

A Case Series of Rare Tumors Encountered in Otorhinolaryngology

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ABSTRACT

Introduction: A variety of masses, benign and malignant, are encountered in otolaryngology practice. Rare lesions are often difficult to diagnose clinically and confirmed diagnosis can only be made with the histopathological examination.

Aim: In this case series, we report four unusual lesions encountered in otolaryngology practice, namely a solitary giant trichoepithelioma of nose, Burkitt lymphoma of tonsil in a child, non-Hodgkin's lymphoma of parotid with facial nerve palsy and a case of laryngeal amyloidosis.

Materials and methods: Following a standard protocol of clinical examination and radiological evaluation, appropriate surgeries were done and histopathological evaluation was carried out to confirm the diagnosis of these rare lesions.

Discussion: Solitary giant trichoepithelioma is usually seen in the groin and thighs and only few nasal cases have been reported so far. Abdomen, mandible or maxilla are common sites for Burkitt lymphoma but its oral occurrences, that too in paediatric age groups, is far uncommon. Similarly primary parotid NHL represents 1-4% of all parotid tumours. Localized laryngeal amyloidosis is another rare disease that often needs surgical intervention and histopathological evaluation for proper diagnosis.

Conclusion: A high index of suspicion, coupled with histopathological and immunohistochemical examination, remains paramount in uncommon masses encountered in ENT practice.

Keywords: Giant solitary trichoepithelioma, NHL of parotid, Tonsillar Burkitt lymphoma, Laryngeal amyloidosis

INTRODUCTION

Otolaryngologists often encounter benign or malignant masses, in the ear, nose or throat that present with common clinical features but histologically, often turn out to be rare entities. Surgeons often get biased while

making a diagnosis, when a rare disease mimics clinical features of commonly diagnosed lesions like polyps, haemangiomas, papillomas in nasal cavity, pleomorphic adenoma or Warthin's tumour over parotid region and polyps, nodules,

cysts or malignant lesions in larynx. We report a case series of such rare tumours encountered in ENT practice. A case of nasal giant solitary trichoepithelioma, Burkitt lymphoma of tonsil, non-Hodgkin's lymphoma of parotid and a case of laryngeal amyloidosis are presented in this series. These rare entities frequently mimic common conditions, leading to potential delays in diagnosis and management. Only few cases of these lesions are reported in literature till this date. If the treating surgeon is not well versed with their clinical details management of such cases might become challenging. By documenting these rare presentations, we hope to expand the existing literature and provide clinicians with a broader differential framework for unusual ENT masses.

CASE PRESENTATION

Case 1: Solitary giant trichoepithelioma of nose

A 66-year-old male patient presented in ENT OPD with complaints of mass in the left nasal cavity for 10 years and left sided nasal obstruction for the last 1 year. When he first noticed the lesion, it was like a small mole which gradually progressed to the size of a peanut. He was on anti-hypertensive and anti-platelet medicines as he had undergone angioplasty 2 years ago. On anterior rhinoscopy, a firm, globular, pedunculated, pale to skin coloured mass of approximately 2cm diameter, was seen in the left nasal cavity, arising from the lateral aspect of the left nasal vestibule. It was non-tender and didn't bleed on touch. The rest of the nasal & ear and throat examinations were normal. (Fig. 1 showing globular mass in left nasal cavity) Diagnostic nasal endoscopy showed no other lesion in bilateral nasal cavities. There was no history of trauma preceding the lesion and no past and family history of tuberculosis. Dermatological examination showed no other or similar lesions on the body.



Fig.1. A globular mass in left nasal cavity

Provisional clinical diagnosis of lupus vulgaris or sarcoidosis was considered. Mantoux test, sputum for acid fast bacilli and chest x-ray were done to rule out tuberculosis. As these tests came negative, surgical excision of mass was planned. Anti-platelet medications were withheld 5 days prior to the surgery.

MANAGEMENT

After all routine blood and radiological investigations, excision of nasal mass with 5mm of healthy margins was done under local anaesthesia. The left nasal cavity was packed with merocel for 24 hours and the wound was allowed to heal with secondary intention. The mass was sent for histopathological examination. (Fig.2 showing excised nasal mass).



Fig. 2 Excised left nasal mass.

Histopathology examination showed a well circumscribed dermal mass lesion lined by

stratified squamous epithelium. The dermis showed a tumour composed of superficial nests of basaloid cells with leaf-like or frond-like architectural patterns with keratin horn cysts of varying sizes. Few cysts were ruptured with multinucleated giant cells along with keratin. The fibrous cellular stroma showed mononuclear cell infiltration. Based upon these findings a final diagnosis of giant solitary trichoepithelioma was made (Fig.3 & Fig. 4 showing histopathology of nasal mass with keratin horn cysts under microscope.)

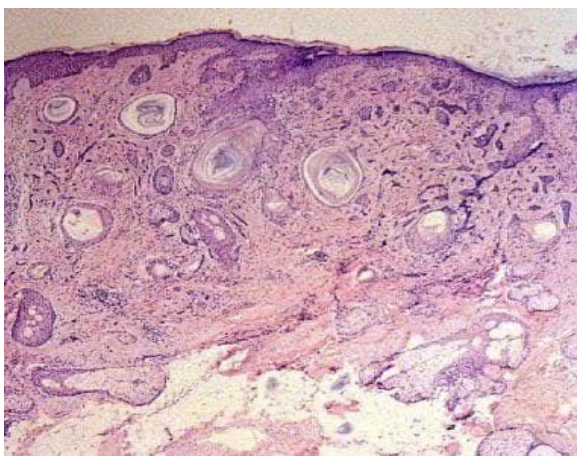


Fig. 3 HPE image showing keratin horn cysts

Immunohistochemistry for CD34, CD10 and PLHDA were advised, but the patient was denied due to poor financial status. 6 months follow-up of the patient showed good healing and no recurrence.

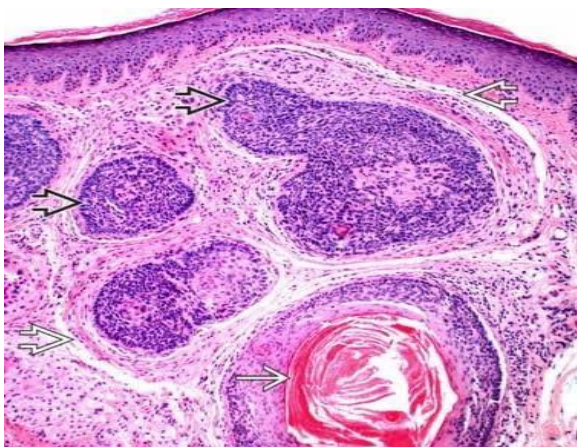


Fig 4. HPE image showing nest of basaloid cells (blue) and keratin horn cyst (red)

Case 2: Burkitt lymphoma of tonsil

A 13-year-old male child came to ENT OPD with chief complaints of difficulty in swallowing for one month. There was no history of odynophagia, dyspnea or restricted mouth opening. Recurrent episodes of fever, sore throat or change in voice were also not reported. Parents informed that the child has developed a unilateral left sided mass in the throat since 1 month which was gradually increasing in size and was associated with difficulty in swallowing, more from the left side. The patient had lost weight over the last 2 months. On clinical examination a unilateral approximately 3cm X 3 cm, smooth, globular mass with an ulcer over its surface, was seen in the left tonsillar fossa. It was approaching midline and touching the uvula. The mass was firm and non-tender on palpation. (Fig. 5 showing left tonsillar mass). On the right-side Grade II tonsillar hypertrophy was noted. Left Level II B and Level V lymph nodes were palpable, firm, mobile and non-tender.

A clinical diagnosis of left tonsillar mass under evaluation was considered and all routine blood investigations were done. Blood profile was normal with mildly raised PT and INR values. (PT -19.2, INR 1.8)



Fig.5 Image showing mass in left tonsillar fossa

A CT skull base to mediastinum (Plain + Contrast) was advised which showed a well-defined soft tissue density lesion in the left tonsillar fossa arising from left palatine tonsil & measuring approximately 23 x 21 x

25 mm (AP x TR x CC). It showed mild post contrast enhancement. There was no evidence of fat density /air pockets/ calcification or obvious collection within the lesion. (Fig.6 showing CT image of coronal section of oropharynx with mass originating from left tonsillar fossa)

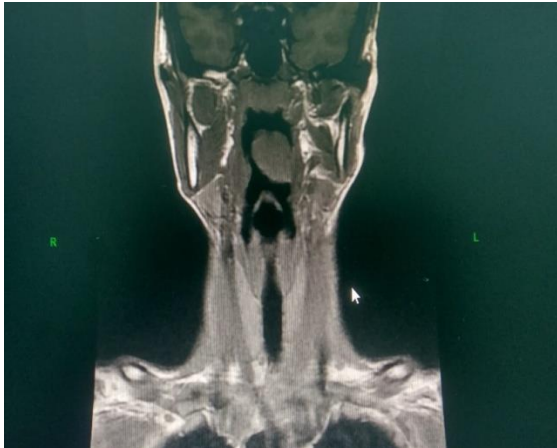


Fig.6 CT neck showing mass arising from left tonsillar fossa

MRI neck (P+C) was also done which showed enlarged left palatine tonsil with subtle post-contrast enhancement with similar features described in CT scan.

Coblator assisted left tonsillar mass excision (Fig.7) under general anaesthesia was done and tissue was sent for HPE. (Fig. 8 showing excised tonsillar mass)



Fig. 7 Coblator assisted left tonsillar mass excision under GA



Fig 8. Excised left tonsillar mass specimen

Histopathological examination showed a tissue lined by stratified squamous epithelium with areas of ulceration & granulation tissue. Subepithelium showed completely effaced tonsillar architecture by a tumour with diffuse infiltration of monotonous lymphoid population & intermixed tangible body macrophages giving a Starry Sky appearance. (Fig. 9 showing 'Starry sky appearance' in histopathological image of excised tonsillar mass under microscope). Individual tumour cells were intermediate sized having high N:C ratio with uniform round nuclei and mildly clumped vesicular chromatin. Single to multiple prominent nucleoli & scanty cytoplasm was seen. Numerous abnormal mitotic figures were also noted.

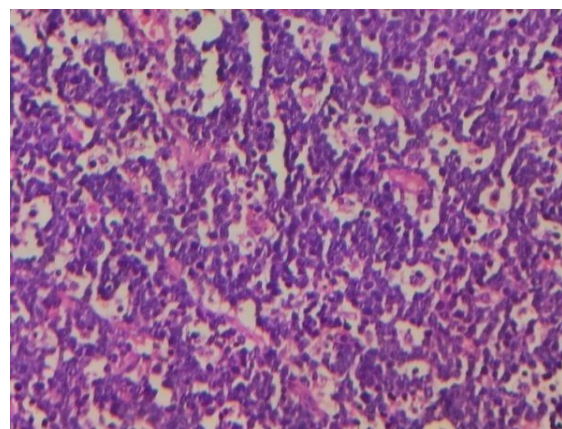


Fig. 9 HPE image showing 'Starry sky' appearance

Above findings were consistent with Non-Hodgkin's Lymphoma. The possibility of Burkitt Lymphoma was highly suspected.

IHC markers CD20, CD10, CD3, Ki67, BCL2, Cyclin D1, c-MYC & BCL6 were advised for further confirmation. CD10 and CD20 IHC markers came to be positive, which confirmed the diagnosis of Burkitt Lymphoma.

Chemotherapy was advised post-operatively and patient has shown no recurrence till now

Case 3: Non-Hodgkin's lymphoma of parotid gland

A 66-year-old male came to ENT OPD with complaints of right sided facial weakness for 7-8 months and swelling on right infra-auricular region for 15-20 days. His facial weakness had aggravated during the last few months and was associated with facial deformity. He had noticed the swelling below his right ear about 3 weeks back which was increasing in size and gradually progressed to the present size of a lemon.

The swelling was not associated with pain or erythema but there was difficulty in opening the mouth. History of loss of appetite and weight loss were also present. There was no history of headache, earache, odynophagia or dysphagia. Any history of trauma or fever was also denied by the patient. The patient had no known comorbidities but he had a habit of tobacco-chewing since the last 15-17 years. 8 months back he was hospitalized for right facial palsy and where intravenous steroids were given in tapering doses after which the patient had slight improvement in facial weakness.

On examination, approximately 6 x 3x 2 cm solitary, firm, non-tender, ovoid swelling was seen in the right parotid region with smooth surface and irregularly defined margins. Superiorly it was extending 1cm above the level of tragal cartilage and inferiorly it was extending 3cm below the angle of mandible. (Fig. 10 & 11 showing clinical photographs of patient with the right parotid swelling)

Posteriorly the swelling was lying along the anterior border of sternocleidomastoid and anteriorly it was extending upto the zygomatic prominence. Skin over swelling was normal and mobile. A single, firm, non-tender and mobile A 2x 1cm Level IIa lymph node, was palpable on right side. Grade V right facial nerve palsy was present. Mouth opening was restricted to 2 & ½ finger breadth.

USG of the swelling suggested a well-defined multi-lobulated, heterogeneous predominantly hypoechoic lesion in right parotid space, involving the right parotid gland and measuring approximately 39 x 20 x 55 mm (APxTRxCC).

Colour Doppler images showed mild internal vascularity.

There was no evidence of calcification noted within the lesion.

Few enlarged necrotic lymph nodes were seen in the right level II neck region, largest measuring 18 x 11mm. Multiple, prominent lymph nodes were noted in the right level IB region with no evidence of necrosis/calcification within.



Fig. 10



Fig. 11

Fig.10 & Fig 11. Showing anteroposterior and lateral images of patient with right parotid mass.

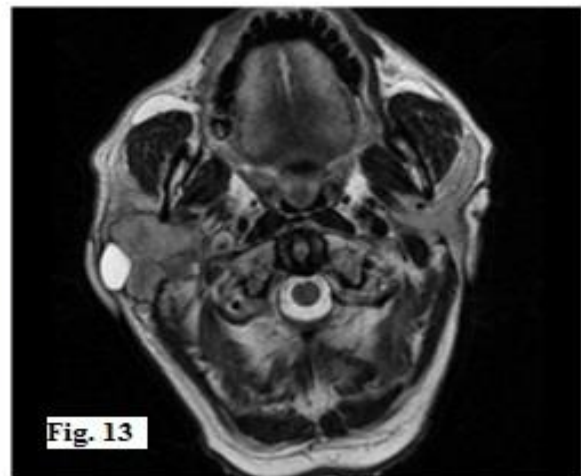
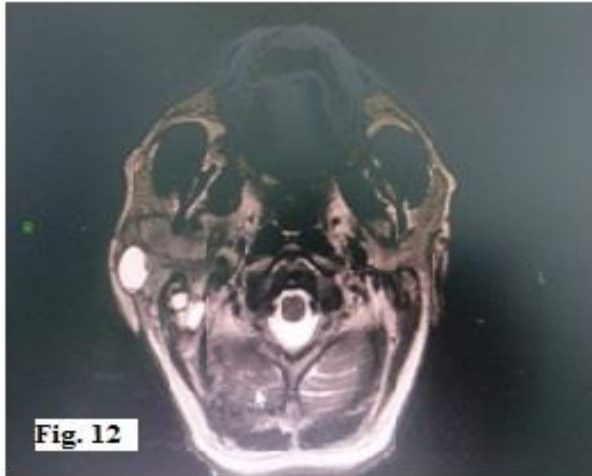


Fig. 12 & 13 Showing axial view of CT (contrast enhanced) & MRI of head with right parotid mass

USG findings were confirmed with Contrast enhanced CT scan which showed involvement of the both superficial and deep lobes of the parotid gland, with few non-enhancing areas within suggestive of necrosis. Mild subcutaneous perilesional fat stranding was also seen. (Fig. 12 & 13

showing CT & MRI images of right parotid mass)

FNAC of mass showed large lymphoid cells with high N:C ratio, pleomorphic, vesicular to hyperchromatic nuclei & scanty to moderate cytoplasm with prominent vacuolations.



Fig. 14-17. Showing step-wise excision of right parotid mass

Abnormal mitotic figures were also noted. Background showed lymphocytes, neutrophils, macrophages, RBCs and lymphoglandular bodies. These observations were suggestive neoplastic lesions. Right total parotidectomy with modified radical neck dissection was planned under

general anaesthesia. Modified Blair's incision was taken and, identifying and preserving all the branches of facial nerve, total parotidectomy was done along with right modified radical neck dissection (Fig14-17 showing stepwise dissection and excision of right parotid mass). Closure was

done in two layers. The specimen was sent for histopathological examination.

Sections studied showed a salivary gland parenchyma whose normal acinar and ductal architecture was completely effaced by a dense, diffuse, and monotonous infiltration of lymphoid cells. The tumor cells were arranged in a solid, sheet-like pattern, extensively infiltrating the inter-lobular septa and perineural spaces.

Intermediate-sized lymphoid cells with high Nucleo Cytoplasmic (N:C) ratio & with uniformly round to oval nuclei with "coarse" or "clumped" vesicular chromatin were seen. Multiple, prominent, peripherally located nucleoli with scanty basophilic cytoplasm with occasional distinct vacuoles was observed. Atypical/abnormal mitotic figures were noted. Numerous macrophages containing ingested apoptotic debris intermixed with the tumor cells, creating a classic starry-sky appearance (Fig. 18 showing histopathological image of parotid mass under microscope with typical 'starry-sky' appearance) suggested non-Hodgkin's lymphoma of Parotid gland.

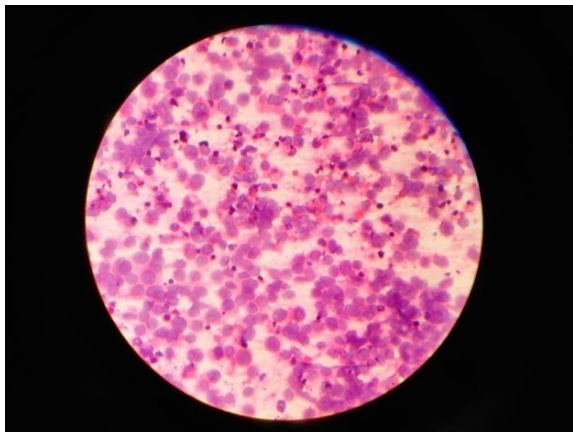


Fig.18. HPE image of NHL of right parotid mass

Immuno-histochemical markers like CD-10, CD-20, and c-MYC were advised for further evaluation but couldn't be done due to non-affordability of patients.

The patient was referred to an oncologist for chemotherapy and radiotherapy for further management. PET-CT on 6 month follow up showed no recurrence till now.

Case 4: Laryngeal amyloidosis

A 61 years old female presented in ENT OPD, with complaints of change in voice since the last 10 years. The change in her voice was insidious in onset and had progressively worsened over the last 10 years. Her voice was hoarse, breathy and was associated with dyspnea on exertion. There were no associated symptoms like dysphagia, hemoptysis, anorexia, weight loss, fatigue or cough during swallowing. She was a housewife with no history of addiction to tobacco or its products. She had hypertension and diabetes mellitus with uncontrolled blood sugar levels. There was no history of chronic diseases like tuberculosis and asthma. Right dacryocystorhinostomy was done a few months ago due to nasolacrimal duct block. She had undergone microlaryngoscopy and biopsy/excision of laryngeal mass about 9 years back, details of which were not available. Her voice improved partially following that surgery but worsened again gradually thereafter.

After clinical ENT examination, flexible laryngoscopy was planned, which revealed a 1 cm x 1cm globular, smooth mass arising from the anterior half of the right false vocal cord. It was sessile with a broad base, partially encroaching the anterior commissure. The right vocal cord was partially obscured by the mass (Fig. 19 showing a sessile mass arising from the right false vocal cord). Vocal cord movements were normal and symmetrical on both sides. Rest of the laryngeal structures were found to be normal.



Fig.19. Laryngoscopic image of right false vocal cord

Computed tomography of the neck revealed a mildly thickened right vocal cord with a small enhancing nodule of size 4.6 x 7.5 x 4.8mm arising from the right false vocal cord. Anteriorly it was extending upto the anterior commissure and abutting the right vocal cord.



Fig. 20. CT image of right false vocal cord mass

Mild fat stranding was noted in right paraglottic space (Fig. 20 showing axial view of CT neck showing mass over right false vocal cord).

A clinical diagnosis of granulomatous lesion of larynx was made and based upon above findings and microlaryngoscopic excision of the mass under general anesthesia was planned.

On microscopic examination under GA, the mass was found to be smooth, sessile, firm and yellowish in appearance, arising from the right vestibular fold reaching up to anterior commissure. The mass was excised from the base using Diode laser and sent for histopathological examination.

Histopathological evaluation showed tissue lined by pseudostratified tall columnar epithelium and partially lined by stratified squamous epithelium. Subepithelium showed amorphous proteinaceous eosinophilic material (Fig.21 showing histopathological image of false vocal cord mass with amorphous proteinaceous eosinophilic material).

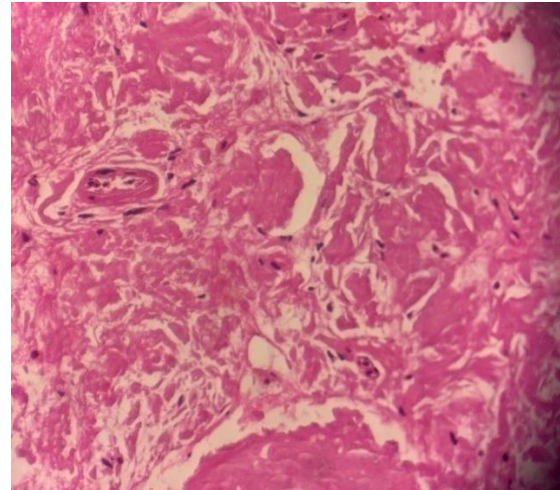


Fig. 21. HPE image of right false vocal cord mass showing amorphous proteinaceous eosinophilic material.

Staining with Congo-red dye sample was positive for amyloid (Fig. 22 showing birefringence of amyloid protein with Congo-red dye). Thus, a final diagnosis of laryngeal amyloidosis was confirmed.

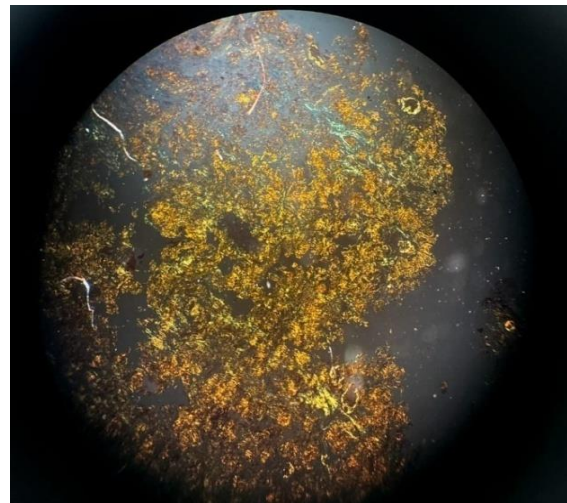


Fig. 22. Birefringence of amyloid protein on staining with Congo red

Postoperatively the patient was given intravenous steroids for 3 days followed by oral steroids for 10 days to reduce laryngeal inflammation. Absolute voice rest was advised for 15 days and speech therapy was given after a period of one month.

One (Fig. 23) and 6 month follow up was done to check recurrence. Stroboscopy on follow up showed adequate healing and no recurrence.



Fig 23. Post operative (one month follow up) image showing healed mucosa of right false vocal cord

Urine electrophoresis for light chain & immunoglobulin detection and abdominal fat biopsy was advised for ruling out systemic amyloidosis but the patient was not compliant for it due to poor financial status.

DISCUSSION

Trichoepithelioma (TE) is a rare benign tumor originating from the undifferentiated pluripotent cells of the follicular-sebaceous-apocrine unit¹. Different variants of TE have been reported in literature which includes: Multiple familial trichoepithelioma (MFT), solitary, giant solitary and desmoplastic. MFT is the most common manifestation. Histopathologically they are similar but not identical.

Solitary trichoepitheliomas are rare benign tumours of skin. They are usually sporadic and commonly seen in elderly patients with male predominance². They can vary in size from a few millimeters to a few centimeters. Telangiectasia may be seen over their surface³. Solitary trichoepithelioma measuring more than 2 cm are termed as giant solitary trichoepithelioma⁴. They can appear on any part of the body but are commonly seen around the groin or thighs. Very few cases have been reported in the nose.

Giant solitary trichoepithelioma presents as solid, firm and lobulated mass with a broad base. However, they may be pedunculated, ulcerated or may be cystic. Though they are

benign but, in few cases, malignant transformation into basal cell carcinoma (BCC) has been reported⁵. Radiological investigations are not routinely helpful but MRI might be helpful for depicting extensions and depth or where diagnosis is uncertain.

Haematoxylin and eosin-stained sections show uniform basaloid cells with scanty cytoplasm and darkly stained nucleus arranged in a nest and adenoid pattern with epithelial islands. Fibromyxoid or fibrocellular stroma may be seen. Mitotic figures are frequent, but abnormal mitoses are not seen. Horn cysts having a fully keratinized centre, surrounded by basophilic cells are characteristic features of trichoepitheliomas. Horn cysts in BCC or trichilemmal carcinoma show gradual and incomplete keratinization of horn cells (trichilemmal keratinization)⁶. Amyloidosis, granulomas or giant cell reactions might also be seen in a few cases.

Immunohistochemistry is the best diagnostic tool to differentiate BCC from trichoepitheliomas. The follicular stem cell marker pleckstrin homology-like domain, Family A, member 1 (PHLDA 1), CD10 and CD34 are positive for trichoepithelioma but not in BCC⁴.

Treatment of giant solitary TE includes surgical excision with 3-4mm safe margin and plastic reconstruction of wound with flaps wherever needed. Cryotherapy, radiotherapy or erbium-YAG lasers can be used for cosmetic reasons. Chances of recurrence are there but rare. Sometimes malignant transformation into BCC has been reported.

About 13% of primary extranodal NHL involve oral cavity and oropharynx with 70% fraction of these occur in tonsils⁷. Most common rapidly growing NHL is diffuse large B cell lymphoma which accounts for 80% of these cases⁸. Burkitt lymphoma is one of the variants of non-Hodgkin's lymphoma which is divided into three subgroups: sporadic, endemic and immunodeficiency related⁹. Abdomen, mandible or maxilla are the usual sites for

Burkitt lymphoma, however in rare instances it can involve oral cavity. Even its sporadic form is more often found in adults, as an intra-abdominal mass rather than oropharyngeal lesion¹⁰. Paediatric cases with tonsillar Burkitt lymphoma are extremely rare. Burkitt lymphoma is high grade lymphoma with high risk of tumor lysis syndrome, which can lead to acute kidney injury and fatal cardiac arrhythmias. Management involves radiological evaluation with MRI and biopsy for confirmation, which shows starry sky appearance on HPE. Disease can be treated completely with four cycles of chemotherapy and two years survival rate is around 89%.

Clinical manifestation of parotid lymphomas is non-specific and thus they cannot be differentiated from other neoplastic lesions of salivary glands¹¹. FNAC plays a key role in evaluating salivary gland tumors but not in cases of malignant lymphomas¹². In our case, histopathological examination of the specimen after surgical excision, confirmed the diagnosis of non-Hodgkin's lymphoma. Primary parotid NHL represents 1%-4% of all parotid tumors. NHL of parotid may be classified as extra-nodal if they arise from mucosa associated lymphoid tissue (MALT) or nodal if their true origin is from lymph nodes. Salivary gland lymphomas have been found associated with Sjogren's syndrome¹³.

Chemotherapy is considered useful adjunctive treatment in these patients¹⁴. A long survival rate is reported by a majority of authors, ranging from 50 % to 75%.

Localized laryngeal amyloidosis (LA) is a rare disease characterized by deposition of amyloid protein in the larynx without systemic involvement. It is a benign disease and slowly progressive. It accounts for 0.2 to 1.2% of benign tumors of larynx¹⁵. LA can be of two types: 1) Discrete tumor nodule or 2) Diffuse subepithelial deposits¹⁶. It typically presents with hoarseness of voice and may be associated with dyspnea, stridor and globus sensations.

Laryngeal amyloidosis is mostly localized in ventricle, false vocal cords, true vocal cords, aryepiglottic folds and subglottis in that order of frequency. Other common sites of localized amyloidosis are trachea-bronchial tree, paranasal sinuses, eyes, orbit and nose. Kidneys and heart are the most common sites for systemic amyloidosis.

Common types of amyloidosis include: 1) AA type- also known as reactive amyloidosis and characterized by deposition of plasma cells and amyloid tissue as inflammatory reaction to antigens. 2) AL type: characterized by deposition of light chains like kappa and lambda due to inability of body to clear them. 3) AB type- characterized by deposition of beta-2 microglobulin in haemodialysis-associated amyloidosis^{17,18,19}.

Diagnosis of laryngeal amyloidosis can be made on fiberoptic laryngoscopy, where yellowish mucosa covered lesions. MRI could be helpful to know the extent of disease. Final diagnosis is made on histopathological examination when it displays apple green birefringence under polarized light on staining with congo red. To rule out systemic amyloidosis abdominal fat biopsy or urine light chain electrophoresis is advisable.

Treatment includes microlaryngoscopic excision with cold steel instruments or CO₂ Laser. Depending upon extension of disease, sometimes partial laryngectomy might be required. Regular follow up with laryngoscopy is advised.

CONCLUSION

This case series highlights the diagnostic challenges posed by presentation of rare lesions in the head and neck. The solitary giant trichoepithelioma demonstrates benign adnexal tumours, though rare but must be confirmed with histopathology to rule out malignancies like BCC. Isolated tonsillar Burkitt lymphoma and primary parotid non-Hodgkin's lymphoma underscore the need for early diagnosis and management. Furthermore, laryngeal amyloidosis, though benign, needs proper histological evaluation

for diagnosis. A high index of suspicion, coupled with histopathological and immunohistochemical examination, remains paramount in such cases. These cases reinforce that uncommon pathologies must remain in the differential diagnosis to avoid treatment delays in anatomically critical sites.

Declaration by Authors

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Authors' contribution:

Dr Sandeep Chahande & Dr. Lisha Sarode conceptualized the concept of case series of rare tumours encountered in otorhinolaryngology. Dr. Sandeep Chahande performed the surgical procedures, and drafted the manuscript. Dr. Manaswi Kadel analyzed the data and revised the manuscript critically for intellectual content.

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