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Case Report

Pleomorphic adenoma of dorsum of the nose: a rare occurrence

Rajiv Kumar Jain[1], Chultim D. Bhutia[1], Deepak Kumar Gupta[1], Ashvanee Kumar Chaudhary[1*], Gagan Rangari[2], Anjali Singh[3]

[1]Department of Otorhinolaryngology, [2]Department of Pathology, Institute of Medical Sciences, Banaras Hindu University, Varanasi, Uttar Pradesh, India
[3]Department of Otolaryngology, Kalpana Chawla Government Medical College, Karnal, Haryana, India

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***Correspondence:**
Dr. Ashvanee Kumar Chaudhary,
E-mail: drashvaneebhu21@gmail.com

ABSTRACT

Pleomorphic adenoma are common benign salivary gland tumours, which are found in majority in major salivary glands such as parotids and submandibular glands. However, Pleomorphic adenoma to originate from dorsum of the nose is a rare entity. In rare cases, it can be found in unusual sites such as upper aero digestive tracts, palate and lacrimal glands. Complete surgical resection is the treatment of choice. Though, the evolution to malignancy and recurrence is not usually encountered, still a long-term follow-up is recommended. Here, we report a 53 year old female , complained of swelling in the left side of dorsum of nose for 10 years with a feeling of heaviness over the left side of face and difficulty in vision on the side of swelling due to the enlarged size of the swelling which gave a feeling of vision disruption , also had a prior history of incision and drainage 4 years ,done elsewhere. On clinical examination, nodular mass was palpated on left side dorsum of nose which was freely mobile, and Skin over the swelling had blackish pigmentation. Anterior rhinoscopy revealed no abnormalities. Complete surgical excision via a lateral rhinotomy incision was performed. Cytological and histological evaluation revealed the presence of pleomorphic adenoma. We observed a decent cosmetic outcome with no evidence of recurrence.

Keywords: Pleomorphic adenoma, Salivary Gland tumour, Mixed tumor, Sinonasal, Nasopharyngeal, Myoepithelial cells, Epithelial cells

INTRODUCTION

Pleomorphic adenoma (PA) as defined in 1972 by world health organization (WHO) is a circumscribed tumour which is characterized by its pleomorphic or mixed appearance, clearly recognizable epithelial tissue being intermingled with tissue of mucoid, myxoid and chondroid appearance. Amongst all salivary gland tumours, Pleomorphic adenoma is the most frequently encountered lesion (50%). It is a common benign salivary gland tumour, which is also known as mixed tumor, found majority in major salivary glands namely, parotid followed by submandibular glands.[1] Besides that, it is a rare presentation to be found in the upper aero-digestive tract and lacrimal glands. Many reasons have been postulated for the origination of nasal pleomorphic adenoma to the lateral wall.

Ersner and Saltzman at el hypothesized that the progenitor cells of the septal pleomorphic adenomas are ectopic embryonic epithelialized cells found in nasal septum mucosa during the nasal bud's migration.[2]

Stevenson at el postulated that the reason for the appearance of these tumors in the nasal cavity was due to the degeneration of an epithelium- lined duct in the

cartilaginous nasal septum which was degenerated in early foetus.[3]

Clinically, it is described as a slow growing nodular swelling, and histopathology is the main stay in the diagnosis of mixed salivary tumor. Pleomorphic adenoma appearing in the skin of head and neck is an extremely rare presentation. The most prevalent site of pleomorphic adenoma of the minor salivary glands, is the palate which is followed by lip, buccal mucosa, floor of mouth, tongue, tonsil, retromolar trigone and nasal cavity.[4,5] Pleomorphic adenoma is rare in the sinonasal and nasopharyngeal areas, with the majority occurring more frequently in women in their third to sixth decades of life.[6,7]

CASE REPORT

A 53-year-old lady, presented to our outpatient department with complaints of swelling in the left side of dorsum of nose since 10 years. The swelling was insidious in onset, gradually progressive in size to attain the present size. She also complained of feeling of heaviness over the left side of face and difficulty in vision on the side of swelling due to the enlarged size of the swelling which gave a feeling of vision disruption. She gave a history of incision and drainage of the same swelling 6 years back in a government hospital in Gaya, Bihar. There was no history of pain, discharge from the swelling, nasal blockage, nasal discharge or epistaxis.

On examination, 2 x 2 cm nodular mass, present on left side dorsum of nose, non-tender, freely mobile swelling was affirmed. Skin over the swelling had blackish pigmentation. Anterior rhinoscopy revealed no abnormality. Clinically, it was diagnosed as a superficial cyst.

Fine needle aspiration cytology revealed features strongly suggestive of pleomorphic adenoma of minor salivary gland. Cect-nose, PNS and Orbit revealed a well-defined enhancing soft tissue density lesion of size 2.1 x 1.8 x 2.3 cm in subcutaneous plane of the lateral aspect of the nose on the left side showing a few foci of internal calcifications (Figure 1).

Figure 1: (A) Axial section (B) Sagittal section (C) Coronal section

Patient underwent complete excision of the mass under local anaesthesia. The gross appearance of the mass was an oval encapsulated mass of 2.5 x 2 cm and the cut surface appearance showed a solid grey white appearance. Skin is closed with 4.0 prolene (Figure 2), sterile dressing is applied.

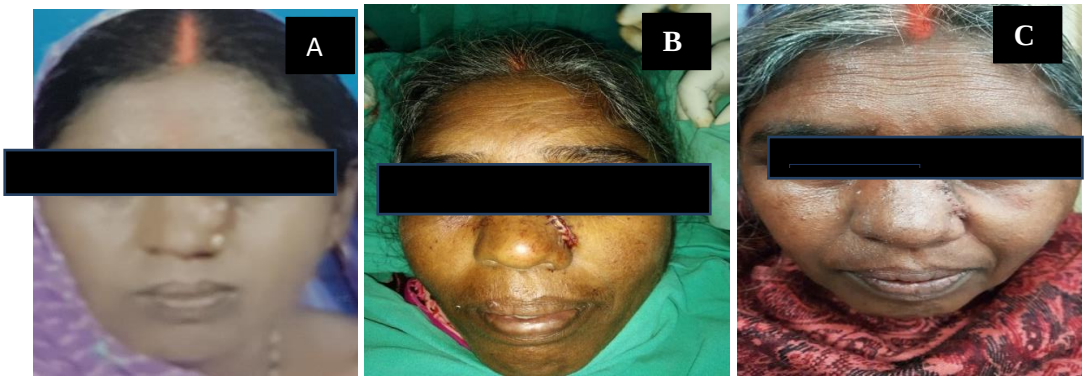

Figure 2: (A) Pre-operative image of the patient, arrow depicting the nasal mass (B) Immediate post- operative, arrow depicting incision after complete excision of the mass (C) Post op day 10th, arrow depicting healed incision after suture removal.

The mass was then sent for histopathological examination which further revealed features consistent with pleomorphic salivary gland adenoma.

Section examined under 10 x and 40 x showed a tumor comprised of epithelial cells arranged in sheets and tubules along with myoepithelial cells comprised of plasmacytoid cells, spindle cells and epitheloid cells arranged in a chondromyxoid stromal background, consistent with features to that of pleomorphic adenoma of salivary gland. Suture was removed on the 10th day and the incision site was healthy with minimal scar visible. (Figure 3) The patient was doing well during the follow up.

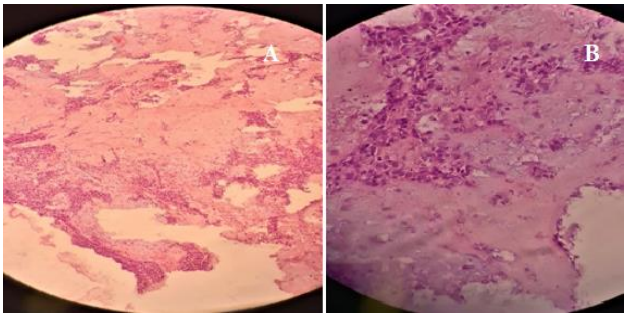

Figure 3: (A) Histopthological microsection under 10X (B) Histopthological microsection under 40X.

DISCUSSION

Pleomorphic adenoma is a benign slow growing tumour which mainly affects the major salivary glands namely, parotid and submandibular gland. Besides that, it can also affect minor salivary glands but is a rare entity. Although rare, but in case of the upper respiratory tract, the commonest site for occurrence of pleomorphic adenoma is the nasal cavity. Usually intranasal pleomorphic adenoma usually arises from the nasal septum, though most of minor salivary glands are located in the lateral wall.

There have been many previous theories that have been proposed for the origin of Pleomorphic adenoma of the nasal cavity. They may arise from residues in the vomeronasal organs, the epithelial lining ducts found in the septa regenerated in early embryonic life. Pleomorphic adenoma consists of epithelial and mesenchymal components. Sinonasal pleomorphic adenomas have more cytoplasm and predominant epithelial components which differs them from mixed neoplasms of major salivary glands, also they are devoid of capsules and have few stromal components.[8] Vento et al reported cases of benign Pleomorphic adenoma of the nasal cavity, which were surgically resected and presented with no recurrence during various follow-up periods.[8,10]

Another study stated that the abnormal origin of Pleomorphic adenoma from the nasal septum mucosa may be caused by dislocated embryonic epithelial cells which arises from the ectoderm and carried into the septal region via the nasal pits.[9]

In our case report, the swelling was located on the left side of dorsum of the nose, therefore we made a clinical diagnosis of a superficial external mass upon consideration. The further advancement of the management for pleomorphic nasal adenoma should depend on the size, location and extension of the tumour. In this case, we opted for the complete excision via a lateral rhinotomy method because the nasal tumour was anteriorly located. Later, A complete mass was retrieved during the excision, which was not in the favour of the mass originating intranasally.

Irfan et al., 2010, reported a similar case in a 16-year-old girl with one-year history of swelling over left side dorsum of nose who further underwent complete excision of the mass which showed features of pleomorphic adenoma.[10]

Suzuki et al., reported a rare case of pleomorphic adenoma which developed from the lateral wall of right nasal cavity in a 43-year-old woman. In Japan, 41 cases of pleomorphic adenoma originated in the nasal cavity had been reported. Age of onset of these cases were 16-74 years, averaging 44.6 years and showed Sex ratio of female predominance. Laterality of affected sites did not have any significant differences.[11]

Compagno and Wong et al concluded that intranasal mixed tumours presents with a relatively low rate of recurrence (10%) compared with recurrence rates as high as 25% for intraoral mixed tumors and 50% for parotid gland mixed tumors.[12]

The diagnosis of Pleomorphic adenoma in nasopharyngeal regions are challenging because symptoms are not characteristic and radiologic findings are usually nonspecific. CT scans generally shows bony alterations and extensive lesions or destructive bony changes, providing reliable indications for differentiating between benign and malignant lesions. They usually present with well-defined, homogeneous soft tissue masses and expansile bony changes. In cases of benign pleomorphic adenoma amongst the sinonasal/ nasopharyngeal regions, recurrence is infrequent.

Regardless of where the tumour arises from, it is a well-versed fact that there is always risk of recurrence. The recurrence rate depends almost on the sufficiency of the initial excision; hence, it is very important to have a good exposure of the mass to remove the tumor completely along with its margins. Since, this is a rare entity, there is no preset standard treatment algorithm. However, follow up of the patient for examination to look out for any signs and symptoms of recurrence is important.

CONCLUSION

Pleomorphic adenoma is a rare sinonasal minor salivarygland tumour. For an ENT surgeon, it is important to consider nasal pleomorphic adenoma as a differential diagnosis whenever a patient presents with a slow growing unilateral mass in the lateral wall of nose, even though it is rarely encountered and we tend to overlook the actual diagnosis. Early diagnosis and management help in a complete excision and avoid any chance of malignant transformation. Complete surgical excision of the tumor under direct vision is preferred, as it allows complete resection along with the margins with bare cosmetic defect. Recurrence is a rare entity, yet follow up post-surgery is important to look for any signs of recurrence or malignant changes.

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