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ADENOID CYSTIC CARCINOMA (CRIBRIFORM VARIANT) OF HARD PALATE: - A CASE PRESENTATION WITH REVIEW

Dental Science

Dr. Jigna S. Shah M.D.S Prof. and Head Department of Oral Medicine and Maxillofacial Radiology, Govt. Dental College and Hospital Ahmedabad - 380016

Dr. Anand J Patel* M.D.S Post Graduate Dept of Oral Medicine and Maxillofacial Radiology Govt.Dental College and Hospital Ahmedabad - 380016 *Corresponding Author

ABSTRACT

Adenoid cystic carcinoma (ACC) of salivary glands is a slow-growing malignant tumor, characterized by wide local infiltration, perineural spread, a propensity to local recurrence and late distant metastasis.

KEYWORDS

Adenoid cystic carcinoma, cribriform pattern, parotid gland

INTRODUCTION WITH REVIEW

Malignant neoplasms of the salivary glands in the head and neck are relatively rare, accounting for only less than 7% of all neoplasms, among which about 10% were adenoid cystic carcinomas. Stell PM were the first to describe the cylindrical appearance of this tumour. Theodor Billroth, in 1856, described ACC as "cylindroma" because of its long amorphous compartments which look like cylinders. The term "basalioma" was coined by Krompecher in 1908, who considered this type of tumor to be of analogous nature to the basal cell growths of the skin. It is a relentless tumor that is prone to local recurrence and eventual distant metastases.

Interestingly, Spies was the first to use the term, "adenoid cystic carcinoma" in 1930. In 1943, Mehta and Parikh emphasized the malignant nature of this tumour.[1] under the name cylindroma attributing to its cribriform appearance formed by the tumor cells with cylindrical pseudo Lumina or pseudo spaces. The term "adenoid cystic carcinoma" was introduced by Ewing (Foote and Frazell) in 1954.[2]

It is a relentless tumor that is prone to local recurrence and eventual distant metastases. It is clinically innocuous by virtue of its small size and slow growth potential which depicts its extensive subclinical invasion and marked ability for early metastasis, the factors which make the prognosis of the neoplasm questionable. Fifty per cent of all ACCs occur intraorally, frequently seen in the hard palate region.[3] The tumor extends well beyond the visible and palpable limits of the salivary gland region. This infiltrative capacity is hallmark of ACC. It typically presents as a slow growing submucosal mass/ swelling with constant low-grade dull pain and is well known for infiltration of nerves with following perineural invasion causing paraesthesia. They behave differently with slow growth and local invasion with recurrences and metastasis seen many years after treatment.[4]

Pain and perineural spread along nerve sheath are usually noted. Majority of the tumors arise in the major salivary glands, minor salivary glands of the oral cavity and mucous glands of upper respiratory tract. Other primary sites are breast, lacrimal glands, lung and prostate. Lymph node metastases are unusual; hematogenous spread, often to the lungs is quite characteristic metastasis to kidney being extremely rare. ACC is known for its prolonged clinical course, multiple recurrence rates and delayed onset of distant metastasis.[5] ADCC has a poor prognosis especially in minor salivary glands compared to major salivary glands.[6]

Here, we report a case of Adenoid cystic carcinoma (Cribriform Variant) on the right side of the Palate, in a 45-year-old woman.

Case Report

A 45-year-old female patient visited to Oral Medicine and Radiology department with chief complain of swelling on Posterior right hard palate since 6 months which gradually increases from 1 x1cm associated with continuous dull aching type of pain to attain the present size of 3 x2cm (Figure 2) with gradually decrease in mouth opening from 4 fingers to 2 fingers.(Figure 3) Medical history was unremarkable. Patient was averagely built, malnourished but calm and cooperative with stable vital signs.

On intra oral examination a single ill-defined swelling size of 3 x 2cm was appreciated on right side of posterior part of hard palate which extends antero-posteriorly from mesial of 16 to maxillary tuberosity and medio-laterally from mid palatine raphe to 3cm lateral to it. Overlying mucosa and surrounded mucosa were normal without any kind of discharge. All teeth were present without any caries. (Figure 2) On palpation number, site, size margins and extensions were confirmed. Swelling was firm in consistency, nontender, non-compressible, non-fluctuant, nonreducible without any kind of discharge. Sub mandibular lymph nodes on right side were enlarged, tender, and mobile in antero-posterior direction. History and above clinical findings, were suggestive of swelling from minor salivary gland of the palate. Grade 1 Mobility i.r.t 16,17 were noted.

Radiological examination included intraoral periapical radiographs, Maxillary cross-sectional occlusal radiograph, orthopantomogram revealed bone loss distal to 17 with discontinuity in floor of right maxillary sinus in 16, 17 teeth region suggestive of malignancy arising from palatal salivary gland. (Figure 4 & 5) Computed tomography revealed mildly enhancing mixed density lesion seen in right maxillary sinus causing erosion and destruction of anterior, postero-lateral and medial wall of right maxillary sinus with extension into retro antral fat pad with involvement of medial and lateral pterygoid muscle and extension into nasopharynx on right side with destruction of floor of right orbit with intra orbital soft tissue component. Thinning of lamina papyracea with extension of lesion along medial aspect of orbit. Extension of lesion seen in nasal cavity with erosion and destruction of middle and inferior turbinate and hard palate in laterally. Erosion of floor of right maxillary sinus was supporting diagnosis as malignancy of palatal salivary gland. (Figure 6 & 7)

Patient underwent Right total maxillectomy and the excised mass was sent for histopathological examination that shows H & E stained section with basaloid cells arranged in glandular pattern and swiss cheese like appearance. Few true glandular lumens are seen which are lined by low cuboidal cells having eosinophilic cytoplasm (Figure 8) suggestive of Adenoid cystic carcinoma involving right hard palate and extending to involve the maxillary sinus.

DISCUSSION

The presented case of Adenoid cystic carcinoma was in 45year old female patient at hard palate region. According to Evesson & Cawson Adenoid cystic carcinoma found in women (F:M = 1.2:1) with age varying from 24 to 78 year. Adenoid cystic carcinoma occurs on the palate with the frequency of 8-15% of all the palatal salivary neoplasm. These were similar finding as found in our case. In reported case a slow growing mass with early local pain without facial nerve involvement, local invasion and fixity to deeper structure were present. These all finding were similar to literature except facial nerve involvement, local invasion and fixity to deeper structure.[7]

The radiographic appearance is important in the diagnosis which determines irregular destruction of bone. Tumors arising in the palate or maxillary sinus may show radiographic evidence of bone destruction. In extensive lesion Computed tomography that revealed

soft tissue involvement with maxillary sinus involvement with destruction of floor of orbit. Similar finding was noted in presented case.[8]

Salivary gland malignancies share similar imaging features, and these cannot be differentiated from each another by imaging alone. Imaging is mainly used to detect malignant features, demonstrate local and distant involvements, define nodal status and guide FNAC, if needed. The incidence of regional lymph node metastasis is relatively rare. Distant metastases occur in 25-50% of the individuals, particularly to the lungs and bone. Also, they are more common and unpredictable. In presented case distant metastasis was not seen.[1]

HISTOMORPHOLOGY AND GRADING OF THE TUMOR: [9]

Various growth patterns and cytological details in Adenoid cystic carcinomas establish the diagnostic criteria for this neoplasm. The primary growth patterns are of three types in adenoid cystic carcinoma.

a.CRIBRIFORM VARIANT: Extensive sheets, uniform bands, or cribriform nests usually composed of relatively small, darkly stained, slightly separated basal/myoepithelial cells and small, at times inconspicuous ductlike structures, which may contain secretory products. Round to oval, often fairly uniformly sized intercellular spaces, termed pseudocysts, containing pale grayish blue to pinkish granulofibrillar material at times with a reticular pattern, which develop in relation to the basal/myoepithelial cells.

b.**TUBULAR VARIANT**: Presence of bilayered duct like structures generally composed of an inner layer of cuboidal to columnar ductal cells with moderate amounts of eosinophilic cytoplasm and outer, smaller darker staining cells.

c.SOLID VARIANT: Arranged as variable, at times fairly uniformly sized groups or as sheets of small, darkly stained tumor cells, those are excess proliferations of the basal/myoepithelial cell component. Small duct like structures must be identifiable among the basaloid cells. Nests or sheets of basaloid cells with the above features from 30% or more of the neoplasm

GRADING OF THE TUMOR:

a. GRADE I: The tumor consisting only of cribriform and tubular histomorphology.

b. GRADE II: A mixture of cribriform, tubular and solid growth patterns, with solid growth pattern less than 30% of the tumor.

c. GRADE III: Tumors with predominantly solid features (>30% or more of the tumor).[9]

According to above classification, our case belongs to Cribriform Variant with grade 1.

Optimal treatment of ACC has not yet been fully established. Although most authors advocate the use of surgical excision and postoperative radiotherapy. Some series have found no statistically significant difference between patients treated with combination therapy and those treated with surgery alone. Careful tumor staging and grading with documentation of perineural invasion and margin status continue to be important prognostic tools.[10] As in our case Right total maxillectomy was done and patient was on post-operative radiotherapy. Adenoid cystic carcinoma of minor salivary gland has been reported to have worse prognosis than those major salivary glands. 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.[8] According to literature 5 years survival rate after effective treatment in 75%[9] but unfortunately, in our case patient died due to patient's negligence, malnourished body, poor physic and locally aggressive malignancy. Delayed diagnosis worsens the prognosis as in our case patient consulted after 6 months.

CONCLUSION

Adenoid cystic carcinoma is uncommon salivary gland malignancy. Early diagnosis is important for patient's survival. Therefore, proper physical examination followed by surgery and radiotherapy with close surveillance and careful attention to quality of life issues are the cornerstones of management of this disease.

Legend of figure:

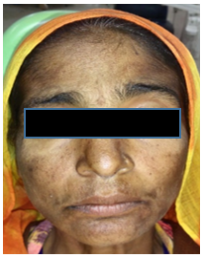

Figure 1. 45-year-old female patient

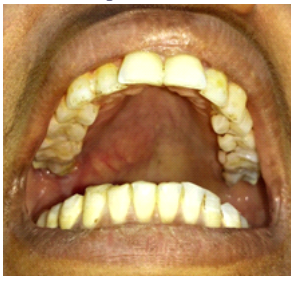

Figure 2. A single diffused swelling present on Maxillary Right Posterior hard palate

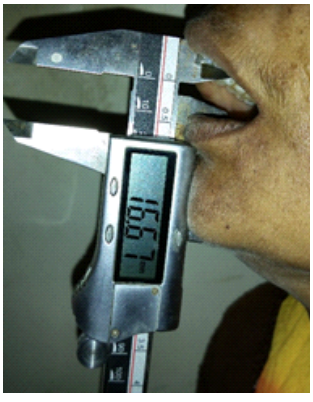

Figure 3. Reduced mouth opening

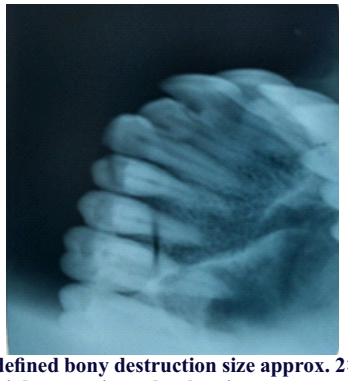

Figure 4. Ill-defined bony destruction size approx. 2×2cm present irt maxillary right posterior palatal region

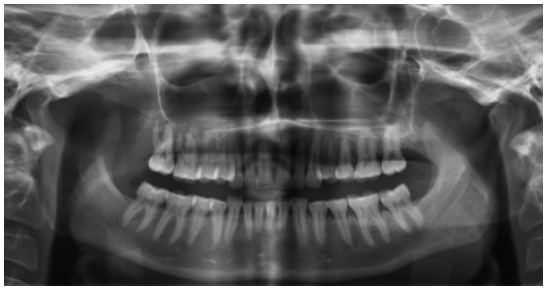

Figure 5. Ill-defined bony destruction size approx. 2×2cm present irt maxillary right posterior teeth region irt 16,17,18 with discontinuity of floor of maxillary sinus.

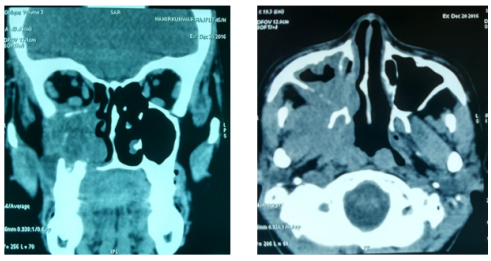

Figure 6 & 7 CECT Shows mildly enhancing mixed density lesion size of 5×4 cm present in maxillary right sinus causing erosion and destruction of anterior, postero-lateral and medial wall of right maxi sinus with extension into retroantral fat pad with involvement of medial and lateral pterygoid muscle and extension into nasopharynx.

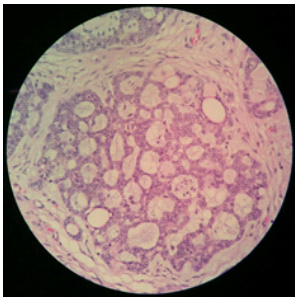

Figure 8. Shows 40x view H & E stained section shows basaloid cells arranged in glandular pattern and swiss cheese like appearance. Few true glandular lumens are seen which are lined by low cuboidal cells having eosinophilic cytoplasm.

REFERENCES

1. Vidyalakshmi, S., & Aravindhan, R. (2014). Adenoid cystic carcinoma of the buccal mucosa: a case report with review of literature. Journal of Clinical and Diagnostic Research: JCDR, 8(3), 266.
2. Chundru, N. S. V., Amudala, R., Thankappan, P., & Nagaraju, C. D. (2013). Adenoid cystic carcinoma of palate: A case report and review of literature. Dental research journal, 10(2), 274.
3. Agrawal, A., Ghatage, D., Patil, S., Chaudhary, M., Gawande, M., Hande, A., ... & Lohiya, P. ADENOID CYSTIC CARCINOMA: CASE REPORT.
4. Naik, K. L., Shetty, P., & Hegde, P. (2013). Adenoid cystic carcinoma of buccal mucosa with extensive hyalinization: A unique case report. Annals of Tropical Medicine and Public Health, 6(5), 571.
5. Singh, S., Jain, J., Pathak, S., & Singh, K. T. (2010). Adenoid cystic carcinoma of buccal mucosa. Journal of maxillofacial and oral surgery, 9(3), 273-276.
6. Dalirsani, Z., Mohtasham, N., Pakfetrat, A., Delavarian, Z., Ghazi, A., Rahimi, S. A., & Anaraki, Z. (2016). Adenoid Cystic Carcinoma of the Buccal Mucosa with Rare Delayed Frontal Bone Metastasis: A Case Report. Journal of Dental Materials and Techniques, 5(4), 208-212.
7. Godge, P., Sharma, S., & Yadav, M. (2012). Adenoid cystic carcinoma of the parotid gland. Contemporary clinical dentistry, 3(2), 223.
8. Thorawat, A., Shetty, P. K., & Tarakji, B. (2016). Minor salivary gland carcinoma of hard palate with CT findings-report of a case. Journal of clinical and diagnostic research: JCDR, 10(8), ZJ10.
9. Mahajan, A., Kulkarni, M., Parekh, M., Khan, M., Shah, A., & Gabhane, M. (2011). Adenoid cystic carcinoma of hard palate: A case report. Oral Maxillofac Pathol J, 2, 127-31.
10. Jaso, J., & Malhotra, R. (2011). Adenoid cystic carcinoma. Archives of pathology & laboratory medicine, 135(4), 511-515.