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Case Report

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PLEOMORPHIC ADENOMA AS NASAL MASS: A RARE CASE REPORT

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ABSTRACT

Pleomorphic adenoma is the most common benign tumor of the major salivary glands with highest incidence in the parotid gland. They may also be seen arising from the minor salivary glands present in the upper aerodigestive tract. Other rare sites for the occurrence of pleomorphic adenoma are the larynx, pharynx, paranasal sinuses, nasal cavity and lacrimal glands. Here we report a case of intranasal pleomorphic adenoma arising from the nasal septum causing nasal obstruction which was clinically diagnosed as nasal mass and excision biopsy was done. Histopathology confirmed the diagnosis of pleomorphic adenoma.

KEYWORDS: Pleomorphic adenoma, nasal mass, histopathology.

INTRODUCTION

Pleomorphic Adenoma (PA) is a slow growing benign tumour of salivary glands, found most commonly in the parotid gland followed by submandibular gland and minor salivary glands. In minor salivary glands, they are seen mainly in the hard palate and soft palate followed by upper lip and lacrimal gland. 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, maxillary sinus and nasopharynx. Mostly intranasal pleomorphic adenomas are found in the septum, few are found in the lateral wall or turbinate. Definitive diagnosis of the entity is made by histopathological examination.

PA usually occurs in the third to sixth decade of life and are more common in women. Etiology of PA is unknown but studies have shown that it has an epithelial origin and comprises cells from epithelial and mesenchymal differentiation. Chromosome abnormalities with aberrations of 8q12 and 12q15 have been described in the literature. For most patients, the chief complaint is a unilateral nasal obstruction and/or occasional epistaxis. The recommended treatment for PA is wide local excision of the growth, the periosteum and involved bone. A recurrence rate of 2 – 44 % after excision has been reported in studies. The risk of the PA becoming malignant is about 1.5% when the duration is less than 5 years but the risk increases to 9.5% when the duration increases to more than 15 years. We report and discuss a rare case of pleomorphic adenoma as nasal mass, arising from the nasal septum of an 61-year-old female.

CASE REPORT

A 61 years old female presented to the outpatient department of ENT with complaints of right nasal obstruction and three episodes of epistaxis for the last six months duration. On examination, single pinkish fleshy mass was observed which was attached to the septum. CT Paranasal sinuses showed moderate sized expansile heterodense soft tissue lesion in the mid right nasal cavity obliterating adjacent middle meatus region with lateral deviation and erosion of medial wall of right maxillary sinus. With clinical diagnosis of right nasal mass, endoscopic nasal mass excision done and excised mass was sent for histopathological examination. We received multiple, gray white, soft tissue bits measuring 3.5cc in aggregate. Few hemorrhagic areas seen, all embedded. Light microscopy of the hematoxylin and eosin (H&E) stained sections showed fragments of stratified squamous epithelium and respiratory epithelium overlying a cellular neoplasm composed of epithelial and myoepithelial cells arranged in acini, ducts and strands in a background of pseudo-cartilagenous and hyalinized stroma. Few polypoidal lesions covered by respiratory epithelium enclosing vascular channels are also seen. The diagnosis of pleomorphic adenoma was made.

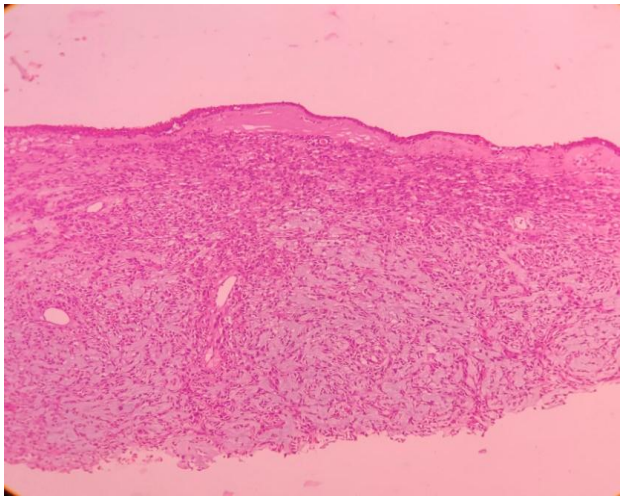

Fig. 1: Scanner view showing respiratory epithelium overlying neoplasm composed of epithelial and myoepithelial cells arranged in acini, ducts and strands in a background of pseudo-cartilagenous and hyalinized stroma.

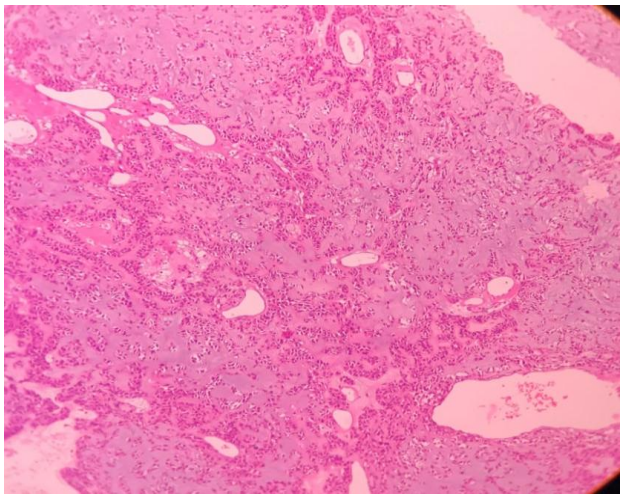

Fig. 2: Low power view showing epithelial and myoepithelial cells arranged in acini, ducts and strands in a background of pseudo-cartilagenous and hyalinized stroma.

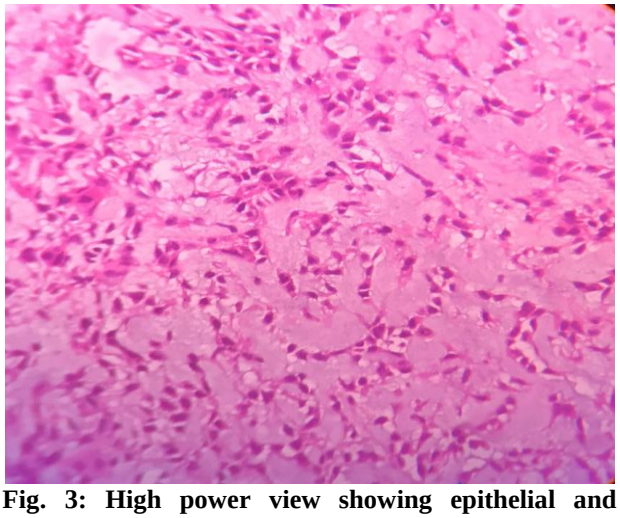

Fig. 3: High power view showing epithelial and myoepithelial cells in a pseudo-cartilagenous stroma.

DISCUSSION

Salivary gland tumors account for 2-4% of the total head and neck neoplasms. Most (70%) salivary gland tumors originate in the parotid gland. The remaining tumors arise in the submandibular gland (8%) and minor salivary glands (22%). Majority of the minor salivary gland tumors are malignant. Minor salivary gland tumors can be located almost anywhere in the upper aerodigestive tract, though the most frequently affected location is the oral cavity and particularly the palate, where the concentration of minor salivary glands is greater. Such lesions also can be found in the cheek mucosa, in the region of the retromolar trigone, the lips, the oropharynx or rarely in the nasal cavities and paranasal sinuses. The majority of intranasal PAs arise from the nasal septum (80%). PA originating from the lateral wall and inferior turbinate accounts for 9.8%–20%, although most of the minor mucous and serous glands are located in the lateral nasal wall and turbinates.

Various theories have been proposed to explain this observation. According to Stevenson, remnants of the vomeronasal organ, an epithelium-lined duct in the cartilaginous nasal septum degenerated in early foetus, could be the reason for the appearance of these tumours in this particular region. According to Ersner and Saltzman, in 1944, the precursors of the septal pleomorphic adenoma are ectopic embryonic epithelialised cells on the nasal septum mucosa, found during the migration of the nasal buds. According to Evans and Cruikshank, it originates directly from the matured salivary glandular tissue; Dawe, in1979, proposed a viral aetiology from polyoma virus.

The first reported case in the literature of a PA of the nasal cavity was in 1929. Larger studies of intranasal pleomorphic adenoma include 40 cases reported by Compagno and Wong and 59 cases reported by Wakami et al. The majority of cases occurs between the third to sixth decades of life and is seen more frequently in women. This finding is similar to our study. The main presenting feature is painless, unilateral nasal obstruction, a mass within nasal cavity and epistaxis. In our study also patient presented to the outpatient department with the complaint of unilateral nasal obstruction with epistaxis.

Histologically, Pleomorphic adenomas of the nasal cavity contain both epithelial and mesenchymal components. Epithelial component produces glandular or ductal pattern. Myoepithelial cells are responsible for the production of abundant, extracellular matrix with chondroid, collagenous, mucoid, and osseous stroma. Immunohistochemical expression of cytokeratin and S-100 are used to confirm the diagnosis. PA associated with other features such as squamous metaplasia and keratin pearl formation could be mistaken for mucoepidermoid carcinoma and squamous cell carcinoma, both being malignant tumors.

A neoplasm originating from the nasal septum has a higher risk of malignancy compared to other sites in the nose. Malignant transformation of pleomorphic adenoma of the nasal cavity has been reported in 2.4 to 10% of cases. The most common variant is carcinoma ex pleomorphic adenoma which has potential to metastasise. A less common form of malignant transformation is the malignant mixed form. The clinical features, such as absence of superficial ulceration, no bleeding either on touch or spontaneously and lack of invasion of surrounding structures suggest a benign nature of the mass. Compared with pleomorphic adenomas of the parotid, intranasal lesions are associated with greater cellularity (more epithelial components), a more benign course overall, and a lower rate of recurrence.

Differential diagnosis of intra-nasal pleomorphic adenoma includes both malignant and benign tumours such as squamous cell carcinoma(the most common intra-nasal malignancy), adenocarcinoma, adenoidcystic carcinoma, mucoepidermoid carcinoma, melanoma, olfactory esthesioneuroblastoma, polyps, papillomas (including inverted papilloma), angiofibromas and osteomas.

Computerized Tomography(CT) scan help in locating the tumor, it's size, tumor extension to the adjoining areas and to asses bony involvement or destruction. In CT, nasal pleomorphic adenoma appears as well-defined, homogeneous mass because of their high cellularity. Magnetic Resonance Imaging (MRI) is used to assess the epithelial and stromal components of the neoplasm, as well as the surrounding soft tissue. MRI of the stromal components reveals a low signal intensity on T1-weighted imaging and an intermediate to high signal intensity on T2-weighted imaging; epithelial components have a low signal intensity on T2-weighted imaging.

The recommended treatment for PA is wide local excision of the growth, the periosteum and involved bone. PA is an encapsulated tumor with the capsule showing varying thickness and at times these tumors show lateral extension into the capsule. Since PA has a high risk of implantability, capsule rupture and incomplete excision would result in recurrence. Studies have shown a recurrence rate of about 2-44%, thereby requiring proper follow up.

CONCLUSION

Pleomorphic Adenomas are slowly progressive, benign tumors but show wide diversity in morphological features which may challenge pathologists. Their occurrence in the minor salivary glands in unusual locations, should be kept in mind as these can be mistaken for other non-neoplastic to neoplastic lesions. This also make surgeons aware of pleomorphic adenoma, as a differential diagnosis of swelling in nasal cavity, because in the management, surgical treatment should consist of complete excision of the mass with clear margin and with a long term follow-up, both endoscopic and radiologic, to exclude malignancy which is mandatory, even if the tumour appears to be clinically benign and resected completely.

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