

ADENOID CYSTIC CARCINOMA: UNUSUAL CAUSE OF GLOBUS PHARYNGEUS

DR SWATI TANDON¹, DR.VASUN BATRA², DR. ISHWAR SINGH³

¹M.S(ENT),DNB(ENT), REGISTRAR ENT, MAULANA AZAD MEDICAL COLLEGE, NEW DELHI,INDIA

²MBBS, RESIDENT,ENT. MAULANA AZAD MEDICAL COLLEGE, NEW DELHI, INDIA

³MS(ENT), PROFESSOR,ENT. MAULANA AZAD MEDICAL COLLEGE, NEW DELHI, INDIA

Email - Drsawatitandon86@gmail.com, vasun58@gmail.com, Drisingh62@gmail.com

Abstract: Adenoid cystic carcinoma comprises about 10% of salivary gland malignancies. Most of them arise in minor salivary glands. It is known for its late presentation, perineural invasion and local recurrence. Adenoid cystic carcinoma of base tongue is very rare. We report a rare case of adenoid cystic carcinoma of base tongue in a female who presented as globus pharyngeus.

Key Words: Adenoid cystic carcinoma, salivary glands, tumor, globus pharyngeus, perineural invasion.

INTRODUCTION:

Adenoid cystic carcinoma (ACC) is a malignant tumor presenting 1-2% of all head and neck tumours. They generally present as slow growing masses. The tumour has marked propensity for neural invasion. In oral cavity, palate is the most common site followed by tongue. The most common site of distant metastasis is lungs. The treatment of choice is surgery followed by radiotherapy. We report a rare case of ACC of base tongue in a 50 year old female managed by surgery and radiotherapy.

CASE REPORT:

A 50 year old female presented with chief complaint of foreign body sensation in throat for 3 months. There were no complaints of dysphagia, odynophagia or change in voice. Patient had presented earlier to private practitioners who diagnosed her as globus pharyngeus and prescribed antacids. Oral examination was unremarkable. On indirect laryngoscopy, a bulge was seen in base of tongue (approx.2x2cm) on left side. On palpation, it was firm, non tender and overlying mucosa was intact. There was no bleeding from the mass. There was no associated cervical lymphadenopathy. A provisional diagnosis of minor salivary gland benign tumor base tongue was made. Biopsy from the mass revealed normal salivary gland tissue. CECT neck revealed heterogeneously enhancing mass lesion in base of tongue on left side(Fig.1). Patient underwent hematological investigations and CXR, which were normal. Patient was planned for surgical excision of the tumor via mandibulotomy approach. Paramedian mandibulotomy was done between lateral incisor and canine and tumor exposed in toto(fig.2). Tumor was excised taking adequate margins and defect was closed primarily. Postoperatively, feeding tube was put which was removed after 5 days. Intravenous steroids were given for 48hours to avoid tongue edema. Postoperative period was uneventful. Histopathology report revealed adenoid cystic carcinoma with perineural infiltration(fig.3). Patient underwent radiotherapy and is free of local recurrence or distant metastasis at 6 months.

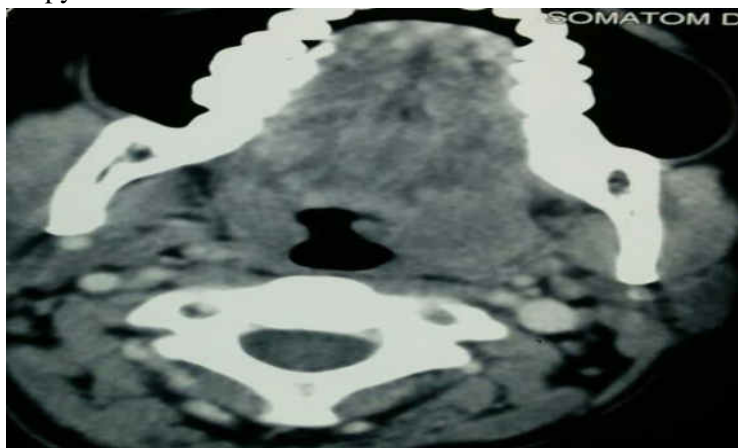


FIG.1 CECT NECK SHOWING ENHANCING MASS IN BASE OF TONGUE LEFT SIDE



FIG.2 INTRAOPERATIVE PHOTOGRAPH

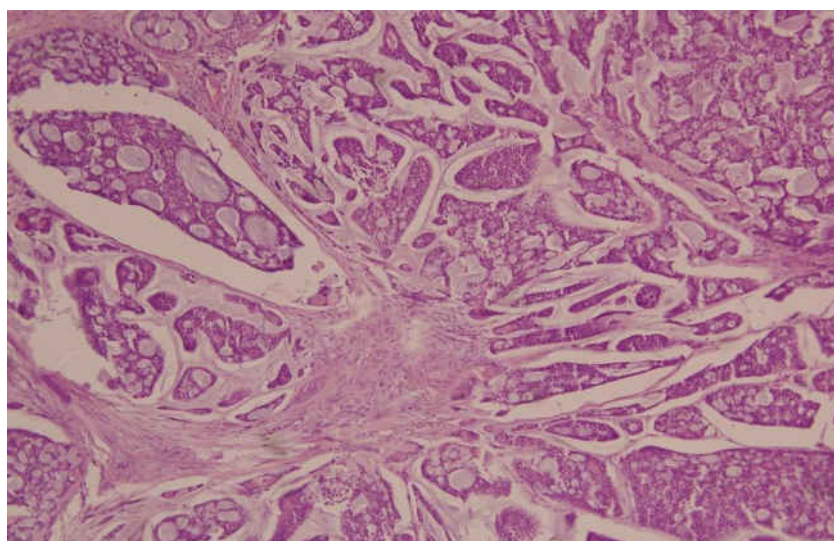


FIG.3 HISTOPATHOLOGICAL PHOTOGRAPH

DISCUSSION:

Majority of minor salivary gland tumours are malignant. Most common is mucoepidermoid followed by adenoid cystic carcinoma. ACC accounts for approximately 10% of all neoplasms of the salivary glands¹. The most common intraoral site for minor salivary gland tumors is the hard palate, followed by the base of the tongue². According to the AFIP (Armed Forces Institute of Pathology) ACC predominates in the 5th and 6th decade of life². The tongue is the site of origin in 3.4-17.1% of cases³. Our patient was a 50 year old female.

One of the peculiar features of ACC is local as well as distant metastasis. The rate of loco-regional recurrence in ACC of the head and neck is approximately 4% to 20% whereas distant metastasis is seen in 20% to 50% cases⁴. The lungs have been reported as the most common sites of distant metastasis⁵. Other sites are bones, brain, liver and even pancreas⁶. In a study of 67 patients diagnosed with ACC of oropharynx at Sloan Kettering Hospital, it was found that T stage and margin status were independent predictors for locoregional recurrence-free survival. Also, it was concluded that clinical T stage, anatomic subsite, and margin status were independent predictors for overall survival⁷.

Perineural invasion is a pathognomic feature of ACC and has been reported in almost half of the cases⁸. Clinically, it can be diagnosed preoperatively only if there is associated nerve palsy. Imaging can now be used as an important tool for detecting perineural invasion preoperatively, thereby calling for aggressive treatment. In a study of 13 patients of ACCs of head and neck, CECT and CEMRI were performed and the images were retrospectively evaluated for the detection of perineural invasion. They concluded various features of perineural invasion as abnormal density/signal intensity, contrast enhancement or widening of the pterygopalatine fossa,

palatine foramen, incisive canal, mandibular foramen and mandibular canal, and enlargement or excessive contrast enhancement of a nerve⁹. However, no such findings were observed in present case.

Histopathologically, ACC may present in three different patterns: solid, tubular, and cribriform; solid pattern is associated with poor prognosis. In our patient, cribriform pattern with perineural invasion was seen.

The surgical management of ACC should be aggressive as tumour is known for local recurrence due to perineural invasion. Various surgical approaches for ACC of base of tongue are: transoral resection, mandibulotomy approach, lateral pharyngotomy and Robotic surgery¹⁰. We used paramedian mandibulotomy approach in our case. Ultrasound guided transcutaneous interstitial photodynamic therapy (PDT) has also been used as a salvage treatment for ACC of base tongue¹¹.

CONCLUSION:

We present this rare case of ACC of base tongue wrongly diagnosed as globus pharyngeus. It is very essential to perform indirect laryngoscopy when patient presents with non specific symptoms to avoid misdiagnosis. Aggressive management and long term follow up is required in ACC as they are known for late recurrences.

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