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Intraosseous Mucoepidermoid Carcinoma Of Mandible: A Case Report And Review Of Literature

Case Report

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Introduction

Mucoepidermoid carcinoma is usually associated with salivary glands and comprises 5–10% of all salivary gland tumours [1, 3]. In 1945, Stewart and associates described its mucous-secreting and epidermal cellular elements thus establishing it as a distinct pathologic entity [4]. Eversole reviewed 815 cases and found that of the major salivary gland tumours, 89.6% involved the parotid, 8.4% submandibular and 0.4% sublingual gland [1]. The palate was the most common site for minor salivary gland involvement, accounting for 41.1% of intraoral lesions [1].

Aberrant salivary gland neoplasms arising within the jaws as primary central bony lesions are extremely rare comprising 2–3% of all mucoepidermoid carcinomas reported [2]. Lepp in 1939 first reported an intraosseousmucoepidermoid carcinoma of the mandible in a 66-year-old woman [5] and Bhaskar [6] in 1963 reported two cases discussing the criteria for their origin, histological composition and possible explanations for tumour pathogenesis. In 1991, after a systematic review of its histology and degree of differentiation the WHO classification recommended that the term "mucoepidermoidtumour" be changed to "mucoepidermoid carcinoma"[7]. Waldron and Mustoe [8] suggested that intraosseousmucoepidermoid carcinoma be included in primary intraosseous carcinoma of jaw as type 4.A very rare location of MEC is the jaw bones. The mandible is affected more commonly than the maxilla, and the posterior part of the mandible is the most common location of intraosseous MECs.

In this report, we present three patients with the diagnosis of intraosseous MEC treated with surgery and a literature review. Informed written consent was obtained from all the patients.

Case Report

A 53 year old female patient reported to our department with swelling in the left lower jaw since 5 months.The patient noticed the swelling 5 months back which was smaller in size. The swelling gradually grew to the present size. The patient does not give any relevant family history. There is no relevant medical history and is not under any medication. On extra oral examination swelling was noted on the left side of the mandible of size 5x5 cm .The swelling was hard in consistency fixed,non compressible non febrile. Intraorally swelling was seen in 36 to 38 region. There was no tenderness on palpation.The patient had no complaints of paresthesia. An incisional biopsy was done and an OPG was taken. The OPG revealed a well defineunilocular radiolucency in the left angle region of the mandible. The incisional biopsy was reported as Central low grade mucoepidermoid carcinoma. Segmental Mandibulectomy + Supraomohyoid neck dissection and Primary closure was planned. The Surgery went according to plan.The specimen was sent for Histopathologicalevaluation. The detailed report was suggestive of low grade mucoepidermoid carcinoma of mandible with no presence of metastatic tumor in the lymph nodes.The patient is being followed up and is disease free for a year.

Discussion

The mandible, particularly the posterior part, is the most common location of intraosseous MECs. Women are more commonly affected than men, and although the most common age group for this tumor is the fourth and fifth decades, there were patients be-

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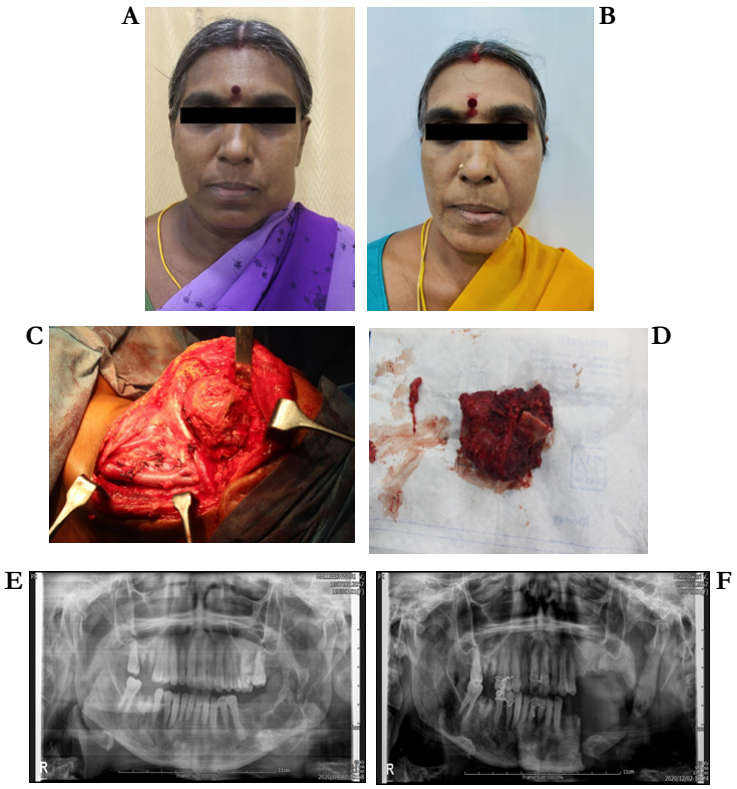
Figure 1. IMAGES:(a)PRE OP image of the patient(b)Post Op image of the patient (c) &(d)Intra OP images(e)Pre op OPG(f) Post OP OPG.



tween the first and seventh decades reported in literature (Maremonti et al. 2001; Gnepp 2009; Gingell et al. 1984).

The criteria that acknowledges the diagnosis of intraosseous MEC is entailed as: (a) intact cortical plates while the cortical perforation does not exclude the diagnosis, (b) radiological evidence of bone destruction, (c) exclusion of an alternative primary tumor, which its metastasis could histologically resemble the central MEC, (d) exclusion of an odontogenic tumor and (e) histopathologic confirmation (Maremonti et al. 2001; Gnepp 2009)

The radiological features are diverse and presumably non-diagnostic, usually presented as a unilocular or multilocular radiolucency. Even though it is frequently scalloped; the margins of the lesion are often well defined(Waldron and Koh 1990). To the best of our knowledge, only a few cases of mixed radiolucent-radiopaque lesions have been reported in the literature(Sherin et al. 2011; Shimizu et al. 2004)(Sherin et al. 2011). Chan et al.(Chan et al. 2013) stated that the common radiological features of these tumors would be the presence of a well-defined sclerotic boundary, internal amorphous sclerotic bone and many small loculations. Moreover, bordering septa in many of these loculations is absent and the outer cortical plate is expanded and perforated with extending into the surrounding soft tissue. Tooth displacement and root resorption is also present. All reported cases of their study exhibited some common diagnostic imaging aspects with other multilocular-appearing lesions of the jaws. Though, the presence of amorphous sclerotic bone and malignant features can be advantageous in the differential diagnosis (Chan et al. 2013)

The presence of calcifications was reported in the clear-cell variant and the conventional MEC, occurring in enduring neoplasms. The dystrophic calcification of the amorphous-eosinophilic material secreted by intermediate basal cells may perhaps produce these features (Sherin et al. 2011)(Gnepp 2009). Eversole et al. (L. R. Eversole, Sabes, and Rovin 1975) found that 50% of mandibular central MEC were associated with dental cysts or impacted teeth; whereas in the study of Brookstone and Huvos(Lucas et al. 1998) and in the research of He et al (He et al. 2012)., no significant relationship was found between central MEC and odontogenic cysts or impacted teeth. Likewise, no relationship was detected between impacted tooth and central MEC in our study. Microscopic examination of MEC revealed a neoplasm composed of nests and islands of epidermoid, mucous, and intermediate cells embracing cystic spaces with various sizes in a fibrous connective tissue (Simon et al. 2003). A considerable number of central MEC have been reported to be mainly low-grade cystic lesions +-. The presenting two cases were also low grade.

Histopathologically, GOC may be confused with intraosseous MEC (Lewis R. Eversole 2001)-(Sciubba, Fantasia, and Kahn 2001)]. This cyst is lined by stratified squamous epithelium with variable thickness and surface cuboidal or columnar ciliated cells. Small microcysts and clusters of mucous cells are also depicted (Woo 2016). Islands, resembling intraosseous MEC, were noted in the GOC wall; which may possibly cause diagnostic drawback (Sciubba, Fantasia, and Kahn 2001)Therefore, molecular assays, explicitly targeting the MEC-like islands in the GOC fibrous wall, may figure out whether these islands signify true malignant transformation or not [6, 8, 10]. In the current study, the second case was primarily diagnosed as GOC. Inevitably, the incorrect diagnosis proceeds to patient complications and overdue treatment. This is imperative since different treatments are demanded for patients with a GOC compared to low- grade MEC. Supplementary implements and methods such as immunohistochemistry and gene abnormality assessments might be supportive in yielding a conclusive differential diagnosis (Woo 2016). One of these markers is Maspin which is expressed in MEC and would be expedient in

the differential diagnosis of MEC from GOC, particularly in ambiguous cases and in small incisional biopsy samples .(Woo 2016) Piresetal.stated that the origin of central MEC is still controversial. GOC is a newly defined entity whose association with low-grade central MEC has been described in the literature. Moreover, the study of Pires et al. was aimed to evaluate the cytokeratin (CK) profile of central MEC and GOC, matching the outcomes with the expression of CK in MEC of salivary glands and odontogenic cysts and tumors. They concluded that all central MECs expressed CKs 5, 7, 8, 14, and 18 and all GOCs expressed CKs 5, 7, 8, 13, 14, and 19

They compared CK expression in GOC and central MEC and noticed dissimilarities in CKs 18 (30% versus 100%) and 19 (100% versus 50%). Central MEC and GOC are perhaps distinct lesions with CK profiles comparable to lesions that have glandular and odontogenic origins, respectively. Moreover, expression of CKs 18 and 19 could be beneficial in their subsequent differential diagnosis .

TORC1/MAML2 and MECT1:MAML2 gene fusion in intraosseous MEC was reported by studies of Khan et al. (Khan et al. 2010)and Fowler et al. (Pindborg, Kramer, and Torloni 1971; Lewis R. Eversole 2001) and it has been confirmed that they can be employed as a diagnostic marker (Khan et al. 2010) -

The foremost treatment for intraosseous MEC is wide local resection, enblock resection or hemi-mandibulectomy(He et al. 2012), . Selective or therapeutic neck dissection has been introduced in the instances of cervical lymphatic metastasis .

Radiotherapy seems to be a useful supplementary aid in cases represented with close surgical margins and high grade tumors (Weber et al. 2007). Microscopic grading appears to have a strong influence on survival rate; so that a low-grade tumor without perineural invasion and with tumor-free margins designates a better prognosis (House Ear Institute 1982).

In He et al.'s study (He et al. 2012), all patients presented low-grade tumors without any evidence of nodal metastasis. The current case similarly did not demonstrate any cervical lymphadenopathy. Brookstone and Huvos have proposed a staging class for intraosseous MEC. Lesions with intact cortical plates with no evidence of bone expansion are categorized as stage I; neoplasms with intact plates, but intrabony expansion are branded as stage II and finally, lesions associated with cortical perforation or nodal disease are classified as staged III. According to these categories, in the current study, the case 1 can be sorted in stage III and case2 can be fitted in stage II.

The recurrence rate of this entity varies from 13-50% in different studies . Metastases are reported in 9% of central MEC primarily to the regional lymph nodes and infrequently to the ipsilateral clavicle, lung and brain, hence, long term follow up is recommended.

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