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A RARE CASE OF RECURRENT ADENOID CYSTIC CARCINOMA OF THE BUCCAL MUCOSA

Plastic Surgery

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ABSTRACT

Adenoid cystic carcinoma is a malignant neoplasm most commonly originating in the minor salivary glands of head and neck region. Among intra oral adenoid cystic carcinoma, buccal mucosa is one of the rarer sites. Here, we report a case of recurrent adenoid cystic carcinoma of the right buccal mucosa in a 33 year old female. As this is an uncommon site for adenoid cystic carcinoma, it should be considered as a differential diagnosis of mass of buccal mucosa. It is imperative that we identify such cases and plan for early surgical excision with adequate margins.

KEYWORDS

Adenoid Cystic, Minor Salivary Glands, Perineural, Recurrence

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a low-grade malignant tumor arising within the salivary glands that accounts for fewer than 5 % of all head and neck tumors and approximately 10–15 % of salivary gland tumors.[1,2] These can arise from all major salivary glands including the parotid, sublingual, submaxillary glands and minor salivary glands. Minor salivary gland of the oral cavity is the most common site of this but involvement can occur in almost any site with a secretory gland component and reported in sites such as the lacrimal gland, breasts, uterine cervix, esophagus, lungs, and prostate.[3] ACC is known for its indolent clinical course, perineural invasion, multiple recurrence rates and delayed onset of distant metastasis. Lymph node metastases are uncommon, but hematogenous spread, especially to the lungs is quite characteristic and metastasis to kidney being extremely rare.[4-6]

Case Report

A 33 year old female came with a complaint of pain and swelling on the right side of face since 7 months. It started after surgery for a similar swelling 2 years back. Initially it was small in sixe but gradually progressed to the present size. Pain is of a constant dull aching pain. There was no history of trauma to the cheek, fever or discharging sinuses. On examination, there was a diffuse swelling over the right cheek region of sixe 9 x 7cm with ill-defined margins. The skin appeared smooth over the swelling with no ulcerations. Facial nerve function was intact. Intraoral examination also revealed a diffuse lesion of the right buccal mucosa measuring 7 x 6cm. It was not tender and the mucosa was not ulcerated. **(Fig. 1)**

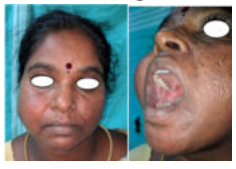

Fig. 1 – Clinical photograph showing the extra-oral and intra-oral views

There was no regional lymphadenopathy. Previous histopathology report showed that the first swelling in the same site was an adenoid cystic carcinoma. MRI showed the extent of the swelling and there were no signs of metastasis. An incisional biosy under local anaesthesia was done and this confirmed the diagnosis of adenoid cystic carcinoma. After obtaining fitness for surgery and explaining to the patient the complications of facial nerve resection in the involved region, surgery was planned. Under general anaesthesia, surgical resection with modified radical neck dissection was performed by the surgical oncology team and sent for histopathological examination which confirmed as adenoid cystic carcinoma. Free anterolateral thigh flap was planned from left thigh. Anamolous origin of the perforator was identified from within the vastus lateralis muscle. The pedicle was dissected for about 9cm length and a flap of size 23 x 10cm was harvested. Inset was given to oral aspect, de-epithelialised for lip and cover for the cheek. Arterial anastomosis was done to the facial artery and vein to the facial vein. Haemostasis was secured and all incisions were closed with 2-0 polyglactin and 3-0 nylon sutures over corrugated rubber drains. **(Fig. 2,3,4)** Patient underwent adjuvant radiotherapy later.

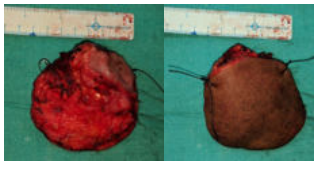

Fig. 2 – Specimen after excision

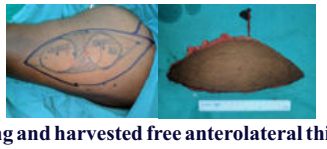

Fig. 3 – Marking and harvested free anterolateral thigh flap

Fig. 4 – Late post-op extra-oral and intra-oral picture, post-radiotherapy status

DISCUSSION

Theodor Billroth in 1856 first described adenoid cystic carcinoma as cylindroma. Stell PM were the first to describe the cylindrical appearance of this tumour.[6] Spies was the first to use the term, "adenoid cystic carcinoma" in 1930. Mehta and Parikh, in1943 emphasized the

malignant nature of this tumour.[7] It is a rare tumor accounting for <1 % of all head and neck malignancies and 10 % of all salivary gland neoplasms.[8] ACC is an uncommon form of malignant neoplasm that arises most commonly in the major and minor salivary glands of the head and neck. ACC make up about 6 % of all salivary gland tumors. They make up 15–30 % of submandibular gland tumors, 30 % of minor salivary gland tumors, and 2–15 % of parotid gland tumors.[9] ACCs are mostly reported in the minor salivary glands, and less frequently in the major salivary glands and is the second most common type of carcinoma which arises in the salivary glands, after mucoepidermoid carcinoma. It usually occurs in females between 5th to 7th decades of life. The most characteristic clinical feature of this malignancy includes its slow growth rate, delayed onset of distant metastasis, increased local recurrences, and peri-neural invasion.[10] Most ACCs are submucosal and they appear as smooth, domed swellings without overlying ulcerations.[10] Bone invasion or a perineural spread can sometimes cause pain or hypoesthaesia. The World Health Organization (WHO) has defined ACC as "a basaloid tumor containing epithelial and myoepithelial cells in diverse morphological configurations, such as tubular, cribriform, and solid patterns. Its clinical course is relentless and usually has a fatal outcome".[11] There are three histopathological types for adenoid cystic carcinoma: cribriform, tubular, and solid patterns.[12,13] Since polymorphism is a common phenomenon in ACC, MD Anderson introduced a pathological grading system which is accepted worldwide: Grade I: tubular and cribriform together, without a solid pattern, Grade II: mostly cribriform, with less than 30% of solid pattern, Grade III: solid being the predominant subtype.[14] Perineural invasion associated with adenoid cystic carcinoma, is defined as "a form of direct primary spread of neoplasm which may not necessarily be macroscopically continuous with the main focus of the tumor, but is usually microscopically continuous." The second and third branches of the trigeminal nerve are most commonly affected. The descending branches of the facial nerve and smaller cranial branches may also get involved. Perineural involvement is claimed to be an indicator of poor prognosis increasing the chances of recurrence and can therefore can have a higher impact on survival rate.[12,13] The primary goal of treatment is local control of the tumor, normal function, and preventing distant metastases.[4] Radical surgery with wide resection margins may not be adequate achieving disease‒ free margins due to perineural invasion. Radiation may be considered for cases having low‒ grade tumors in association with perineural invasion, or evidence of tumor seeding during surgery. A lower radiation dose has also been recommended for patients with tumors located in lymphatically rich areas.[15] Using radiotherapy as primary treatment is proposed when surgery is not feasible and for alleviating bone and brain metastases.[12, 16] Chemotherapy for ACC are usually given for advanced disease statuses, as a single agent or in combinations. Commonly used single-agents are 5-Fluorouracil (5-FU), Gemcitabine, Doxorubicin, Vincristine, Cisplatin, Mitoxanthrone, Epirubicin, Paclitaxel.[17] Neutron, proton or carbon ion radiations may be given for some unresectable or deep tumours. Molecular targeted therapy is a promising tool in cancer treatment. It uses certain drugs that target some specific pathways which play an important role in the proliferation, angiogenesis, metastasis, and/or apoptosis of cancer cells such as imatinib (against c-kit or CD 117), lapatinib, gefitinib (against EGFR), cetuximab, and bortezomib. The roles of immunotherapy, gene therapy and other systemic therapy, such as hormonal treatment for ACC, are still undergoing clinical trials.[18]

CONCLUSION

Adenoid cystic carcinoma is a malignant tumor, commonly originating from minor salivary glands, characterized by features such as indolent growth, perineural infiltration and hematogenous distant metastasis. ACCs should be treated aggressively to achieve negative surgical margins to prevent recurrence to obtain a better prognosis.

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