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Case Report: Adenoid cystic oral carcinoma of the buccal mucosa: a case report study

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ABSTRACT

Adenoid cystic carcinoma is a rare tumor with epithelial cells origin in the jaw and face area. Here, we reported a case of this malignancy in the palate of a 43-years-old man. In this case report study, a 43-years-old man came to Shohada Qaen Hospital in January 2022 with a complaint of a sore in his mouth. An adenoid cystic based hypo-echo lesion was reported in the ultrasound. The pathological examination of the biopsy obtained by outpatient sampling method showed malignant neoplastic tissue with proliferation, hyperchromatic nucleus with false cyst appearance and cribriform glands, and the diagnosis of oral adenoid cystic carcinoma was confirmed. The patient was treated surgically. The prognosis of this tumor is different depend on the tissue of the mass, the clinical stage of the tumor, the site of involvement and the presence of vascular or perineural invasion.

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Introduction

Adenoid cystic carcinoma (ACC) is a malignant and rare neoplasm of the epithelium, which occurs in the minor and major salivary glands (1). After anaplastic thyroid carcinoma, ACC is the second most common malignant tumor of the endocrine system (2). This malignancy can originate from other areas such as the tongue, hard palate, nasopharynx, lacrimal glands and also the external auditory canal. The highest incidence of ACC in the head and neck area is reported in the fifth to sixth decade of life (3). According to the reports of the Institute of Pathology (AFIP), in the cases of ACC diagnosis, 26.8% in the parotid, 24% in the submandibular gland, 20.5% in the palate, 5% in the tongue, about 4% in the lips and buccal mucosa and 1.2% occur in the sublingual gland. The incidence of ACC is in the 4th to 6th decade of life and it occurs slightly more in women than in men. The malignant tissue usually grows slowly and is clinically observed as a hard nodule or a lump with intact mucus (4, 5). Adenoid cystic carcinoma consists about 7.5% of all salivary gland neoplasms. More frequent developing in minor salivary gland, this is a slow-growing tumor with a long-lasting natural evolution, quite aggressive locally, but which has a tendency toward local recurrence and even for distant metastasis. The literatures have reported a high rate of distant metastasis (about 40%), that lung reported as the most commonly affected site. Also, for patients receiving surgical resection, the survival rates of 5 and 25-years were 77.3% and 25.5%, respectively (6, 7). This tumor is diagnosed late due to its slow growth, and because of this late diagnosis, perineural and hematological spread and the risk of cyst recurrence and metastasis exist even several years after initial diagnosis and treatment (8).

Case Report

The patient is a 43-years-old man who is addicted to opium and alcohol, and in his history, he mentions migraines and hemodialysis due to high creatinine, as well as increased liver enzymes. The patient had a history of hospitalization due to headache, decreased level of consciousness (GCS = 9), shortness of breath, and increased creatinine in 2022. So, He was diagnosed with TTP and underwent dialysis. There is no significant familial history. He has a history of using sertraline, chlordiazepoxide, Inderal and sodium valproate. In January 2022, he went to Shohada Qaen educational and medical center with a complaint of a sore inside his mouth. In the initial examinations, a mass was felt in the right buccal mucosa, measuring 2x2 cm, with a leukoplasia mucosal surface from a few months ago, and the depth of the mass to the muscles was 2 cm. The mass was without pain and bleeding and cervical lymphadenopathy was not observed. An ultrasound was requested for the patient and a mass-like hypo-echo lesion with the size of a lobule, approximately 22 x 20 x 15 mm, deep to the right muscles of mastication, based on adenoid cystic was reported. An outpatient sampling of the lesion on 2022/11/01 was performed in the office and malignant neoplastic tissue with proliferation, hyperchromatic nucleus with false cyst appearance and cribriform glands were observed, and the diagnosis of oral adenoid cystic carcinoma was confirmed.

On 04/02/2022, the patient underwent a wide excision surgery of the malignant tumor of the palate and buccal mucosa. Three days after the operation, due to the chest pain reported by the patient, an X-ray imaging was performed and pneumothorax was diagnosed. A chest tube was inserted for the patient. The pathology report of the sample performed in 16/02/2022 was as follows: in the microscopic view, sections of the mucosa with non-dysplastic squamous epithelial lining in depth with mucinous salivary gland acini and striated muscles with tumoral involvement consisting of cells with Large hyperchromic nuclei and vesicles with little mitosis and little cytoplasm with the formation of more cribriform structures and fewer tubules was observed and sometimes causing desmoplasia reactions and nerve invasion with infiltrative tumor margins along with the spread to the muscle layers and involvement of the harvest margins were evident. The final diagnosis was

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adenoid cystic carcinoma of the mouth, right buccal. The dimensions of the tumor were 2.1 x 2 x 1.5 with a cribriform pattern and less than 5% of the solid component, and the depth of invasion was at least 20 mm, along with perineural invasion and without invasion to the vascular lymph and molars of the upper and lower margins. The patient was discharged on 14/02/2022 with a good general condition.

Discussion

Adenoid cystic carcinoma is a tumor of salivary glands, head and neck, and it includes 1-2% of all head and neck malignancies (8). Typical clinical and pathological findings of this tumor include slow growth, perineural invasion and potential local recurrence and metastasis with high mortality. Up to 50% of these tumors occur in the intraoral minor salivary glands usually in the hard palate. Perineural invasion is seen in 59% of cases(9). Pain is an important symptom due to its propensity for perineural spread. This disease has a long clinical course and questionable prognosis, Although minor salivary gland ACCs have a worse prognosis than those of the major salivary glands. Diagnosis is usually made by CT-scan, histological examination and biopsy(10).

Histopathologically ACC presents as three patterns: cribriform, tubular and solid. The most important pattern is the “cribriform” pattern that nests of tumor cells have a sieve-like configuration. The second major pattern observed in ACC is the “tubular” pattern in which elongated tubular structures with a central lumen are seen. “solid” pattern is the third pattern which the tumor islands are completely filled with basaloid tumor cells without cystic spaces. All of these three patterns can usually be observed in most cases of ACCs, although the distribution varies greatly between different lesions. Tumor is graded according to Szanto et al staging scale, : cribriform or tubular (grade I), less than 30% solid (grade II), or greater than 30% solid (grade III) (11). Other factors that determine the prognosis of this type of tumor include the clinical stage of the tumor, the site of involvement, presence of vascular or perineural invasion, positive surgical margins, duration of disease symptoms, tumor recurrence or metastasis. Early diagnosis with a 10-years survival rate is estimated 75%, while the survival rate gradually decreases with the progression of the disease. ACC has a special tendency to invade the tissue around the nerve. Perineural invasion is associated with a decrease in survival and an increase in distant metastasis (12, 13). In our patient, who presented with ulcer symptom without pain and absence of paranesthesia, it seemed that he has no perineural invasion, but the pathology report stated that the tumor is along with perineural invasion and without invasion of vascular lymph and molar upper margins and it has been lower. In this case we reported, no metastasis had occurred at the time of diagnosis. After surgery and additional treatments, the patient's follow-up continues.

Conclusion

In our patient, who presented with symptom of lesion without pain and absence of paresthesia, it seemed that he has no perineural invasion, but the pathology report stated that the tumor is along with perineural invasion and without invasion of vascular lymph and molar upper margins and it has been lower. In this case we reported, no metastasis had occurred at the time of diagnosis. After surgery and additional treatments, the patient's follow-up continues.

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Authors’contributions

Maryam Sadat Katebi: Conceptualization, Methodology, Writing - Review & Editing **Davood Smailpour:** Resources, Investigation, Visualization **Razie Sadat Javadzadeh:** Methodology, Visualization **Mina Ghalenoei:** Writing - Original Draft, Data Curation **Masumeh Daliri:** Funding acquisition, Project administration, Supervision

Conflict of Interests None

The authors declare no conflict of interest.

Ethical declarations

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This research is approved by the ethics committee of Birjand University of Medical Sciences

Availability of data and material

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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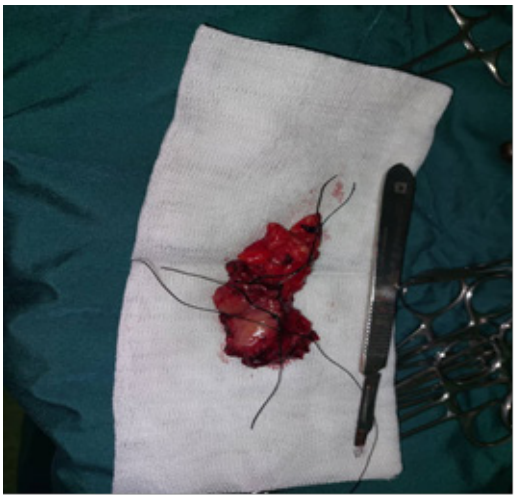

Figure 1 Tumor size and shape after surgery

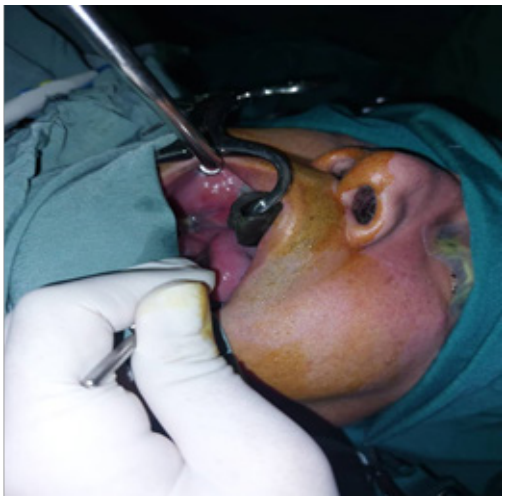

Figure 2. Location of the malignant tumor

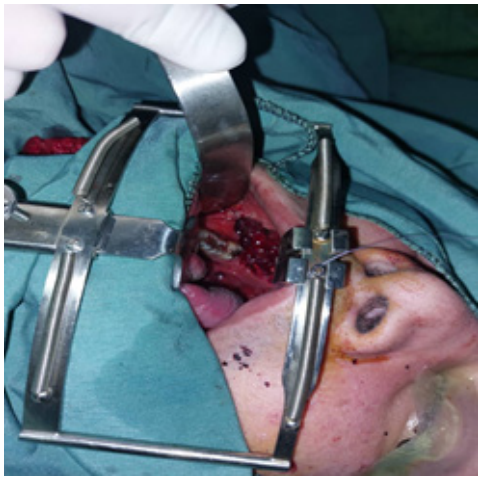

Figure 3. Location of the malignant tumor

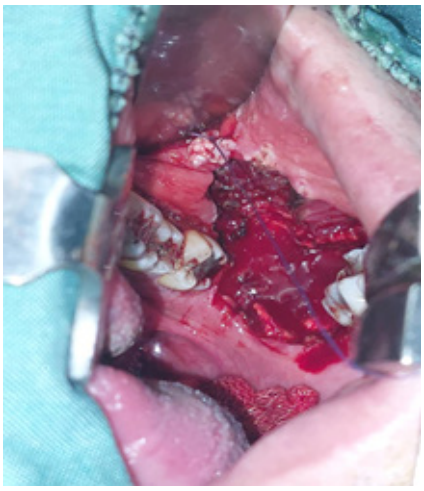

Figure4. Location of the malignant tumor