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Case Report

Adenoid cystic carcinoma of soft palate

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ABSTRACT

Adenoid cystic carcinoma is a slow growing and relentless salivary gland malignancy composed of epithelial and myoepithelial neoplastic cells that form various patterns, including tubular, cribriform, and solid forms. It is a rare tumor constitutes for less than 1% of head and neck malignancies and 10% of all salivary gland tumors. Palate is the most common site to be involved in the oral cavity followed by parotid gland and submandibular gland. They are usually asymptomatic, characteristically shows infiltrative growth and perineural invasion. In this paper a case of adenoid cystic carcinoma of soft palate in 52 year old male is presented.

1. Introduction

Adenoid cystic carcinoma is a malignant neoplasm that may affect either the major or minor salivary glands of the oral cavity.[1,2] It was first described by three Frenchmen (Robin, Lorain, and Laboulbene) in two articles published in 1853 and 1854. It was they who described the cylindrical appearance of this tumor. Billroth, in1859, first described ACC under the name "cylindroma", for its cribriform appearance formed by tumor cells with cylindrical pseudolumina or pseudospaces and described that ACC had a "great tendency to recur.[1]" It accounts for about 5% to 10% of all salivary gland neoplasms, representing 2% to 4% of malignant occurrences of the head and neck area. Approximately 31% of lesions affect minor salivary glands, particularly the palate, though they can also be observed in the submandibular and parotid glands.[2]

2. Case Report

A 52 year male presented with complaints of ulcer over the soft palate since 3 months associated with local bleeding ,h/o pain on swallowing solid, liquid and saliva present since 3months. Clinical examination:- 3x4 cm ulcer noted over the soft palate, 0.5cm above the uvula with everted edged and irregular margins. On palpation, tender, bleeds on touch, firm and everted edges.

Elevated blood sugar levels and ESR noted. Rest all parameters are within normal limits. C.T. scan of neck and upper chest showed mild thickening noted in soft palate on left side. Few lymph nodes noted at bilateral levels 1b, II and III, measuring 7 to 10mm. The structures of the nasopharynx and oropharynx are normal. The structures of the larynx Le., the vocal cords, anterior and posterior commissures and the paralaryngeal spaces display normal morphology. The cartilages namely the epiglottis, thyroid. cricoid and the arytenoids show no abnormalities. The sub-glottic area, the trachea and the oesophagus are also normal. The hypopharynx i.e., the pyriform fossae, post cricoid region and the posterior pharyngeal walls are normal. The thyroid

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and salivary glands show normal configuration. Both the lungs show a normal bronchial and vascular branching pattern. The heart and mediastinal vascular structures have a normal anatomical configuration. The thoracic aorta and its branches are normal and show no evidence of calcification. There is no evidence of mediastinal adenopathy. Under GA soft palatal local rotation flap by undermining the edges was done. Sutured and left for secondary healing. Radiation therapy – Received 66Gray in 33 fractions. HPE: Sections show tumor tissue with overlying hyperplasic squamous epithelium arranged in tubular, acinar and cribriform pattern. The tumor cells are biphasic with luminal ductal epithelial cells and abluminal myoepithelial cell. The ductal epithelial cells are low cuboidal cells with round nuclei, indistinct nucleoli and eosinophilic cytoplasm surrounding myoepithelial cells have hyperchromatic, angulated nuclei in coarse chromatin, small nucleoli and clear cytoplasm. Scant mitotic figures seen (0-1/HPF). Surrounding abundant hyalinised stoma is noted. Focally tumor is seen infiltrating into the adjacent skeletal muscle resected margin. Periphery shows normal lobules mucinous minor salivary gland. Features are of Adenoid cystic carcinoma of soft palate.

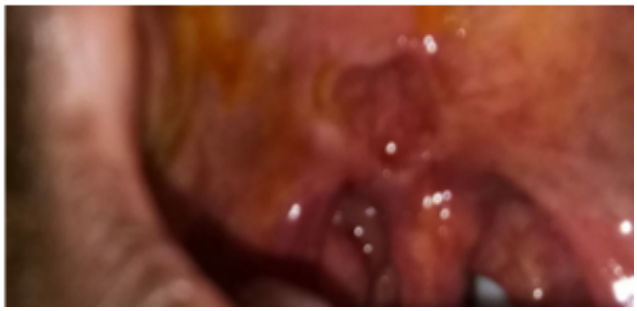


Fig. 1: (Preoperative image)

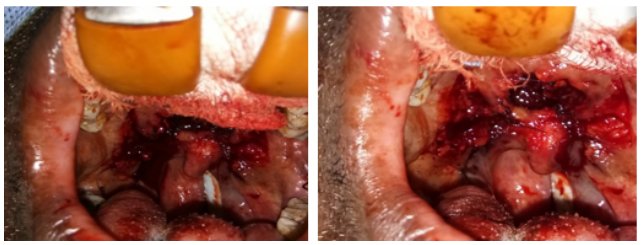
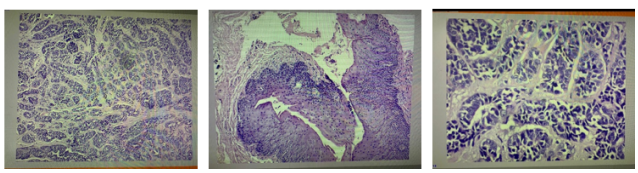


Fig. 2: (Intraoperative images)



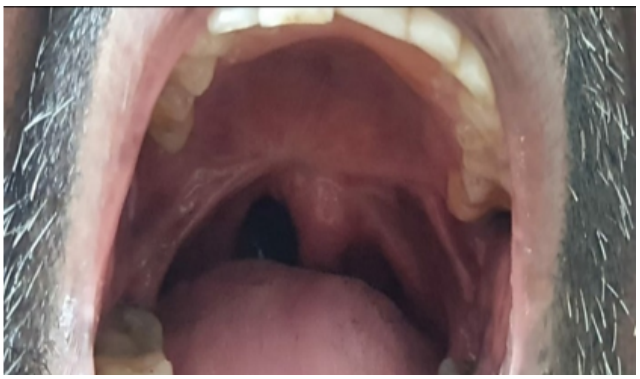


Fig. 3: (Post op and post RT image after 3years)

3. Discussion

Adenoid cystic carcinoma most often presents a diagnostic and treatment challenge owing to the rarity of the lesion. Most findings regarding ADCC are actually based upon studies with a small number of patients and there is a need for further information regarding its clinical behaviour as well as treatment modalities and their results.[3] ADCC may occur at any age although in most cases the patients' age ranged from 24 to 78 years. The age of patients affected with major salivary gland tumors has been shown to be younger (mean 44 years) compared to the age of those who developed tumors of the minor glands (mean 54 years) and shows female predilection (F:M – 1.2:1). Pain is a common and important finding, occurring early in the course of the disease before there is a noticeable swelling.[4] Neoplastic cell neurotropism causes pain. Among the minor salivary glands, palate is more commonly involved generally the area of greater palatine foramen. Among the malignant neoplasms of minor salivary glands, the most common was mucoepidermoid carcinoma (21.8%) followed by polymorphous low grade adenocarcinoma (PLGA) (7.1%) and ADCC was the third most common (6.3%).[5,6] ADCCs of the minor glands have been reported to have worse prognosis than those of the major salivary glands.[7,8] Tumors involving the nose, paranasal sinuses and maxillary sinus have the worst prognosis as they are usually detected with higher stages at the time of diagnosis.[9] Tumors of minor salivary glands usually have the tendency to infiltrate extra glandular soft tissues and bone thereby allowing increased dissemination of the tumor. Lymph node involvement is uncommon (< 5% of cases) and is usually due to contiguous spread rather than lymphatic permeation or embolization.[10]

Histopathologically, it presents three patterns, cribriform, tubular and solid; the most common variant is the cribriform pattern, in which the epithelial cells are arranged in multiple cylindrical spaces, having a pseudo cystic appearance, and many of these pseudo cysts contain a hyaline material. The tubular type is made up of ducts that can be formed by one or two

layers of cells similar to the myoepithelial cells. The solid variant is composed of solid epithelial islands with central areas of necrosis; the cells are small, basophilic and hyperchromatic with a densely granulated nucleus and scarce mitotic figures.[11] The solid type has a poor prognosis contrary to the cribriform type which has a better prognosis. Surgical excision with wide margins is the treatment of choice and, when it metastasizes to the lymph nodes, post-surgical radiotherapy is recommended. The most important prognostic factors include primary lesion size (T), anatomical localization, presence or absence of metastasis (M) at diagnosis time, invasion of the facial nerve and the histopathology grade (G).[11] Possible treatments of ACC include four different modalities: surgical therapy, radiotherapy, chemotherapy and combined therapy (surgery and radiotherapy, radiotherapy and chemotherapy), being the latter in most cases, the treatment of choice.[11,12] Only surgical removal or radiotherapy in isolation may fail to eliminate the possibility of recidivation in surgical margins, as well as the occurrence of metastasis in cervical lymph nodes, lungs, bones and brain.

4. Conclusion

ADCC is rather an uncommon salivary gland malignancy. It is unique for its peculiar histopathological features and tendency for PNI. Prognostic factors of ADCC are the anatomic site, histologic subtypes and metastasis. ADCC with a solid histopathologic pattern is associated with a worse prognosis than those with a cribriform or tubular arrangement. Therefore, early detection, prompt treatment and long-term follow-up are essential in the clinical management of this tumor.

5. Patient Perspective

Satisfactory

6. Informed Consent

Separate informed consent was taken prior to CT scan and surgery.

7. Source of Funding

None.

8. Conflict of Interest

None.

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