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CaseReport: Adenoid Cystic Oral Carcinoma of the Buccal Mucosa: A Case Report Study

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ABSTRACT

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

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1. Introduction

Adenoid cystic carcinoma (ACC) is a malignant and rare neoplasm of the epithelium, which occurs in the minor and major salivary glands (1). ACC is the second most common malignant tumor of the endocrine organ after anaplastic thyroid carcinoma (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 cases of ACC diagnosis, 26.8% of cases are 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% occurs in the sublingual gland. The incidence of ACC is in the 4th to 6th decade of age, and it is seen slightly more in women than in men. It usually grows slowly and is clinically seen as a hard nodule or a lump with intact mucus (4, 5). Adenoid cystic carcinoma 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 literature has reported a high rate of distant metastases (about 40%), that lung reported, as the most commonly affected site. Also 5 and 25-year survival rates for patients receiving surgical resection were 77.3% and 25.5%, respectively (6, 7). This tumor has a slow growth and is diagnosed late due to its slow growth, due to late diagnosis, perineural and hematological spread, and the risk of cyst recurrence and metastasis exists even years after initial diagnosis and treatment (8).

2. Case Report

The patient is a 43-year-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. He was diagnosed with TTP and underwent dialysis. No significant family history. He has a history of using sertraline, chlordiazepoxide, Inderal and sodium valproate. In January of 2022, he went to Shohada Qaen educational and medical center with a complaint of a sore inside his mouth. In the initial examination, 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 without cervical lymphadenopathy. An ultrasound was requested for the patient. According to the ultrasound report, the msee-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 cystic adenoid, an outpatient sampling of the lesion on 2022/11/01 was performed in the office., malignant neoplastic tissue with proliferation, hyperchromatic nucleus with false cyst appearance and cribriform ego and glands were observed, and the diagnosis of oral adenoid cystic carcinoma was made.

On 04/02/2022, he underwent a wide excision surgery of a malignant tumor of the palate and buccal mucosa. Three days after the operation, due to chest pain, an X-ray was performed and pneumothorax was diagnosed. A chest tube was inserted for the patient. The pathology report of the sample dated 16/02/2022 is 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 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 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.

3. Discussion

Adenoid cystic carcinoma is a tumor of salivary glands and head and neck, and it includes 1-2% of all head and neck malignancies (8). The clinical and pathological findings typical of this tumour include slow growth, perineural invasion and potential local recurrence and metastasis with high mortality. Up to 50% of these tumours 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 of the condition due to its propensity for perineural spread. , this disease have a long clinical course and questionable prognosis, also minor salivary gland ACCs having a worse prognosis than those of the major salivary glands Diagnosis is made by CT-scan, histological examination and biopsy(10).

Figure 1. Tumor size and shape after surgery

Figure 2. Location of the malignant tumor

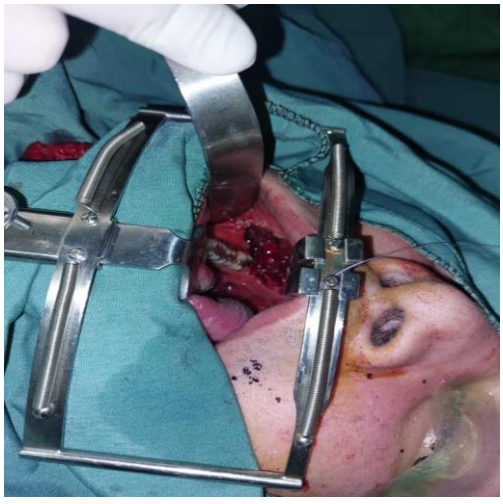

Figure 3. Location of the malignant tumor

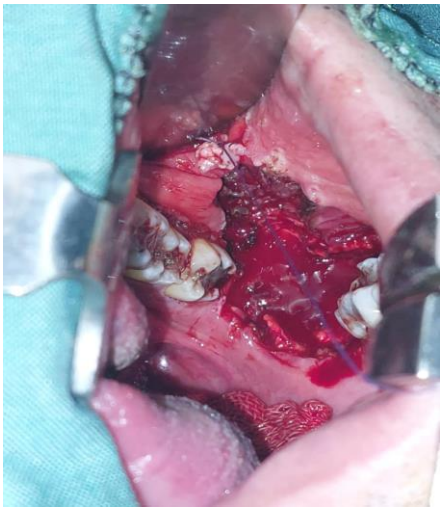

Figure 4. Location of the malignant tumor

Histopathologically ACC presents as three patterns_cribriform, tubular and solid. The most important feature of it is the "cribriform" pattern where nests of tumor cells have a sieve-like configuration. A second major pattern observed in ACC is the "tubular" pattern in which elongated tubular structures with a central lumen are seen. The third pattern is the "solid" pattern where the tumor islands are completely filled with basaloid tumor cells without cystic spaces. In most ACC, all three patterns can usually be observed, although the distribution varies greatly between different lesions. Tumor is graded according to Szanto et al.–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-year survival rate is 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 paraesthesia, it seemed that he had no perineural invasion, but the pathology report stated that along with perineural invasion and without invasion of vascular lymph and molar upper margins and It has been lower. In our reported case, no metastasis had occurred at the time of tumor diagnosis. After the surgery and additional treatments, the patient's follow-up continues.

4. Conclusion

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

Ethical Considerations

Compliance with ethical guidelines

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

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

The authors declare no conflict of interest.

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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