MUCOEPIDERMOID CARCINOMA - A RARE OCCURRENCE ON THE GINGIVA -A CASE REPORT

Anuradha.Ch*, Lalith Prakash.K, Chandrasekhar. P***, Shamala R****, Reddy B.V.R.***** Madhusudhan Kasipathi******

Mucoepidermoid carcinoma (MEC) is a malignant salivary gland neoplasm which exhibits a widely diverse biologic behavior, well correlated with the histological grade of the tumor. About 6% of the major and 19% of the minor salivary gland tumors is mucoepidermoid carcinoma. It is commonly seen on palate, buccal mucosa, tongue, and the retromolar area. It is a rare entity on the gingiva, because of the absence of salivary glands in this location. Here, we present a case report of Mucoepidermoid carcinoma of the mandibular lingual gingiva. It was clinically diagnosed as Pyogenic granuloma. Based upon the histopathological findings and special stains like Periodic Acid Schiff's and Mucicarmine a confirmatory diagnosis of Mucoepidermoid carcinoma was given. Careful examination and histopathological correlation are mandatory, for better prognosis of the patient.

Key words: Mucoepidermoid carcinoma; Salivary glands; Gingiva

INTRODUCTION:

Most malignant tumors arising in the gingiva are squamous cell carcinomas. Salivary gland tumors occurring in the gingiva are rare [1]. Mucoepidermoid carcinoma is a common malignant lesion of the major and minor salivary glands [2]. It is defined as "a malignant glandular epithelial neoplasm characterized by mucous, intermediate and epidermoid cells, with columnar, clear and oncocytoid features"[3]. It was first described by Masson and Berger in 1924 [4]. In 1945, Stewart and associates described its mucous-secreting and epidermal cellular elements thus establishing it as a pathologic entity [5].

This carcinoma of the salivary glands accounts for 5% of all salivary gland tumors. About 2/3 arise within the parotid gland and 1/3 arise within the minor salivary glands. It is also the most common malignant tumor to arise in children and adolescents under 20yrs of age [6]. The female to male ratio was 1.3: 1. Although the palate was the most frequent site for mucoepidermoid carcinoma (37.5%), tumors were frequently found in the gingiva (18.8%), floor of the mouth (18.8%), the buccal mucosa (12.5%), the lip (6.2%) and the tongue (6.2%) [7]. The characteristic presentation is that of a painless, slow –growing mass [8].

Microscopically the tumor is composed of combination of mucinous, squamous and intermediate type cells. Mucinous cells are intensely positive for mucicarmine and PAS-Alcian blue [9].

Standard treatment for MEC is surgical resection, with an adequate margin. MEC has been considered a radioresistant tumor, though postoperative radiation is thought to be effective [10]. The prognosis for low-grade lesions is excellent, with a cure rate of over 90 %. Intermediate-grade lesions are more aggressive, whereas for high-grade lesions the survival rate is only 30%-54% [11].

CASE REPORT:

A 39 year old female patient reported to the department of Oral and Maxillofacial Pathology with an asymptomatic swelling in the right lower back tooth region, which was noticed 3 months back. Past medical and dental history was non contributory.

** Senior Lecturer ** Post graduate student *** Senior Lecturer **** Professor & Head, Department of Oral Pathology ***** Professor, Department of Oral Patholgy ****** Madhusudhan Kasipathi, DDS, (USA), Overseas contributor*

Extra oral examination revealed no facial asymmetry & no lymphadenopathy.

Intra oral examination revealed a well circumscribed, pink colored, non ulcerative, 3x2.5cm sessile mass on the gingiva extending to the alveolar mucosa lingually, in relation to 45 & 46 (Fig. 1). On palpation, it was soft to firm, non tender, not compressible, showing blanching on pressure, bleeding on probing and no associated tooth mobility.

Radiographs reveal no bony erosion. The complete blood picture also showed normal results. Based upon the clinical and radiological information, a provisional diagnosis of Pyogenic granuloma was given. An excisional biopsy was performed under local anesthesia.

Microscopic examination revealed a tumor composed of numerous nests of epidermoid cells admixed with few clear cells and mucous cells in some areas, within a connective tissue stroma. The epidermoid cells are characterized by squamoid features demonstrating a polygonal shape and few areas of keratinization. Dysplastic features like hyper chromatism, prominent nucleoli, altered nuclear – cytoplasmic ratio and mitotic figures were observed. Some areas showed mucous cells with foamy cytoplasm and clear cells with vacuolated cytoplasm. (Fig. 2)

Special staining procedures like "Periodic Acid Schiff's stain" (PAS) and "Mucicarmine stain", were performed, both of which showed positive results (Fig.3 & 4).

Based upon the histopathological features and results of special stains a confirmatory diagnosis of "Mucoepidermoid carcinoma – High grade" was given.

DISCUSSION:

Mucoepidermoid carcinoma is the most frequently diagnosed malignancy of the salivary glands and has the second highest frequency of occurrence. The term "Mucoepidermoid tumor" was first used by Stewart, Foote, and Becker in 1945. In most of the cases, the etiology is unknown, the most common probability being therapeutic radiation [12, 13]. Specific etiologic factors for MEC were not evident in our study.

Although MEC accounts for less than 10% of all tumors of the salivary gland, it constitutes approximately 30% of all malignant tumors of the salivary gland [13]. Mucoepidermoid carcinomas account for 50% of malignant salivary gland tumors in children [8].

The prevalence of MEC is noted to be highest during the 3rd and 5th decades of life (14). It shows a slight female predilection with a ratio of 3:1[12]. In our case also the age and gender predilection of the patient are consistent with the above mentioned findings.

The most common intraoral site is palate (17.9%), followed by cheek (5.5%), lip (3.4%), tongue (3.4%), floor of the mouth (3.0%) and gingiva (0.6%) [12, 15]. One case of an intraoral MEC occurring on the lower gingiva was reported and it showed metastasis to cervical lymph nodes, which was confirmed by CT findings [16]. Ide et al., reported a case of a firm, polypoid, non-ulcerated mass tethered to the gingiva, which was histopathologically diagnosed as Sclerosing mucoepidermoid carcinoma with eosinophilia [17]. In our case, the lesion occurred on the gingiva which is a rare site.

It is a very rare entity on the gingiva, due to the absence of salivary glands in this location. Though gingival metastases are rare, a case of Mucoepidermoid Carcinoma of the oesophagus which showed an unusual metastasis to gingiva & pharynx was reported by Dimri et al [18].

Similarly, Hentati et al., reported a case of a 47 year old man presenting with a gingival metastasis from non-small cell lung carcinoma [15]. So in our case, we confirmed that, there was no evidence of any primary lesion in the above mentioned areas, with a chest x-ray and abdominal scan.



Fig 1: A painless, slow growing lingual mass i.r.t. 45; 46

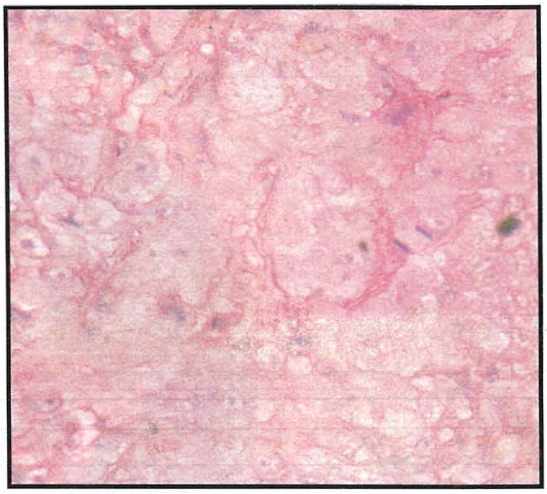


Fig 2: H & E stained section showing prominent nucleoli (A); Mitotic figure (B); & Altered nuclear-cytoplasmic ration (C) (High power view)

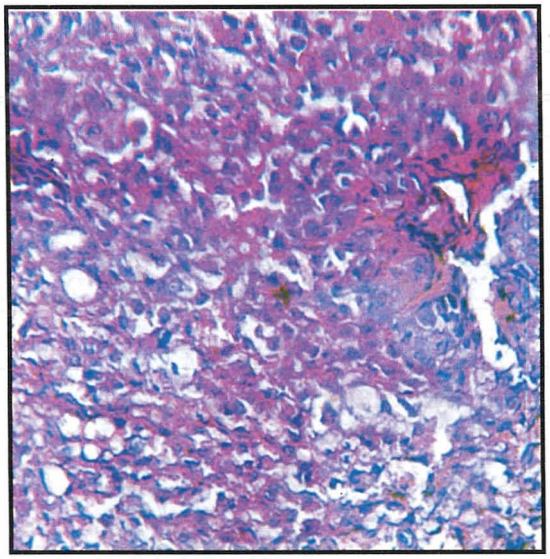


Fig 3: Periodic Acid Schiff Staining showing positivity

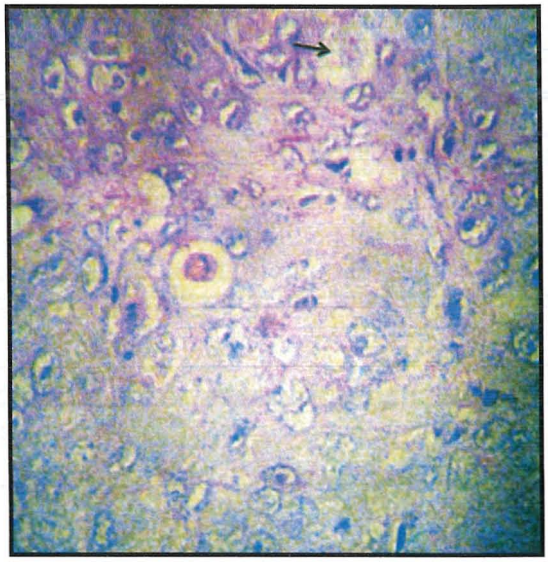
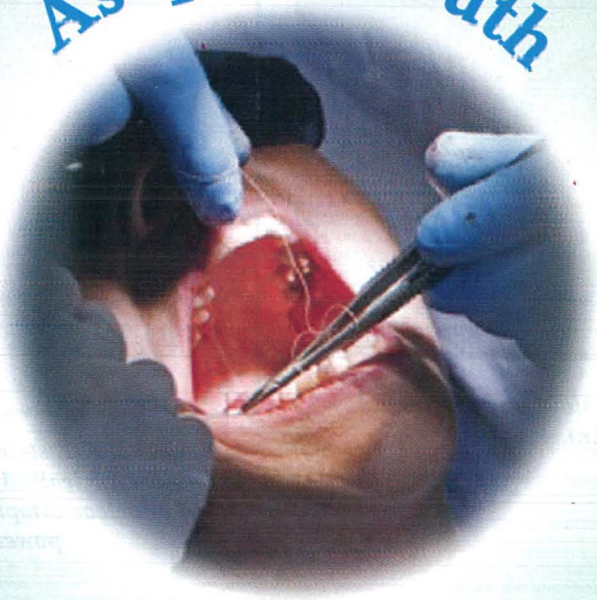


Fig 4: Mucicarmine stain showing positivity

Aseptic Mouth



HEXIDINE

(Chlorhexidine gluconate 0.2% w/v)

- Broad Spectrum of activity against Gram negative, Gram positive, vegetative bacteria, yeasts, dermatophyte, fungi & lipophilic viruses
- Binds strongly to mucosa



- **prevents plaque in absence of brushing**
- **prevents & treats gingivitis**
- **aids in the treatment of mouth & throat infections**

The Gold Standard
Antiplaque, Antimicrobial

ICPA

ICPA HEALTH PRODUCTS LTD.

E-mail : icpahealth@eth.net

Earlier, an intra osseous Mucoepidermoid carcinoma arising from salivary gland inclusions in the mandible was reported by M. Dorman et al [19]. Interestingly, Bernard et al. have reported about 6 cases of choristoma arising from ectopic salivary glands on gingiva [20].

From the foregoing, as the tumor was located on the gingiva, and since there was no evidence of any primary lesion that would have metastasized, we arrived at a point that it would have originated from an ectopic salivary gland. Ectopic salivary gland tissue occurs in many sites within the head and neck region [21].

Microscopically the tumor is composed of combination of mucinous, squamous and intermediate type cells. Mucinous cells are intensely positive for mucicarmine and PAS Alcian blue [9,22]. In our case also, similar results were observed.

Studies of Ki-67 and PCNA expression demonstrate the difference in immunoreactivity for these proliferation markers between low- and high-grade MECs. Clinicopathological studies have shown increased Ki-67 and PCNA immunoreactivity in MECs. Relatively few tumor cells show PCNA or Ki-67 immunoreactivity in low-grade MECs, while intermediate-grade MECs show a moderate increase [23].

Radical surgery is the treatment of choice for all high-grade MECs, or low/intermediate-grade tumors that are large and involve the bone. It is contraindicated in small, localized tumors with low- to intermediate-grade histological appearance [23].

Radiation therapy is used with surgery although this carcinoma seems to be radioresistant. Radiation therapy should be added in high-grade tumors and for patients with unclear surgical margins or for patients with positive lymph nodes. It is generally accepted that for high grade MECs, radiotherapy combined with surgery should achieve local control of the disease and good survival rates [24].

For the MECs, the histopathological stage of the tumor is to be associated with the clinical findings (rapid or slow development, symptomatic / asymptomatic, the location of the tumor, in the palate, in the floor of the mouth, the clinical staging T1 – T4), before any treatment decisions are made [2].

In our case, surgical excision of the lesion was done, healing was good and there was no evidence of any recurrence even after 6 months.

Prognosis of MEC's is a function of the histological grade, adequacy of excision, and clinical staging.

CONCLUSION:

In most of the cases, gingival lesions might appear clinically benign, but sometimes may show a malignant histological course. Hence, dental and medical practitioners who treat oral lesions should consider biopsy as an important diagnostic tool which aids in early diagnosis and therapeutic intervention for better prognosis.

References:

1. Urade et al. Unusual presentation of carcinoma of the mandibular gingiva: report of a case. J Oral Maxillofacial surgery 55:1007-1010, 1997.
2. Paul L. Auclair; Robert K. Goode; Gary L. Ellis; Mucoepidermoid carcinoma of intraoral Salivary glands - Evaluation and application of grading criteria in 143 cases; CANCER April 25, 1992; Volume 69; No. 8; 2021-2030.
3. R.K. Goode; A.K. El Naggar: in: World health organization classification of Tumors; Pathology & Genetics; Head & Neck tumors (2005); IARC; Mucoepidermoid carcinoma: 219-220.
4. Nagao T, Sugano I, Ishida Y et al. Mucoepidermoid carcinoma arising in warthin's tumor of the parotid gland: report of two cases with Histopathological, ultrastructural and immunohistochemical studies.histopathology 1998; 33: 379-386.
5. Deepti Simon, Thara Somanathan, K Ramdas and Manoj Pandey.central mucoepidermoid carcinoma of mandible-a case report and review of the literature.world j surg oncol 2003; 10: 1186-1477.
6. Margaret S.Brandwein, Katya Ivanov, Derrick I et al. Mucoepidermoid carcinoma a cinicopathologic study of

80 patients with special reference to histological grading. Am J Surg Pathol 2001; 25: 835-845.

7. R. Rajendran, B. Sivapatha Sundaram, Shafer. Hine. Levy – "Shafer's text book of Oral Pathology"; 5th edition; Elsevier; 2006.
8. Reza Rahbar, J.Fredrik Grimmer, Sara O.Vargas et al. Mucoepidermoid Carcinoma of the Parotid Gland in Children. Arch Otolarynol Head Neck Surg.2006; 132:375-380.
9. Hacer Haltas, Zehra Bozdag, Nasuhi Engin Aydin, Bulent Mizrak. Cerebral metastasis of mucoepidermoid carcinoma as the first clinical presentation: detailed immunohistochemical evaluation. Turkish Journal of Cancer 2008;38 (2):83-86.
10. Hiroyuki Ozawa, Toshiki Tomita, Koji Sakamoto et al. Mucoepidermoid Carcinoma of the Head and Neck: Clinical Analysis of 43 Patients. Jpn J Clin Oncol 2008; 38(6): 414-418.
11. Brad W. Neville; Douglas D. Damm; Carl M. Allen; Jerry E. Bouquot; "Oral and Maxillofacial Pathology"; 2nd edition; Elsevier; 2007
12. Gary l. Ellis; Paul l. Auclair; douglas R. Gnepp; Surgical pathology of the salivary glands; vol.25; W.B.saunders company
13. Derek Kofi O.Boahene, Kerry D.Olsen, Jean E.Lewis et al. Mucoepidermoid Carcinoma of the parotid gland. Arch Otolaryngol Head Neck Surg.2004; 130:849-856.
14. Joseph A. Regezi; James J. Scuibba; Richard C.K. Jordan – Oral pathology clinical pathologic correlations; 4th edition – Saunders, 2003: Mucoepidermoid carcinoma: 203-207.
15. Hentati D., Chraiet N., Kochbati L., Maalej M. – Gingival metastasis from the lung through a needle and a pin: A Case report; J. Cancer Radiother 2007, Dec, 11 (8): 500-502.
16. Ichiro Ogura, Tohru Kurabayashi, Teruo Amagasa, Takehito Sasaki; CT findings of cervical lymph node metastasis from intraoral mucoepidermoid carcinoma: Histopathological correlation; Oral radiology 2002, Vol. 18, No. 2: 9-15.
17. F. Ide, K. Obara, K. Enatsu, K. Mishima, I. Saitò - Sclerosing mucoepidermoid carcinoma of the oral cavity – Case report; J Oral Pathol Med 2005, 34: 187-189.
18. Dimri K; Rastogi N; Lal P – Carcinoma of the oesophagus with unusual metastasis to gingiva and phalanx; Indian journal of Medical sciences 2003, Vol. 40, Issue 1, pg: 37-38.
19. M. Dorman and D. Pierse - Ectopic salivary gland tissue in the anterior mandible: a case report; British dental journal 2002, vol. 193, No. 10: 571-572.
20. Bernard S. Moskow; Ernest Baden – Gingival salivary gland Choristoma – Report of a case; Journal of clinical periodontology 2005, vol. 13, Issue – 7, pg: 720-724.
21. Seifert G, Miehlke A, Haubrich J, Chilla R. Diseases of the Salivary Glands. New York Georg Thieme, 1986; 237-238.
22. John D. Bancroft; Marilyn Gamble; "Theory & practice of Histological techniques"; 5th edition; Churchill Livingstone; 2005: 177-188.
23. K. Triantafillidou; J. Dimitrakopoulous; F. Iordanidis; D. Koufogiannis, "Mucoepidermoid carcinoma of minor salivary glands: a clinical study of 16 cases and review of the literature"; Oral Diseases (2006) 12: 364-370.
24. Eversole LR, Rovin S, Sabes WR (1972). Mucoepidermoid carcinoma of minor salivary glands: report of 17 cases with follow-up. J Oral Surg 30: 107-112.

CHANGE OF ADDRESS

Note new address of Chief Editor

PROF. V P JALILI, KALA NIWAS,
5 MISHRA VIHAR COLONY
NEAR GITA BHAWAN MANDIR,
INDORE-452 001 (M.P.) INDIA