CASE REPORT

Mucoepidermoid carcinoma on dorsum of tongue: An unusual presentation in Nigerian patient

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Abstract
The aim of this study was to report an unusual presentation of mucoepidermoid carcinoma (MEC) on the dorsum of tongue and to add to the body of knowledge on the various sites of occurrence of the lesion. MEC is a malignant, locally invasive tumor of salivary glands composed of varying proportions of mucous-secreting and squamous cells, with the presence of intermediate cell differentiation between these other two cell types. More than half of reported cases involve major salivary glands and it is seen rarely on the dorsum of tongue. We present an unusual case of a low-grade MEC on the dorsum of tongue of a 40-year-old man. Discussion on clinical and microscopic types and immunohistochemical features of this tumor is presented in relation to pertinent literature. Biologic behavior and clinical manifestations of MEC usually correlate with its histological grade, which are required for appropriate treatment.

Keywords: Histological grade, minor salivary gland, mucoepidermoid carcinoma, tongue tumor

Introduction
Mucoepidermoid carcinoma (MEC) is a malignant epithelial neoplasm of salivary gland origin that contains varying proportions of squamous, mucus-secreting, and intermediate cells. Although a primary disease of the salivary gland, intraosseous MEC has also been reported. It is the most common malignant tumor of the major salivary glands and the most common malignant salivary gland tumor in children. MEC affects more commonly the parotid gland and the palate is the most common site of occurrence in minor salivary gland with few cases reported on the retromolar area, floor of mouth, buccal mucosa, lip, and tongue. Although MEC of the tongue is reported rarely in literature, about 22 cases have been documented with none from Nigeria until date. The previous reports of MEC on the base of tongue exist in literature; it has been documented in a 12-year-old child and several adult patients while reports on the dorsum are sparse. We report a case of a 40-year-old male who presented with a low-grade MEC of the dorsum of tongue.

Case Report
A 40-year-old male reported at a private dental clinic in Lagos state, Nigeria, with a 14-month history of an asymptomatic tongue swelling [Figure 1]. Examination revealed a well-circumscribed nodular mass on the dorsum of the tongue. The mass was restricted to the tongue’s musculature, sparing the mucosa. There was no associated ulceration around the tongue. It measures about 5 cm by 4 cm. The tongue mass was firm in consistency, non-tender with no related paresthesia or anesthesia. There were no palpable lymph nodes on neck examination. We considered a provisional diagnosis of fibroma and a differential diagnosis of nerve sheath tumor. Computed tomography and magnetic resonance imaging were not available due to the patient’s financial constraint. Submitted incisional biopsy specimen was evaluated. Microscopic analysis of hematoxylin and eosin-stained sections of tissue revealed a circumscribed tissue mass with proliferating mucus-producing cells, cystic spaces, and intermediate cells. In addition, few squamous cells were disposed mainly as glandular structures and in areas as solid nests surrounding the mucus-producing cells. The neoplastic cells were suspended in mature fibrous connective tissue stroma [Figure 2]. Individual tumor cells showed features of mild cellular atypia; however, there was no evidence of perineural invasion, mitotic figures, or areas of necrosis. The tumor cells were restricted to the lamina propria and muscular layer of the tongue, sparing the resection margins, and overlying epithelium. Fibrosis and dense mononuclear inflammatory cell infiltrates were present around an area of
extravasated mucin. A diagnosis of low-grade MEC was made after a grade score of 4 was awarded using the criteria by Auclair et al., 1992, for minor salivary gland MEC.\(^9\) Immunohistochemistry showed positivity for cytokeratin 7 (CK7), P63, and epithelial membrane antigen (EMA) [Figures 3-5].

Partial glossectomy and neck dissection under general anesthesia were performed to excise the tumor. Post-operative recovery was uneventful. No additional adjunct therapy was employed in the management of this patient and the patient has remained tumor-free 4-year post-operative.

**Discussion**

This is a case of low-grade MEC, which has occurred on the dorsum of the tongue in Nigerian patient. It is considered an unusual occurrence because no MEC on the tongue has been reported from Nigeria and especially from the dorsum of the tongue. While most of the reports in literature agree with MEC as the most common malignant primary salivary gland tumor, Nigerian studies have shown that adenoid cystic carcinoma is the most common.\(^{10}\) The presentation of this case on the dorsum of the tongue is contrary to previously reported MEC on the base of tongue and posterolateral tongue.\(^{6-8}\) The low-grade histological grading of this lesion is similar to reports of MEC on the base of tongue.\(^{6,7}\) This may account for its clinical presentation as an asymptomatic slow-growing lesion.\(^6\)

Alanizi\(^7\) reported a change in voice inpatient with MEC of base of tongue, but there was no associated change in voice of our patient, this may be accounted for by the occurrence of this lesion on the dorsum of tongue. The age of this patient is in agreement with documented age of the occurrence of MEC, usually the fifth decade with slight female predilection.

Immunoperoxidase staining showed strong positivity for CK7 and 14 [Figure 3] demonstrating the presence of both the mucus and squamous cells in the lesion, although this does not reflect the
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Figure 5: Histologic features demonstrate neoplastic cells exhibiting a positive staining for epithelial membrane antigen (at ×4)

biologic behavior of MEC. Lesion was also immunoreactive for p63 and EMA [Figures 4 and 5].

The patient was treated with a wide local excision intraorally with adequate tumor-free margins.

This conforms to the accepted treatment of cases of MECs of minor salivary glands with low- to intermediate-grade that there was no adjunct therapy in the form of post-operative radiotherapy and chemotherapy in the treatment of this patient as recommended for low-grade tumors.

Conclusion

MEC of the dorsum of tongue is a rare lesion.

Clinical Significance

Atypical locations and innocent appearance of MEC such as the one reported can mislead the clinician and can lead to an erroneous diagnosis. Therefore, proper investigations are required for appropriate treatment.

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References