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CASE REPORT

Adenoid Cystic Carcinoma of Maxillary Sinus: An Usual Tumor at Unusual Site

Margaret Theresa J, Jeyashambavi J, Senthil Kishore, Swapna N, Jawahar R

Abstract

Adenoid cystic carcinoma is a relatively uncommon tumor of malignant behavior arise in the maxillary sinus. The tumor usually occurs in the salivary gland. Adenoid cystic carcinoma is a slow growing mass associated with pain. Female preponderance is noted. Microscopically the tumor exhibits various growth patterns tubular, cribriform and solid pattern. Tumor is composed of dual type of cell population, ductal epithelial cells and myoepithelial cells that line the aforementioned ducts. Immunohistochemistry plays a vital role in diagnosis of Adenoid cystic carcinoma.

Key Words

Adenoid Cystic Carcinoma, Maxillary Sinus, Cribriform Pattern, Ductal Epithelial Cells, Abluminal Myoepithelial Cells

Introduction

Adenoid cystic carcinoma (AdCC) is a malignant tumor. Incidence of adenoid cystic carcinoma inparanasal sinus is uncommon, accounting 0.3- 1.0% of all paranasal sinus tumors.[1] Adenoid cystic carcinoma was first documented in 1859 by Bill Roth.[2] Tumor presents as a slow, indolent growing mass, which commonly involves the salivary glands. The tumor is more common from 5th to 6th decades.[3] Female to male ratio is approximately 3:2.[4] Clinical presentations are nasal discharge, nasal obstruction and tenderness. Tumor is locally invasive with the evidence of perineural spread.[5] High rate of recurrences is documented in adenoid cystic carcinoma.

Case Report

42 years old male presented with the complaints of nasal discharge associated with mild nasal obstruction since one month. Aggravation of nasal obstruction associated with pain over the maxillary sinuses was noted since one week. Physical examination revealed subcutaneous swelling around the right maxillary sinus. Cervical lymph node was not palpable. Computed tomography showed a polypoidal, heterogenous, hyperintense mass occupying the supero- posterior aspect of the right maxillary sinus measuring 24 x 22 x 17 cms, causing erosion of the roof of orbit and postero-lateral wall. Patient had no history of allergy, chronic respiratory illness and metabolic disease. Patient was not on any medication. Clinical and radiological examination were in favor of malignancy in maxillary sinus. Under local anesthesia biopsy was obtained from the right maxillary sinus mass and sent for histopathological examination. Grossly we received 2 soft tissue fragments, the largest measuring 2 x 2 x 1.5 cms and the smallest measuring

Department of Pathology, Aarupadai Veedu Medical College and Hospital, Vinayaka Mission's Research Foundation (Deemed to be university), Kirumampakkam, Puducherry, India.

Correspondence to: Dr. J. Margaret Theresa, Associate Professor Department of Pathology, Aarupadai Veedu Medical College and Hospital, Vinayaka Mission's Research Foundation (Deemed to be university), Kirumampakkam, Puducherry-607402, India.

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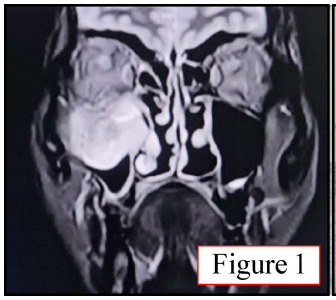


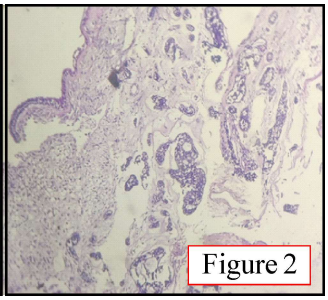
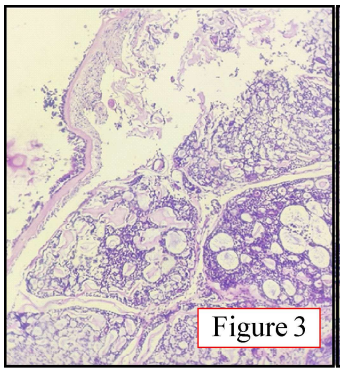


Figure 1: Shows the radiological image (Computed tomography) a polypoidal, heterogenous, hyperintense mass occupying the supero-posterior aspect of right maxillary sinus.

Figure 2: Shows the respiratory epithelium, sub-epithelium shows invasive basaloid neoplasm.



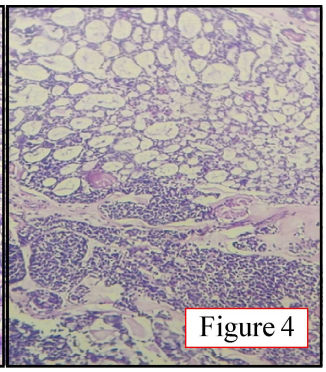


Figure 3: Shows the basaloid tumor cells arranged in various growth patterns-cribriform, tubular pattern and solid pattern.

Figure 4: Shows the tumor cells composed of ductal (luminal) and myoepithelial (abluminal) cells in a myxohyalinestroma.

2x1x1cm, which was gray white to gray brown in color. Microscopic examination revealed respiratory epithelium, subepithelium showed an invasive basaloid neoplasm. The basaloid cells were arranged in various growth patterns cribriform (50%), tubular pattern (30%) and solid pattern (20%) composed of luminal and abluminal cells in a myxohyaline matrix. Some of the tubules showed mucoid and hyaline material. No evidence of vascular, perineural and bony invasion was seen. Special stain Periodic Acid–Schiff (PAS) was performed and showed positivity for hyaline material. Histopathological examination revealed to be Adenoid cystic carcinoma, Grade II in right maxillary sinus.

Immunohistochemical markers, including CD117 (c-KIT) highlighting the luminal epithelial cells and S100 Highlighting the myoepithelial cells were perormed and showed disffuse postivity, supporting the diganosis of Adenoid cystic carcinoma.

Discussion

Adenoid cystic carcinoma is a malignant neoplasm. It is frequently found in salivary glands such as the parotid, palate, and submandibular glands.[6]Literature shows documentation of Adenoid cystic carcinoma in the maxillary sinus is seldom.[7] Female predominance is identified. Adenoid cystic carcinoma manifests as a painful, slow growing and expanding tumor mass. Facial nerve palsy is one of the complications of adenoid cystic carcinoma which was reported in few patients.[8] Grossly the tumor is unencapsulated, solid, gray-white to gray-brown mass and firm in consistency. Microscopic features display a variety of growth patterns like cribriform, cylindrical, solid, and tubular in pattern. Cribriform pattern is the most common growth pattern whereas solid pattern is least common. Adenoid cystic carcinoma is composed of two types of cells, ductal epithelial cells and abluminal myoepithelial cells. Tumor cells exhibit homogeneity in their chromatin distribution, oval, basophilic nuclei and sparse eosinophilic cytoplasm. Adenoid cystic carcinoma is characterized by a “swiss cheese” appearance and is comprised of transformed myoepithelial cells with a solid and pseudocystic region in a hyalinized stroma.[9]The tumor usually shows pseudocystic spaces filled with myxoid or hyaline material. Adenoid cystic carcinoma is graded based on the growth pattern, Grade I tubular pattern, Grade II cribriform pattern and Grade III solid growth pattern. Mixed pattern is also common is adenoid cystic carcinoma, However if the tumor exhibits more than 30% of solid pattern it is classified as solid variant of AdCC.[9] It is observed that the solid pattern is aggressive in their behavior. Adenoid cystic carcinoma shows a high tendency for perineural invasion. Special stains like PAS and Alcian blue can be performed to show positivity for basal laminar components.[10]Adenoid cystic carcinoma exhibits various morphological patterns, hence it is essential to differentiate from other malignant tumors like Polymorphous low-grade adenocarcinoma, Basaloid squamous cell carcinoma and Epithelial and myoepithelial carcinomas. Immunohistochemical markers provide a strong evidence in confirming the adenoid cystic carcinoma. Markers likecytokeratins including CK5/6, epithelial membrane antigen and C-Kit are known to highlight the epithelial cells. Myoepithelial cells are showcased by p40, p63, SOX10 and S100 protein.[10]

Conclusion

Adenoid cystic carcinoma is common in salivary glands. Incidence of adenoid cystic carcinoma in paranasal sinus is a rare entity. we report a case of adenoid cystic carcinoma in paranasal sinus, which was supported by radiological finding and confirmed with immunohistochemistry.

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