

# Central mucoepidermoid carcinoma of the mandible: a rare case report with diagnostic dilemma and review of the literature

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## Introduction

Primary central mucoepidermoid carcinoma of jaws is a rare lesion comprising 2–3% of all mucoepidermoid carcinomas reported in the literature. It affects the mandible as unilocular or multilocular radiographic lesions.

## Case report

The case presented here is that of a 55-year-old woman with expansile osteolytic lesion of the mandible who was managed with en-bloc resection considering ameloblastoma as preoperative diagnosis.

## Conclusion

Mucoepidermoid carcinoma of the jaw is a rare tumour. The speculation on its aetiopathogenesis is mainly centered on the pluripotential capabilities of the epithelial lining of odontogenic cysts.

## Keywords:

intraosseous mucoepidermoid carcinoma, mandible, odontogenic epithelium

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## Introduction

Mucoepidermoid carcinoma (MEC) is a commonly occurring malignant salivary gland neoplasm, comprising 2.8–15% of all salivary gland tumours. Aberrant salivary gland neoplasms arising in jaws as primary central bony lesions are extremely rare, comprising 2–4.3% of all MECs reported [1]. A thorough review of the English literature yielded about 100 reported cases of MEC arising in the mandible [2,3].

## Case report

A 55-year-old hypertensive woman attended the OPD with chief complaints of pain and swelling on the left lower jaw of 4 months' duration associated with intraoral swelling (Fig. 1). There was no associated trismus or loose tooth. Past history of extraction of lt. lower last two molars 10 years back was noted. On examination, the swelling was diffuse and bony-hard, involving the ramus and along the superior aspect of the angle of the mandible with expansion of the buccal and lingual cortices with crackling and bluish discoloration of the overlying mucosa (Fig. 2) and no cervical lymphadenopathy. CT scan revealed a unilocular osteolytic expansile lesion involving ramus, coronoid process and sigmoid notch (Fig. 3). Fine needle aspiration cytology (FNAC) through an intraoral route was suspicious of ameloblastoma. Marginal mandibulectomy was performed. Histopathological examination revealed intraosseous MEC (Fig. 4). Postoperatively, the patient

received 40 Gy of radiation in 20 fractions and doing well until 4 months of follow-up.

## Discussion

The first case of central mucoepidermoid carcinomas (CMEC) was reported in the mandible of a 66-year-old woman, by Lepp, in 1939. Waldron and Mustoe [4] suggested that intraosseous MEC be included in the primary intraosseous carcinomas of the jaws (PIOC) as type 4. PIOC was classified as follows [4]:

- (1) Type 1, PIOC ex-odontogenic cyst.
- (2) Type 2a, malignant ameloblastoma.
- (3) Type 2b, ameloblastic carcinoma arising *de novo*, ex-ameloblastoma or ex-odontogenic cyst.
- (4) Type 3, PIOC arising *de novo*:
  - (a) Keratinizing type and
  - (b) Nonkeratinizing type.
- (5) Type 4, intraosseous MEC.

Its origin remains controversial and possibilities considered are as follows:

- (a) Metaplasia of the odontogenic cyst epithelium;

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Figure 1



Patient with left jaw swelling.

Figure 2



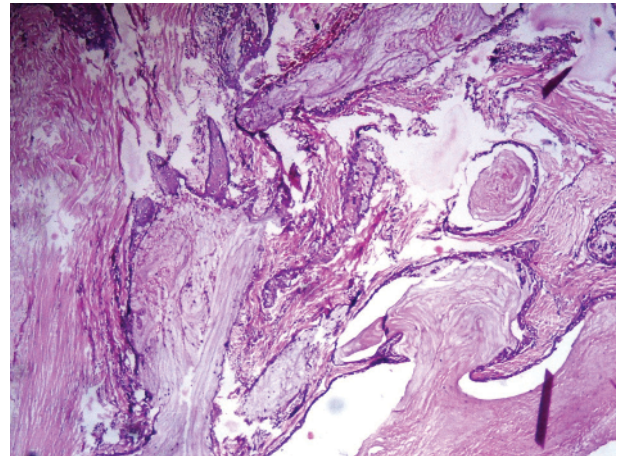
Excised specimen with cortical expansion and bluish discoloration of the mucosa.

Figure 3



Computed tomography (CT) scan showing expansile osteolytic lesion at ramus and coronoid process.

Figure 4



HPE× slide showing nests of neoplastic squamous cells along with mucous secreting cells.

- (b) Entrapment of salivary tissues from the submandibular, sublingual or minor salivary glands and from the retromolar area during development
- (c) Maxillary sinus epithelium,
- (d) Iatrogenic entrapment of minor salivary glands (chronic osteomyelitis, sinusitis),
- (e) Odontogenic remnants of dental lamina [5].

CMEC affects women twice more than it does men, with the majority occurring in fourth and fifth decades of life, and involves the mandible (the molar angle region) twice more often than the maxilla. In our case, the patient is female with involvement of the ramus and the molar angle region.

The main symptoms are swelling and pain, with trismus, paraesthesia and tooth mobility being noted occasionally [2]. Previous history of a cyst or impacted tooth indicate towards

neoplastic transformation to MEC [5]. The periphery of the lesion is mostly well-defined, corticated and often crenated or undulating in nature, which is similar to a benign odontogenic lesion [6]. The multilocular lesion resembles a soap bubble or honeycomb.

The clinical and radiographic differential diagnosis should include an odontogenic keratocyst, ameloblastoma, dentigerous cyst, ameloblastic fibroma and metastatic lesions to the jaw [7].

Criteria for diagnosing CMECs include the following:

- (a) Presence of a radiographic, distinct, osteolytic lesion;
- (b) Positive mucicarmine staining;
- (c) Absence of rupture of one or more cortical plates.
- (d) Clinical and histological exclusion of a metastasis or an odontogenic lesion;

- (e) Exclusion of the origin from a soft tissue salivary gland; and
- (f) Histological confirmation [6].

Brookstone and Huvos [2] had put forward a staging system based on condition of the overlying bone.

- (1) Stage I disease: lesions with intact cortical plates with no bony expansion;
- (2) Stage II disease: surrounded by intact cortical bone with some degree of expansion;
- (3) Stage III disease: cortical perforation, breakdown of the overlying periosteum or nodal spread.

The case presented here showed expanded, eroded buccal and lingual cortices and could be imputed as stage III disease.

The primary treatment modality for patients with central intraosseous MEC is surgery. As a rule, even as low-grade tumours, MEC should be managed with wide local resection, en-bloc resection or hemimandibulectomy [2,8] or partial/total maxillectomy. Neck dissection is performed in cases where metastasis to the cervical nodes is suspected [8]. Adjuvant radiotherapy is recommended for high-grade tumours [9].

### Conclusion

CMEC in the jaws is a rare entity. The posterior part of the mandible is the common site. Surgery such as en-bloc resection is the treatment of choice with adjuvant radiotherapy in high-grade cases.

### 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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Nil.

### Conflicts of interest

There are no conflicts of interest.

### References

- 1 Bouquot JE, Gnepp DR, Dardick I, Hietanen JHP. Intraosseous salivary tissue: jawbone examples of choristomas, hamartomas, embryonic rests, and inflammatory entrapment: another histogenetic source for intraosseous adenocarcinoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000; 90:205–217.
- 2 Brookstone MS, Huvos AG. Central salivary gland tumors of the maxilla and mandible: a clinicopathologic study of 11 cases with an analysis of the literature. *J Oral Maxillofac Surg* 1992; 50:229–236.
- 3 Maremonti P, Califano L, Mangone GM, Zupi A, Guida C. Intraosseous mucoepidermoid carcinoma. Report of a long-term evolution case. *Oral Oncol* 2001; 37:110–113.
- 4 Waldron CA, Mustoe TA. Primary intraosseous carcinoma of the mandible with probable origin in an odontogenic cyst. *Oral Surg Oral Med Oral Pathol* 1989; 67:716–724.
- 5 Eversole LR, Sabes WR, Rovin S. Aggressive growth and neoplastic potential of odontogenic cysts with special reference to central mucoepidermoid and mucoepidermoid carcinomas. *Cancer* 1975; 35:270–282.
- 6 Simon D, Somanathan T, Ramdas K, Pandey M. Central mucoepidermoid carcinoma of mandible: a case report and review of literature. *World J Surg Oncol* 2003; 1:1.
- 7 Darling MR, Wehrli BM, Ciavarrro C, Daley TD. Pericoronary radiolucency in posterior mandible. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008; 105:139–431.
- 8 Caccamese JF Jr, Ord RA. Pediatric mucoepidermoid carcinoma of the palate. *Int J Oral Maxillofac Surg* 2002; 31:136–139.
- 9 Freje JE, Campbell BH, Yousif WJ, Clowry LJ. Central mucoepidermoid carcinoma of the mandible. *Otolaryngol Head Neck Surg* 1995; 112:453–456.