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Managing Some Rare Parotid Masses

Keshav Gupta¹, Mohit Srivastava², Ashu Bhati³

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ABSTRACT

Background: The parotid gland is a major salivary gland in the human body located in both the cheeks. It accounts for approximately 3-6% of all head and neck masses. Three fourths of all parotid masses are benign. However, they are very diverse in nature and in intimacy with important anatomical structures. An accurate diagnosis and surgical management of some rarely presenting parotid masses is further challenging to the medical team because of limited literature and experience with these masses.

Methods: A retrospective study was done at our Tertiary Care Institute studying different approaches towards rarely presenting parotid masses during last two years.

Results: A giant pleomorphic adenoma with 14 cm diameter managed with total superficial Parotidectomy, a difficult to diagnose parotid mass at angle of mandible which was confirmed to be Schwannoma post excision and a large Paediatric lymphangioma managed with Sclerotherapy are discussed here.

Conclusion: All were managed without mortality, with minimal morbidity and no recurrence noted till date. Our experience will be of great use to physicians and surgeons while dealing with rare benign parotid masses.

Keywords: Diagnosis Management Mass Parotid Rare Surgery Swelling.

INTRODUCTION

Human body houses many salivary glands which release their secretions through the ducts opening into the oral cavity. These secretions are composed of saliva, enzymes, minerals, water and immunoglobulins and hence responsible for the

mechanical, defensive, digestive, immunological and pH regulatory functions of the oral cavity. There are larger paired major salivary glands and smaller numerous minor salivary glands. Parotid gland is the largest of all salivary glands which houses both the cheeks. Its secretions are watery and reaches the oral cavity through its duct named as Stenson's duct.

The parotid gland is inverted pyramidal in shape with its apex towards the angle of mandible and its base at the zygomatic arch anterior to the external auditory canal. Anteriorly it is in relation with buccal pad of fat and posteriorly it curves around the posterior border of mandible.¹ The gland is covered by a tough, non-yielding false fascia which is formed by condensation of deep fascia of neck and it splits to surround the gland. The gland is composed of a superficial lobe and a deep lobe. Important anatomical structures like Facial Nerve, retromandibular plexus of veins, Facial Vein,

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external carotid artery and its branches passes through the substance of the gland. Facial nerve enters the gland as soon as it leaves the stylomastoid foramina either in form of a single trunk or its branches and gives off its terminal branches within the substance of the gland. The Stenson's duct emerges at the anterior part of the gland, passes horizontally through masseter, pierces buccinator and opens in oral cavity through papillae on both sides opposite the upper second molar teeth.²

Parotid masses are quite common and accounts for approximately 3-6% of all head and neck masses. Majority (almost three-fourths) of all parotid masses are benign masses and rest are malignant.³ Benign parotid masses also consist of a large variety of masses and have been classified on various basis. One of the largest studies on benign parotid masses were conducted by Bradley and Mc Gurk⁴ (1065 cases) and Everson and Cawson⁵ (2410 cases). The incidence of benign parotid masses was reported to be 5.3-6.2 per lakh population. No age group is spared. Sixth decade of the life is most likely to face a benign parotid mass. Females are more likely to present with this condition.⁶ Diagnosis as well as treatment needs good clinical as well as technical expertise. Pleomorphic adenoma (PA) is one of the commonest parotid masses. PA has a definitive anatomical relation between Facial Nerve (FN) and its branches and hence its surgical management may be challenging. A large PA is more challenging as it may be in close relation with many or all branches of FN.⁷ Schwannoma are rare benign parotid masses. There are limited studies available in literature about Parotid Schwannoma (PS). Large Bradley- Mc Gurk and Everson-Cawson studies did not report a single case of PS. Parotid Lymphangioma (PL) are another rare benign parotid mass. Management of paediatric parotid masses is always challenging as they require dedicated and specialised medical care right from presentation till the last step in management. Surgery may be quite challenging in this age group.

There are various factors which needs to be considered while managing a case of rare benign parotid mass. Prompt diagnosis in a systematic and cost-effective manner in a single sitting may be challenging in such cases. After a diagnosis is reached, the next step is to decide the best treatment which may be medical or surgical or both. Surgical cases require choosing the best approach, meticulous planning, good surgical exposure, good surgical skills, perfect completion and team work. We have addressed all these factors for the management of benign parotid mass in detail in the present paper.

AIM

To study the management of rare benign parotid masses.

OBJECTIVES

- To study the best diagnostic modalities in prompt, systematic and cost-effective diagnosis of rare benign parotid masses
- To study the treatment options available in management of benign parotid masses
- To decide the best surgical approach in surgical management of rare benign parotid masses

MATERIAL AND METHODS

We conducted an observational retrospective study at our Tertiary care Hospital after taking due permission from the Institutional Ethics Committee to study the management of rare benign parotid masses in indoor patients in Department of Otorhinolaryngology and Head and Neck Surgery during last two years. Informed and written consent for publication for using the clinical data in good faith were taken. Patient's right to confidentiality has been maintained and patient's identity is revealed nowhere. The clinical knowledge hence gathered has been reviewed with knowledge from the literature and present protocols to conclude the right management approach for management of rare benign parotid masses.

Inclusion criteria: Patients of all sexes and all age groups who were admitted in Department of Otorhinolaryngology and Head and Neck Surgery at our Tertiary Care Centre from April 2021- March 2023 diagnosed with rare benign parotid masses were included in the study

Exclusion criteria: Patients with common parotid swellings, infectious conditions and malignancy were excluded from the study.

OBSERVATIONS

The first case was that of a 34 years male skilled labourer by occupation who presented with a right parotid mass of 12 years duration (Fig. 1). The swelling was of gradual onset and slowly increasing in size. It was a single painless swelling in the region of right cheek. There was no history of sudden increase in size, any episode of decrease in size, bursting, discharge, change in overlying skin, abnormal sensations. There was no history of trauma, radiation exposure, exposure of long duration to heat, heavy metals, dyes and toxins.

However, there was long duration exposure to bright sunlight almost 8 hours a day for last 20 years. There was no previous history of occurrence and/or resolution of a noticeable long duration swelling in same or any other region of the body. There was no history of fever and weight loss. There was no history of systemic illnesses. Patient is a chronic smoker and smokes 2-5 bidis a day for last 10 years. The mass was measured to be an ovoid mass with maximum 14 cm diameter. It had a smooth surface, no edges, no pulsations and no impulses. Temperature of overlying skin was normal. There was no tenderness, well defined regular margins, no fixity to skin or other structures. It was non-compressible and non-reducible. The mass was dull on percussion with no auscultatory sounds. There were no palpable lymph nodes. Bilateral Facial Nerve was intact.

An ultrasound guided FNAC followed by CECT Neck (Fig. 2) were done in a single sitting. All investigations suggested PA and demanded histopathological confirmation.

The second case was that of an 18 years male student who presented with a very slowly progressive left infra-auricular mass over last 5 years (Fig. 3). Rest of the mass was freely mobile but there was slight indentation on skin at a single point at its periphery. There were no overlying or surrounding fistula or sinus. However, there was no history of tuberculosis in patient and close relatives but it still could not be ruled out at presentation. A FNAC done 3 months back suggested a benign inflammatory lesion with epithelioid cells? Tubercular. However, diagnosis could not be confirmed. We did a USG and core biopsy at our centre. USG showed a hypoechoic lesion in superficial lobe of parotid gland with a solid cellular structure without a cavity. Histopathology was recommended to find its exact nature. Histopathology showed cells which were well stained with eosin only without any nuclear material. No signs of malignancy, necrosis and inflammation were seen. Excision was advised to find its exact nature.

The third case was that of a 2 years male child with a huge swelling of left cheek (Fig. 4). It was present almost since birth and gradually increasing in size since then. It blenched on digital pressure and had no pulsations. USG with colour doppler, USG guided FNAC and CECT neck were all suggestive of PL.

RESULTS

The first case was a large parotid PA. Contrast enhanced computerised tomography (CECT)

showed that the mass was limited to superficial lobe of the parotid gland. It was in intimate relationship with four branches of the FN on its deep surface. It was excised in toto by total superficial parotidectomy (Fig. 5-6). The intra-operative period was uneventful. There was transient FN dysfunction in immediate post operative period which was probably due to handling of the branches of the FN. The FN function completely recovered within 72 hours after surgery spontaneously. Histopathology confirmed the diagnosis of PA. There has been no recurrence till date.



Fig. 1: A large parotid mass of 12 years duration in a 34 years old male patient



Fig. 2: Contrast enhanced computerised tomography (CECT) shows a large mass in superficial lobe of parotid in close contact with important neurovascular structures



Fig. 3: A small infra-auricular with a skin indentation at a single point in an 18 years old male patient for last 5 years



Fig. 4: A large parotid swelling in a 2 years old male patient present and enlarging since birth

Pre-operative diagnosis was not clear in the second case. It was a small mass away from the major neurovascular bundles. Partial superficial parotidectomy was done in this case and the mass was completely excised with 5 mm strip of normal surrounding tissue and sent for histopathology (Fig. 7). There were no intra-operative and post-operative complications. Histopathology revealed presence of compact hypercellular Antoni A and myxoid hypocellular Antoni B areas suggestive of PS.

The third case was a paediatric case with PL. USG doppler was done to rule out arteriovenous

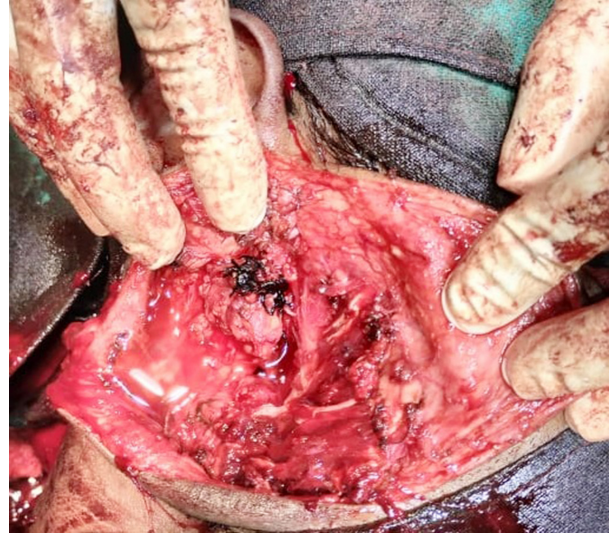


Fig. 5: Total superficial parotidectomy after complete removal of superficial lobe of parotid gland. Facial Nerve (FN) and its branches are intact in the bed

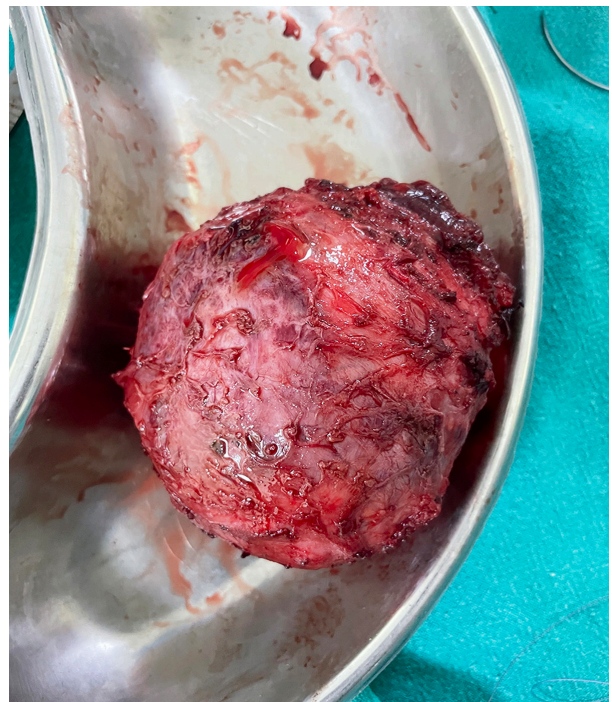


Fig. 6: The Pleomorphic Adenoma (PA) of maximum 14 cm diameter was excised intact and complete, and sent for histopathological evaluation

malformation, presence of blood flow within the mass and significant direct vascular contribution to it by the surrounding blood vessels. CECT Parotid was done to note topographic and angiographic details. The child was taken to the paediatric operation theatre where under sedation, anaesthetic monitoring and after sensitivity testing under CT guidance, a 27 Gauge needle was passed into the mass perpendicular to the skin in pre

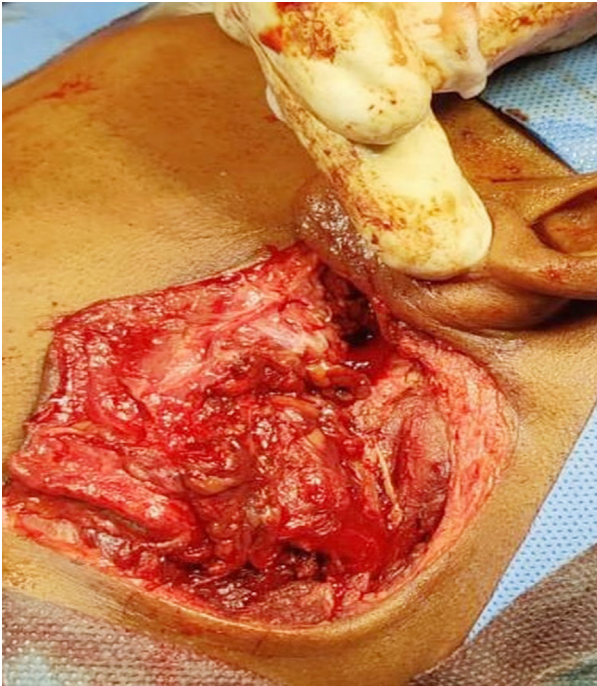


Fig. 7: Partial superficial parotidectomy was done with 5 mm strip of normal tissue around the mass and sent for histopathology

auricular region, thick red blood was aspirated from the collection of immature vessels and 1 mL of sclerosing agents were injected. The child was kept under monitoring for 24 hours and discharged the next day with a plan for repeat procedure after a month for the residual mass. There was no residual



Fig. 8: The mass completely disappeared on first follow up 7 days after the first session of sclerotherapy

mass when the patient came for follow up after 7 days (Fig. 8). The child is under regular follow up for last 3 months and no recurrence is seen during this period. Also, no repeat procedure was done during this period.

DISCUSSION

Aetiology of benign salivary gland tumours is not well understood. There have been concerns regarding petrochemicals, dyes⁸, radiation⁹, mobile phones¹⁰ and Human Papilloma Virus (HPV)¹¹ but none has a proven association with benign parotid tumours. Benign parotid tumours are slow going and usually devoid of other symptoms other than the presence of a mass. Presence of pain, abnormal sensations, increase in growth rate, weakness, skin involvement, fixity or irregularity should alert the physician for possibility of malignant transformation. It is advisable to do an ultrasonography (USG) and USG guided FNAC at presentation in a single sitting. It saves time, money and provides a more accurate diagnosis. USG is very useful as it increases accuracy of FNAC, fulfils most of the requirements of imaging,¹² distinguishes a lump from a node and from diffuse enlargement of the gland. A CECT/ Magnetic Resonance Imaging (MRI) should be done in cases of tumours larger than 3 cm, involvement of deep lobe and parapharyngeal space and suspicion of malignancy. FNAC has an accuracy of 80-90%¹³ and a core biopsy increases sensitivity to 100% and specificity to 92%.^{14,15} In our case of PS, a single point of indentation on skin may be attributed to previous FNAC which was done 3 months back. Common differentials of parotid masses are PA, Warthin's tumour, Oncocytoma, Monomorphic masses, basal cell carcinoma (BCC), cystadenoma and myoepithelioma.¹⁶ Surgery is indicated in most of the parotid masses for definitive histology, presence of continued growth in persistent mass and probability of malignant transformation. Enucleation is not performed now because of high recurrence rate.¹⁷ Either total superficial parotidectomy or partial superficial parotidectomy with a cuff of normal tissue should be done.¹⁸ A parotid tumour will always be found near the FN and its branch¹⁹ and hence carries high risk of iatrogenic FN injury. Overall recurrence rate of PA is 2% and usually years after the surgery.⁷

CONCLUSION

All parotid masses should be managed in a systematic and a comprehensive manner. History

taking and clinical examination are the cornerstone of the management. USG is the most valuable imaging modality. USG with FNAC should be done in a single sitting. However, CECT and MRI may be required in select cases. Core biopsy may add on to histopathological evaluation. Total and partial superficial parotidectomy are the preferred surgical techniques for the masses of superficial lobe. Surgery has to be done very carefully as in most of the cases FN or its branch will be in close relation. Sclerotherapy is a promising novel technique for angiogenic masses. Long term follow up is to be done as recurrence may be seen decades after surgery.

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Submucosal Globular Tongue Swelling: A Rare Presentation of Adenoid Cystic Carcinoma

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ABSTRACT

Introduction: Adenoid cystic is an infrequent malignancy of the salivary glands. Some of its unique features include slow growth, the tendency for perineural invasion, local recurrence, and the ability to metastasize. The mobile tongue's adenoid cystic carcinoma (ACC) is a rare reported case in the literature.

Report: We report a rare case of adenoid cystic carcinoma, in a 55-year-old female arising in the dorsal and lateral aspect of tongue. Histopathology after complete excision of the mass confirmed the diagnosis of adenoid cystic carcinoma. There has been no evidence of disease following excision with tumour-free margins.

Discussion: The pathophysiology of ACC is an understudied area due to the condition's rarity. Chromosomal abnormality and dysregulated MYB oncoprotein have to be thought to play a key role in the pathophysiology and proliferation of ACC.

Conclusions: Adenoid cystic carcinoma of the tongue is exceedingly rare but should be considered in the differential diagnosis in patients with tongue swelling. These tumours are malignant and treatment requires surgical excision and neck dissection followed by adjuvant radiotherapy.

Keywords: Adenoid cystic carcinoma; Carcinoma tongue; Oral cavity; Perineural invasion; Lingual tumours.

INTRODUCTION

Adenoid cystic carcinoma (ACC) is an infrequent malignancy that affects secretory glands, most

commonly the salivary glands. Although rare, it is an essential differential to consider for a painless swelling in the head and neck region because of its high likelihood to metastasis. ACC has a slight predominance in females, with a peak incidence in the fifth and sixth decades of life. It represents 1 to 2% of all malignant tumours of the head and neck with a reported incidence of 2.9% of all cases in the mobile tongue.¹ We present a case of a 55-year-old female with ACC of mobile tongue treated with surgery followed by adjuvant radiotherapy and a brief review of the literature.

CASE REPORT

A 55-year-old female patient presented to the OPD with complaints of slow-growing swelling in the tongue for the past one year before the

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consultation. She also complained of associated difficulty chewing and speaking for 2 months. There was no addiction history.

On intraoral clinical examination and palpation, an obvious smooth firm mass of about 2.5 cm in diameter on the left side of the tongue with the same colour as the surrounding mucosa was observed without other oral lesions. (Fig. 1) The cervical lymph nodes were not palpable.



Fig. 1: Pre-operative image showing the swelling on the dorsal surface of left side of the tongue.

Contrast-enhanced magnetic resonance imaging was done for the patient which revealed a T2/STIR hyperintense enhancing tongue lesion of size 2.1 x 2.8 x 2.6 cm involving the left anterior and lateral border of the tongue, not involving the base and root of the tongue. (Fig. 2) The lesion appeared benign on clinic-radiological correlation.



Fig. 2: MRI showing hyperintense enhancing lesion in the tongue (A) Axial view, (B) Coronal view & (C) Sagittal view.

She underwent an excisional biopsy under general anaesthesia. Intraoperatively a 2.5 x 2 x 1 cm well-encapsulated swelling was excised in toto (Fig. 3) and sent for histopathological examination.



Fig. 3: (A) intraoperative picture showing the excision of the tongue mass. (B) Image showing the excised mass.

A detailed morphological examination revealed a basaloid tumour comprised of varied patterns,

amounting to 75% cribriform, 20% solid, and 5% tubular architecture. On immunohistochemistry, the ductal epithelial cells were positive for CK7 and c-kit, while the abluminal/myoepithelial cells were

highlighted by p63 and focally by S100. Hence, a diagnosis of Adenoid Cystic Carcinoma, Grade 2 was rendered. No necrosis or features of high-grade transformation were noted. (Fig. 4)

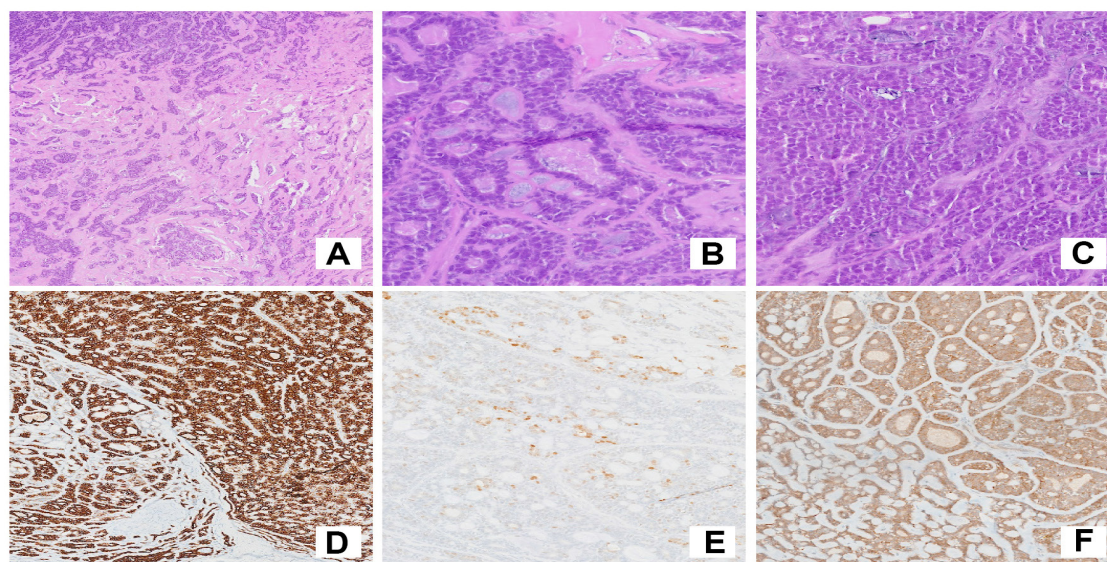


Fig. 4: A) Basaloid tumour with cribriform and tubular pattern (Haematoxylin and eosin, 20x) B) Cribriform and tubular pattern (Haematoxylin and eosin, 400x) C) Solid area (Haematoxylin and eosin, 400x) D) Immunohistochemistry for CK 7, highlights the luminal and abluminal cells (400X) E) Immunohistochemistry for S-100 highlights few myoepithelial cells (400X). F) Immunohistochemistry for CD 117 highlights luminal cells (200X)

This unexpected diagnosis of malignancy, required a margin revision of the tongue lesion and left selective neck dissection (levels 1 to 4). The final histopathological report revealed scanty residual viable adenoid cystic carcinoma with all margins free of tumour. Total regional lymph nodes were sixty-six, negative for metastasis (0/66). No nodal metastasis was identified.

DISCUSSION

Billroth defined ACC as “cylindroma” and explained its recurring nature in 1859. In 1953, Foote and Frazell introduced the term “adenoid cystic carcinoma”.²

Adenoid cystic carcinoma is a rare malignancy arising from the secretory glands. It most commonly affects the salivary glands. It accounts for about 1 % of the head and neck malignancies. It primarily affects the minor salivary glands, accounting for over 10% of salivary gland malignancy.³ The tongue is the site of origin for ACCs in the head and neck region, accounting for 3.4% to 17.1% of instances, with the mobile tongue accounting for 2.9%. (1) The majority of cases are reported to be in the base of the tongue.⁴ Nerve invasion is a hallmark of the tumour, which also implies a poor prognosis.

Histologically three distinct patterns are noted in ACC: tubular, cribriform, and solid, with cribriform pattern being the most common and associated with a better prognosis. Immunohistochemistry considerably contributes in the diagnosis of ACC since the tumor cells stain positive for smooth muscle actin, S100, vimentin, as well as for MYB and CD 117 (receptor tyrosine kinase c-KIT), which helps differentiate it from other malignancies.⁵

The treatment approach for adenoid cystic carcinoma of the tongue is surgical resection followed by radiotherapy. Adenoid cystic carcinoma is known to cause cervical lymph node metastases with an incidence of 17.6% in mobile tongue as reported by Carrasco *et al.*⁶ Level 1b and II are the most commonly involved lymph nodes. Thus, selective neck dissection should be considered in patients with clinical N0 neck with ACC of the tongue. A combined approach in the management of ACC of the tongue includes wide local excision of the ACC of the tongue with adequate margins, with or without reconstruction, selective neck dissection, and adjuvant radiotherapy. Adjuvant radiotherapy is generally considered in advanced T-stage cases and the presence of positive tumour margins. Some authors have suggested radiotherapy in cases of advanced non resectable tumour.

Adenoid cystic carcinoma of the tongue has been rarely reported in the literature. Goldblat et al reported 5 cases of ACC of the tongue in 1987 with no mention of the specific site demarcation in the tongue.⁸ In 2009, Luna Ortiz et al reported 2 cases of mobile tongue ACC in a retrospective study of 68 patients of head and neck ACC from 1986 to 2006.⁷ Xi Tang et al reported a case of ACC of the mobile tongue treated with surgery, reconstruction with anterolateral thigh flap, and adjuvant radiotherapy in 2019.⁹ Kumar S et al reported a case of ACC of the tongue in 2016, along with a review of literature demonstrating the rare presentation of adenoid cystic carcinoma in the mobile part of the tongue.¹⁰ Thus, unusual presentations of adenoid cystic carcinoma of the mobile tongue have been recorded infrequently in the literature.

Postoperative management typically involves close surveillance to monitor for recurrence, as it carries a tendency for local recurrence and also distant metastasis.

CONCLUSION

We present a rare case of adenoid cystic carcinoma arising in the dorsal aspect of the tongue. Adenoid cystic carcinoma though rare, should be considered as a differential in growths identified in the tongue. Understanding the clinical presentation, diagnostic approach, histopathological characteristics, and treatment options for adenoid cystic carcinoma is crucial for achieving optimal patient outcomes and minimizing the risk of recurrence.

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Paediatric Subglottic Mucocele: Early Intervention and Recovery

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ABSTRACT

Subglottic mucocele is a rare entity in the pediatric population, often associated with intubation-related complications. We present a case of a 1.5-year-old male child with a history of repeated hospitalizations for respiratory distress, ultimately diagnosed with subglottic mucocele. Despite being born at full term without comorbidities, the patient required mechanical ventilation shortly after birth. Imaging revealed an ill-defined lesion in the subglottic region, confirmed histologically as a mucocele. Following initial airway compromise during excision, emergency tracheostomy was performed, followed by successful removal of the mass via coblation. Postoperative follow-up showed significant improvement in respiratory symptoms, and the patient was successfully decannulated. This case highlights the importance of considering subglottic mucocele as a differential diagnosis in pediatric airway obstruction, necessitating prompt diagnosis and appropriate surgical management. We discuss the etiology, differential diagnosis, and surgical techniques for managing subglottic lesions in pediatric patients, emphasizing the significance of a multidisciplinary approach for optimal outcomes.

Keywords: Subglottis; Mucocele; Coblation; Stridor; Airway.

INTRODUCTION

Subglottic mucocele is a rare but potentially life-threatening condition in pediatric patients, often arising as a consequence of endotracheal intubation.¹ Despite its infrequency, the

management of subglottic mucocele demands a prompt and comprehensive approach to prevent airway compromise and ensure favourable outcomes.² In our case, early intervention played a pivotal role in expediting the patient's recovery. Prompt recognition of the subglottic mucocele and its potential for airway compromise led to immediate surgical intervention. Despite encountering a challenging intraoperative airway event necessitating emergency tracheostomy, decisive action allowed for continued surgical excision and subsequent resolution of the lesion. Additionally, close postoperative monitoring enabled timely adjustments in the management plan, facilitating successful decannulation and ensuring a smoother transition to respiratory autonomy. This emphasis on early recognition and intervention underscores the critical role of proactive management strategies in mitigating complications and promoting optimal patient outcomes in pediatric airway disorders.

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CASE PRESENTATION

A 1.5-year-old male child presented to the paediatrics emergency in our tertiary care centre with a history of shortness of breath and noisy breathing for the past three months. The child had a history of mechanical ventilation 2 hours after birth following feeding and required mechanical ventilation for 48 hours. Till 1.5 years of age, the child had a history of repeated hospital admissions for breathing difficulty and received treatment for pneumonia. History revealed that he had a term delivery, developmental milestones as per age and

no other history of comorbidities. On examination, the child had a normal BMI (body mass index) and he had occasional biphasic stridor with suprasternal retractions.

Investigations

A contrast-enhanced computed tomography from the base of the skull to the diaphragm revealed an ill-defined, polypoidal, non-enhancing, hypodense lesion, likely granulation tissue or inflammatory aetiology in subglottis (mainly on the right), maximum thickness 6.5mm, narrowest diameter 4mm (TR), 14mm (AP) at the level of C5 (Fig. 1).

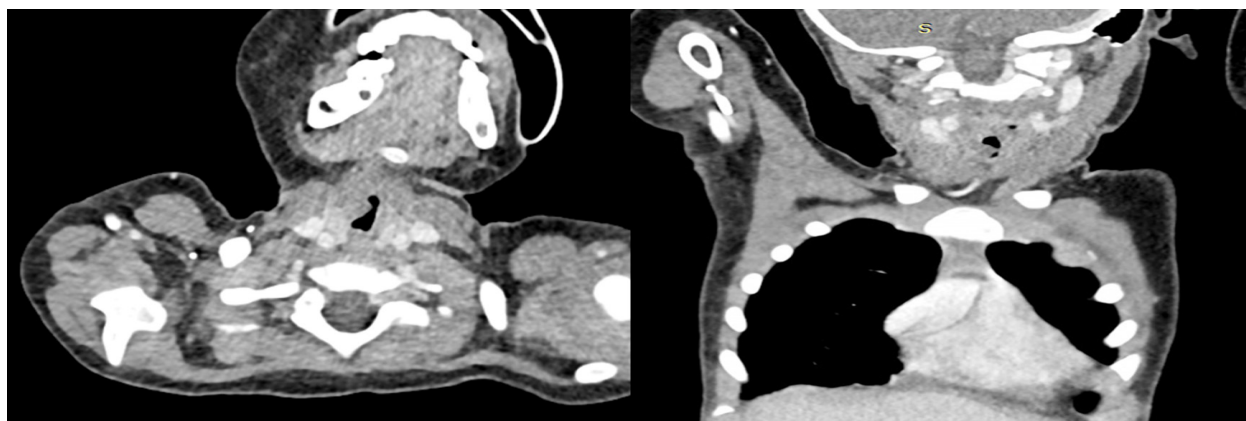


Fig. 1: Axial and coronal CECT neck showing soft tissue narrowing airway in subglottis

Treatment

Direct Laryngoscopy under Monitored anaesthesia noticed a smooth pinkish mass in the subglottis arising from the posterolateral wall of the right subglottic region. (Fig. 2A) Intraoperative sudden fall in SpO_2 necessitated emergency tracheostomy. The patient was kept on ventilator for 6 days and after stabilization on the eighth day after the first procedure, he was taken up under GA via

tracheostomy tube (4.5mm). Mass was excised with the help of a coblator. The histopathology report came out to be mucocele. The child was discharged after advising tracheostomy tube care. Twenty-five days later, the child was re-admitted, and a direct laryngoscopic assessment was done, which showed no recurrence and healthy, healed mucosa was seen in the postoperative bed. Decannulation has been done uneventfully (Fig. 2B).

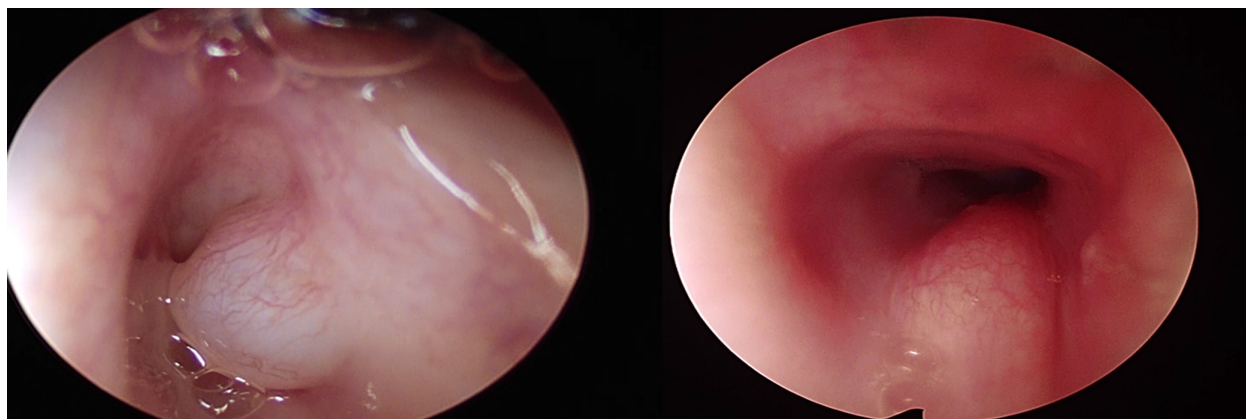


Fig. 2: a) Endoscopic picture of subglottic mass

b) 1 month post excision picture

Table 1: Differentiating features of different subglottic masses

Cause	Differentiating feature	Treatment
Subglottic haemangioma	Biphasic stridor at 2-3 months of age, asymmetric narrowing of trachea on imaging	Open excision, endoscopic laser excision or microdebrider, propranolol. ⁵
Subglottic cyst	History of endotracheal intubation, prematurity, cystic nature-air/fluid content on imaging	Endoscopic marsupialization with laser/cold instruments. ⁹
Respiratory papillomatosis	History of anogenital warts in mother, 'cauliflower-like' exophytic growth lesions, solid/cavitated pulmonary nodules in cases of lung involvement	Excision with microdebrider, laser or coblator, interferon alpha, bevacizumab. ⁶

Outcome and follow-up

After discharge, the child remained clinically stable. His noisy breathing subsided. The child was followed up every two weeks post-excision, and no signs of respiratory distress. He gained weight from 11kg(post-operative period) to 16kg (1 month postoperative).

DISCUSSION

In literature, most subglottic cysts are related with prematurity due to requirement of intubation. Our report, with an average term of 38 weeks of amenorrhea at birth, is inconsistent with the literature. Obstructive lesions of the pediatric airway can occur at any level, from the nose and nasopharynx through the pharynx and supraglottis to the glottis, subglottis, trachea, and lower airway structures. At any level, the lesions can be congenital or acquired; expansile, dynamic, or static; and partially to progressively to entirely obstructive. The subglottis is particularly injury-prone in neonatal and pediatric populations because it is a significant point of contact for the life-saving measures of endotracheal intubation.¹

There appears to be a clear emergence of subglottic mucus retention cysts as a complication following intubation in pediatric population. The presence of the endotracheal tube causes erosion, inflammation, and subsequent scarring in the subglottic region, leading to the obstruction of submucous gland ducts and the formation of cysts.³ The subglottic submucosa has a more significant proportion of glandular soft tissue than the rest of the airway, making it the narrowest part of the neonatal airway as opposed to the glottis in the adult. The clinical importance of subglottic cysts depends on the degree of airway compromise, as small and solitary cysts may be an incidental finding on endoscopy, and large or multiple cysts may cause catastrophic airway obstruction. Subglottic cysts should be differentiated from hemangioma, stenosis, and

respiratory papillomatosis, which are more common causes of paediatric stridor and rare subglottic lesions such as thyroglossal cysts and ectopic thymic cysts.⁴ The different causes of subglottic masses and the differentiating features are given in Table 1.

Contingency plans should be delineated while planning for excision, including a plan for emergency tracheostomy in case of difficult ventilation or intubation. Excision of the cysts will often be sufficient to correct the obstructed airway; however, the recurrence rate is as high as 40%.⁶ Endoscopic removal allows a clear view of the lesion, which aids the excision of cyst content, de-obstructing the airway and causing no damage to the underlying mucosa. This approach could explain the low recurrence rate observed compared to other modalities, such as CO2 laser.^{3,6} The use of microdebrider has been reported to allow better control of the excision of the cysts. Steehler et al. also reported promising results of the post-marsupialization application of Mitomycin-C in a recent study. It could prevent recurrence and reduce post-marsupialization scarring.⁷

CONCLUSION

- Timely diagnosis and surgical intervention are paramount in managing pediatric subglottic mucocele to prevent airway compromise and minimize morbidity.
- Early recognition of subglottic lesions, coupled with decisive surgical excision and vigilant postoperative monitoring, facilitates successful resolution of symptoms and avoidance of long-term complications.
- Multidisciplinary collaboration among otolaryngologists, paediatricians, anesthesiologists, and other healthcare professionals is essential for optimizing outcomes in pediatric patients with subglottic mucocele.

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Kidd EAM, editors. Dental caries: The disease and its clinical management. Oxford: Blackwell Munksgaard; 2003. p. 7-27.

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[8] World Health Organization. Oral health surveys - basic methods, 4th edn. Geneva: World Health Organization; 1997.

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