

CASE REPORT

Mucoepidermoid carcinoma of the posterior-lateral border of tongue: a rare presentation

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SUMMARY

Mucoepidermoid carcinoma (MEC) is the most common malignant tumour of the major and minor salivary glands. Minor salivary glands are scattered in different areas of the oral cavity such as palate, retromolar area, floor of the mouth, buccal mucosa, lips and tongue, but so far, only a few lingual MEC cases have been documented in the literature and most of the studies have shown a predilection for base and dorsum of the tongue. We report a rare case of MEC involving the posterior-lateral border of the tongue.

BACKGROUND

Salivary gland malignancies are rare neoplasms and account for only 5% of all head and neck malignancies.¹ Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumour representing 35% of malignant tumours from major and minor salivary glands.² It is most common in the parotid gland and the minor salivary glands constitute the second most common site with a site predilection for palate followed by buccal mucosa. MEC of tongue is quite rare and only less than 30 cases have been documented so far.^{1,3} We report a rare presentation of MEC in the posterior-lateral border of the tongue and discuss the potential risk of inadequate management of such lesion as it may often be misdiagnosed as foliate papillitis.

CASE PRESENTATION

A 45-year-old woman came to us complaining of swelling with intermittent pain on the left side of the tongue that had afflicted her for 2 years. It started as a small lesion and grew gradually to reach the current size. She had a history of thyroid cancer, which was removed 15 years ago. The patient was moderately built and nourished and there was no pallor, icterus, cyanosis or clubbing. There was no generalised lymphadenopathy. Intraoral examination revealed an ovoid well-circumscribed lesion measuring 2×2 cm on the posterior-lateral border of the left side of the tongue (figure 1). The overlying mucosa was erythematous and the surface was smooth. The lesion was tender on palpation and hard in consistency.

DIFFERENTIAL DIAGNOSIS

1. Foliate papillitis
2. Traumatic fibroma
3. Benign vascular/neural tumours
4. Metastatic thyroid cancer.

TREATMENT AND FOLLOW-UP

Excisional biopsy of the lesion was done and histopathological examination of the tissue sections showed numerous cystic spaces lined by epithelial cells and islands and sheets of epidermoid cells in the connective tissue (figure 2). The neoplasm consisted of three populations of cells: the epidermoid, and the intermediate and mucous cells. The epidermoid cells and the intermediate cells showed vesicular nuclei, prominent nucleoli, nuclear pleomorphism and increased mitosis (figure 3). The epithelial cells lining the cystic spaces and forming the solid islands were interspersed by Per iodid acid Schiff (PAS) -positive mucous cells (figure 4). Based on these features, the lesion was diagnosed as low-grade MEC. The excretory duct opening into the oral mucosa also was lined by neoplastic cells showing cellular atypia and mucous cells (figure 5). However, the tumour margins were free of neoplastic cells.

OUTCOME AND FOLLOW-UP

Following surgical excision, the healing was uneventful and a regular follow-up of the patient for a period of 3 years did not reveal any evidence of recurrence.

DISCUSSION

MEC was first described by Volkman in 1895, which was further elaborated on by Stewart in 1945



Figure 1 An ovoid well-circumscribed swelling, measuring 2×2 cm on the posterior left lateral border of tongue.



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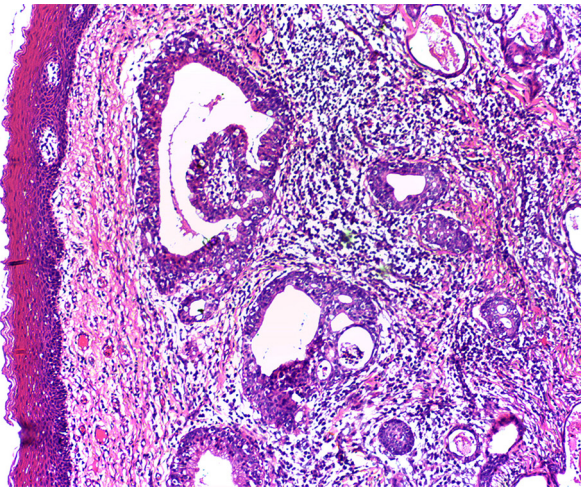


Figure 2 Histopathological examination of the tissue sections showing numerous cystic spaces lined by epithelial cells and solid epithelial islands in a fibrous connective tissue stroma (H&E, 10×).

as mucoepidermoid tumour⁴ due to its highly variable biological behaviour. Later, the term ‘mucoepidermoid tumour’ was replaced by ‘mucoepidermoid carcinoma’, a more preferred designation, as it was recognised that even low-grade tumours could occasionally exhibit malignant behaviour.

MEC is the most common salivary gland malignancy with the second highest frequency of occurrence among all salivary gland tumours. Though it shows a predilection for parotid gland with 45% of the cases occurring at this site,⁵ it is the most common malignancy involving the minor salivary glands. Minor salivary glands are distributed throughout the oral mucosa, except in the anterior region of the hard palate and the gingiva. The minor salivary gland tumours account for 14%–22%⁶ of all salivary gland carcinomas with MEC being the most common type of intraoral adenocarcinoma,⁷ showing predilection for the palate (35.5%) followed by buccal mucosa (14%).⁸ Its occurrence in other intraoral sites, including tongue, are extremely rare and a PubMed search of related English-language literature revealed only 16 case reports of lingual MEC (table 1). Though lingual MEC can occur at any of the sites where salivary gland tissue

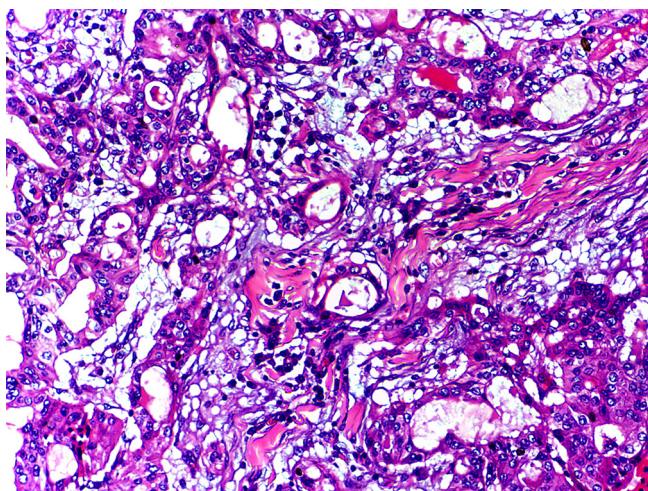


Figure 3 The epidermoid cells and the intermediate cells showing vesicular nuclei, prominent nucleoli, nuclear pleomorphism and increased mitosis (H&E, 20×).

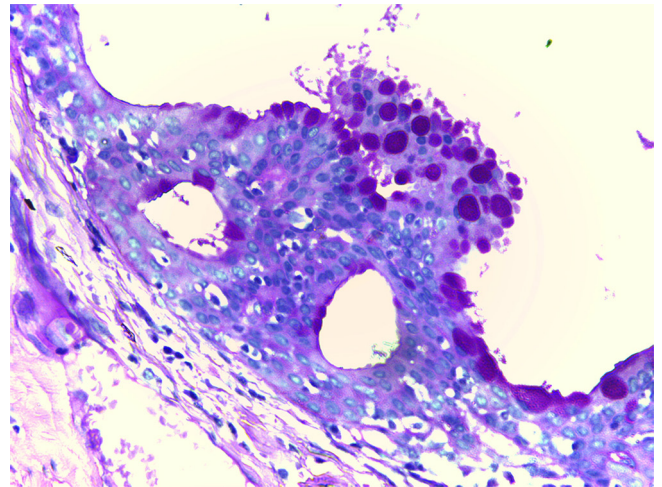


Figure 4 The epithelial cells lining the cystic spaces (stained blue) interspersed by PAS-positive mucous cells (stained magenta) (H&E, 40×).

is normally distributed, most cases are seen in the tongue’s base, representing more than 50% of all tumours.² The review of previously reported cases showed that most of them developing in the base and dorsum of the tongue were, in fact, located in the tongue’s midline. Different from that, the present case occurred in the posterior-lateral border of the tongue, anterior to the circumvallate papillae, so it can be considered the first case published in the literature out there. Importantly, as this site mainly comprised foliate papilla, MEC may be mistakenly diagnosed as foliate papillitis, thus being ignored in the early stages of its development by the clinicians. Since our patient had a history of thyroid carcinoma, and as metastatic thyroid carcinoma was considered as a possible diagnosis in this case, we went ahead with excisional biopsy. As with most documented cases of lingual MECs, microscopically, the present case also showed features of low-grade mucoepidermoid carcinoma.

MEC is thought to arise from pluripotent reserve cell of excretory duct^{9–11} from the salivary gland that potentially can differentiate into squamous, columnar, mucous and clear cells. In the

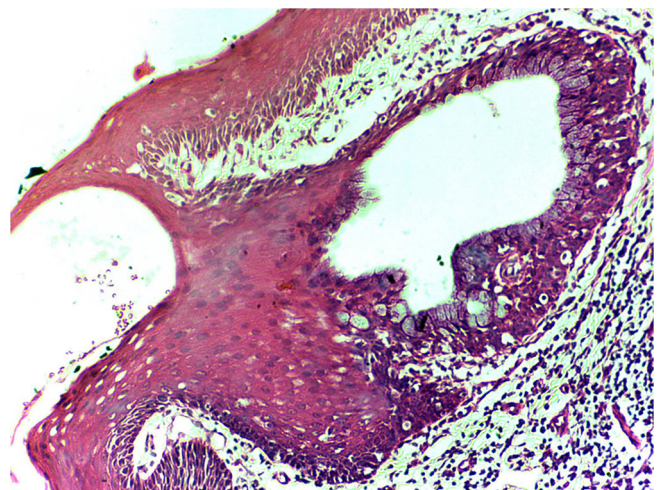


Figure 5 The excretory duct opening into the oral mucosa, lined by neoplastic epithelial cells showing cellular atypia and mucous cells suggestive of the origin of the neoplasm from the excretory duct (H&E, 20×).

Table 1 Summary of the reported cases of mucoepidermoid carcinoma of tongue

Serial No	Authors	Gender	Site	Tumour grade
1	Heidelberger and Batsakis ¹⁷	F	Base of the tongue	High grade
2	Adkins and Putney ¹⁸	M	Base of the tongue	Low/intermediate
3	Pickell ¹⁹	M	Base of the tongue	Low grade
4	Pfendler ²⁰	F	Base of the tongue	Low grade
5	Varghese <i>et al</i> ²¹	M	Base of the tongue	Low grade
6	Pires <i>et al</i> ²²	F	Oral tongue (right anterior border)	Low grade
7	Andrews and Eveson ²³	M	Base of the tongue	High grade
8	Leong <i>et al</i> ³	M	Base of the tongue	High grade
9	Liu <i>et al</i> ²⁴	F	Base of the tongue	High grade
10	Sobani <i>et al</i> ²⁵	M	Base of the tongue	Low grade
11	Martellucci <i>et al</i> ²⁶	F	Base of the tongue	Low grade
12	Kalogirou <i>et al</i> ⁷	M	Posterior dorsal tongue	Low grade
13	Mesoilella <i>et al</i> ²	F	Base of the tongue	Low grade
14	Bollig <i>et al</i> ²⁷	F	Base of the tongue	Intermediate
15	Su <i>et al</i> ²⁸	M	Base of the tongue	Low grade
16	Rubin <i>et al</i> ²⁹	F	Base of the tongue	Low grade

F, female; M, male.

case presented here, MEC's tumour cells were also evident in the corresponding excretory duct opening into the superficial oral epithelium, suggestive of its origin from the excretory duct. The tumourigenesis of MEC is not clearly understood. The translocation of gene material, t(11;19)(q21;p13.1), has been identified as a solitary abnormality in some cases of MEC.¹² The occurrence of MEC, among the survivors of the Hiroshima and Nagasaki atomic bomb explosions,¹³ and in patients after radiation therapy for thyroid carcinoma or leukaemia¹⁴ confirms the aetiological role of ionising radiation in MEC. Recently, the presence of transcriptionally active human papillomavirus (HPV) 16/18 was demonstrated in MEC, implicating high-risk HPV in its pathogenesis.¹⁵

MEC is a unique malignancy since it demonstrates a broad spectrum of aggressiveness ranging from an indolent tumour, which is cured by surgery, to aggressive neoplasm, which is prone to local invasion, recurrence and metastasis.¹⁶ Tumour location, clinical stage, tumour grade and adequacy of treatment are the main prognostic factors for MEC-affected patients. The treatment is usually surgical excision with free margins, and neck dissection is indicated in the presence of lymph node metastases or those classified as grade III.

Learning points

- ▶ Mucoepidermoid carcinomas (MECs) are rare salivary gland neoplasms showing varied biological behaviour.
- ▶ Due to its typical location, MEC occurring in the posterior-lateral border of the tongue may often be misdiagnosed as foliate papillitis.
- ▶ As minor salivary glands are widely distributed in the oral mucosa, MEC can occur in any locations inside the mouth; hence, it is essential that it should be considered as a differential diagnosis for all intraoral soft tissue swellings.

Contributors All authors were responsible for the concept and design; definition of intellectual content; drafting/revision of intellectual content; data acquisition, analysis and interpretation; final approval of version published; agreement to be accountable for the article.

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