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CLINICAL REPORT

Common Tumor in an uncommon location: Pleomorphic Adenoma of nasal cavity – a case Report

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Abstract
Pleomorphic adenoma (PA) is the most common benign salivary gland tumor of which parotid gland involvement is the most common. PA may arise from minor salivary glands as well, however, PA is very rare in the sinonasal and nasopharyngeal areas. It usually affects middle aged females. They are frequently misdiagnosed due to high cellularity and myxoid stroma, leading to delay in diagnosis & further appropriate management. Here we present a case report of a female who presented with gradually progressive nasal obstruction, on examination found to have a nasal mass in right nasal cavity. Imaging was done and the nasal mass was excised. Histopathological report revealed a PA.

Common tumor in an uncommon location: Pleomorphic adenoma of the nasal cavity – a case report.

Keywords Pleomorphic adenoma · Nasal mass · Nasal obstruction · Parotid tumor · Myxoid tumor · Mixed tumor

Introduction

Pleomorphic adenoma is the most common benign salivary gland tumor. Salivary gland neoplasms are uncommon and make up less than 5% of all head and neck neoplasms. 1 About 60% of these tumors arise from the Parotid gland. PA is very rare in the sinonasal and nasopharyngeal areas, with the majority of cases occurring more frequently in women in their third to sixth decades of life. Highly cellular and few myxoid stroma cause frequent misdiagnosis, leading to delay in diagnosis and further appropriate management. Here is a case presented as gradually progressive nasal obstruction with intranasal mass in a middle-aged female diagnosed to be a rare presentation of pleomorphic adenoma in the nasal cavity.

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Case Report

A 57-year-old female, presented with complaints of unilateral (R) sided nasal obstruction of 03 years duration, which was insidious in onset, persistent, and gradually progressive and associated with a nasal mass in the (R) nasal cavity. No history of pain, bleeding from the nose, nasal discharge, recurrent sneezing, or facial pain. No history of pain in the ear, diminished hearing, or ear discharge. No other complaints.

On examination, Anterior rhinoscopy revealed a mass filling the right nasal cavity and impinging over the septum(Fig. 2), causing a broadening of the external nasal framework on the (R) (Fig. 1). The mass was firm to hard in consistency and was bleeding on probing. The left nasal cavity appears normal. No Paranasal sinuses tenderness. The scope could not be pass beyond the mass on Diagnostic nasal endoscopy.

CECT Neck and PNS revealed a well-defined homogenously enhancing soft tissue lesion measuring $24 \times 15 \times 23$ mm noted in the anterior part of the right nasal cavity inferior to the middle turbinate, displacing the nasal septum towards the left side(Fig. 3) and extending into the lateral wall of the nose. No erosion of any bony nasal septum and turbinates.

Biopsy of the nasal mass (R) nasal cavity was inconclusive.

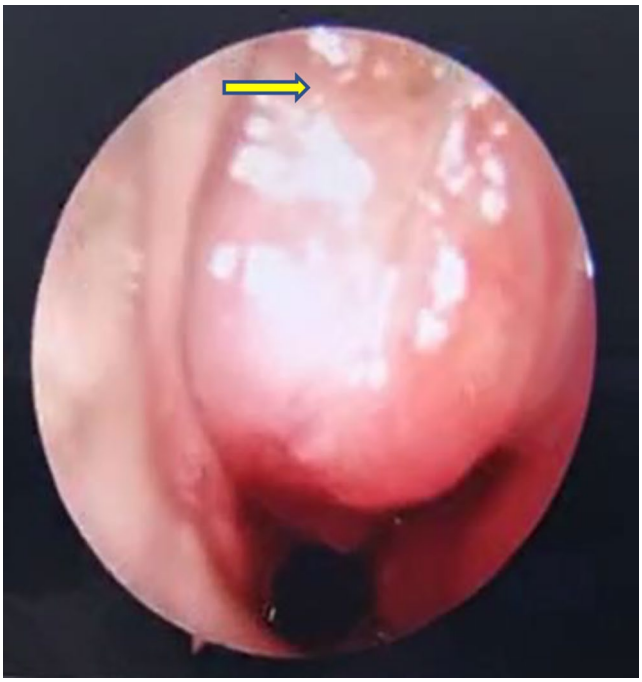

Fig. 2 Soft tissue lesion in right nasal cavity present in middle meatus area(arrow)

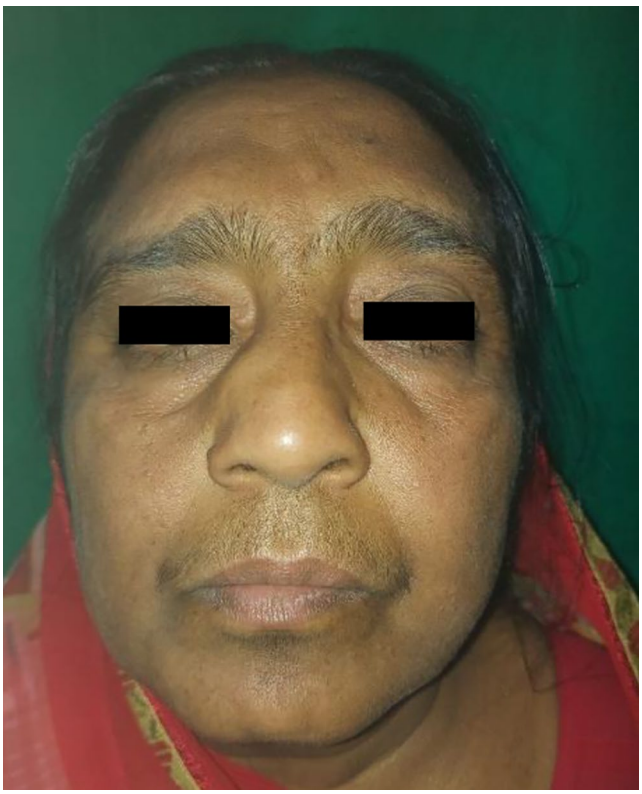

Fig. 1 Broadening of the nose over the right side

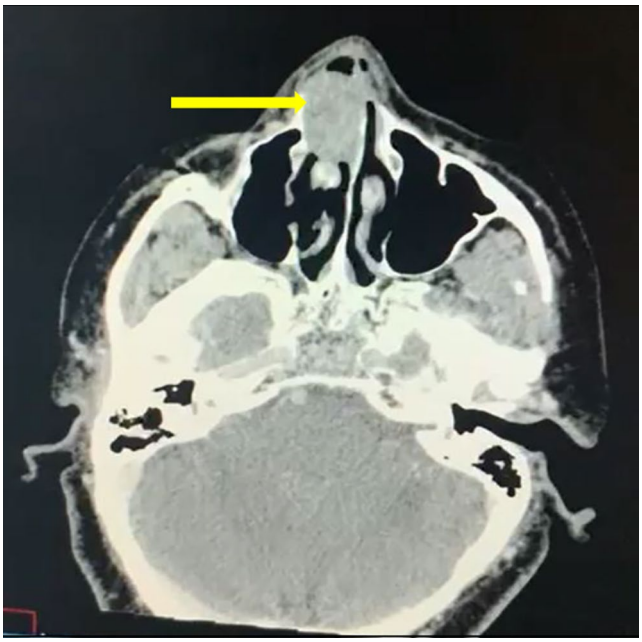

Fig. 3 CECT Neck & PNS a Well-defined homogenously enhancing soft tissue density measuring 24 × 16 × 23 mm was noted in the anterior aspect of the right nasal cavity (Marked arrow)

The tumor was resected completely by endoscopic approach under GA. The mass was found attached to the anterior part of the middle turbinate, the medial part of the inferior turbinate, and (R) of the cartilaginous septum. After confirming its attachment the mass was cauterized at the base and resected completely.

The HPE showed features of Intranasal pleomorphic adenoma with myoepithelial proliferation(Fig. 4). IHC was done which showed P63, CK 7, & vimentin positive. S100 positive in myoepithelial cells and negative in ductal component, Ki67 is < 5%. Desmin and CD 34 were negative. Over 06 months follow up there were no signs of local recurrence.

Discussion

Pleomorphic adenoma or mixed salivary gland tumor is a benign tumor, arising mainly in the major salivary glands (65%), especially in the parotid and, less frequently, in accessory salivary glands (35%). Rare cases have been reported in the lip, the hard and soft palate, the lacrimal gland, and the external auditory canal. It is extremely rare to find these in the respiratory tract. The incidence is even lower in the upper respiratory tract, such as the nasal cavity and nasopharynx. 2 Denker and Kahler were the first to report a case of pleomorphic adenoma in the nose. 3 Intranasal PA is so uncommon that there are only three reports

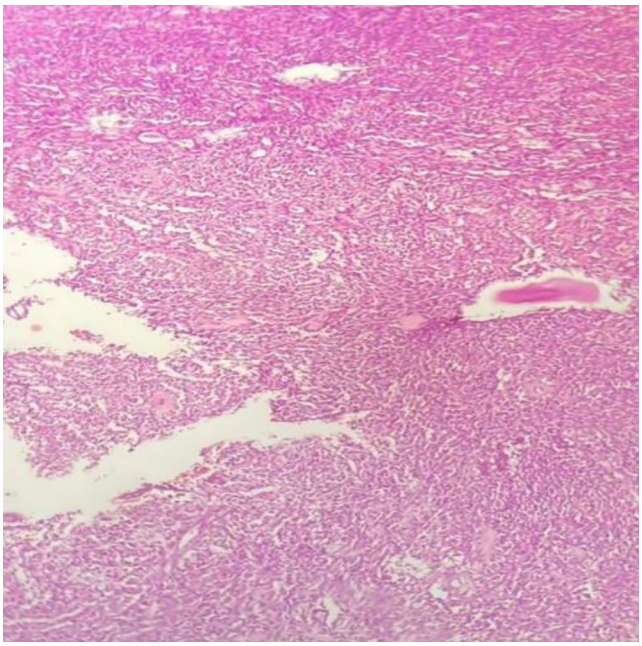

Fig. 4 Histopathological appearance of pleomorphic adenoma with myoepithelial proliferation

of statistically sizable series presented by Compagno and Wong, 4 Suzuki et al. 5 and more recently by Spiros et al. 2.

PA of the nasal cavity typically presents with complaints of unilateral nasal obstruction and epistaxis seen in 71 and 56% respectively and other complaints include nasal swelling, epiphora, and mucopurulent rhinorrhoea. 4 PA is usually a slow-growing painless mass, therefore clinical symptoms only appear after a long silent period and when tumor mass reaches a relatively large size, an external swelling of the nasal pyramid may be present as is seen in this case. On examination, PA appears as a smooth, pink-pale lobulated mass and soft to firm in consistency. The absence of ulceration and lack of invasion of surrounding structures suggest a benign nature of the mass. 5 Hence imaging may help in identifying the extent of the lesion, and invasion of surrounding structures. Differential diagnosis includes both benign and malignant tumors such as nasal polyps, papillomas, angiofibroma, osteomas, squamous cell carcinoma, adenocarcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, melanoma, and olfactory esthesioneuroblastoma 6.

Histopathological examination with Immunohistochemistry(IHC) provides a definitive diagnosis and differentiate benign from malignant lesion. Intranasal PA is different from major salivary gland PA as they contain higher cellularity and epithelial component with a low stromal component and these tumors also lack a capsule. 7.

The histological diagnosis of PA can be confirmed by immunohistochemical staining for positive expression of cytokeratins, Vimentin, S100 protein, smooth muscle actin (SMA), and glial fibrillary acidic protein (GFAP) the "mixed" nature of neoplasms, namely, mesenchymal and epithelial lines. Malignant changes are targeted by, overexpression of the p53 protein, HER-2, and proliferation marker Ki-67 (MIB-1) in areas of PA. 8.

The main risk of PA is malignant transformation and local recurrence after resection. After surgical resection recurrence rate ranges from 0 to 8%, with multiple recurrences increasing the risk of malignancy. 9 The risk of malignant transformation is 6% (2) and is estimated at 1.5% within 5 years in the absence of resection. Risk factors for recurrence are predominantly myxoid stroma, an irregular or invaded capsule, and multinodularity. 10 This case is managed by Endoscopic resection of the tumor with tumor negative margins as it is the treatment of choice nowadays which have the advantage of less morbidity, hospital stay, avoiding external scar, and unnecessary excessive resection. 11.

Conclusion

PA is very rare in accessory salivary glands, they are rarer in the nasal cavity where they generally originate from the nasal septum. The malignant transformation potential of PA should not be underestimated. There is also a chance of recurrence after resection with an increased chance of malignant transformation with each recurrence. Hence appropriate early diagnosis, complete tumor resection with tumor negative margins, and prolonged follow-up is therefore mandatory.

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Declarations

Conflict of Interest Nil.

Ethical Approval NA.

Research Involving Human Participants and/or Animals Nil.

Informed Consent Informed consent was taken from the patient for publishing the case report.

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