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Mucoepidermoid Carcinoma – A Case Report and Review of Literature.

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Abstract

Mucoepidermoid carcinoma of jaws is a rare condition and is usually associated with salivary glands. It comprises of 5-10% of all the salivary gland tumors which shows considerable histologic variability and unusual clinical course. Mucoepidermoid carcinoma of jaws is more commonly seen in mandible than maxilla. Any lesion to be described as PIOC, should have no histological origin similarity to oral mucosa, antral or nasal mucosa (for intramaxillary lesions), overlying skin. These lesions can also be termed as odontogenic carcinoma as they are considered to arise from epithelium involved in odontogenesis. It is important to clinically detect malignant signs and histopathological features of PIOC of mandible to confirm diagnosis. Surgery remains the mainstay for treatment, although studies suggests that relapses are more with conservative procedures. But with segmental resection with or without adjuvant treatment has less relapse rate.

Keywords- *PIOC, mucoepidermoid carcinoma, primary intraosseous carcinoma of mandible.*

Introduction

Mucoepidermoid carcinoma of jaws is a rare lesion and is usually associated with salivary glands.**[**1,2**]** It comprises of 5-10% of all the salivary gland tumors which shows considerable histologic variability and an unusual clinical course.**[**2**]** Due to its unpredictable clinical course and histological variations, it forms a distinct pathological entity. Mucoepidermoid carcinoma of jaws is more commonly seen in mandible than maxilla. Evans in his case series of 69 cases have found that occurrence of mucoepidermoid carcinoma is more in parotid gland (44cases) as compared to that of submandibular gland (2cases) rest 23 were present in minor salivary glands.**[**3**]** Eversole also reviewed 815 cases and found same results. 89.6% involved the parotid, 8.4% submandibular and 0.4% sublingual gland.**[**4**]** Salivary gland neoplasms occurring in within jaws are extremely rare finding. The first reported intraosseous mucoepidermoid carcinoma of mandible was reported by Lepp in 1939 in a 66-year-old female.**[**5**]** Pathogenesis of tumour is not clear, many studies have explained the possible reasons of mucoepidermoid carcinoma. Bhaskar **[**6**]** in his study in 1963 discussed the histological composition and the feasible causes for tumour pathogenesis. Waldron and Mustoe **[**7**]** suggested that intraosseous mucoepidermoid carcinoma be included in primary intraosseous carcinoma of jaw as type 4 (Table 1). A review of the English literature

revealed about 100 reported cases of mucoepidermoid carcinoma arising in mandible.**[**8**]** Here we report another case of intraosseous mucoepidermoid carcinoma within the bony mandible.

Case Presentation

A 60-year-old male patient reported to the OPD with chief complaint of swelling in the lower left jaw region for 2 months. The swelling was associated with pain especially while eating. The patient took over the counter medication from local medical store for pain relief for initial few days.

On initial presentation, there was no gross extraoral swelling. Intraoral examination revealed a swelling measuring nearly 5cm x 3cm in the left lower jaw region extending from lateral incisor till retromolar region. Overlying mucosa was ulcerated and there was expansion of buccal and lingual cortical plates. Overlying teeth were also missing (history of spontaneous fall a month back). Clinical examination of neck revealed palpable node at left level 1B, which was firm, mobile and non-tender. No medical comorbidities were reported. MRI scan (skull base to clavicle) was advised for detailed evaluation and a biopsy of lesion was done.

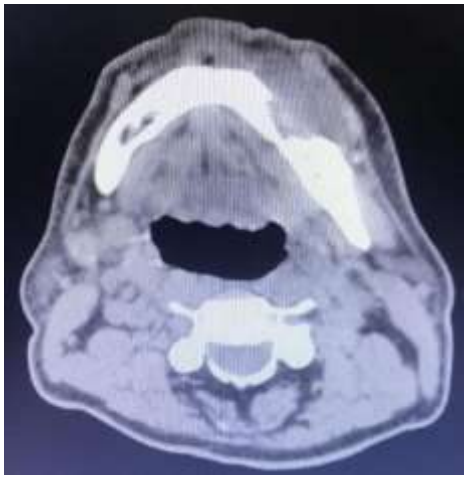

Fig. 1- axial scan showing the site of tumor

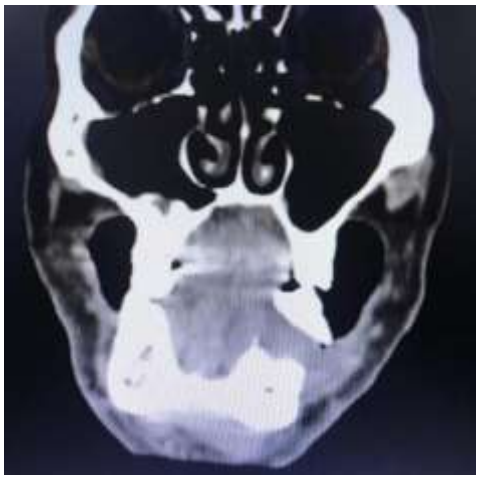

Fig. 2- coronal section

MRI scan showed ulcero-infiltrative heterogeneously enhancing lesion seen involving left lower bucco-alveolar sulcus from central incisor upto the lower 3rd molar teeth, involving lower gingivobuccal sulcus. (figure 1 & 2) There is associated erosion and marrow infiltration with exposed superior wall of inferior alveolar nerve canal from lower lateral incisor teeth up to the angle of mandible. The lesion also extends

to involve lingual mucosa of lower alveolus abuts myelohyoid muscle and sublingual gland. Hetergenously, enhancing left level 1B node is seen measuring 10 X 7 mm.

Biopsy was suggestive of Well Differentiated Squamous Cell Carcinoma.

Type 1: PIOC exodontogenic cyst Type 2A: Malignant ameloblastoma
Type 2B: Ameloblastic carcinoma arising de novo, exameloblastoma or exodontogenic cyst
Type 3: PIOC arising de novo a) Keratinising type b) Nonkeratinising type
Type 4: Intraosseous mucoepidermoid carcinoma.

Reprinted from International Journal of Oral and Maxillofacial Surgery, Volume No.30, Thomas G et al, Primary intraosseous carcinoma of the jaw. Report of two new cases and pooled analysis of world literature, 349–55, 2001, by permission of the publisher Churchill Livingston.

Table 1: Classification of Primary Intraosseous Carcinoma

No history of any previous surgical or medical intervention was reported. Final diagnosis of squamous cell carcinoma was made. The patient & his relatives were explained about the nature of disease and its treatment. With due consent from patient & his relatives, surgery was planned.

As a planned procedure, patient underwent COMPOSITE RESECTION wide excision of buccal mucosa and lingual soft tissue with segmental mandibulectomy extending from 43 to ipsilateral subsigmoid region, and selective neck dissection (level 1 to 4) was performed. Primary specimen & bony marrow from cut ends was sent for frozen section. All the margins & base and bony marrow were reported negative for malignancy. Microvascular reconstruction of the defect was done with free fibula osteocutaneous flap. Post-operative hospital stay of the patient was uneventful. The patient was discharged on 6th post-operative day.

Final histological report suggests a neoplasm composed predominantly of cycstic spaces and an epidermoid component in a fibrous stroma. mucin stain shows pink colored intra cytoplasmic mucin in tumor cell nests. H&E staining reveals the solid nests, composed of intricately mixed squamous and mucinous cells and separated by scanty stroma. Although it was N0 but high grade mucoepidermoid carcinoma. (Figure 3)

As per Brookstone & Huvos classification, the tumor confirms to stage 3 with cortical perforation. In view of the bone involvement the final designation of the tumor is revised to Central Mucoepidermoid carcinoma of mandible.10 This classification is now used to determine the prognosis.11 (Table 2)

Stages	Description	Prognosis
1.	The lesions with an intact cortex layer and without any bony expansions	Good
2.	Lesions that expand the bone but do not disturb the integrity of the cortex	Fair
3.	Lesions that disrupt the integrity of the periosteum or cause cortical perforation with or without nodalinvolvement and masses with nodal involvement	Poor

Table 2: Brookstone & Huvos classification.

Pertaining to the high-grade variant of mucoepidermoid carcinoma, patient was referred to radiation oncologist for opinion of adjuvant radiotherapy. Patient was reviewed after 1month post operatively. The surgical site was healed and healthy.

On arriving at a final diagnosis of mucoepidermoid carcinoma of jaw, we reviewed literature for the same. It revealed that there are very few cases reported for mucoepidermoid carcinoma of the jaws.

Figure 3 a. mucin stain shows pink colored intra cytoplasmic mucin in tumor cell nests.

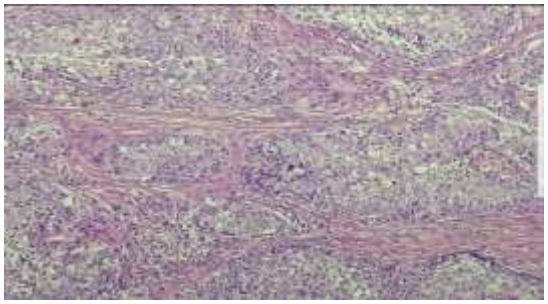

Figure 3 b H&E staining reveals the solid nests, composed of intricately mixed squamous and mucinous cells and separated by scanty stroma.

Discussion

Mucoepidermoid carcinomas occurring within the jaws as primary bony lesions are termed as central mucoepidermoid carcinomas and are extremely rare, comprising 2–3% of all mucoepidermoid carcinomas.[1,2,8,9]

Mucoepidermoid carcinoma has a female predilection. It affects females twice more than males, but the case presented here is a 60-year-old male.

In children it occurs more in mandible than maxilla. According to the literature, most common site is premolar-angle region, this is in accordance with the presented case.

No age specification is reported. It has been reported in pediatric as well geriatric patients but seen more in 4th and 5th decade, which is in accordance with the presented case.

The origin of these lesions in the jaws is thought to be due to neoplastic transformation of the sinus epithelium; entrapped retromolar mucous glands and developmental embryonic remnants of the submandibular gland within the mandible or neoplastic transformation of the mucous- secreting cells commonly found in the pluripotential epithelial lining of dentigerous cysts associated with impacted third molars.[1,8] It is also reported that 83% of cases of CMEC occurred in the molar region which is the most probable location of developing dentigerous and odontogenic keratocysts. Accompanying cysts and impacted teeth were reported in 40% of the cases.[8]

Diagnosing CMEC is difficult. Many lesions are listed as differential diagnosis. Based on the clinicopathological reports, the diagnosis is made. Imaging plays a pivotal role in the detection and differentiation of MEC because of peripheral sclerosis and mixed internal structure, consisting of a unilocular and/or multilocular pattern. Its radiologic pattern is similar as the imaging characteristics to other lesions, such as ameloblastoma, glandular odontogenic cyst, and keratocystic odontogenic tumour (Figure 3).

OPG and CT are routinely prescribed for evaluating the maxillofacial area.[13] It is described as a radiolucent image with well-defined scleral periphery and numerous small loculations. The presence of tooth dislocation and root resorption are common findings. Its aggressive behavior is revealed by cortical bone perforation and extension to surrounding soft tissues. Studies suggest that fine needle aspiration (FNA) is considered to be effective for high-grade or intermediate-grade as compared to that of low-grade lesion.[14]

Although surgical treatment remains the mainstay for treating CMEC, but few studies suggests that relapses are more if conservative procedures like marsupialisation, enucleation, marginal resection etc. are undertaken. But with segmental resection with or without adjuvant treatment has very less relapse rate.**[**11**]**

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