Adenoid Cystic Carcinoma of the Palate Involving Maxillary Sinus- A Case Report

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Abstract

Adenoid Cystic Carcinoma accounts for nearly 10% of all salivary gland neoplasms. The majority of tumors arise in the minor salivary glands (60%). The lesion is most common in 5Th to 6th decade and is rare in individuals younger than 20 years of age. The tumor usually appears as a slow growing mass. Pain is a common and important finding, occasionally occurring early in the course of the disease.

A 17 year old female presented with pain and swelling in the left maxillary posterior region. CT scan showed large expansile soft tissue mass present in the left maxillary sinus causing bony destruction of the inferior orbital wall, alveolar process of the maxilla and posterior wall of the maxillary sinus. On incisional biopsy, sections showed basaloid tumor cells arranged in small groups, cords and sheets interspersed with basophilic material. No cellular atypia and mitotic figures were evident. Immunohistochemistry showed positivity for cytokeratin. FMA, calponin and a diagnosis of low grade myoepithelial tumor was made following which hemimaxillectomy was done. Histopathology revealed tumor cells arranged in tubular, cribirifonn and solid patterns with perincural and intraneural invasion. A final diagnosis of high grade ACC was made and a high dose of radiation therapy was recommended.

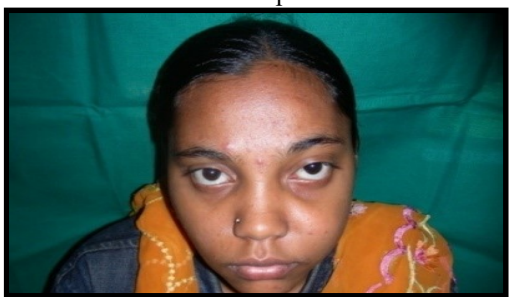
Keywords: Adenoid cystic carcinoma (ACC), cribriform pattern, perineural invasion, epithelial membrane antigen (EMA).

Introduction

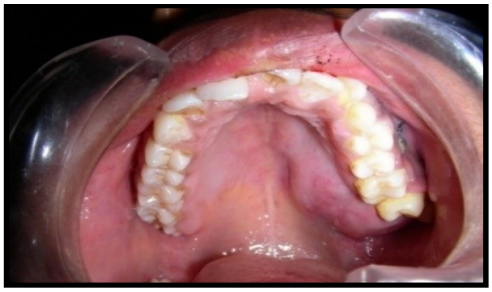
Adenoid cystic carcinomas (ACCs) account for approximately 10% of all salivary gland neoplasms. The majority of tumors arise in the minor salivary glands (60%) most frequently in the palate. They occur more commonly in females and in 5th to 6th decade of life and is rare in individuals younger than 20 years of age. The tumor usually appears as a slow growing mass. Pain is a common and important finding, occasionally occurring early in the course of the disease.[1, 7, and 8]

Case Report

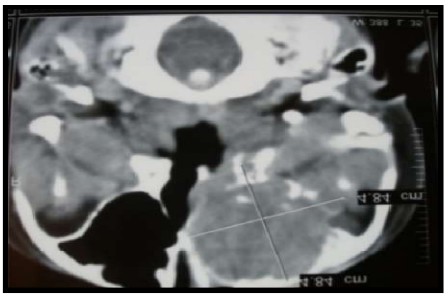
We report a case of 17 year old female who presented with pain and swelling in the left maxillary posterior region. CT scan revealed a large expansible soft tissue lesion in the left maxillary sinus. Medially the lesion was extending into the left nasal cavity and causing its obstruction and right sided deviation of the nasal septum. Superiorly the tumor was extending into the orbit and causing expansible erosion/destruction of the inferior orbital wall. Inferiorly lesion was extending in oral cavity by causing destruction of the alveolar process of maxilla and hard palate and displacing left 2nd molar laterally. Posteriorly the lesion was causing destruction of the posterior wall of maxillary sinus and extending in infratemporal fossa and anteriorly subcutaneous extension was present.



Extra oral view

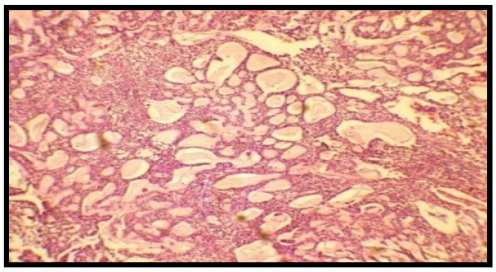


Intraoral view

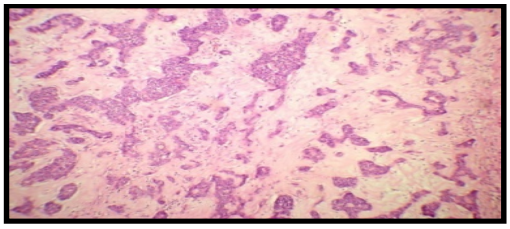


CT scan

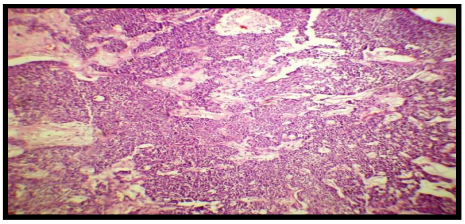
On incisional biopsy, sections showed basaloid tumor cells arranged in small groups, cords and sheets interspersed with basophilic material. No cellular atypia and mitotic figures were evident. Immunohistochemistry showed positivity for cytokeratin, E.MA, calponin and a diagnosis of low grade my epithelial tumor was made following which hemimaxillectomy was done. Histopathology revealed tumor cells arranged in tubular, cribriform and solid patterns with perineural and intraneuralinvasion. A final diagnosis of high grade ACC was made and a high dose of radiation therapy was recommended.



Cribriform pattern



Tubular pattern



Solid pattern

Discussion

Adenoid cystic carcinomas (ACCs) comprise of 20-25% of all salivary gland tumors, with a predilection for occurrence in the palate. It occurs mainly in fifth or sixth decade of life and slightly more common in females. It is characterized by slow growth, diffuse invasion and potential to produce distant metastases, mainly to the lungs and bones. In contrast to its usual presentation, our case occurred in a young patient showing rapid growth in a short duration, with extensive involvement of the maxillary sinus and its surrounding structures [7].

Histopathologically, we noted a predominant solid pattern with perineural and intramural invasion in our case and previous studies suggest that tumors with such a presentation have a poor clinical behaviour [8]. As compared to other cancers of the head and neck surgical excision of ACC is difficult due to its proximity to important neural and vascular structures. Therefore, radiotherapy is recommended as an adjunct to surgical resection. Patients with advanced-stage ACC have higher rates of distant metastasis even after receiving postoperative radiotherapy, so distant metastasis is still the main problem in the management of ACC [11].

The presented case had the following peculiarities:-

1. Age of presentation at less than 20 years.
2. Rapid growth and short clinical course, highlighting its high grade nature.
3. The relatively innocuous biopsy finding, prompting it to be labelled as low grade neoplasm.
4. The presence of all three patterns of ACC, laying emphasis on multiple sampling of excision specimen for accurate histotyping.

Conclusion

The solid variant of ACC is the most aggressive form of this neoplasm. Successful treatment and patient survival are related to the histological grade, tumor location, size and the early diagnosis of the lesion. This is an unusual presentation of a tumor in a young patient with rapid growth and short duration of presentation. These findings suggested an unfavourable prognosis for the patient. Hence a close follow up was recommended in this case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the

patient has given her consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest: There are no conflicts of interest

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