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A case report of a rare nasopharyngeal myoepithelial dominant pleomorphic adenoma

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ABSTRACT

INTRODUCTION: Pleomorphic adenoma is the most common benign salivary gland neoplasm. The majority of cases occur in the major salivary glands; however, they can also originate from the minor salivary glands. The nasopharynx is an uncommon site, but it has been reported in the literature. The characteristic difference of our report from the literature is the young age of our patient and his previous unremarkable medical or surgical history.

The objective of this report is to present a case of salivary gland pleomorphic adenoma, discuss radiological and histopathological findings and treatment options.

CASE PRESENTATION: Herein we report a case of a 25-year-old male patient presenting with a long-term history of snoring, mouth breathing, and progressive left nasal obstruction. After examining the patient and confirming the diagnosis, patient successfully underwent tumor resection using a combined transnasal/transoral endoscopic approach with no complications following surgery and significant improvement of the previously reported symptoms.

DISCUSSION: Pleomorphic adenoma of the minor salivary glands can occur anywhere throughout the distribution sites of these glands such as: along the upper aerodigestive tract, parapharyngeal fat spaces, soft palate, the sinonasal, and nasopharyngeal areas.

CONCLUSION: The occurrence of pleomorphic adenoma in uncommon sites has been reported in the literature, and the nasopharynx is considered to be one of these uncommon sites. The mainstay of treatment for nasopharyngeal pleomorphic adenoma is surgical excision as they can grow to giant sizes if left untreated.

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1. Introduction

Pleomorphic adenoma is the most common benign salivary gland neoplasm [1]. The majority of cases occur in the major salivary glands, however; they can also originate from the minor salivary glands which can be found anywhere throughout their distribution sites such as along the upper aerodigestive tract and in the parapharyngeal fat spaces [2]. Histologically pleomorphic adenoma has components of both epithelial and mesenchymal cells with a wide variety of histopathological appearance due to difference in proportion between the two cell types. one variant of pleomorphic adenoma is myoepithelioma which is characterized by myoepithelial elements and lack of ductal structures and is composed of three cell types: plasmacytoid, spindle, and clear cells [3]. and in our

case, the tumor was almost pure plasmacytoid myoepithelioma (Fig. 3).

This Case report has been written in line with the 2020 SCARE Criteria [4].

2. Case report

A 25-year-old male, medically free. Was referred to our otolaryngology clinic at a tertiary care academic hospital with complaints of snoring, mouth breathing, and progressive left nasal obstruction. These symptoms have been occurring for the past ten years with no history of nasal bleeding or ear fullness. The endoscopic examination revealed an approximately 4 × 4 cm pedunculated smooth mass, covered with normal mucosa, not fragile, occupying the entire nasopharynx and extending to the soft palate. The rest of the examination was unremarkable. The patient underwent a computed tomography angiogram (CTA) (Fig. 1) and magnetic resonance imaging (MRI) (Fig. 2) scan which showed

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Fig. 1. CT angiogram, sagittal view showed a hypo-vascular nasopharyngeal mass that is extending to the level of the soft palate, not associated with the aggressive appearing of the bone destruction.



Fig. 2. MRI, sagittal view showed a nasopharyngeal mass extended to the level of the soft palate.

hypovascular mass filling the nasopharyngeal airway, extended to the level of the soft palate, not associated with the aggressive appearing of bone destruction. Biopsy under local anesthesia could not be done as the patient refused. The patient underwent surgical resection by a team of rhinologists and the nasopharyngeal tumor was removed entirely with the pedicle through a transnasal endoscopic approach combined with a transoral approach releasing the adhesions of the mass from the soft palate. A microscopic histopathological examination of the tumor revealed that it was a myoepithelial dominant pleomorphic adenoma and was almost pure myoepithelioma (Figs. 3–8). At one year follow-up after surgery, the patient was examined in the clinic and reported significant improvement of his previously symptoms. The nasopharynx was disease-free, apart from mild secretions.

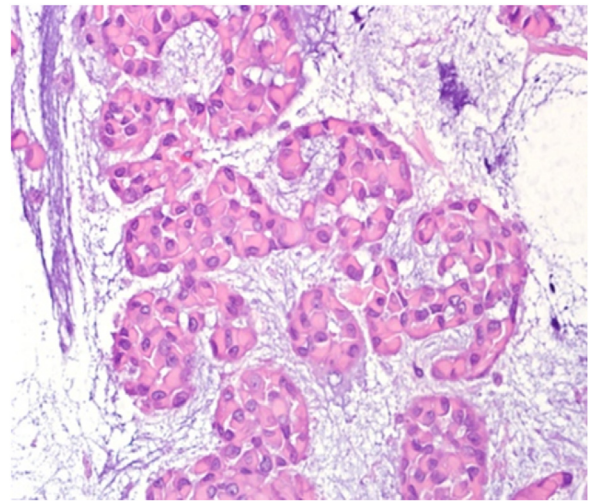


Fig. 3. 40X H&E showing plasmacytoid myoepithelial cells (which were dominant, over 95 % in the tumor).

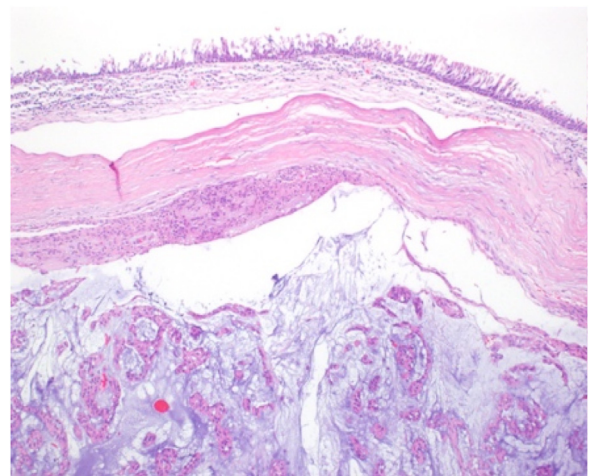


Fig. 4. 10X H&E to show surface respiratory epithelium of the nasopharynx. Subepithelial minor salivary gland tumor with myxoid matrix and plasmacytoid myoepithelial cells.

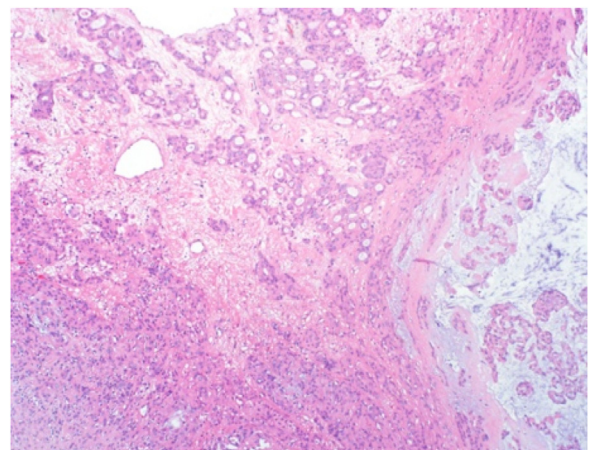


Fig. 5. 10X focal ductal differentiation in background of dominant myoepithelial component.

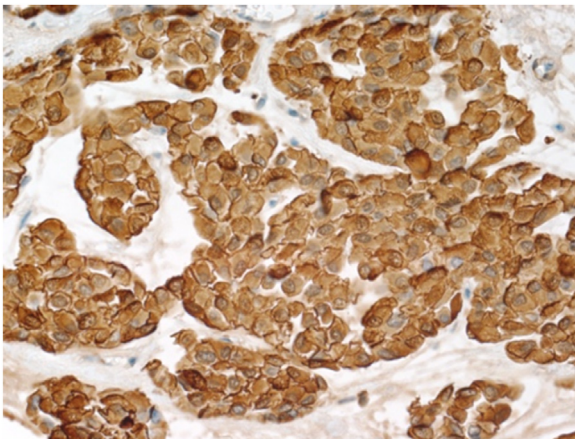


Fig. 6. 40X s100 to show diffusely strongly positive myoepithelial cells.

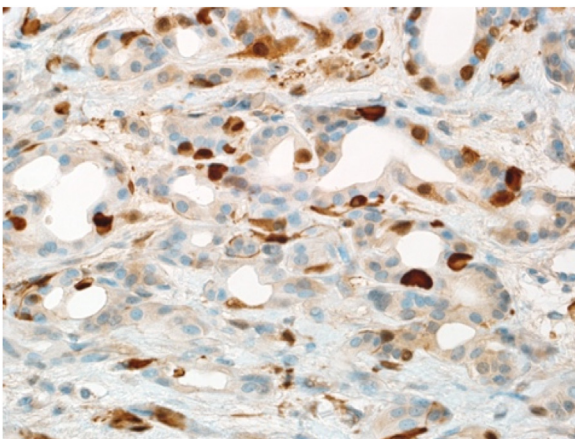


Fig. 7. 40X in the ductal differentiation area the S100 is much less positive.

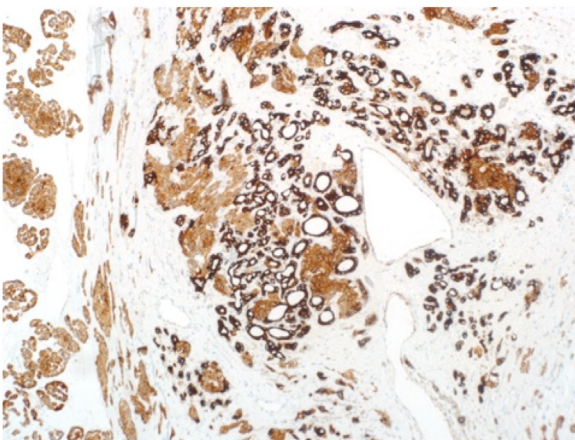


Fig. 8. 40X in the ductal differentiation area the S100 is much less positive.

3. Discussion

Pleomorphic adenoma of the minor salivary gland can arise anywhere along with the distribution sites of these glands with reports of the palate being the most common site (10 %), followed by the lips (4%), larynx, sinonasal spaces, and epiglottis [3]. In another case series of 39 patients with pleomorphic adenoma of the sinonasal spaces and skull base, the authors reported that the lateral nasal wall and nasal septum were the most common locations, followed by the nasopharynx [5]. A different study reported

that the most common primary site for pleomorphic adenoma of sinonasal and nasopharyngeal spaces is the nasal septum, followed by the lateral wall of the nasal cavity and nasopharynx [6]. Pleomorphic adenoma of minor salivary glands is more common in females compared to males. Moreover, it was found that pleomorphic adenoma was common during the 3rd to 6th decades [7]. Radiological imaging cannot certainly distinguish between benign and malignant salivary neoplasms; However, it is worth mentioning that imaging plays an important role in the diagnostic workup of nasopharyngeal tumors providing a roadmap to plan the next step in the management of pleomorphic adenoma patients, but only histopathological analysis has the superiority to determine the exact nature of the disease. One modality to further investigate superficial salivary gland tumors is fine-needle aspiration cytology which depends on the size and the site of the tumor however, one limitation to fine-needle aspiration is the difficulty to differentiate pleomorphic adenoma from adenoid cystic carcinoma and polymorphous low-grade adenocarcinoma [3]. Ultrasound, Magnetic resonance imaging (MRI), and computed tomography (CT) scans are commonly used as part of the assessment of pleomorphic adenoma [8,9]. Ultrasound may be used depending on the location of the tumor and pleomorphic adenoma typically appear hypoechoic, homogenous well-circumscribed mass with posterior acoustic enhancement [8]. On MRI, it has variable enhancement patterns that are due to the tumor myxoid and cellular composition [9], and appears either isointense or hypointense on T1, and hyperintense on T2-weighted images [10]. And on CT scan, pleomorphic adenoma typically appears as a well-circumscribed mass of soft tissue density with homogeneous contrast enhancement [11].

The mainstay of treatment for nasopharyngeal pleomorphic adenoma is surgical excision [12] as they grow to giant sizes if left untreated. Variable surgical approaches for benign nasopharyngeal tumor were mentioned, such as lateral rhinotomy, transnasal, and transpalatal approaches [13], however, the choice of endoscopic transnasal approach accompanied by transoral approach was chosen in our case due to the advantage of visualization the tumor margins at the attachment sites and the surrounding normal tissue endoscopically which can be challenging in other surgical methods [5].

4. Conclusion

Majority of pleomorphic adenoma arise from the major salivary glands but tumors arising from the minor salivary glands have been reported as well. The mean age of presentation of pleomorphic adenoma is during 4th to 6th decades of life however, in our case the patient was a 25-year-old with unremarkable past medical and surgical history. Surgical excision is the mainstay of treatment for pleomorphic adenoma as they can grow to giant sizes if left untreated.

Declaration of Competing Interest

The authors report no conflict of interest.

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Ethical approval

This study has been approved by Research Advisory council at King Faisal Specialist hospital in Riyadh, Saudi Arabia (RAC# 2200308).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

1. Kholoud AlAmari: First author, writing and editing – original draft, data collection and finalized the manuscript.
2. Abdullah M. Zahlan: Writing – review, editing and finalizing the manuscript for submission.
3. Esam Albawardi: Writing and reviewing manuscript.
4. Mohammed Dababo: Writing and reviewing manuscript.
5. Naif H. Alotaibi: Corresponding author, writing – reviewing & Editing manuscript, supervising.

Registration of research studies

Not applicable.

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