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An Unusual Presentation of Swelling in the Palate-An Interesting Case Report

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Abstract:- Adenoid cystic carcinoma is an infrequent tumor that forms less than 1% head and neck malignancy and 10 % of all salivary gland tumors and the common location is the palate. Here is a rare case report of adenoid cystic carcinoma of the palate with a poor prognosis. The late diagnosis resulted only in palliative treatment. Awareness of signs and symptoms should be elaborated to all primary physicians and among common people.

***Keywords*:-** *Adenoid Cystic Carcinoma, Perineural Invasion, Palatal Swelling.*

I. INTRODUCTION

Adenoid cystic carcinoma[ACC] is a malignant salivary gland neoplasm that was first defined by three Frenchman [Robin, Lorain, and Laboulbene]subsequently published in 1853 and 1854.[1]Adenoid cystic carcinoma is a rare tumor that forms less than 1% of head and neck malignancies and 10% of all salivary gland tumors and the common location is palate.[2]The margin between tumor tissue and surrounding healthy tissue is indistinct which makes the early diagnosis and early treatment mandatory. [3] Advanced diagnostic modalities play a vital role in accurate diagnosis. Here is a rare case report of ACC of the palate which was provisionally diagnosed as minor salivary gland malignancy of palate and histopathological examination confirmed ACC and treatment was planned by contrast-enhanced computed tomography [CECT].[4]

II. CASE REPORT

A 46 -year old male patient reported to the department of oral medicine and radiology with the chief complaint of pain in the right upper back tooth region since 10 days and swelling in the same region since 8 days. The patient gave a history of extraction of the right upper back tooth 3 months back. The pain was sudden in onset, throbbing type and radiates to forehead region on the right side and relieved on medication. The swelling was primarily small in size and progressed to its current size. He also reported associated symptoms of nasal blockage and difficulty in breathing during night time. He was hypertensive since 10 years and was under medication.

On extraoral examination, no visible facial asymmetry was noted. Single right submandibular lymph node was palpable which was 0.5 x 0.5 mm in size, hard in consistency, fixed to underlying structures, and tender on palpation. Paresthesia was noted on the right malar region.

On intraoral examination, diffuse reddish-pink swelling of size, 4x3 cm was noted on the right side of the palate adjoining 16, 17, 18 regions to the midline of the palate [figure 1] The margin of swelling was irregular. The swelling was hard in consistency and tender on palpation. On the basis of history and clinical examination, malignancy of minor salivary gland was provisionally diagnosed.

The patient was advised for orthopantomogram [OPG] which revealed ill-defined hazy radio-opacity on the right maxillary sinus. Discontinuity was noted in the medial wall, lateral wall, and posterior wall of the maxillary sinus, and also on the palatal roof. Medially radiopacity was seen extending into the right nasal fossa. Root resorption was present in the distobuccal and mesiobuccal root of the maxillary right first molar[figure 2]. Investigation of incisional biopsy was done which revealed nests of basaloid epithelial tumor cells arranged in cylindrical cyst- like patterns suggestive of adenoid cystic carcinoma CECT revealed a soft tissue density lesion measuring 4.9 x 4 x 3.6 cm noted with epicenter in upper alveolus on the right side causing erosion of upper alveolus extending anteriorly abutting anterior wall of the maxillary sinus, inferiorly eroding hard palate and involving posterior oral cavity on

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the right side with extension to the retromolar trigone, Medially eroding medial wall of the maxillary sinus, right middle and inferior turbinates abutting nasal septum, posteriorly eroding posterior wall of the right maxillary sinus, pterygoid plates, and extending to the right pterygopalatine fossa.[Figure 3] This revealed an inoperable tumor extending to the skull baseand radiotherapy was the treatment plan. The patient was allotted to radiation oncology for palliative treatment.

III. DISCUSSION

ACC is a malignant neoplasm that occurs commonly in head and neck salivary glands. [5]ACC arises from epithelial cells of mucus-secreting glands especially salivary glands. [3] It constitutes 10 % of all salivary gland neoplasm, 22% of salivary gland malignancy with a median age between 47 and 56 years. The common sites are the palate, tongue, floor of the mouth, and lips. Most commonly minor salivary gland is involved. [2]

As ACC is a rare malignancy, scarce data studies are available regarding its etiology. Deletion of chromosome IP 35-36 is frequently seen in ACC. The translocation between chromosome 6q and 9p is another ACC-specific chromosome abnormality and creates the MYB: NFIB gene fushion that leads to overexpression of MYB oncoprotein.This promotes tumorigenesis. Dysregulated MYB genes play a major role in carcinogenesis.[6]

ACC is usually seen in the fourth to sixth decade of life. Generally, ACC does not show specific signs and symptoms. Classic presentation is asymptomatic mass. As the tumor grows it has a propensity for invading nerves and perineural invasion occurs. [1]

Perineural invasion is a distinguishing feature of ACC. Maxillary and mandibular nerves are usually affected by perineural invasion which indicates a poor prognosis. It increases the chance of reccurence.[5]The case presented has pain in the upper right back tooth region and paresthesia in the right malar region which is suggestive of perineural invasion. Regional lymph node metastasis is rare compared to distant hematogenous metastases. However, the case presented had lymph node metastasis.[5]

Differential diagnosis of adenoid cystic carcinoma includes mucoepidermoid carcinoma, low-grade polymorphous adenocarcinoma, basal adenocarcinoma as these are the common malignant salivary gland neoplasm. The tubular, cribriform, solid, are 3 major histological subtypes of adenoid cystic carcinoma and the oral cavity is the most common site. Grading of ACC is done according to histological patterns Tumor grade, tumor size, and perineural invasion determine the prognosis of ACC. The case presented which is of solid variant had a poor prognosis.[4]

Treatment depends on location, extent, size, metastasis of tumor, and age of the patient. The main goal of treatment is to control the size of the tumor and prevent distant metastasis. [5] The main clinical modality for ACC is surgical resection.[7]Radiotherapy is used as a primary treatment when surgical resection is not feasible.[5]Here in our case surgical resection was not feasible which was revealed by CECT. Thus advanced imaging modality proved to be of immense importance in the treatment plan.The main prognostic factors depend on the perineural invasion, patient's age, histological subtype, the extent of the tumor, type and duration of symptoms, treatment, clinical stage, and histological state of surgical resection.[7]

IV. CONCLUSION

Early diagnosis and treatment are imperative for a promising prognosis and improved quality of life for the patient. Advanced diagnostic modalities aids in early diagnosis and better treatment planning. The late diagnosis resulted only in palliative treatment. Awareness of cancer signs and symptoms should be elaborated to all primary physicians and among common people. Further larger prospective studies are required for better diagnosis of a disease which can aid in treatment outcome.

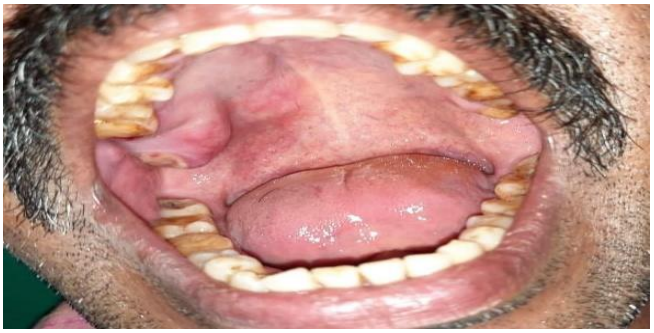

Fig 1 Diffuse Swelling on Right Retromolar Region

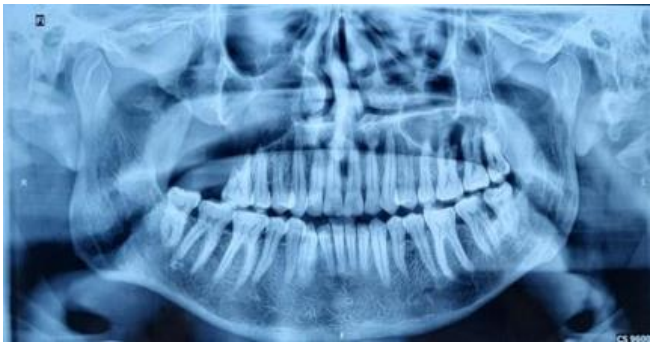

Fig 2 OPG Reveals Ill-Defined Hazy Radiopacity on Right Maxillary Sinus

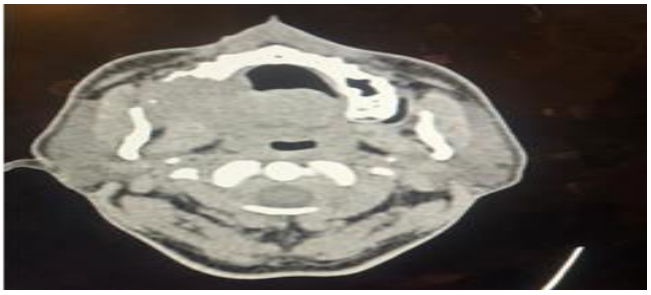

Fig 3 Reveals Erosion of Upper Alveolus Extending Anteriorly Abutting Anterior Wall of the Maxillary Sinus, Inferiorly Eroding Hard Palate

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