

Case Report

A classic triphasic pleomorphic adenoma of nasal cavity mimicking as malignancy: a rare case report

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ABSTRACT

Pleomorphic adenoma (PA) is the most common benign tumor of major salivary glands and intra nasal PA is unusual which may be misdiagnosed as malignancy. A 65-year-old female presented with unilateral nasal bleeding and nasal obstruction in right nasal cavity for 6 months. Local examination showed polypoid mass which surgically excised. HPE showed classic triphasic component of epithelial, myoepithelial with cartilaginous stromal elements and pathological diagnosis of PA made. High cellularity of tumor and predominant squamous epithelial component with keratin pearl created diagnostic confusion of malignancy and it confirmed by immune-histochemistry using p63 to demonstrate presence of myoepithelial component. Highlighted this case report for its unusual location and rare triphasic component of tumor tissue with keratin pearl formation which mimic as malignancy.

Keywords: PA, Nasal mass, Minor salivary glands, P63

INTRODUCTION

Pleomorphic adenoma (PA) is a mixed benign tumor composed of epithelial and mesenchymal components. It arises commonly from the major salivary glands especially in parotid (70%), and rarely from the minor salivary glands (6-7%) that are seen in nasopharynx, pharynx, larynx, hypopharynx, lacrimal glands and nasal cavity (0.4%).¹ Cases have been reported in lip, hard and soft palate, lacrimal gland and external auditory canal.² Here we report a case of classic triphasic PA arising from sero mucinous glands of nasal mucosa which is presented clinically as bleeding nasal polyp.

CASE REPORT

A 65-year-old female presented with complaints of nasal obstruction in the right nasal cavity with unilateral nasal bleeding and for 6 months. These symptoms were worsened during episodes of upper respiratory tract infection, touching the mass, and exposure to cold. The patient was a known asthmatic patient and used inhalers

on episodes. The patient had a past history of oral mass excision from the floor of the mouth 20 years ago and histopathology of that mass was not known. Anterior rhinoscopy examination revealed a polypoid mass covered with blood crust in the right nasal cavity, as well as hypertrophy of the inferior turbinate in the left nasal cavity. Surgical excision was done with a clinical diagnosis of nasal polyp for evaluation. Gross examination showed well defined polypoid mass of size 2×1.5×1cm size and cut surface showed solid gray-white, gray-brown and glassy areas (Figure 1). Histopathological examination showed well circumscribed, pseudo encapsulated tumor tissue with increased cellularity showing classic triphasic components of epithelial, myoepithelial and cartilaginous stromal components. Epithelial component showed predominantly benign squamous cell nests, clusters with keratin pearls, mucinous glandular cells and sheets of small band spindle myoepithelial cells which created a pathological diagnostic confusion of malignancy (Figure 2). But the presence of pseudo encapsulation (Figure 3), lack of cellular atypia, presence of benign cartilaginous stromal elements favor diagnosis of PA (Figure 4).

Additionally, immunohistochemical confirmation of myoepithelial component by p63 justified pathological diagnosis of PA in this unusual site (Figure 5).



Figure 1: Excised nasal polypoid mass.

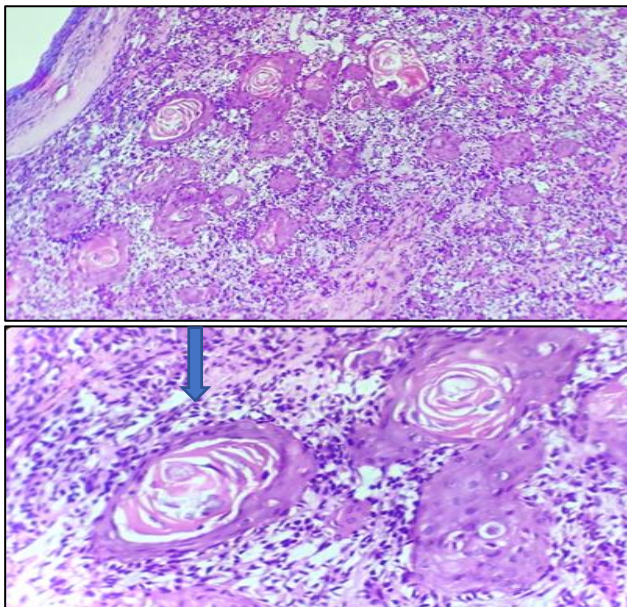


Figure 2: Nests of squamous cells with keratin pearl (Arrow)-hematoxylin and eosin stain x400.

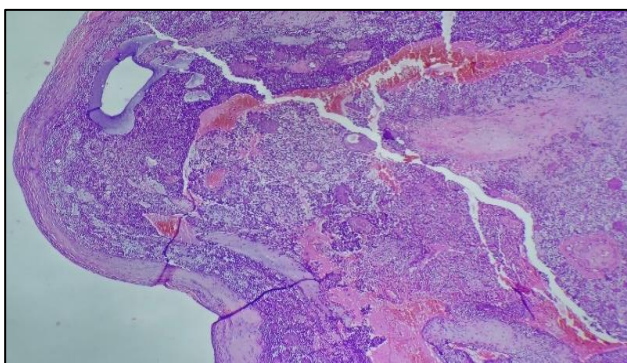


Figure 3: All components of pleomorphic adenoma with capsule-hematoxylin and eosin stain x400.

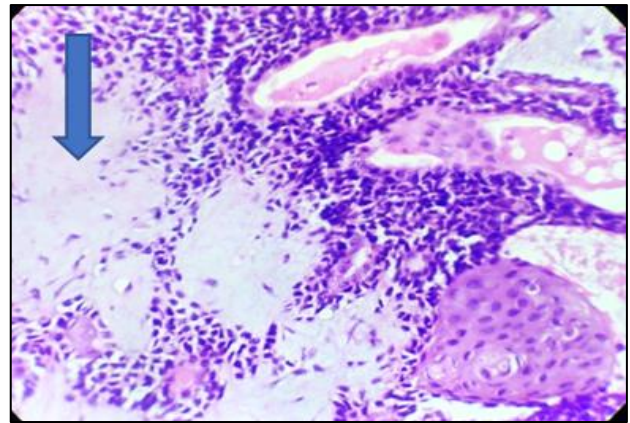


Figure 4: Cartilaginous component (Arrow)-hematoxylin and eosin stain x400.

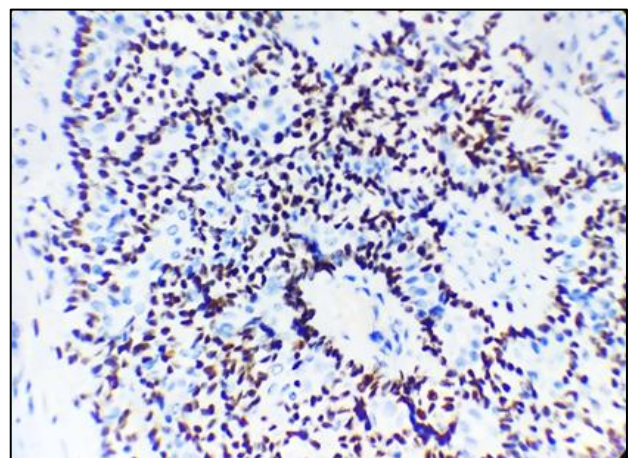


Figure 5: P 63 positive myoepithelial component-immunohistochemistry x400.

DISCUSSION

PA is the most common benign salivary gland tumor. It can arise from parotid glands, submandibular glands or other minor salivary glands. It can also rarely occur in the upper respiratory tract via the minor salivary glands (6-7%).¹ In upper respiratory tract, few cases have been reported in hard palate and lips, tongue in the oral cavity and nasal cavity followed by maxillary sinus and nasopharynx.² Nasal PAs (0.4%) most often arise from the nasal septum followed by lateral nasal wall. Intranasal PA mostly occur at the age of 30-60 years and the incidence is more common in females.³

Most common clinical presentation in unilateral nasal obstruction is epistaxis. Other signs and symptoms are nasal mass, epiphora and mucopurulent rhinorrhoea.⁴ Intra nasal mass has many differential diagnosis and it can be either benign or malignant. Differential diagnosis of benign intranasal mass includes inverted papilloma, squamous papilloma, schwannoma, PA and chondroma. Malignant intranasal mass includes squamous cell

carcinoma, malignant melanoma, lymphoma, cartilaginous tumors as differential diagnosis.³

Our case is 65-year female with chronic unilateral nasal obstruction and nasal bleeding in right nasal cavity. Histopathological examination of resected nasal mass revealed the diagnosis of PA. PA is a circumscribed tumor characterized by its pleomorphic or mixed appearance consisting of proliferation of epithelial and spindle shaped myoepithelial cells in variable stroma. In our case, there is predominant epithelial component composed of benign squamous cell nests, clusters with keratin pearls, mucinous glandular cells and sheets of small bland spindle myoepithelial cells and scanty benign cartilaginous stromal elements. The pathogenesis of keratin pearl formation in nasal PA is exposure of the minor salivary gland duct epithelium to various sources of irritants.⁵ Histologically, PA in the upper respiratory tract may resemble aggressive epithelial tumors due to their high cellularity and absence or presence of minimal stromal component. It is important to note that this histological feature is distinct from that observed in the major salivary glands, which exhibit lower myoepithelial cellularity. In our case there is predominant squamous epithelial component along with keratin pearl which created a pathological diagnostic confusion of squamous malignancy. In addition, mixed components of squamous cells, mucinous cells and myoepithelial spindle cells which can be interpreted as intermediate cells can be mistaken as Mucoepidermoid carcinoma. But the presence of cartilaginous stromal elements in the tumor contradicts the diagnosis of malignancy. So p63 marker was used to identify the myoepithelial component for confirming the diagnosis. The histopathological diagnosis of PA in an unusual site of nasal cavity was confirmed by immunohistochemistry.

The chance of malignant transformation of PA is about (2-6%), and 2.4% cases of nasal PA have been reported to undergo malignant transformation.^{4,6} Occasionally, PA can present as a malignant mimic, the most common variant being carcinoma ex PA which has a potential to metastasis. The predominant metastatic site is bone, but spread to lungs, regional lymph nodes and liver has been documented.⁷ Therefore, margins of surgical resection should be carefully identified and examined. Wide local resection with histological clear margin is generally agreed as the treatment of choice for benign salivary gland tumors.⁸ This case of nasal PA is reported for its rare site, peculiar morphology and its chance for misdiagnosis as malignancy.

CONCLUSION

In summary, PA s are rare tumors of the nasal cavity and often can be misdiagnosed as malignancy clinically. In addition, unusual morphology distinct from those of

salivary gland origin like high cellularity of the tumor, predominant squamous epithelial cell component with epithelial pearls can mislead Pathologists to misdiagnose this as malignancy in histopathological evaluation too. So clinical correlation, careful evaluation of specimen with adequate sampling, margin status and if necessary immunohistochemical confirmation are needed to avoid over diagnosis and aggressive management.

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