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Case Report

Cribriform Variant of Adenoid Cystic Carcinoma of the Hard Palate: A Case Report

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ABSTRACT: Adenoid cystic carcinoma (ADCC) is an uncommon malignant tumor of the salivary glands, often involving the palate. It is notorious for its indolent yet infiltrative behavior, characterized by perineural invasion and late recurrences. Its presentation may overlap with benign odontogenic lesions, creating diagnostic dilemmas.

A 40-year-old male presented with a 6-month history of swelling, pain, and tooth mobility in the left maxillary region, accompanied by occasional bleeding. Computed tomography revealed a multilocular hypodense lesion extending from the left maxilla to the palate with bone destruction. Radiological impression suggested an odontogenic tumor. Histopathological examination of an incisional biopsy confirmed adenoid cystic carcinoma, cribriform type. The patient underwent partial maxillectomy with adjuvant radiotherapy.

ADCC in the minor salivary glands can radiographically resemble ameloblastoma or other multilocular lesions, leading to potential misdiagnosis. Histopathology remains the cornerstone for diagnosis, grading, and prognosis determination. Radical surgical excision with tumor-free margins combined with postoperative radiotherapy offers the best control. Long-term surveillance is essential due to its unpredictable late metastasis.

Palatal lesions with multilocular radiolucency warrant histopathological confirmation before definitive management. Early detection and multidisciplinary treatment planning are vital for improving survival outcomes.

Keywords: Adenoid cystic carcinoma, Hard palate, Minor salivary gland, Cribriform, Maxilla

INTRODUCTION

Adenoid cystic carcinoma (ADCC) is a malignant salivary gland neoplasm first described by Theodor Billroth in 1856 as "cylindroma.". [1-3] The term ADCC was introduced by Ewing in 1954. It represents approximately 5–10% of all salivary gland neoplasms and accounts for 2–4% of malignant head and neck tumors. Minor salivary glands, particularly the palate, are affected in about 31% of cases, followed by the submandibular and parotid glands.[2-4]

ADCC commonly affects middle-aged adults with a slight female predominance. It is characterized by slow growth but aggressive biological behavior due to its high recurrence rate and tendency for perineural invasion. Histologically, it presents in three patterns: cribriform, tubular, and solid, graded as follows:[1-3]

Grade I: tubular and cribriform without solid components

Grade II: cribriform with <30% solid areas

Grade III: predominantly solid pattern.

Due to its indolent course and paucity of early symptoms, diagnosis is often delayed, and patients

frequently present with advanced-stage disease associated with poor prognosis.[3,4]

CASE HISTORY

A 40-year-old male presented with a 6-month history of pain and swelling over the hard palate, associated with mobility of teeth 21 to 26 and occasional bleeding from the palatal region at night. The patient had a significant history of tobacco chewing for three years but no history of loss of consciousness, vomiting, ENT bleed, seizures, or amnesia. On general examination, Glasgow Coma Scale was 15/15, and facial symmetry was maintained. Mouth opening and temporomandibular joint movements were within normal limits, with no evidence of facial nerve palsy. Local examination revealed gross facial swelling involving the maxillary region, although symmetry was preserved.

Laboratory investigations including hemoglobin (16.2 g/dl), TLC (8600/mm³), bleeding time (2 min), clotting time (6 min), prothrombin time (14 sec), INR (1.0), and serum creatinine (1.04 mg/dl) were within normal limits. HIV status was non-reactive.

On computed tomography (CT) of the face, an ill-defined hypodense, multilocular soft tissue lesion was observed involving the left maxillary sinus and nasal cavity, with extension into the palate. The lesion exhibited a multilocular appearance with associated bony involvement. Based on these imaging features, a provisional diagnosis of ameloblastoma or an odontogenic tumor was suggested. Ameloblastoma typically manifests as a slow-growing, expansile, multilocular radiolucent lesion, often described as having a "soap-bubble" or "honeycomb" appearance on imaging. While it predominantly affects the mandible, involvement of the maxilla has also been reported.

Based on clinical and radiological findings, a provisional diagnosis of an aggressive hard palate lesion was made. Differential diagnoses considered included ameloblastoma, odontogenic keratocyst, central giant cell granuloma, and maxillary sinus carcinoma with palatal extension, considering the patient's tobacco habit. Ameloblastoma was strongly suspected due to the multiloculated radiolucent pattern and soap-bubble appearance. Further confirmation by incisional biopsy and histopathological examination was advised which revealed Grade 1 adenoid cystic carcinoma, cribriform pattern (Figure 1). No evidence of peri-neural invasion. There was maxillary bone involved by the tumor. The patient was then subjected to partial maxillectomy with planned adjuvant radiotherapy followed by subsequent prosthetic rehabilitation.

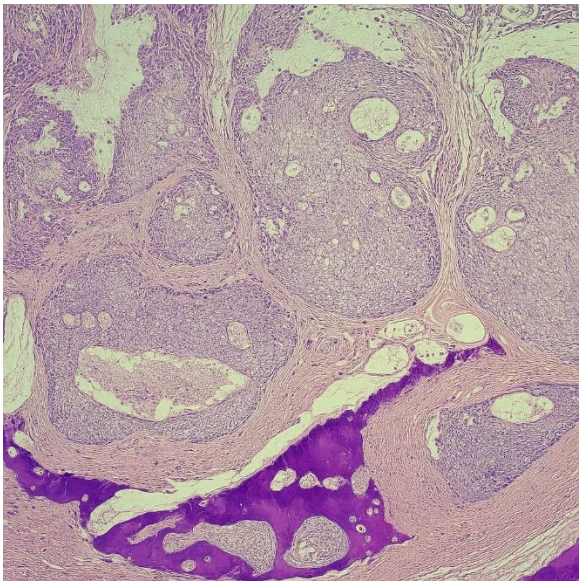

Figure-1: Microphotograph of biopsy-Histopathological section showing cribriform pattern of adenoid cystic carcinoma (H&E stain, 40×).

DISCUSSION

ADCC is a malignant salivary gland tumor with distinct histopathological subtypes influencing prognosis. The cribriform variant, as in this case, is the most common and generally exhibits a better prognosis than the solid type.[3,4] Despite its slow growth, ADCC is notorious for perineural invasion, leading to local recurrence and delayed distant metastasis, particularly to the lungs and bones.[5]

Radiological imaging such as CECT and MRI are essential for assessing tumor extent, bone involvement, and planning surgical resection.[6] However, imaging lacks specificity in distinguishing ADCC from benign odontogenic tumors, as highlighted by the initial provisional diagnosis in this case. Histopathological evaluation remains the 'gold standard' for definitive diagnosis.[7]

Management of palatal ADCC involves wide surgical excision with negative margins, often requiring partial or total maxillectomy, followed by adjuvant radiotherapy.[8] Chemotherapy has limited efficacy and is primarily used in palliative settings.[9]

Prognostic factors include tumor stage, histological subtype, margin status, and presence of perineural or lymphovascular invasion.[5,7] The cribriform pattern, early diagnosis, and combined modality treatment in this patient favor a better prognosis, though long-term follow-up remains crucial due to the risk of late recurrence.

Emerging molecular insights, such as MYB–NFIB gene fusion and expression of CD117 (c-KIT), have been identified in ADCC and may offer future therapeutic targets.[10]

Differential Diagnosis in Histopathology:
Histopathologically, cribriform adenoid cystic carcinoma (ADCC) should be differentiated from polymorphous adenocarcinoma (PAC), basal cell adenocarcinoma, mucoepidermoid carcinoma, and pleomorphic adenoma.[11,12] PAC often demonstrates a more uniform cellular morphology with perineural invasion but lacks the true cribriform pseudocystic architecture of ADCC.[13] Basal cell adenocarcinoma may contain basaloid cells but shows peripheral palisading and does not exhibit the hyaline globules typical of ADCC.[14] Mucoepidermoid carcinoma is characterized by mucous, epidermoid, and intermediate cells, which are absent in ADCC.[15] Pleomorphic adenoma, though benign, can mimic ADCC in small biopsies but is generally encapsulated with chondro-myxoid stroma.[16] Immunohistochemistry (CD117/c-KIT, MYB, p63) can help confirm ADCC and exclude its histological mimics.[17]

CONCLUSIONS

Cribriform variant of adenoid cystic carcinoma of the hard palate can present as a slow-growing lesion mimicking benign odontogenic tumors, leading to potential diagnostic delay. Accurate diagnosis requires clinicoradiological and histopathological correlation. Early radical surgical excision with adjuvant radiotherapy remains the mainstay of treatment to reduce recurrence risk and improve long-term outcomes.

REFERENCES

1. Datta M, Sinha R, Sarkar S, Shome S. Adenoid cystic carcinoma of palate: A unique case report. J Indian Acad Oral Med Radiol. 2022;34(1):116–9.

2. Adnane S, El Bouhairi M, El Khalifa K, Yahya IB. Adenoid Cystic Carcinoma of Palate: A Case Report and Literature Review. Open Access Library Journal. 2024 Jul 4;11(7):1-7.

3. Lakshmi JS, Thomas BT, Jose DS. Case report on adenoid cystic carcinoma of palate with review of literature. Indian J Case Reports. 2022;8(6):182-184.

4. Bharti JN, Dey B, Dey P, Yadav S, Rajwanshi A. Cribriform adenoid cystic carcinoma of minor salivary gland: A cytological and histopathological correlation. Diagn Cytopathol. 2017;45(3):234–7.

5. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, et al. WHO Classification of Head and Neck Tumours. 4th ed. Lyon: IARC; 2017. p.149–50.

6. Kapoor C, Ohri N, Vaidya S, et al. Adenoid cystic carcinoma of hard palate: Case report with emphasis on diagnostic approach. Saudi J Health Sci. 2015;4(1):65–70.

7. Lloyd S, Yu JB, Wilson LD, et al. Determinants of survival in adenoid cystic carcinoma of the head and neck: A population-based analysis. Am J Otolaryngol. 2011;32(5):441–7.

8. de Lima Flavio DE, Aro K, Ekanayake K, Budu V, Mäkitie AA, et al. Clinicopathologic features of adenoid cystic carcinoma of minor salivary glands: A ten-year single-center retrospective study. Eur J Surg Oncol. 2023;49(4):1023–30.

9. Xu Z, Majumder D, Jaber MA, Wieckiewicz M, et al. Adenoid cystic carcinoma of the minor salivary glands of the retromolar pad region: A rare case report. Oral Dis. 2024;30(2):350–4.

10. Laurie SA, Ho AL, Fury MG, Sherman E, Pfister DG, et al. Systemic therapy in the management of adenoid cystic carcinoma of the head and neck: Current and future directions. Cancer Treat Rev. 2023;117:102536.

11.Seethala RR, Stenman G, Nagao T, Skalova A, El-Naggar AK, Lewis JS, et al. Polymorphous adenocarcinoma vs adenoid cystic carcinoma: Histological distinctions. Head Neck Pathol. *2015;9(2):123–31.*

12. Skalova A, Stenman G, Simpson RHW, Hellquist H, Slouka D, Lewis JS, et al. Differential diagnosis of salivary gland carcinomas: An update. Virchows Arch. *2018;472(5):653–64.*

13. Cheuk W, Chan JKC, Fletcher CDM, Thompson LDR, Michal M, Kuo TT, et al. Polymorphous low-grade adenocarcinoma: A clinicopathologic study. Am J Surg Pathol. 2012;36(3):343–50.

14. Barnes L, Eveson JW, Reichart P, Sidransky D, He Y, Skalova A, et al. Basal cell adenocarcinoma of the salivary glands. Histopathology. 2011;59(5):776–84.

15. Goode RK, El-Naggar AK, Grandis JR, Takata T, Slootweg PJ, Nagao T, et al. Mucoepidermoid carcinoma: Diagnosis and prognosis. Mod Pathol. *2017;30(S1):S102–S115.*

16. Speight PM, Barrett AW, Slootweg PJ, Triantafyllou A, Stenman G, Skálová A, et al. Pleomorphic adenoma: Diagnostic challenges and pitfalls. Oral Oncol. *2018;82:132–9.*

17. Bell D, Hanna EY, Weber RS, El-Naggar AK, Stenman G, Skalova A, et al. Immunohistochemical differentiation of adenoid cystic carcinoma: Role of CD117 and MYB. Mod Pathol. *2013;26(8):1041–7.*

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