

Mucoepidermoid carcinoma of the lower lip: A rare site of occurrence

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy. However, MEC arising in the minor salivary glands of the lower lip is extremely rare, and to date, only a few have been reported. In this study, we are reporting a case of MEC of the lower lip in a 60-year-old male patient. The patient presented with a slow growing mass in the lower lip for about three years with a recent history of an increase in size. Surgical excision and histopathological examination of the resected mass confirmed the diagnosis of low-grade MEC. We are presenting this case to highlight that although rare, MEC should be kept in mind in the differential diagnosis of any lip mass.

Key words: Histopathological examination, lower lip, mucoepidermoid carcinoma

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy, which is commonly seen in the parotid gland.^[1] However, MEC can also occur in other locations, such as, the respiratory and digestive tracts.^[2] Minor salivary gland malignancy is an uncommon form of cancer of the oral cavity consisting of about 0.5% of all malignancies of the upper aerodigestive tract.^[2] Although MEC accounts for a majority of minor salivary gland malignancies, MEC of the lip is a rare entity.^[3-5] In this study, we report a case of MEC on the internal aspect of the lower lip in a 60-year-old male patient, which presented as an asymptomatic mass, with a recent history of an increase in size, which was diagnosed on histopathological examination of the resected specimen.

CASE REPORT

A 60-year-old male patient presented to the Surgical Outpatient Department of our hospital with a globular,

slightly oval mass, on the internal aspect of his right lower lip. The swelling was present for the last three years and was gradually increasing in size. On physical examination, the size of the swelling was approximately 1.5 cm × 1 cm, with a soft-to-firm consistency, and an indurated border. There was no associated cervical lymphadenopathy.

As the swelling was progressively increasing in size, surgical excision of the swelling was planned. Preoperative routine blood examination and a chest x-ray were within normal limits. The mass was removed under general anesthesia and sent to the Pathology Department for histopathological examination.

On gross examination, the specimen was round-to-oval, measuring about 1.5 cm × 1 cm in size. The cut surface was pale yellow, with both the solid areas and multiple small cystic spaces being filled with mucin. The histopathological examination showed typical features of MEC, with intermediate and squamous cell islands and many mucin-filled cystic spaces of variable size, lined by mucous cells [Figure 1]. Immunohistochemical staining was done for MUC4, cytokeratin (CK) 7, 14, and 20. MUC4, CK 7, and 14 showed positivity, while CK 20 was negative. Considering the histological features and immunohistochemical staining pattern, a definitive diagnosis of low-grade MEC was made.

On the first postoperative check up, the wound was healthy. However, after this check up, on the request of the

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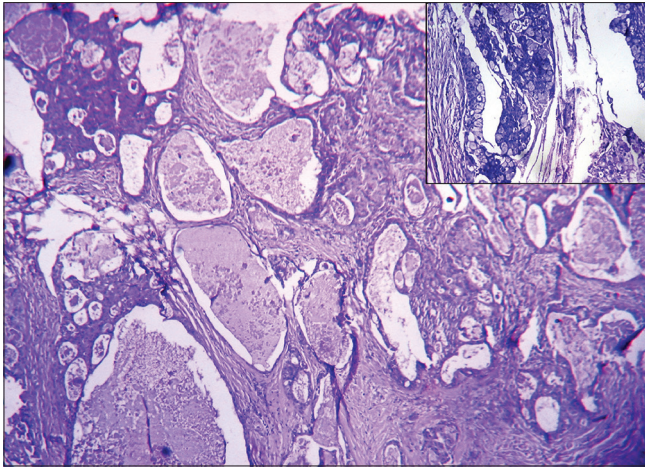


Figure 1: Histopathological section showing a tumor composed of multiple cystic areas lined by mucous cells and islands of squamous and intermediate cells. (H and E, $\times 200$); Inset: Cystic spaces lined by mucous cells with ample clear cytoplasm and a small, basally placed nucleus (H and E, $\times 400$)

patient, he was referred to Kolkata for further evaluation and follow up. Thus, we lost the patient to further follow up.

DISCUSSION

Most commonly, MEC arises in the head and neck region, and is mainly located in the parotids. Intraorally, the palate and buccal mucosa are the most common sites of MEC. The skin, upper respiratory tract, breast, pancreas, and thyroid are the rare sites where MEC can arise.^[4] MEC can also occur in the minor salivary glands of the lip, but this is very rare. As in our case, MEC had arisen in the inner aspect of the right lower lip, which is a rare entity.

Mucoepidermoid carcinoma represents about 5% of all salivary gland tumors, in which, about one-third occurs in the minor salivary gland. MEC is the third most frequently encountered minor salivary gland tumor, preceded only by a mixed tumor and adenoid cystic carcinoma.^[6] The affected minor salivary glands are most frequently located in the palate, followed by the lower lip.^[7]

Histologically, MEC is composed of various types of cells in varying proportions, like mucin-producing or mucous cells, squamous cells, intermediate cells, and clear cells. The squamous cells usually form solid nests, often with individual cell keratinization, which is more common in high-grade tumors. Mucin-producing cells or mucous cells can be dispersed or may form small clusters, partly lining the cystic spaces.^[6] Mucous cells are usually predominant in low-grade tumors. The intermediate cells have the characteristics of basal cells and squamous cells in terms of size and appearance.^[8] On occasions, MEC can be predominately composed of clear cells or oncocytic cells.^[6] MECs can be classified into low-grade, intermediate-grade,

and high-grade by histological examination using various parameters, namely, intracystic components, neural invasion, cellular anaplasia, mitosis, and necrosis.^[9] In low-grade MECs, well-formed glandular structures are usually present, which are lined by a single layer of mucous secreting columnar cells, which have intermediate cells and well-differentiated squamous cells, as in our case. Grossly, these low-grade tumors are well-circumscribed and cystic. High-grade tumors are usually poorly delineated, solid, and can be fixed to the surrounding tissues. These tumors show variation in cell size, increased mitosis, obvious invasion with perineural extension, and focal areas of necrosis.^[9] Immunohistochemically, MEC expresses various mucins like MUC1, MUC2, MUC4, MUC5AC, and so on.^[10] MUC1 and MUC4 are expressed in the apical membrane of glandular tumor cells and the entire membrane of intermediate, clear, and epidermoid tumor cells, while MUC2 and MUC5AC are expressed in the cytoplasm of glandular, mucous, and intermediate tumor cells. Handra-Luca *et al.*, found that expression of MUC4 is related to tumor differentiation and decreases in high-grade tumors, while expression of MUC1 indicates a worse prognosis.^[10] In the cytokeratin (CK) expression profile, various CKs like CK 7, 8, 13, 14, and 19 show positivity, especially in high-grade MECs, out of which CK14 is especially useful, to differentiate from squamous cell carcinoma.^[11] Again the negative expression of CK 20 is a useful finding in MECs, which aids in the diagnosis.^[12]

The biological behavior of the tumor depends on the histological grade, although the clinical stage is the best prognostic indicator.^[13] Metastasis and recurrence are rare in low-grade carcinomas. Wide surgical excision is usually required to ensure no recurrence. In a study, Spiro *et al.* have found that low-grade MEC can be treated by surgical resection alone, and patients have 92 and 90% survival rates after five and ten years of treatment, respectively.^[14] Hence, low-grade MEC does not appear to recur if treated by wide excision. However, it is unfortunate that we have lost the patient to follow up following surgery and cannot comment on the five-year survival.

As the lip is an uncommon site for MEC, a correct clinical diagnosis is not possible. The differential diagnosis of a lip mass is of a wide range, due to the diversity of tissues present in the lip. As the lip contains muscles, adipose tissues, blood vessels, nerves, and salivary glands, any tumor or malformation that arises from these, can occur in the lip.

CONCLUSION

Although malignant minor salivary gland tumors are rare in the lip, they must be kept in mind in the differential diagnosis. As the prognosis of low-grade MEC is excellent

after wide local resection, although rare, the possibility of MEC should be always be considered while dealing with any lip mass.

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