



Research Paper **Medical Science**

Nasal Pleomorphic Adenoma

Dr .Shamendra kumar	assistant professor govt.medical college kota
Dr.vijay Kumar meena	associate professor govt.medical college kota
Dr. Rajkumar jain	professor govt.medical college kota

ABSTRACT Pleomorphic adenomas are the most common benign tumor of the major salivary glands. In addition to the major salivary glands, they may also occur in the salivary glands of the hard and soft palate. Rare cases have been reported in the nasopharynx, oropharynx, hypopharynx, and larynx. Intranasal pleomorphic adenomas are quite rare and may be misdiagnosed because they have greater myoepithelial cellularity and few myxoid stromata compared to those elsewhere. We present a rare case of a pleomorphic adenoma of the nasal ala and discuss the pathologic findings and clinical management. It rarely originates in the nose, and is especially rare in the lateral nasal wall.

KEYWORDS pleomorphic adenoma, lateral nasal wall, FNAC,

Introduction

Pleomorphic adenoma is a benign mixed tumor mainly arising in the major salivary glands. Rarely, they can be found in the minor salivary glands of the upper aerodigestive tract, including the nasal cavity, pharynx, larynx, and trachea. Intranasal pleomorphic adenomas are rare and nasal septum is the most common site of origin.¹⁻³ Here, we present and discuss an unusual case of pleomorphic adenoma arising from the lateral nasal wall.

Mixed tumour of the salivary gland or the pleomorphic adenoma is a benign tumour, arising mainly in the major salivary glands (65%), especially in the parotid and, less frequently, in accessory salivary glands (35%)¹. Rare cases have been reported in the lip², the hard and soft palate³, the lacrimal gland⁴, and the external auditory canal⁵. It is extremely rare to find these in the respiratory tract⁶⁻⁸. The incidence is even lower in the upper respiratory tract, such as the nasal cavity, maxillary sinus and nasopharynx⁸⁻⁹. The largest reported case series of intra-nasal pleomorphic adenomas are those of Spiros et al.¹⁰ with 40 cases, Compagno and Wong¹¹ with 40 cases and, more recently, Suzuki et al.⁴ with 41 cases.

Intranasal pleomorphic adenomas generally arise in the nasal septal mucosa (reported incidence varies between 82.5%¹¹ and 90%¹²), even though the seromucosal glands are mainly located within the lateral nasal wall, in particular in the turbinates¹³⁻¹⁴.

Various theories have been proposed to explain this observation. According to Stevenson¹⁵, remnants of the vomero-nasal 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 buds¹⁶. According to Evans and Cruikshank, it originates directly from the matured salivary glandular tissue⁶; Dawe, in 1979, proposed a viral aetiology from polyoma virus¹⁷.

CASE REPORT

A 52-year-old female presented to an outside institution with a 5-year history of right-sided nose swelling and 2-month history of repeated pain. On seeing by naked eyes revealed a smooth, reddish, not easily hemorrhagic tumor causing pressing of the right nasal cavity. We admit her in 25/4/2016 and operate on 03/5/2016. Dr. shamendra(+919414909984)s, Dr. vijay kumar meena and Dr. rajkumar jain was team members.



(Figure 1).

Her head and neck examination showed no other lesions or lymphadenopathy. On FNAC report was pleomorphic adenoma

The histology was typical of pleomorphic adenoma and comprised an admixture of epithelial, myoepithelial and stromal components with acini or ducts and a myxochondroid stroma (Figure 4). There was no invasion of the adjacent structure microscopically. The patient's postoperative course was uneventful. She healed well without evidence of residual or recurrent disease on follow-up.

Postoperative photo was



Figure 2.

DISCUSSION

Nasal pleomorphic adenoma is seen predominantly in females ⁴⁻¹¹ usually between the third and fifth decades of life ¹⁸. There is no reported correlation with occupational exposure or inhaled toxic chemical compounds. It is generally known to be a slow-growing tumour and, therefore, clinical symptoms appear after a long silent period. Patients commonly present with gradual worsening of monolateral nasal obstruction and occasional epistaxis. Less commonly, when the tumoural mass reaches a relatively large size, to that of the nasal cavity, external swelling of the nasal pyramid as well as pain may be present.

Clinically, it appears as a polypoid, unilateral, sessile, translucent, pinkish-grey mass, with smooth surface and soft consistency. Tumours can range in size from < 0.7 cm to > 7 cm.

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.

Histologically, all pleomorphic adenomas have a collagenous thin capsule, with a clear-cut distinction of the tumour tissue from the surrounding normal connective tissue. The tumours consist of three main structures: tubuloductal structure, solid area, and myxoid area. The tubuloductal structure presents ducts with double cell layers: cuboidal-shaped epithelial cells at the inner layer, and spindle-shaped myoepithelial cells at the outer. Predominantly the solid areas consist of the spindle-shaped cells with high cellularity; the myxoid areas are characterised by their low cellularity ¹⁸.

The intra-nasal pleomorphic adenoma shows a predominance of epithelial rather than stromal elements, as compared with major salivary gland tumours. The epithelial cells are small, oval-shaped and often arranged in cords; sometimes, they are organized in small acinous structures ¹³.

Immunohistochemical stains prove positive for various cytokeratins, S100 protein, glial fibrillary acid protein (GFAP), Vimentine, a smooth muscle actin (SMA). This describes the "mixed" nature of the tumour, namely, the stromal and the epithelial line ¹⁸.

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, adenoid cystic carcinoma, mucoepidermoid carcinoma, melanoma, olfactory esthesioneuroblastoma ²⁰, polyps, papillomas (including inverted papilloma), angiofibromas and osteomas.

Differential diagnosis can also be difficult in the presence of a "neuroesthesioepithelioma" (even though the site of origin of such a lesion, i.e., the ethmoid plate, should suggest its na-

ture, since the lateral nasal wall is extremely rare), as, in the early stage, it presents a small cell proliferation organized like a "rosette", positive for S100 protein. Diagnosis is possible on account of the lack of an extracellular neurofibrillar structure, neurotubules, neurosecretive granules and due to the presence of mucinous material and the rarity of malpighian lobules disseminated on the pleomorphic adenoma ²¹.

Regardless of where the lesion originates, the main treatment modality should be surgical. While complete excision of the tumour with histologically clear margins is mandatory, the surgical approach will depend upon the size, location and extension. A radical and wide resection lowers the risk of recurrence, especially when the capsule is interrupted and a direct contact with the surrounding normal tissue is present. Approaches include lateral rhinotomy ²²⁻²⁴, trans-nasal or mid-facial degloving ¹³⁻¹⁸ and intra-nasal excision. The present patient underwent endoscopic resection since the tumour was small enough to observe under the endoscope. The advantages of endoscopic resection include no external scar, less blood loss. In the presence of large masses, the mid-facial degloving approach is preferred, since it has the advantage of wide exposure of the mass and direct approach to the nasal cavity.

Recurrences are not frequent, Compagno and Wong reported 3 cases of local recurrences in 40 patients (7.5%) ¹¹, probably, as they thought, due to the amount of myxoidstroma of the tumour, which could be split into the surgical field.

The potential risk of malignant transformation of the pleomorphic adenoma is about 6% and is predominantly seen in the female patients ²⁵. The risk is increased by delay in diagnosis.

A histopathologically confirmed case with adenoid cystic and squamous carcinomatous differentiation has been reported ²⁶.

There has also been a report of metastasis to the submandibular lymph-node, in a recurrent septal pleomorphic adenoma, 17 years after the first diagnosis. Even in this case, the microscopic features of both the primary and metastatic lesion were benign. In this regard, a iatrogenic theory has been proposed. This theory suggests that the metastasis occurs as a result of incomplete excision or inadvertent disruption of the tumour with consequent spread through haematogenous or lymphatic routes. Pulmonary, hepatic and bone metastasis have also been reported ²⁷.

Long term follow-up is therefore necessary for early diagnosis of loco-regional recurrences by endoscopic examination followed by imaging (CT or MR) in case of clinical evidence of disease.

Conclusions

In the presence of a slow-growing unilateral mass of the nasal cavity, it is important to consider, among the various diagnoses, the presence of pleomorphic adenoma, even if it is not frequently encountered. Early diagnosis offers the possibility of a more complete excision with adequate care being taken not to disrupt the tumour in order to prevent local and distant spread of neoplastic cells. Long-term follow-up, to exclude malignancy is mandatory, even if the tumour appears to be clinically benign and resected completely.

References

1. Chomette G, Auril M. Histopathologie buccale et cervico-faciale. Paris: Masson; 1986. p. 210-2.
2. Trotoux L, Lefebvre B. Pathologie des glandes salivaires. Enc Med ChirOrl. Paris: Masson; 1979.
3. Campos A, Bagan JV, Zapater E, Martorell M, Basterra J. Adenoma pleomorfo de glandulasalivaraccessoria. ActaOtorrinolaringolEsp 1995;46:331-4.
4. Suzuki K, Moribe K, Baba S. A rare case of pleomorphic adenoma of lateral wall of nasal cavity, with special reference of statistical observation of pleomorphic adenoma of nasal cavity in Japan. Nippon JibiinkokaiGakkaiKaiho 1990;93:740-5.
5. Lahoz MT, Valerio J, Royo J, Yus C, Camora F. Tumormixto de conductoaudi-

- tivoexterno. *ActaOtorrinolaringolEsp* 1990;41,1:53-6.
6. Evans RW, Cruikshank AH. Epithelial tumors of the salivary glands. In: Major problems in Pathology. Vol. 1, Philadelphia: WB Saunders; 1970. p. 281-95.
 7. Fraile Rodrigo JJ, Martinez Berganza Y, Asensio R, YusGotor C, Garcia Ortin M, Garcia Ortin J. Adenoma pleomorfo de tabique nasal. Revision. *ActaOtorrinolaringolEsp* 1993;44:239-41.
 8. Wallace RD. Pleomorphic adenoma of the nose. Clinical and pathologic diagnosis. *Arch Otolaryngol Head Neck Surg* 1990;116:486-8.
 9. Kamal SA. Pleomorphic adenoma of the nose. A clinical case and historical review. *J LaryngOtol* 1984;98:917-23.
 10. Spiro RH, Koss LG, Hadju SL. Tumors of minor salivary origin. A clinicopathologic study of 492 cases. *Cancer* 1973;31:117-29.
 11. Compagno J, Wong RT. Intranasal mixed tumors (pleomorphic adenomas): a clinicopathologic study of 40 cases. *Am J ClinPathol* 1977;68:213-8.
 12. Nonomura N, Nijima H, Kimura O, Karoshi F, Nakano Y, Kimura K. Immunohistochemical study of pleomorphic adenoma of the nasal septum. *AurisNasus Larynx* 1992;19:125-31.
 13. Castello E, Caligo G, Pallestrini EA. Su uncaso di adenoma pleomorfo della fossa nasale. *ActaOtorhinolaryngolItal* 1996;16:433-7.
 14. Carriero E, Almadori G, Cadoni G. Adenoma pleomorfo del turbinato inferiore: caso clinico e revisione della letteratura. *ActaOtorhinolaryngolItal* 1997;17:377-81.
 15. Stevenson HN. Mixed Tumor of the septum. *Ann OtolRhinolLaryngol* 1932;41:563-70.
 16. Ersner MJ, Saltzman M. A mixed tumor of the nasal septum. Report of a case. *Laryngoscope* 1944;54:287-96.
 17. Dawe CJ. Tumors of salivary and lacrimal glands, nasal fossa and maxillary sinus. *IASC Sci J* 1979;23:91-113.
 18. Freeman SB, Kennedy KS, Parker GS, Tatum SA. Metastasizing pleomorphic adenoma of the nasal septum. *Arch Otolaryngol Head Neck Surg* 1990;116:1331-3.
 19. Hirai S, Matsumoto T, Suda K. Pleomorphic adenoma in nasal cavity: immunohistochemical study of three cases. *AurisNasus Larynx* 2002;29:291-5.
 20. Batsakis J. Tumor of the head and neck. Baltimore: Williams and Wilkins Publishers; 1984. p. 76-99.
 21. Couloigner V, Julien N, Molas J, Sterkers O. Adenome pleomorphe de la cloison nasale. A propos d'un cas. *Ann OtolaryngolChirCervicofac* 1993;110:230-3.
 22. Bergstrom B, Biorklund A. Pleomorphic adenoma of the nasal septum. Report of two cases. *J LaryngolOtol* 1981;95:179-81.
 23. Haberman RS, Stanley DE. Pleomorphic adenoma of the nasal septum. *Otolaryngol Head Neck Surg* 1989;100:610-2.
 24. Majed MA. Pleomorphic adenoma of the nasal septum. *J LaryngolOtol* 1971;85:975-6.
 25. Shaheen OH. Benign salivary glands tumors. *Scott Brown's Otolaryngol* 1997;5:1-18.
 26. Freeman SR, Sloan P, De Carpentier J. Carcinoma ex-pleomorphic adenoma of the nasal septum with adenoid cystic and squamous carcinomatous differentiation. *Rhinology* 2003;41:118-21.
 27. Wermuth MJ, Mann CH, Odere F. Metastasizing pleomorphic adenoma arising in the soft palate. *Otolaryngol Head Neck Surg* 1988;99:505-8.