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Case Report

Adenoid Cystic Carcinoma of Salivary Glands – A Duplet of Cases

Shalini Subramanyam[1]*, Peeyush Shivhare[2] and Amit Ashok Basannavar[3]

[1]Oral and Maxillofacial Pathologist, Family Dentist and General Dentistry, Independent Researcher, Bangalore, Karnataka, India

[2]Oral Medicine Consultant and Oral Radiology Specialist, Nobel Medical College and Teaching Hospital, Nepal

[3]Oral and Maxillofacial Surgeon, Associate Professor, Vidyapeeth's Dental College and Hospital, Sangli, India

***Corresponding Author:** Shalini Subramanyam, Oral and Maxillofacial Pathologist, Family Dentist and General Dentistry, Independent Researcher, Bangalore, Karnataka, India.

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Abstract

Adenoid cystic carcinoma is regarded as a rare malignant neoplasm of the salivary gland. The clinical as well as pathological features of this neoplasm include slow growth, perineural invasion and the potential for local recurrence. The total incidence rate is about 2 – 4% of head and neck tumors. It involves palate most commonly among the minor salivary glands, and parotid and submandibular salivary glands among the majors. Here, we describe two cases of adenoid cystic carcinoma involving the parotid salivary gland and the palate regions.

Keywords: Malignant Neoplasm; Palate; Parotid Salivary Gland; Salivary Gland Tumor

Introduction

Adenoid cystic carcinoma is regarded as a rare malignant neoplasm of epithelial origin involving the salivary glands [1]. It has an incidence of about 2 – 4% all the head and neck malignant neoplasms, and 5 – 10% of all the salivary gland neoplasms. Lorain and Laboulbene first described this neoplasm in 1853. It was named as cylindroma by Billroth in 1859. The name adenoid cystic carcinoma was given by Spies in 1930 replacing the older name cylindroma. Before 1943 it was regarded as a benign mixed salivary gland tumor, when Dockerty and Mayo first emphasized on its malignant nature [2].

Adenoid cystic carcinoma most commonly (about 31%) involves the minor salivary gland, particularly the palate, although major salivary glands such as parotid and submandibular are also involved in some cases [3]. Its clinical findings include slow growth, perineural invasion, local recurrences, and distant metastasis [1]. We are reporting two cases of adenoid cystic carcinoma involving the parotid salivary gland and the palate regions of the oral cavity.

Case 1

A male 50yr old patient reported to the outpatient department with the complaint swelling of left face in front and below the ear since 5yr. It was sudden in onset and gradually progressed to present size and remains constant since 5 yr. Patients also give history of restricted mouth opening since 1 yr. Past medical, family and personal histories were non- contributory. On extraoral examina-

tion there was presence of diffuse swelling in left parotid region, measured about 5x5cm extending antero posteriorly from 2.0cm front of ear lobe to 3 cm posteriorly, superoinferiorly from ear lobe to lower border of mandible (Figure 1a). Surface of swelling was smooth with normal colour. Skin over swelling was normal. No visible pulsation. On palpation swelling was firm, nonfluctuant, non tender and afebrile, not compressible, and not reducible. Skin over swelling was pinchable and normal. Mouth opening was just 10mm. Provisional diagnosis of benign tumor of parotid gland was given. The reason for mouth opening was questionable as there were no sign of any odontogenic infection or oral submucous fibrosis. Patient was advised for radiological investigations. Conventional radiographs were non-significant. In the T1 (Figure 1b) and T2 weighted (Figure 1c) Magnetic Resonance Imaging scan, the left parotid gland region appeared ill defined and presents with multiple hypointense regions inside the gland structure signifying cystic changes. The anterior margin of the gland was ill-defined and appears to merge with the masseter muscle. Axial section in computed tomography showed several hyperdense areas that are arranged in a way to produce multiple cystic hypodense regions inside the left parotid gland structure (Figure 1d). The anterior margin of the left parotid gland was ill-defined and appeared to merge with the masseter muscle. There was no bony involvement seen in the ramus. Based on radiological features we could able to come an initial conclusion that a tumor arising from parotid had encroached in master muscles which probably is the reason of trismus. Ultrasound guided fine needle aspiration was done. Patient was then referred to department of maxillofacia surgery for excisional biopsy where left radical parotidectomy was done. On histopathological examination, the tumor cells were seen in small and large groups with very few individual cells infiltrating a desmoplastic stroma. The cells had a uniform round to oval nuclei with hyperchromasia and mild pleomorphism. The cells in small groups formed a cribriform pattern with small and large cystic spaces within the groups of cells. A few true acinar structures were also seen. There was also a myxoid change in some groups of cells. The tumour had a strong perineural infiltrate and extension around major nerve trunks (Figure 1e). The tumor infiltrates the surrounding connective tissue and the salivary gland tissue and skeletal muscle (Figure 1f). Later patient was further advised for radiotherapy.

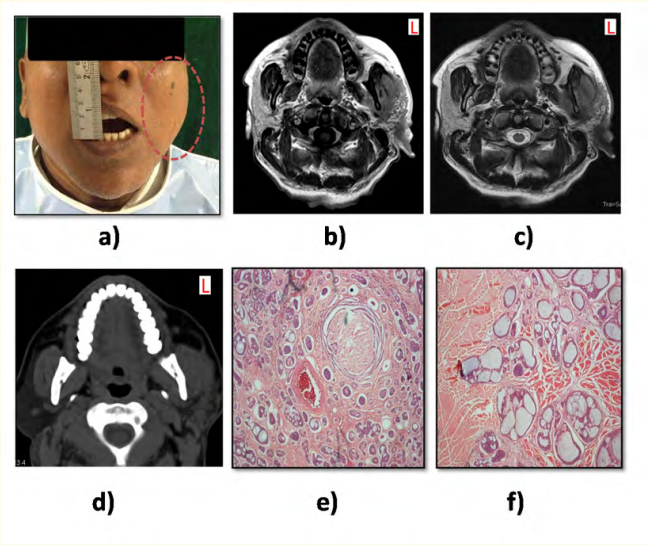

Figure 1: a) Facial profile of the patient showing left parotid swelling and trismus

b) T1 weighted MRI – axial view

c) T2 weighted MRI – axial view

d) Axial section in computed tomography

e) Photomicrograph showing perineural invasion

f) Photomicrograph showing skeletal muscle infiltration.

Case 2

A 27-year-old male patient reported to the Department of Oral medicine and radiology with a chief complaint of pain and sore in roof of the mouth since 2months. The soreness was initially smaller which had progressively increased. Patient's medical, past dental, family and personal history were non-significant. General physical examination was normal. The extra oral examination showed facial symmetry with no evidence of lymphadenopathy. Intraoral examination revealed a solitary ulcero-nodular growth involving the right posterior portion of hard palate and was also seen involving the soft palate on the same side measuring about 3x2.5cm. The swelling was seen extending from mesial aspect of first premolar to 1 cm posterior to tuberosity on left side, involving the palatal area slightly crossing the midline (Figure 2a). On palpation ulcer was painful with soft base but swelling was non-tender and firm in consistency. Provisional diagnosis of malignant lesion arising from minor salivary gland and differential diagnosis of malignancy arising from maxillary sinus was given. The computed tomography scan showed an ill-defined destruction of the palate on the right

side that has involved the entire posterior part of the palate as well as crossed the midline (Figure 2b,2c). The lesion also appeared to involve the left maxillary sinus with the involvement of the medial, lateral, inferior and posterior walls of the sinus (Figure 2d). The lesion has also involved the left nasal cavity. Incisional biopsy revealed islands of hyperchromatic basiloid epithelial cells containing multiple cysts like spaces filled with hyalinized eosinophilic product forming cribriform and tubular structures (Figure 2e). Patient was referred to the oral and maxillofacial surgeon for further treatment.

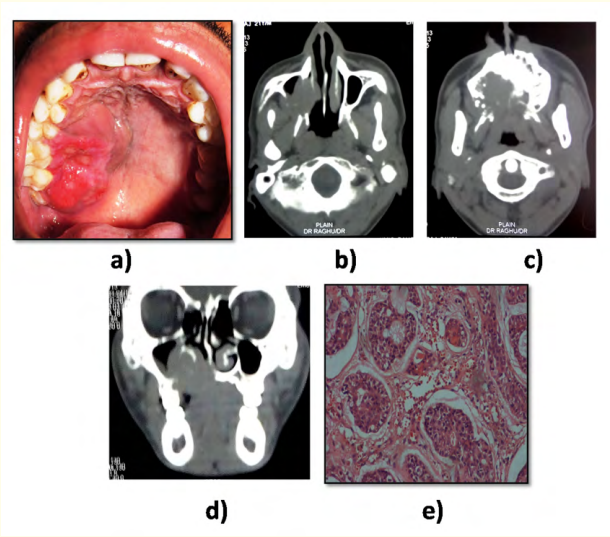

Figure 2: a) Intraoral view of the patient

b) Axial section in computed tomography

c) Axial section in computed tomography

d) coronal section in computed tomography

e) Photomicrograph showing cribriform and tubular structures.

Discussion

The age distribution for Adenoid cystic carcinoma widely ranges from 5th decade to 6th decade of life. It commonly involves the females than compared to the males [4]. It involves the minor salivary glands most commonly, which include palate, floor of the mouth, tongue and lip. It may also rarely present as a primary intraosseous tumor of the maxilla and mandible [2].

The typical clinical presentation of adenoid cystic carcinoma is 2 - 4 cm in diameter when involves the major salivary gland and 3 cm or less in diameter for minor salivary glands [5-7]. It has a low incidence rate of cervical lymph node metastasis, while distant metastasis to lung and bones is via the blood stream. Death may happen with the direct extension of the neoplasm into the base of the skull [7,8].

It is a highly invasive cancer with high recurrence rate although slow growing nature. This tumor can spread by direct invasion, hematogenous, perineural and rarely by lymphatic. Intracranial ACC even is rarer and has been reported as 4 – 22% of ACC. Perineural tumor spread is a poor prognostic indicator. The literature regarding perineural spread of ACC revealed the different possible sites of invasion are divisions of the trigeminal nerve (mainly 2nd and 3rd), the descending portion of the seventh cranial nerve and cavernous sinus etc. The region of Gasserian ganglion to be the most common site of involvement (35.8%), while cavernous sinus was involved in 15.1%. In our first case the tumor had spread directly to adjacent muscles and perineural invasion to facial nerve as well [9].

There are three histologic patterns of adenoid cystic carcinoma have been described, cribriform, tubular, and solid patterns. The cribriform pattern also referred to as Swiss-cheese-pattern by many. It shows histological patterns of cylinder, hence the name cylindroma was given to this neoplasm. Tubular form is less aggressive than the cribriform type. Solid pattern has the worst prognosis [2]. The differential diagnosis of adenoid cystic carcinoma should include both benign and malignant neoplasm of the salivary glands, for e.g. pleomorphic adenoma and adenocarcinoma [1].

Surgical resection followed by radiation therapy is the treatment of choice. Chemotherapy may be used, but its role in the treatment of adenoid cystic carcinoma is controversial [10,11]. Due to the relatively early detection of intraoral lesions or pathologies, the adenoid cystic carcinomas of the minor salivary glands have better prognosis, less chances of advancement to later stages than compared to the neoplasm of the major salivary glands [2].

Conclusion

Adenoid cystic carcinoma is a rare, slow growing but highly invasive malignant neoplasm of the salivary gland. Perineural invasion is one of the special characteristics of this entity, on which

special emphasis should be given during examination and investigations as it is suggestive of poor prognosis.

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