A Rare Case of Mucoepidermoid Carcinoma of Parotid with Mandibular Metastasis

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Abstract

Mucoepidermoid carcinoma is the most common malignant tumor of salivary gland. Majority of the salivary gland tumors arise in parotid gland with maximum being of benign nature. Although Mucoepidermoid carcinoma accounts for less than 10% of all tumors of the salivary gland, it constitutes approximately 30% of all malignant tumors of the salivary gland. It is characterized by a mixed pattern of the two main cell types: epidermoid and mucus-producing cells. Metastasis from salivary gland malignancy is infrequent and predominantly found in bone, lung, liver and lymph nodes. We are presenting a rare case of metastasis of mucoepidermoid carcinoma of parotid gland to the contralateral mandible.

Keywords: Mandible metastasis, mucoepidermoid carcinoma, parotid gland

Introduction

The majority of salivary gland tumors, i.e., about 80%, arise in the parotid glands. Approximately 80% of parotid gland tumors are benign.[1] Although mucoepidermoid carcinoma (MEC) accounts for <10% of all tumors of the salivary gland, it constitutes approximately 30% of all malignant tumors of the salivary gland. It is the most common malignant tumor of the parotid gland and the second-most common malignant tumor of the submandibular gland, after adenoid cystic carcinoma. Approximately 60-90% of these lesions occur in the parotid gland. Men and women are affected equally by this tumor, and the highest incidence occurs in the third–fifth decades of life.[1] In 1945, Stewart *et al*. introduced the term mucoepidermoid to define a distinct salivary gland tumor characterized by a mixed pattern of the following two main cell types: epidermoid and mucus producing cells.[2] It is classified as high grade, intermediate, and low grade, depending on the ratio of epidermal cells to mucous cells. The low-grade tumor has a higher ratio and is a less aggressive lesion. Although low-grade tumors have the ability for metastasis and local invasion, they behave more like benign tumors. The high-grade form is a more malignant tumor and has a poorer prognosis.[1] Metastasis from salivary gland malignancy is infrequent and predominantly found in bone, lung, liver, and lymph nodes.[3]

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In the present case report, we present a rare case of metastasis of MEC of parotid gland to the contralateral mandible.

Case Report

A 38-year-old male patient reported with a chief complaint of a swelling in the left upper part of the neck region for 1 year. The swelling extended from the parotid region to the posterior part of mandible measuring 3 cm × 3 cm at its greatest dimension, was round in shape, and had a smooth surface. Another swelling was present on the right side at the lower border of the mandible extending from the region up to the ear lobe and measuring 6 cm × 2 cm at its greatest dimension, was elliptical in shape, and had a smooth surface [Figure 1]. On palpation, swelling on the right side was found to be tender, but that on the left side was nontender, and the consistency of both was hard with well-defined edges and borders. The patient was a smoker and had been smoking 1 packet of beedi per day for the

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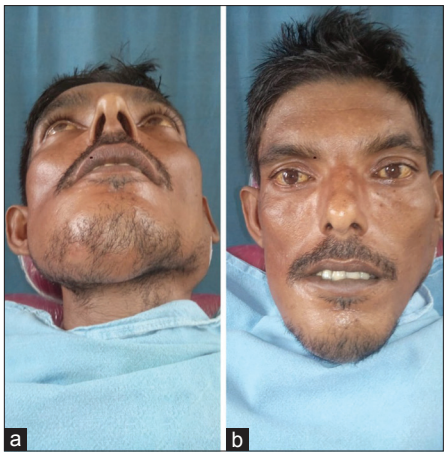

Figure 1: (a) Worm's-eye view, (b) Profile view

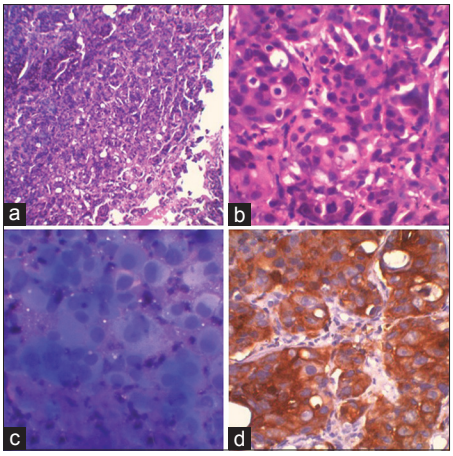

Figure 2: (a) Section showing diffuse infiltration by tumor nests, cords, and singly scattered neoplastic cells (H and E, ×100), (b) showing cells with high N:C ratio, nucleohypochromasia, deep eosinophilic to vacuolated cytoplasm. Few pseudoglandular spaces and mitotic figures seen. (c) Showing diffusely scattered and small cluster of neoplastic polyhedral cells having high N:C ratio and pale blue cytoplasm (MGG, ×400). (d) Strong membranous and cytoplasmic positivity with monoclonal carcinoembryonic antigen (IHC, ×400)

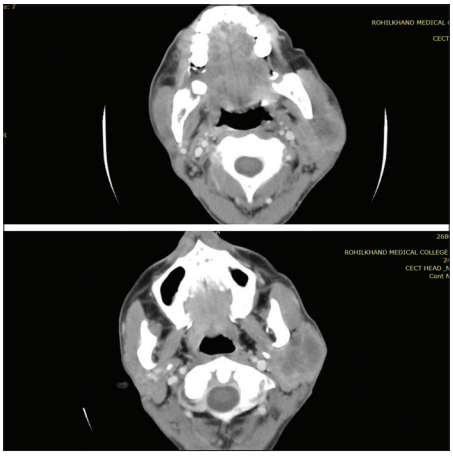

Figure 3: Contrast-enhanced computed tomography of head and neck at the level of parotid gland shows ill-defined hypodense mass lesion involving both superficial and deep lobes of the left parotid gland

past 3 years. Mouth opening was restricted to 10 mm. Bilateral lymphadenopathy of submandibular lymph nodes was present measuring 1 cm × 1.5 cm and was tender, hard, and mobile. On intraoral examination, an ulcer was found on the right gingivobuccal sulcus measuring 6 cm × 1 cm. The margins were well-defined, and edges were everted. It was hard in consistency, tender, and associated with bleeding on palpation. Provisional diagnosis of malignant neoplasm of the right buccal vestibule and left parotid region was made. Differential diagnosis of reactive lesion of the right buccal vestibule and independent benign tumor of the left parotid gland was also considered. Punch biopsy was taken from buccal vestibule and core needle biopsy for the parotid region. The histopathology of parotid showed the presence of infiltrating sheets, nests, and groups of neoplastic cells in subcutaneous connective tissue. Cells had high nucleus: cytoplasm ratio, pleomorphism, and hyperchromasia [Figure 2]. The report was suggestive of high-grade mucoepidermoid carcinoma (MEC) of the right buccal vestibule. Biopsy revealed extensive infiltration of tumor cells in the form of sheets and groups. Individual cells were polygonal, were moderately pleomorphic with hyperchromatic nuclei, and had a high N:C ratio. There was the presence of pseudoglandular spaces within tumor nests. Histopathological diagnosis made was of high-grade MEC.

Contrast-enhanced computed tomography (CECT) of head and neck revealed predominantly peripherally enhancing mass lesion measuring 4.8 cm × 3.2 cm, in the left parotid gland with extensive necrotic cervical adenopathy. A permeative lytic lesion was noted in the mandible, and the soft-tissue component was seen along the intraoral and gingivobuccal aspect of mandible, predominantly on the right side [Figure 3]. The CECT report was suggestive of malignant neoplasm of the left parotid gland metastatic cervical lymphadenopathy and mandibular metastasis.

Since on the basis of history, parotid lesion had appeared first, we made a diagnosis of a primary parotid lesion with mandibular metastasis.

His tumor node metastasis classification was $cT_3N_2M_1$ and was the American Joint Committee on Cancer Stage IVC. After receiving informed consent, the patient underwent chemoradiotherapy, with a total radiation dose of 50-Gy. Chemotherapy was administered in two cycles with a 4-week interval, with a regimen of cisplatin 80 mg/m^2 (day 1) and 5-fluorouracil 800 mg/m^2 per day (days 1–5). The patient was not ready for surgical intervention. The patient was recalled after 3 months of follow-up.

Discussion

Kokemueller *et al*. showed that prognosis of patients with MEC is significantly influenced by grade, stage, and margin status.[4] Minor salivary glands are more likely to have higher grade MECs and more advanced stage tumors, and a minor salivary gland site itself may be associated with worse survival. MECs of the parotid gland characteristically present as a

painless mass, 2–3 cm in diameter at the initial discovery. With high-grade lesions, pain and rapid growth can be prominent. A high rate of metastasis is observed in MEC patients with high histologic grade.[5] Ozawa *et al*.[6] did a clinical analysis of 43 patients and observed regional lymph node metastasis in 24% of the patients with low histologic grade, 30% in those with intermediate grade, and 56% in those with high grade. Lung and breast carcinomas account for the majority that metastasize to oral cavity.[7] It is widely accepted that the jaws do not contain a lymphatic system, and it is believed that metastases there occur through the bloodstream, a fact that is supported by the occurrence of metastatic foci in areas where spongiosa and slowing of the circulation favor the entrapment of metastatic emboli. Metastasis to oral cavity is unusual and constitutes 1% of all oral malignant tumors.[8] The jaw bones are more commonly affected than the oral mucosa, in a ratio of 2.5:1. Some intraoral malignancies, especially from salivary glands, have histologic features similar to that of tumors in distant organs.[9] The initial treatment of MEC, regardless of grade, is essentially based on surgical resection and eventually on adjuvant radiotherapy. Due to the rarity of the disease, current literature is scarce and often reflects data from small and heterogeneous series. Thus, no guidelines are available to support the clinician's decision, and the management of metastatic MEC remains challenging. Local recurrences not amenable to further locoregional treatments and metastatic disease are treated with systemic chemotherapy. Single-agent or combination chemotherapy with cisplatin, fluorouracil, and/or paclitaxel has demonstrated activity in published series, but overall response rates are unsatisfactory and of short duration.[10] Overexpression of the human epidermal growth factor receptor (HER) family of oncoproteins, HER1/epidermal growth factor receptor (EGFR) and HER2, has been described in approximately 70% of salivary gland carcinomas including MEC and adenoid cystic carcinoma,[11] but few studies have evaluated the therapeutic relevance of an anti-EGFR/HER2 strategy in these neoplasms.

Conclusion

Mucoepidermoid tumor rarely presents with metastasis. Mandibular metastasis has not been reported in literature. We want to highlight this case so that clinical suspicion can be made if such situation arises.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand 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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Conflicts of interest

There are no conflicts of interest.

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