smoothly. Radiographic imaging methods, especially computed tomography (CT) and magnetic resonance imaging (MRI), are very important for determination of tumor, surgical margins and postoperative recurrence. While perineural invasion and the extension of the tumor in the soft tissues are best visualized by MRI, bone involvement can be best detected by CT (Dong et al., 2015). Our patient's CECT revealed destruction in the nasal septum, lower nasal concha and maxillary buccal and palatal bone, nasal cavity and medial wall of left maxillary sinus. Right maxillary sinus was intact.

Differential diagnosis of ACC is performed by dental abscess, pleomorphic adenoma, mucoepidermoid carcinoma, adenocarcinoma and Warthin tumor (Uraizee et al., 2018). The dentists should be careful in the palatine areas, especially in the palatal swellings without any infection, and should be aware of the malignant character of the lesions caused by the minor salivary glands.

Large resections should be applied in ACC treatment because of the tendency to local recurrence and distant metastases. The excision of the regional lymph nodes and radical extraction of the tumor is recommended. Postoperative radiotherapy can be applied to reduce the

likelihood of recurrence, radiotherapy and chemotherapy. Combinations can be performed in patients who cannot be operated (Bradley, 2017). In our case, ACC was confirmed by biopsy and the patient was referred to Otorhinolaryngology outpatient clinic. Maxilla resection and then postoperative radiotherapy was performed. After postoperative treatment, our patient was treated with an obturator and prosthetic rehabilitation was achieved and patient was followed.

4. Conclusion

Adenoid cystic carcinomas are rare salivary gland tumors in the head and neck region. The local aggression of the tumor may be fatal even after 10-20 years initial treatment due to local aggressive, high local recurrence and late distant metastasis. Therefore, long-term follow-up is recommended in these tumors (Mendenhall, 2004). Dentists play an important role in the diagnosis and follow-up of tumors located in the mouth.

Conflict of interest

The authors declare that there is no conflict of interest.

Acknowledgements and ethics

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions. Informed consent was obtained from all patients for being included in the study.' If doubt exists whether the research was conducted in accordance with the Helsinki Declaration, the authors must explain the rationale for their approach, and demonstrate that the institutional review body explicitly approved the doubtful aspects of the study. Identifying information of patients or human subjects, including names, initials, addresses, admission dates, hospital numbers, or any other data that might identify patients should not be published in written descriptions, photographs, or pedigrees unless the information is essential for scientific purposes and the patient (or parent or guardian) gives written informed consent for publication. If any identifying information about patients is included in the article, the following sentence should also be included: 'Additional informed consent was obtained from all patients for which identifying information is included in this article.'

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