JOURNAL OF ORAL BIOLOGY AND CRANIOFACIAL RESEARCH XXX (2015) XXX–XXX

Available online at www.sciencedirect.com
ScienceDirect
journal homepage: www.elsevier.com/locate/jobcr

Short Communication

Adenoid cystic carcinoma of the floor of the mouth – A rare presentation

Chandresh Jaiswara [a], *Neeraj Kumar Dhiman* [a], *Akhilesh Kumar Singh* [b,*], *Naresh Kumar Sharma* [c], *Vishal Verma* [d], *Arun Pandey* [d]

[a] *Assistant Professor, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, U.P., India*
[b] *Service Senior Resident, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, U.P., India*
[c] *Professor & Head, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, U.P., India*
[d] *Junior Resident, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, U.P., India*

ARTICLE INFO

Article history:
Received 18 September 2015
Accepted 28 September 2015
Available online xxx

Keywords:
Adenoid cystic carcinoma
Minor salivary gland
Floor of the mouth
Malignant

ABSTRACT

Adenoid cystic carcinoma (ACC) is an unusual salivary gland malignancy that remains poorly understood. It is a slow growing but aggressive neoplasm with a tendency for recurrence. It is characterized by the proliferation of ductal (luminal) and myoepithelial cells in cribriform, tubular, solid, and cystic forms. Standard treatment, including surgery with postoperative radiation therapy, has attained reasonable local control rates, but distant metastases do not allow any improvement in the survival rate. The understanding of the molecular mechanisms driving ACC is quite rudimentary. We present a case of a 55-year-old female diagnosed with ACC involving the floor of the mouth with an aim to present the carcinoma's behavior, immunohistocytochemistry, the staining pattern, its treatment, and prognosis.

1. Introduction

Adenoid cystic carcinoma (ACC) is the fourth most common malignant epithelial salivary gland neoplasm, accounting for approximately 10% of all salivary carcinomas.[1] ACC was first described by three Frenchmen (Robin, Lorain, and Laboulbene) in the years 1853 and 1854. It was later described as cylindroma by Billroth[2] in 1856. The term 'adenoid cystic carcinoma' was coined in the year 1928 by Spies[3] and is in use till date. ACC constitutes less than 1% of all head and neck malignancies with 50% occurring intraorally, commonly in the hard palate.[3,4] Other less frequent intraoral sites are the lower lip, retromolar region, sublingual gland, buccal mucosa, and floor of the mouth.[3] Extraorally, parotid gland (25%) is the single most common site of origin. These are clinically innocuous

* *Corresponding author.*
E-mail address: georgianaks@gmail.com (A.K. Singh).
http://dx.doi.org/10.1016/j.jobcr.2015.09.007

lesions usually characterized by small size and slow growth,[4] but are generally associated with extensive subclinical invasion and distant metastasis.[4] Pain is an important symptom of the condition due to its perineural spread.[3] It occurs mostly in 5th and 6th decades, with a female predilection. Cervical lymph node metastasis is found in 8–13% of cases. Distant metastasis is found in up to 50% cases in lungs and bones. ACCs involving minor salivary glands have worse prognosis than those of the major salivary glands.[3]

2. Case report

A 55-year-old female patient reported to our unit with a chief complaint of intraoral swelling below the tongue for the last one year. She was apparently asymptomatic one-year back when she noticed a small swelling over the right side of the floor of the mouth. The swelling gradually increased in size over this period. No associated pain, bleeding, or discharge was found. Past medical and dental history was not significant. Extraoral examination revealed a firm, diffuse, nontender swelling with slight fullness of the right side cheek and lower lip region. Intraorally, a firm, diffuse, nontender oval-shaped swelling measuring approximately 3 cm × 4 cm was present in the right side of floor of mouth region extending over the edentulous alveolar ridge and displacing the tongue contra laterally (Fig. 1). The tip of tongue deviated toward the right side on protrusion. No loss of taste and general sensation of the tongue were noted. An ulcer was noticed to develop over the swelling following the FNAC from the puncture site. Cervical lymph nodes were not palpable. Routine hematological findings were within normal limits. Computed tomographic scan (coronal view) showed an ill-defined homogenous poorly capsulated mass present in the right floor of the mouth region with infiltration in the adjacent musculature of tongue (Fig. 2).

Fine-needle aspiration cytology was suggestive of ACC of the minor salivary gland. TNM staging was $T_3N_0M_0$. The treatment plan was complete excision of the lesion through intraoral approach under general anesthesia. A longitudinal incision of approximately 4 cm length was placed over the most prominent region of swelling, and then layer-wise dissection was done. On exposure, the tumor was found capsulated and hence complete en bloc excision was done (Fig. 3). Lingual nerve and deep lingual vessels were preserved. The closure was done in layers with 3-0 vicryl suture. After one week, wound healing was found satisfactory. Histopathology study revealed it as ACC of the minor salivary gland (Fig. 4). The patient was advised postoperative radiotherapy. She underwent radiotherapy with 60 Gy Cobalt. Following radiotherapy, oral mucositis was observed, which was managed symptomatically. Patient was advised for regular follow-up visits after every three months and no locoregional recurrence has been found as yet (Fig. 5).

Fig. 1 – Preoperative intraoral photograph showing the swelling over the floor of the mouth.

3. Discussion

ACC accounts for approximately 10% of all salivary gland neoplasms. The parotid gland is the single most common site of origin (25%) in the head and neck regions. Most ACCs arise in the minor salivary glands (60%). ACC of minor salivary gland origin occurs most frequently in the hard palate.[4] Foote and Frazell[5] were the first to describe that ACC was located in the major and minor salivary glands. They also reported that the tumors were usually small with an incomplete capsule and showed the variations in histology that had a propensity to perineural spread. They suggested that relatively conservative surgical approach leads to high failure rates and advocated a more radical surgical treatment. At present, ACC remains an extremely difficult lesion to treat. Conley and Dingman[6] described it as one of the most biologically destructive and unpredictable tumors of the head and neck. It has high recurrence rate in patients with increased survival and even when radical excision has been performed.

Clinically, ACC is a relentless, slow growing, and progressive tumor. Sometimes it is associated with pain in cases of large tumor size and perineural spread.[4] There is a strong positive correlation between site of origin and prognosis. The more favorable prognosis with major salivary gland ACC as compared to minor salivary gland is attributed to

Please cite this article in press as: Jaiswara C, et al. Adenoid cystic carcinoma of the floor of the mouth – A rare presentation, *J Oral Biol Craniofac Res*. (2015), http://dx.doi.org/10.1016/j.jobcr.2015.09.007