

## RESEARCH ARTICLE

## Four cases of Pleomorphic Adenoma of the nasal cavity: An unusual entity

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### Abstract

Pleomorphic adenomas are the most common benign tumor of the major salivary glands. In addition, they may also occur in the minor salivary glands of the hard and soft palate. Nasal masses are often due to antrochoanal or ethmoidal polyps or angiofibromas or haemangiomas or papillomas. Intranasal pleomorphic adenomas are very unusual and may be misdiagnosed because they have greater myoepithelial cellularity and fewer myxoid stromata compared to those elsewhere. Even though pleomorphic adenomas are seen rarely in nasal cavities, we have come across 4 cases in a period of 15 months. Here we present a case series of pleomorphic adenomas of the nasal cavity, out of which three presented as a huge nasal mass, one patient presenting with epistaxis as main symptom. All the nasal masses were unilateral, vascular and were arising from the nasal septum. They were seen to be occupying the nasal cavity, without causing any invasion of the lateral wall or the sinuses. One patient had pressure atrophy of the middle turbinate due the large nasal mass. In all patients the nasal mass was excised in toto, 3 patients through transnasal approach and 1 patient through through a lateral rhinotomy approach. All the patients were followed up regularly and no recurrence was noted in any of the cases. Histopathology revealed the nasal masses to be of mixed histology and pleomorphic adenoma was considered.

**Keywords:** Pleomorphic adenomas, benign tumor, nasal masses, haemangiomas, histopathology.

### Introduction

Salivary gland tumors constitute about 3% of all neoplasms. The majority of these tumors are benign and about 70% are pleomorphic adenomas (Compagno and Wong, 1977; Jassar *et al.*, 1999). A small minority (8%) are located in the oral cavity, neck and nasal cavity. Nasal masses originating from the septum are usually due to squamous papilloma, inverted papilloma, haemangioma or granulomatous diseases. Several benign lesions of the septum such as leiomyoma, osteochondroma and transitional cell papilloma have been reported in literature. The other differential diagnoses may include malignant tumors such as melanoma, adenoid cystic carcinoma and squamous cell carcinoma (Batsakis, 1979). The majority of these tumors arise from the mucosa of the bony and cartilaginous septum.

Nasoseptal swell body is a discrete area of erectile tissue in the submucosa over the anterior nasal septum. In some individuals, it can present as a suspicious lesion. It does not have a significant relevance when considering the differential diagnosis in these patients given the enormous size of the septal mass. However, in smaller septal swellings, it could be given consideration. Intranasal pleomorphic adenomas are rare, benign tumors which typically grow slowly and have a propensity for recurrence after surgical resection (Compagno and Wong, 1977; Jassar *et al.*, 1999; Freeman *et al.*, 1990; Mackie and Zahirovic, 2004).

Computed tomography (CT) and Magnetic resonance imaging (MRI) appearances of intranasal pleomorphic adenomas have been described though a definitive diagnosis of the entity is made by histo-pathological examination. In this study, we present a case series of pleomorphic adenomas of the nasal cavity and their histopathological examination.

### Materials and methods

**Patients:** Totally 4 patients were examined in this study for the presence of pleomorphic adenomas in a period of 15 months in the department of ENT, Adichunchanagiri Institute of Medical Sciences. They are

1. Girijamma, 50 years, female
2. Huchappa, 60 years, male
3. Rajappa, 66 years, male
4. Ranganath, 35 years, male

**Parameters:** History of nasal discharge, facial pain, hawking sensation, headache or watery eye was thoroughly examined. General physical examination was carried out for checking any other associated disease. Rigid endoscopy, X-ray water's view and radiological examination were carried out. The nasal masses were excised through intranasal approach using endoscopic methods, in one patient a lateral rhinotomy approach. In all the four cases, the tumor was excised in toto along with the mucoperichondrium to which the tumor was attached. The excised tumor specimens of the patients were sent to histopathological examination.

**Results**

The first case was a 50 year old woman, Girijamma who presented with a 2 year history of right nasal obstruction and widening of right side of the nose. There was no history of nasal discharge, facial pain, hawking sensation, headache or watery eye. There was no history of bleeding from nose. There was no history of trauma or any previous surgery. Her weight was stable and her general health was satisfactory. Her general physical examination revealed no evidence of any other associated disease. Her external nose showed widening of dorsum of nose on right side extending to root. Anterior rhinoscopy revealed a single, smooth surfaced reddish pedunculated polypoid mass arising from the anterior part of the nasal septum (Fig. 1). Rigid endoscopy of the nose revealed a single, smooth polypoidal mass attached to the anterior part of the nasal septum without any attachments either to the lateral wall or the floor.

Fig. 1. Nasal mass of patient 1.

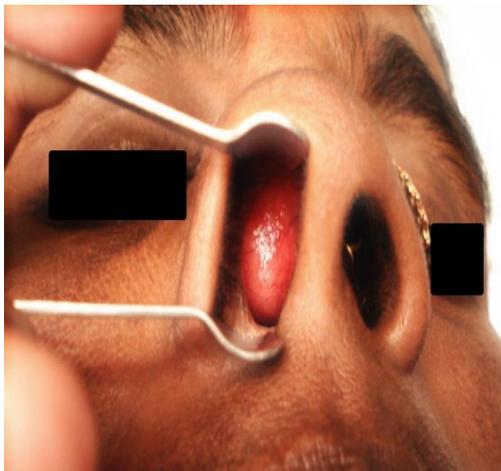


Fig. 2. Tumor specimen.

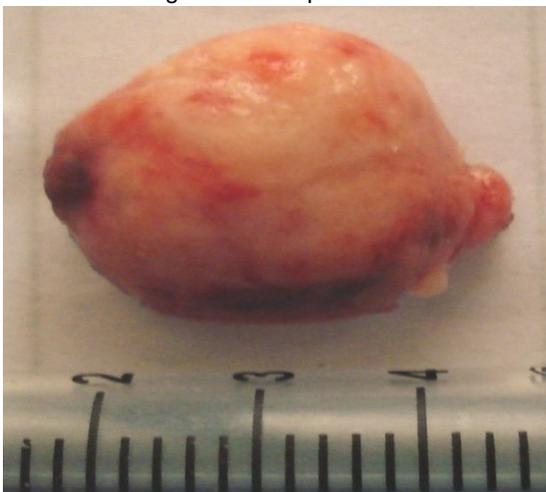
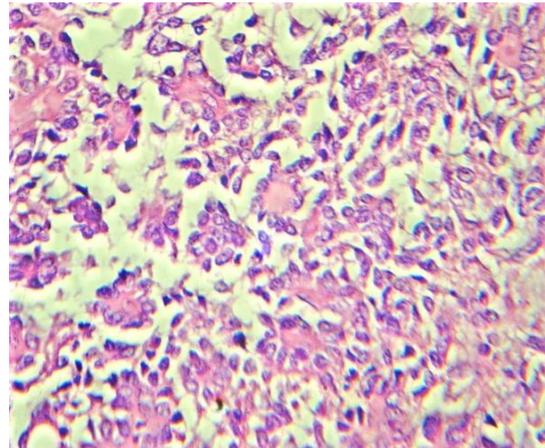


Fig. 3. Histopathology of tumor cells.



X-ray Water's view showed a soft tissue density mass in the right nasal cavity with deviation of septum to left. CT of the nose and sinuses showed a homogenous soft tissue density mass noted arising from the anterior nasal septum extending to lateral wall of nose measuring around 2.5x2x2.2 cm. The smooth surface, preservation of mucosal lining and the localised nature of the mass were consistent with a benign lesion. There was no evidence of involvement of the sinuses. Fine needle aspiration cytology was done and the report revealed pleomorphic adenoma. Under general anaesthesia complete excision of the tumor submucosally was done and the tumor was removed in toto (Fig. 2). Septal cartilage was found to be totally normal, so the cartilage was spared. As the exposed cartilage was small, mucosal grafting was not considered. The excised specimen was sent to histopathological examination which revealed the mucosal integrity as intact and lined by pseudostratified ciliated columnar epithelium to stratified squamous epithelium. It was composed of epithelial and mesenchymal elements. The mesenchymal component was composed of chondromyxoid stroma. The tumor cells were arranged in ductal/acinar pattern. The cells were round to oval with bland nuclei and scanty cytoplasm, also areas of cystic change were seen. All these features were suggestive of pleomorphic adenoma (Fig. 3). Postoperatively the patient had no problems and the septal mucosal defect closed by the growth of mucosa. The patient was followed at regular intervals and no fresh complaints were noted.

The second case was a 60-year-old man named Huchappa, who presented with a 3-year history of right nasal obstruction, right nasal mass and widening of right side of the nose. Clinical examination revealed a huge, reddish mass occupying the right nasal cavity (Fig. 4). The attachments of the mass could not be determined as the bulk of the mass was big. The mass was firm to touch and was appearing very vascular and used to bleed on touch. Posterior rhinoscopy revealed a vascular mass dropping into the nasopharynx.

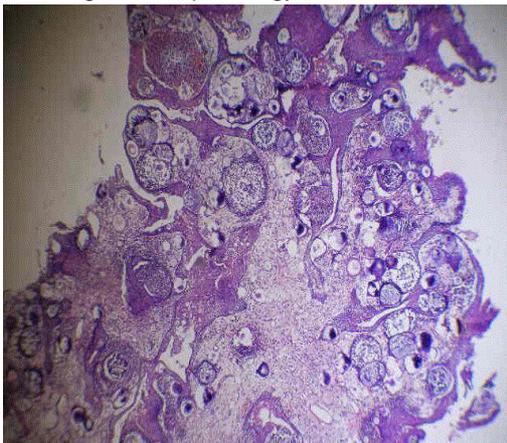
Fig. 4. Nasal mass of patient 2.



Fig. 5. Tumor specimen.



Fig. 6. Histopathology of tumor cells.



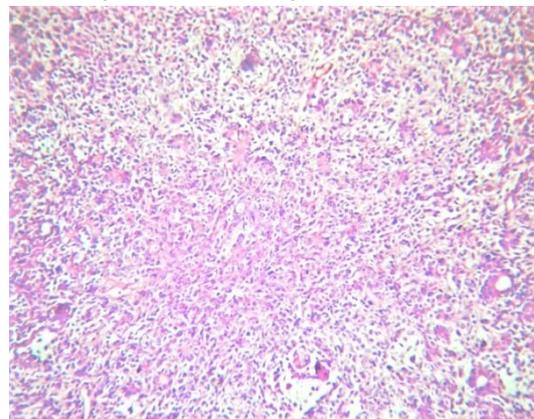
FNAC of the nasal mass was inconclusive suggesting a mixed cellularity. So a lateral rhinotomy approach was used and the mass was found to be attached only to the septum. Due to the firm attachment to the nasal septum, a piece of mucoperichondrium was removed along with the mass in toto (Fig. 5). There was no evidence of attachments or erosions of any the other structures. Only the middle turbinate was found to be atrophied due to pressure atrophy. Histopathology revealed it as pleomorphic adenoma (Fig. 6). Regular followup was done to rule out recurrence. After 3 months the mucosa had healed well and no fresh complaints were noted.

The third case was a patient named Rajappa, aged 66yrs, presenting with a huge nasal mass, epistaxis, nasal obstruction. Examination revealed an angry red mass occupying the nasal cavity (Fig. 7). Due to manipulation of the mass by the patient the mass was appearing inflamed and angry red. Also the adjacent vestibular skin was appearing inflamed. Clinically a provisional diagnosis of rhinosporidiosis was made. CT scan revealed a mass occupying the anterior part of the nasal cavity, appears to be arising from the nasal septum. There was no evidence of any erosion or involvement of the sinuses.

Fig. 7. Huge nasal mass of patient 3.



Fig. 8. Histopathology of tumor cells.



Radiological examination (CT scan) demonstrated a soft tissue mass occupying the right nasal cavity extending from the anterior nares to the choana and nasopharynx with no contrast enhancement. There was no evidence of destruction of the ethmoids or the medial wall of maxilla. There was no extension into pterygomaxillary fissure or infratemporal fossa. All the sinuses were well pneumatized and clear.

Under GA the nasal mass was excised transnasally and the base was cauterised and also the mucoprichondrium was removed at the base area and the mass was sent for histopathology. HPR revealed it as pleomorphic adenoma (Fig. 8). Follow-up was uneventful and the patient was symptom free at the end of 2 months. The fourth case was a patient named Ranganath, 35 years, who presented with 2 episodes of epistaxis. On examination, a small sessile red mass measuring 0.5 cm was seen on the anterior part of the septum (Fig. 9). Diagnostic nasal endoscopy revealed no other cause of nasal bleed and a clinical diagnosis of squamous papilloma were made and the mass was excised under local anaesthesia and sent for histopathology which turned out to be pleomorphic adenoma (Fig. 10).

Fig. 9. Small nasal mass of patient 4.

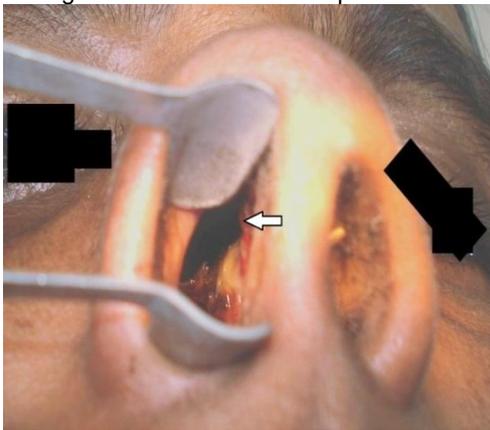
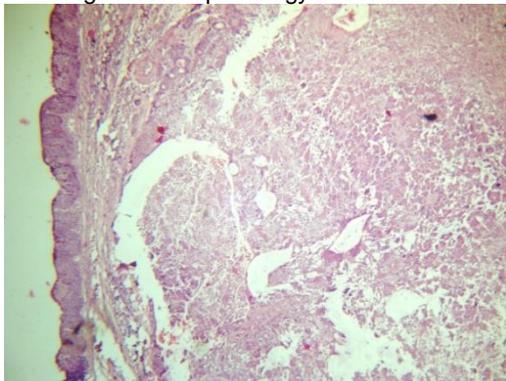


Fig. 10. Histopathology of tumor cells.



## Discussion

The most common tumors of the major salivary glands are pleomorphic adenomas, but in rare instances, they can occur in the respiratory tract (via minor salivary glands). Cases have been reported in the nasal cavity, paranasal sinuses, nasopharynx, oropharynx, hypopharynx, and larynx. In the upper respiratory tract, the most favoured site of origin is the nasal cavity, followed by the maxillary sinus and the nasopharynx. The first reported case in the literature of a pleomorphic adenoma of the nasal cavity was in 1929 (Jassar *et al.*, 1999). Although the vast majority of minor mucous and serous glands are located in the lateral nasal wall, pleomorphic adenomas in the nasal cavity mostly

originate from the nasal septum. Larger studies of intranasal pleomorphic adenoma include 40 cases reported by Compagno and Wong (1977) and 59 cases reported by Batsakis (1979). The majority of tumors presents between the age of 30 and 60 years and are slightly more common in women. Typical presenting features include unilateral nasal obstruction (71%) and epistaxis (56%). Other signs and symptoms include a mass in the nose, nasal swelling, epiphora, and mucopurulent rhinorrhoea. Pleomorphic adenomas are characterised by epithelial tissue mixed with tissues of myxoid, mucoid or chondroid appearance.

Histologically, pleomorphic adenoma of the aerodigestive tract may resemble aggressive epithelial tumors because of the high cellularity and lack of a stromal component. Importantly, this feature is not in keeping with that of the major salivary glands which demonstrate relatively reduced myoepithelial cellularity. Occasionally, pleomorphic adenomas are composed almost entirely of epithelial cells with few or no stromata. This can lead to misdiagnosis as a carcinoma. A fact reflected by Compagno and Wong (1977) wherein 55% of cases were initially not accurate. Complete resection with histological clear margin is generally agreed as the treatment of choice for benign salivary gland tumors. Postoperative radiotherapy has been advocated by some authors in circumstances where residual disease was apparent. In the case of intranasal pleomorphic adenoma, several surgical approaches have been used to achieve wide local clearance and these include intranasal, transnasal endoscopic, external rhinoplasty, lateral rhinotomy and mid facial degloving.

In their reported series of 40 patients, Compagno and Wong (1977) used the lateral rhinotomy approach for excision of tumor in the majority of the patients. Only three patients had a recurrence of disease after 3 years of follow-up. The recurrent lesions constituted more stroma than cellular elements and the former is thought to provide the focus for recurrence. The outlook for intranasal mixed tumors is better than for those in other ectopic sites, because they show early symptoms leading to an early diagnosis. Involvement of the surrounding structures such as bone is rare since the tumors have sufficient space to expand within the nasal cavity. A neoplasm originating from the nasal septum has a higher risk of malignancy compared to other sites in the nose (Mackie and Zahirovic, 2004).

Occasionally, pleomorphic adenoma can behave in a malignant fashion, the most common variant being carcinoma ex pleomorphic adenoma which has a potential to metastasise. The predominant metastatic site is bone but spread to lungs, regional lymph nodes and liver has been documented. Ten cases of metastasising pleomorphic adenoma of the parotid gland and three patients with metastatic pleomorphic adenoma of the minor salivary glands have been reported in the literature (Freeman *et al.*, 1990).

## Conclusion

In summary, pleomorphic adenomas are rare tumors of the nasal cavity. We suggest consideration of this diagnosis if the patient has unilateral nasal mass of septal origin. They have a higher epithelial and lower stromal component compared to their major salivary gland counterparts and may be misdiagnosed at an early stage leading to more aggressive treatment. Complete excision via transnasal or lateral rhinotomy approach is the treatment of choice. In view of the potential for tumor recurrence, long-term follow-up and careful examination of the nose with an endoscope are necessary.

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