

Recurrent Pleomorphic Adenoma of Nasal Cavity: A Case Report

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Abstract: *Pleomorphic adenoma is the most common benign tumour of the major salivary glands. They also arise from minor salivary glands located on hard and soft palate. Ectopic occurrence of pleomorphic adenoma is seen in pharynx, trachea, and larynx and very rarely on nasal septum [1, 2]. There are many differences in the histology of major salivary gland tumours and ectopic minor salivary gland tumours. Hence diagnosis becomes a dilemma. These tumours are known to recur. We present a case of recurrent nasal septal pleomorphic adenoma which was treated with wide local excision and with Rintala's flap.*

Keywords: Pleomorphic adenoma; Nasal septum; Rintala's flap

1. Introduction

Ectopic presentation of pleomorphic adenoma is very rare, but cases have been recorded of tumours found 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 [4]. These tumours are notorious for they are very easily misdiagnosed on the basis of their myoepithelial cellularity and scanty myxoid stroma [3]. Since these tumours have high risk of recurrence, the patient needs to be kept on long term follow up.

2. Case Report

Patient is an 80yrs old male, complaining of a mass on the dorsum of nose since 1 yr. Swelling was painless, gradually progressively increasing with no episodes of nasal bleeding or rapid growth of the swelling or ulcerations. There were no complaints of nasal obstruction or reduced sense of smell. On examination the swelling was firm, 1.5*2 cm globular, fixed on the bony part of nasal dorsum, non tender, not inflamed with overlying and surrounding skin normal. Anterior rhinoscopy revealed septal perforation in the mid cartilagenous part and irregular red mass on both sides attached to remnants of septum.



Figure 1: front profile, showing the recurred mass on nasal dorsum with lines of excision marked



Figure 2: Side profile showing the same mas

Diagnostic nasal endoscopy revealed a 2*3 cm irregular polypoidal red mass attached to the upper part of septal remnants on both the sides. The mass was non tender, not bleeding to touch.

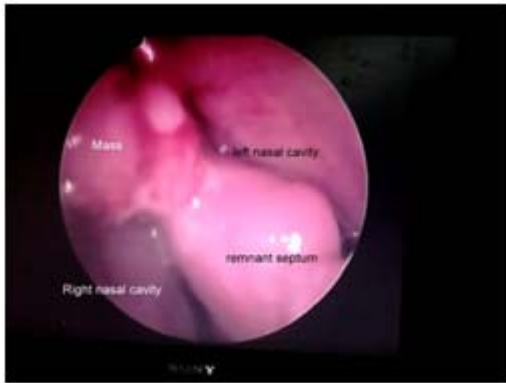


Figure 3: Diagnostic nasal endoscopy-Scope placed in the septal perforation. Both nasal cavities visualised. Mass was seen attached to the remnant of septum

CT PNS was done which showed a 2.4*2.1*1.2 cm, well defined, homogenously enhancing soft tissue mass below the root of nose. There was communication with the adjacent remaining cartilagenous nasal septum, just above the post operative defect, bony septum appeared normal. Anterolaterally it caused a bulge on the overlying skin. No intracranial extension. Bony walls of the sinuses were normal with no expansion noted.

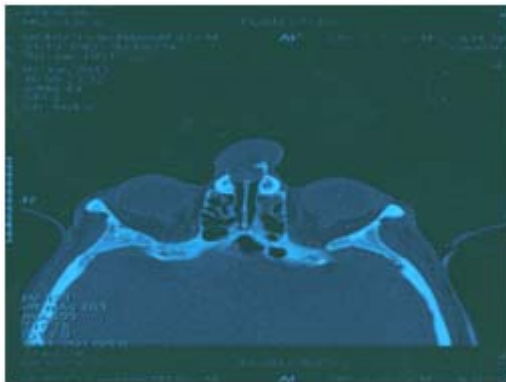


Figure 4: CT PNS is showing the mass arising from the septum and involving the subcutaneous tissue

FNAC from the external specimen was obtained and it proved to be pleomorphic adenoma. Punch Biopsy from the intranasal mass was done, which too suggested the same.

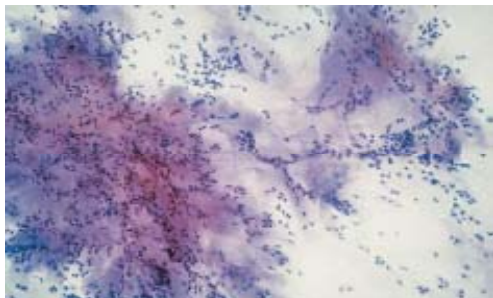


Figure 5: FNAC and punch biopsy suggestive of Pleomorphic Adenoma

Patient was previously operated 2 yrs back for a small bulge on the dorsum of the nose since 1 month and intermittent episodes of fresh, small amount of spontaneous bleeding. Anterior rhinoscopy had revealed an irregular mass in Rt. Nostril, on the mid portion of septum, opposite the middle turbinate, red polypoidal, non tender, but bleeding to touch.

CT PNS at that time had revealed a soft tissue mass in right nostril on the cartilagenous septum with all bony walls intact and no intracranial extension.

But there was discrepancy on tissue diagnosis. The FNAC done from the dorsum suggested it to be anaplastic carcinoma, but the punch biopsy from the nasal mass suggested it to be myoepithelioma. Patient was operated by lateral rhinotomy incision after obtaining anaesthetic fitness. Whole of nasal mass along with cartilagenous septum, subcutaneous layer of skin were removed in situ. Final histopathological diagnosis which was confirmed by two experienced pathologist was pleomorphic adenoma.

Since patient presented again with recurrence 1.5 yrs later, it was decided to reoperate the patient. After receiving necessary anaesthetic fitness, patient was operated by wide local excision including removal of 1 cm of healthy skin, underlying bone and surrounding mucosa. Whole mass was removed and subjected for HPE.



Figure 6: Intraoperative defect after removal of mass and full thickness skin

Reconstruction was done using burrow's triangle and Rintala's flap technique. Initial report came out to be anaplastic carcinoma where as the other pathologist reported it as myoepithelioma.



Figure 7: Burrows triangle excised and advancement incisions taken

Since clinically the swelling was slow growing and never during the course of recurrence, was bleeding encountered, with no erosion of nasal bones, skull base, cervical lymphnode involvement; diagnosis of anaplastic carcinoma was not accepted. Because of advanced age, to prevent further morbidities, radiotherapy was advised but refused by the patient. Post operatively patient's graft uptake was excellent and was discharged and kept on regular follow up.



Figure 8: Post operative appearance on day 1

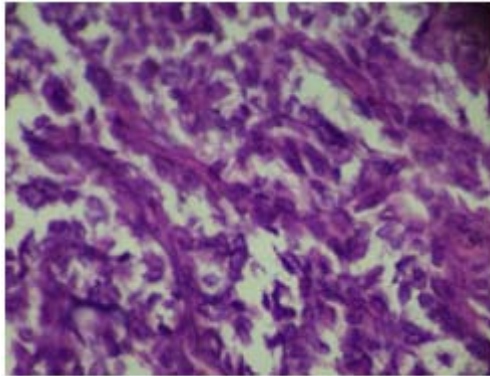


Figure 9: Photograph of the slide prepared from the histopathological Specimen



Figure 10: pt. after complete suture removal, showing good take and contour of the graft

3. Discussion

The first reported case in the literature of a pleomorphic adenoma of the nasal cavity was by Denker and Kahler [5]. Actual incidence of nasal adenoma is not known. Nasal septum forms the major site of origin, but few cases from the lateral nasal wall have too been seen. As compared to the tumour in major salivary glands, those in minor salivary glands have major differences histopathologically. Myoepithelial cellularity is extensive along with reduced myxoid stroma. Sometimes these nasal tumours may be composed of entirely epithelial cells with few or no stroma [5]. The cause of its recurrence was spillage of the stroma into the surrounding tissue during the time of operation [6]. Recurrence rate of nasal tumours was reported in one series to be around 50 % [9]. General recurrence rate of pleomorphic adenoma has been taken as around 10 % [7]. A wide local excision of the skin swelling along with bony septum and surrounding mucosa should be the treatment of

choice. Rintala's flap in old age is easy to harvest because of increased laxity of skin. Being a local advancement flap with adequate perfusion maintained from broad base, the graft survival and take up is very good. The coverage provided is good with excellent colour match and good post op nasal contour [8]. Major confusion regarding histopathological diagnosis is because of biphasic component comprising of epithelial/myoepithelial and fibromyxoid, chondroid stroma. But this is associated with wide variations. Predominance of epithelial cells in FNAC leads to a mistaken diagnosis of monomorphic adenoma, myoepithelioma and adenoid cystic carcinoma. If myxoid material is predominant with scanty or absent epithelial cells it may be erroneously diagnosed as retention cyst, while presence of many metaplastic cells with scanty mucoid material may lead to mistaken diagnosis of mucoepidermoid carcinoma [9].

4. Conclusion

Nasal pleomorphic adenomas are rare tumours, but they are easily misdiagnosed. Even experienced pathologists find it hard to diagnose. Hence the best modality of management is wide local excision. Rintala's flap, a local advancement flap utilising lax forehead skin is ideal graft to cover the defect adequately and also has good take up rate. These tumours are prone to recur, hence regular but long term follow up is necessary.

References

- [1] Spiro RH, Koss LG, Hajdu SI, et al. Tumors of minor salivary origin. A clinicopathologic study of 492 cases. *Cancer* 1973; 31: 117-129
- [2] 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 Jibiinkoka Gakkai Kaiho* 1990; 93:740-745
- [3] Liao BS, Hilsinger RL Jr, Chong E: Septal pleomorphic adenoma masquerading as squamous cell carcinoma. *Ear Nose Throat J* 1993; 72:781-782
- [4] Batsakis JG: *Tumors of the Head and Neck*. 2nd edition. Baltimore: Williams and Wilkins; 1984: 76-99.
- [5] Denker, Kahler: *Handush der Hals. Nasen ohrenheilkunde* 1929; 5:202
- [6] Compagno J, Wong RT: Intranasal mixed tumors [pleomorphic adenomas]: A clinicopathologic study of 40 cases. *Am J Clin Pathol* 1977; 68:213-218.
- [7] Krolls SO, Boyers RC: Mixed tumors of salivary glands. Long term follow up. *Cancer* 1972; 30:276-281
- [8] Chiu LD, Hybarger CP, Todes-Taylor N. The Rintala flap revisited. *Plast Reconstr Surg*. 1994 Nov; 94[6]:801-7
- [9] Verma K, Kapila K. Role of Fine needle aspiration cytology in diagnosis of pleomorphic adenomas, *Cytopathology* 2002; 13:121-127