

Mucoepidermoid Carcinoma of Tongue: A Case Report and Review of Literature

¹Dr. Fatema Saify, Reader, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

²Dr. Revati Raghunath Shinde, Postgraduate Student, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

³Dr. Amit Wasti, Associate Professor & Head of Department, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

⁴Dr. Nidhi Sharma, Senior Lecturer, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

⁵Dr. Rashmi Christin Kerketta, Assistant Professor, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

⁶Dr. Himanta Ghrilahare, Assistant Professor, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

Corresponding Author: Dr. Revati Raghunath Shinde, Postgraduate Student, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G)

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Abstract

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor, typically affecting the parotid gland, with rare occurrence in the tongue. We report a case of a 49 – year - old male presenting with a painful ulcer on the left lateral border of the tongue. Histopathological analysis of an incisional biopsy demonstrated tumor islands composed of intermediate, mucous, and squamous cells with stromal invasion.

Special stains (PAS and mucicarmine) confirmed mucin production, establishing a diagnosis of intermediate-grade MEC. This case highlights the rarity of MEC at the tongue and emphasizes the importance of Histopathological evaluation for accurate diagnosis. Early detection and long-term follow-up are essential due to the risk of recurrence.

Keywords: MEC, PAS and Mucicarmine, Histopathological.

Introduction

Mucoepidermoid carcinoma (MEC) is the second most common neoplasm of the salivary glands, accounting for approximately 5% of all head and neck cancers¹ and most common malignant salivary gland tumor, first studied and described as a distinct pathologic entity by Stewart et al. in 1945.² The tumor was initially titled "Mixed epidermis and mucous secreting carcinoma," later replaced by "MEC."³

MEC commonly affects the parotid gland, particularly in women, and typically occurs in the fifth decade of life.¹

The global annual incidence of malignant salivary gland neoplasms ranges from 0.9 to 2.6 cases per 100,000 individuals.⁴ MEC arises from the pluripotent reserve cells of the excretory ducts of salivary glands.⁵

Mucoepidermoid carcinoma (MEC) usually affecting the parotid and minor salivary glands.⁶ Occasional cases have been reported in the palate, retro molar area, floor of the mouth, buccal mucosa, lips, tongue, lacrimal glands, bronchi, nasal mucosa, esophagus, maxilla, mandible, liver, cervix, penis and anus.⁷ In this case report, we present a malignancy involving the minor salivary glands of the tongue in a 49-year-old male patient.

Case Report

A 49-year-old male patient presented with an ulcer on the left side of the tongue for 3 days. As per the patient's history, he was apparently asymptomatic 2 months earlier, after which he experienced loosening of teeth in the lower left posterior region, leading to continuous tongue biting and subsequent creation on the left lateral border of the tongue. The ulcer progressively enlarged, resulting in severe pain and difficulty in speech. The patient had a history of chronic tobacco, gutkha, and alcohol use for 15 years. He had

been on medication for diabetes mellitus (1 year) and hypothyroidism (3 years).

On extraoral examination, the left submandibular lymph node was palpable and found to be large, fixed, and tender. Intraoral examination revealed a 2 × 2 cm pale ulcer on the left lateral border of the tongue (Figure 1), associated with teeth 35 and 36. The lesion was firm in consistency, fixed to the underlying muscle, and severely tender. A provisional diagnosis of malignancy of the left ventrolateral surface of the tongue was made, following which an incisional biopsy was performed under local anesthesia.

The specimen was received in the Department of Oral and Maxillofacial Pathology and Oral Microbiology, Government Dental College, Raipur (C.G.).



Figure 1: Clinical presentation showing ulcer on left ventrolateral border of tongue.

Microscopy showed tumour cells arranged in the form of islands. These islands showed peripheral low columnar to cuboidal cells with eosinophilic cytoplasm and large hyperchromatic nuclei and central cells appear to be round to oval in shape with large hyperchromatic nuclei (intermediate cells) [Figure 2]. Interspersed within these cells were few clear cells and mucous cells [Figure 3] with pale basophilic cytoplasm and eccentrically placed flat nucleus. Deeper section showed tumour cells invading into the connective tissue stroma in the form of small groups and cords. These cells were polygonal in

shape with eosinophilic cytoplasm and hyper chromatic nuclei (squamous cells) [Figure 4]. The mucinous nature of MEC was highlighted by special stains such as PAS [Figure 5] and mucicarmine [Figure 6] and A diagnosis of MEC intermediate grade was given.

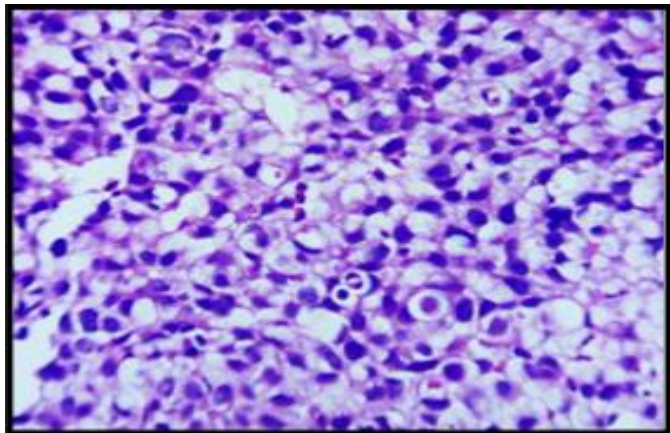


Figure 2: Photomicrograph showing a mixture of small hyper chromatic abundant intermediate cells with scanty cytoplasm (H&E, 40 xs)

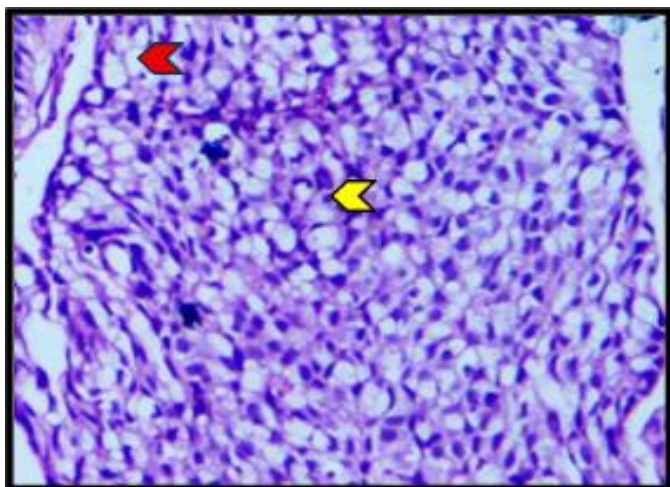


Figure 3: Photomicrograph showing mixture of clear cells (red arrowhead) and mucous cells (yellow arrowhead) (H&E, 40x)

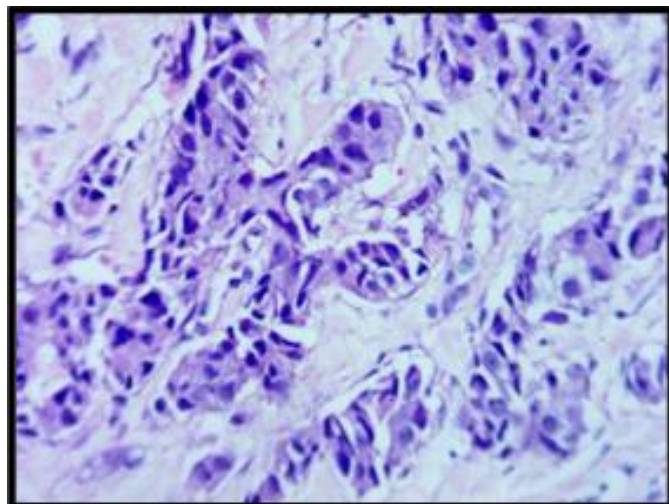


Figure 4: Photo micro graph showing polygonal epidermoid cells (H&E, 40x)

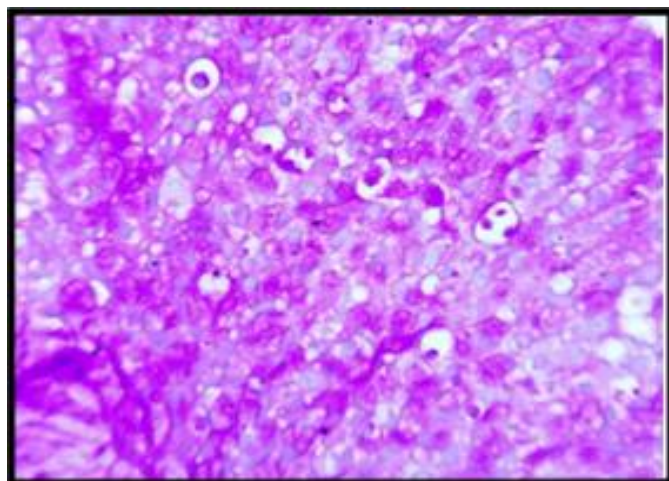


Figure 5: Photomicrograph showing Intracellular and extracellular staining for mucin (PAS, 40X)

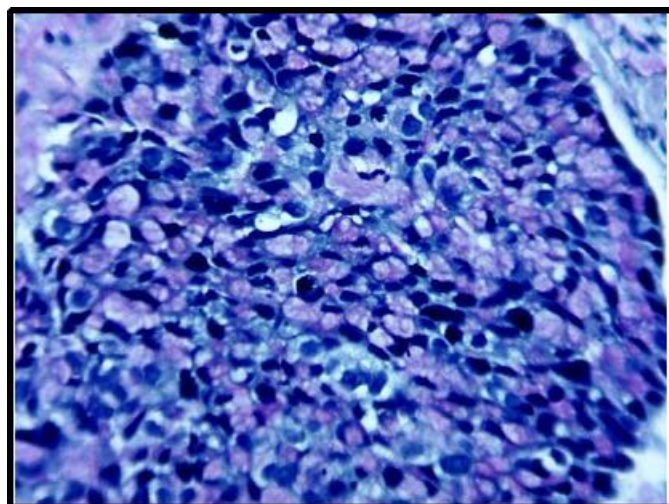


Figure 6: Photomicrograph showing Intracellular and extracellular staining for mucin (mucicarmine, 40X)

Discussion

Salivary gland tumors constitute 5% of head-and-neck neoplasms, and among them, MEC is the most common malignant salivary gland tumor, frequently affecting the parotid and minor salivary glands in adults. Although MEC accounts for 30 % of all salivary gland malignancies, it represents only 10% of overall salivary gland tumors and less than 5% of head and neck cancers.¹ Approximately 50-60% of MEC cases originate in major salivary glands, with the parotid gland accounting for over 80% of cases, the sub mandibular gland 8-13%, and the sublingual gland 2-4%.

Minor salivary gland MEC primarily affects the palate, with less common occurrences in the tongue, retro molar region, and buccal mucosa.^{4, 8, 9}

The etiology of MEC is yet to be established completely. Exposure to ionizing radiations has been suggested in few cases. A gene translocation at t (11; 19) (q21; p13) encoding a transcription factor CREB (Camp response element – binding protein) has been detected in more than 80% of the MEC cases.^{10, 11}

A retrospective study conducted by Li LJ¹² evaluated 3,461 salivary gland neoplasms. The mean age of patients was 41.38 years

for benign lesions and 45.20 years for malignant lesions. The male – to - female ratio was 0.99:1 in benign cases and 1.34:1 in malignant cases. Lesions most frequently involved the parotid gland, followed by the palate and sub mandibular gland. Muco epidermoid carcinoma was identified as the most common malignant neoplasm, while the parotid gland and palate were the predominant anatomical sites of involvement.

A clinic opathological study conducted by Dhanuthai Ketel.¹³ evaluated 311 intra oral salivary gland tumors among 16,358 accessioned cases in the Thai population. Of these, 311 cases (1.90%) were diagnosed as intraoral

salivary gland tumors, including 147 benign lesions (47.27%) and 164 malignant lesions (52. 73%). The palate was identified as the most frequently affected site, and Mucoepidermoid carcinoma was reported as the most common malignant tumor. Buchner A¹⁴ evaluated the relative frequency of intraoral minor salivary gland tumors (IMSGTs). Among 92,860 accessioned cases, 380 (0.4%) were Diagnosed as IMSGTs, including 224 (59%) benign and 156 (41%) malignant lesions.

Mucoepidermoid carcinoma, accounting for 21.8% of cases, was the most common malignancy identified. The study conducted by Wang et al.¹⁵ reported that Mucoepidermoid carcinoma (MEC) was a malignant, locally infiltrative salivary gland neoplasm that predominantly involved the parotid gland. Occurrence at the base of the tongue was considered extremely rare, with fewer than 30 cases identified in the Pub Med and CNKI databases. In a retrospective analysis spanning 50 years, 145 cases of tongue base MEC were identified in the SEER Program database. The study demonstrated that MEC of the tongue base had a very low incidence, occurred more frequently in females, and was predominantly diagnosed in patients older than 50 years, accounting for approximately 75% of cases.

Minor salivary glands were distributed throughout various regions of the oral cavity, including the palate, retro molar area, floor of the mouth, buccal mucosa, lips, and tongue. Among patients with intraoral minor salivary gland tumors, approximately 1% presented with carcinoma of the tongue. Over 30-year period, retrospective study of tongue base tumors conducted, total 22 cases were malignant 45% of them diagnosed as MEC.¹⁶

The study conducted by Alanizi OS¹⁷ reported that Mucoepidermoid carcinoma (MEC) of the tongue base is a rare clinical entity. The first reported case was

documented in 1973. In a retrospective study conducted over a 30-year period on tongue base tumors, 22 cases were identified as malignant, of which 45 % were diagnosed as MEC. Similarly, Goldblatt and Ellis found that 85 % of the tongue base lesions in their study cohort were malignant in nature.

The classification of MECs into low, intermediate and high grade was based on the relative proportion of cell types. MEC of the minor salivary glands is composed histologically of three cell types: mucous cells, epidermoid squamous cells and poorly differentiated intermediate cells that have the ability to differentiate into either mucous producing or epidermoid cells.¹⁸

- High - grade MEC tumour- The predominant epidermoid element
- Low-grade MEC tumour- The mucin-producing cells within a predominantly cystic architecture
- Intermediate-grade MEC tumours- The less cystic and show a greater tendency to form large, more irregular nests or sheets of squamous cells and often have a more prominent intermediate cell population

Auclair et al.¹⁹ studied the grading criteria of minor salivary glands MECs. The Histopathological features that indicated high-grade behaviour were:

- An intra-cystic component of < 20% (+ 2 points);
- four or more mitotic figures per 10 high power field (+ 3 points);
- neural invasion (+2points);
- necrosis (+3points);
- cellular anaplasia (+4 points) According this grading scale, tumours with:
 - score of 0-4 were considered low-grade;
 - scores 5-6 were considered intermediate (between low grade and high-grade);
 - Scores ≥ 7 indicated highly aggressive behaviour.

Kokemueller H et al.²⁰, histologically classified the tumors into low-grade, intermediate-grade, and high-grade categories based on the presence of cystic structures, cellular atypia, and cell type. Low-grade tumors were well-circumscribed, cystic, and rich in mucous cells, whereas intermediate-grade tumors were comparatively more solid and less circumscribed. Low - grade tumor occurred more commonly in the minor salivary glands, while high-grade tumors were observed predominantly in the major salivary glands.

In a retrospective study, Hicks J²¹ evaluated 26 pediatric patients with salivary gland Mucoepidermoid carcinomas (MECs). Only one case involved the tongue base. The median tumor size was 2.5 cm (range: 1.5–5cm). Histologically, nine tumors were classified as low grade, 15 as intermediate grade, and two as high grade.

A study by Goldblatt LI²² analyzed 55 cases of primary salivary gland tumors of the tongue from the files of the Armed Forces Institute of Pathology. Of these, 50 were malignant, and over 85% involved the tongue base. Low-grade Mucoepidermoid carcinoma was the most common malignant tumor, accounting for 38% of cases.

Surgical excision is the treatment of choice for MEC. They have a local recurrence rate varying from 15% to 75 %, usually appearing without change in histopathology, mostly during the 1st postoperative year.⁷

Conclusions

High-grade MEC is an aggressive malignancy requiring a multidisciplinary treatment approach combining surgery, radiotherapy, and, in some cases, chemotherapy. Early diagnosis through Histopathological evaluation is crucial for effective management. There are anatomical location of this case, along with the patient's history of tobacco and gutkha usage, may lead to a diagnostic challenge for both oral pathologists and surgeons. Given the moderate

prognosis associated with MEC, long – term follow –up is essential to monitor for recurrence and metastasis.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/ has given his/ her/ their consent for his/ her/ their images and other clinical information to be reported in the journal. The patients understood that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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