CASE REPORT

Central mucoepidermoid carcinoma of maxilla with radiographic appearance of mixed radiopaque–radiolucent lesion: a case report

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Mucoepidermoid carcinoma is the most common primary malignancy of the salivary glands. Central mucoepidermoid carcinomas are extremely rare in the jaws. Here we report a case of a 19-year-old central mucoepidermoid carcinoma in a young female patient which was asymptomatic throughout her history and which radiographically simulated a fibro-osseous lesion; both features misled the clinical diagnosis. A radiographic picture of mixed radiopaque–radiolucent appearance in central mucoepidermoid carcinoma is extremely rare. The long duration is also uncommon for a malignant lesion. This case report adds a new dimension to the clinical and radiographic picture a central mucoepidermoid carcinoma can reveal. *Dentomaxillofacial Radiology* (2011) **40**, 463–465. doi: 10.1259/dmfr/82151785

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Introduction

A 29-year-old female patient came to the Oral Medicine and Radiology Department for extraction of asymptomatic and grossly decayed molars. On intraoral examination, there were root stumps of the upper right second and third molars (17, 18) and a large swelling palatal to these teeth which was extending to the midline. The swelling was about 5 × 5 cm in size and normal in colour, except in the midline where it was slightly bluish in a single area. The swelling was bony hard in consistency, except in the midline where it was firm in consistency. In the detailed history, the patient had had this swelling since the age of 19 and had been asymptomatic since then. She had a history of previous dental consultation at the age of 10 when the swelling was first detected and was informed of the option of surgical removal if it increased in size. Since there was no increase in swelling (according to the patient) or any symptoms, she did not report for further treatment. There was no previous history of trauma, toothache, paraesthesia, mobility or any other findings. A panoramic radiograph revealed a radiopaque–radiolucent lesion at the apical region of the molars overlapping the right maxillary sinus (Figure 1). The maxillary occlusal view showed minute specks of calcification in the region adjacent to molars (Figure 2). It

was approximately 6 × 6 cm in size with ill-defined borders and faint radiopacities, and septae in the internal aspect of the lesion. Vitality tests of the teeth in the quadrant, except the root stumps, showed that all teeth were vital. Axial plain and contrast CT examinations showed a moderately enhancing soft-tissue density lesion extending medially into the medial wall and floor of the maxillary sinus, posteriorly destroying the nasopharynx and medial pterygoid plate; superiorly extending up to the middle turbinate; and inferiorly destroying the hard palate and the alveolar process (Figure 3). The CT report was suggestive of fibrous dysplasia. Based on the history, clinical and radiographic features, differential diagnosis included fibro-osseous lesion, benign salivary gland neoplasm and vascular lesions.

An incisional biopsy was taken from the palate adjacent to the molars after removal of bone adjacent to the molars. The histopathology proved to be an intermediate-grade mucoepidermoid carcinoma (Figure 4). The histopathological picture did not provide any evidence of calcifications since the biopsy was done superficially. The patient was referred to a regional cancer centre for further treatment where radical excision of the lesion with adjuvant radiotherapy was carried out. Microscopic examination showed diffuse dystrophic calcifications in the lesion and confirmed the radiographic picture. The post-operative period was uneventful and the patient is under strict follow-up.

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Figure 1 Panoramic radiograph

Discussion

Mucoepidermoid carcinomas arising within the jaws as primary central bony lesions are termed central mucoepidermoid carcinomas and are extremely rare, comprising 2–3% of all mucoepidermoid carcinomas.^{1–4}

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.^{1,2} The origin of the lesion in our case could be from ectopic salivary gland tissue of

the maxilla or from the sinus epithelial lining since the lesion was grossly involving the maxillary sinus with minimal involvement of the palate, which was confirmed on CT imaging. The possibility of origin from sinus epithelial lining cannot be entirely ruled out.

The inclusion of the present case as a central lesion satisfies the criteria by Simon *et al*.¹ The findings that satisfied the criteria for central lesions were radiographic evidence of bone destruction; exclusion of another primary tumour; exclusion of an odontogenic tumour; histopathological confirmation; and detectable

Figure 2 Maxillary occlusal view

Figure 3 CT image of the lesion

Figure 4 Histopathology showing mucous secreting cells and intermediate cells

intracellular mucins.^{1,4,5} Some authors have mentioned the exclusion of cases with extension into the alveolus from central lesions.^{3,4} In our case, considering the large size and longstanding nature, it could be possible that the lesion would eventually extend to the alveolus.

According to a staging system proposed by Brookstone *et al*⁵ based on the condition of the overlying bone, our case was stage III as the lesion demonstrated cortical perforation in the middle of the palate.^{4,5}

Central mucoepidermoid carcinoma is a rare lesion and so the treatment protocol is still not clear. Various authors have discussed the treatment modalities and

have confirmed that radical resection is the treatment of choice.^{1,2,4,6} The lesion in this case was extensive and was treated with radical resection.

According to the literature, radiographic pictures of the lesions have always been of a multilocular or cystic-like radiolucency, except in a single case reported recently where the lesion appeared with radio-opaque masses in the radiograph and bone in histopathology.^{2,7} In our case, histopathologically, the calcifications appeared as dystrophic. Only a few cases of calcification have been reported histopathologically. Cases of calcifications in clear cell variants of mucoepidermoid carcinomas and in palatal minor salivary gland mucoepidermoid carcinomas were reported by some authors, and they thought them to be due to dystrophic calcification of the amorphous eosinophilic material secreted by intermediate basal cells.^{8–10} Longstanding cases can manifest with atypical clinical appearances.¹¹ The present case supports the data suggesting that calcifications can occur in longstanding lesions and that central mucoepidermoid carcinoma should be included in the differential diagnosis of all mixed lesions.

Radiographic appearance of central mucoepidermoid carcinoma as a mixed radiolucent–radio-opaque lesion is very rare, as indicated by the literature. The long duration is also uncommon for a malignant lesion. Clinically and radiographically, since the lesion's behaviour was different, it is important that the clinician be aware of the various clinical presentations of a particular disease process. This case report adds a new dimension to the clinical picture revealed by a central mucoepidermoid carcinoma.

References

1. Simon D, Somanathan T, Ramdas K, Pandey M. Central mucoepidermoid carcinoma of mandible: a case report and review of the literature. *World J Surg Oncol* 2003; **1**: 1–5.
2. Kochaji N, Goossens A, Bottenberg P. Central mucoepidermoid carcinoma: Case report, literature review for missing and available information and guideline proposal for coming case reports. *Oral Oncol Extra* 2004; **40**: 95–105.
3. Gingell JC, Beckerman T, Levy BA, Snider LA. Central mucoepidermoid carcinoma: review of the literature and report of a case associated with an apical periodontal cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1984; **57**: 436–440.
4. Namin AK, Moshref M, Shahoon H, Mashhadi A, Khojasteh A. Intraosseous mucoepidermoid carcinoma of the maxilla in a teenager: a case report and review of literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005; **100**: e93–e96.
5. 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.
6. De Mello-Filho FV, Brigato RR, Mamedea RCM, Alves Ricz HM, Saggiaro FP, Xavier SP. Central mucoepidermoid carcinoma: report of 2 cases. *Br J Oral Maxillofac Surg* 2008; **46**: 239–241.
7. Johnson B, Velez I. Central mucoepidermoid carcinoma with an atypical radiographic appearance. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008; **106**: e51–e53.
8. Maiko S, Tooru T, Masaki F, Ken A, Kaoru K, Jun S. A case of mucoepidermoid carcinoma of the hard palate with calcifications. *Jap J Oral Maxillofac Surg* 2004; **50**: 392–395.
9. Do Prado RF, Lima CF, Pontes HAR, Almeida JD, Cabral LAG, Carvalho YR. Calcifications in a clear cell mucoepidermoid carcinoma: a case report with histological and immunohistochemical findings. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; **104**: e40–e44.
10. Yoon JH, Ahn SG, Kim SG, Kim J. Calcifications in a clear cell mucoepidermoid carcinoma of the hard palate. *Int J Oral Maxillofac Surg* 2005; **34**: 927–929.
11. 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.