

## Case Report

# A case of central mucoepidermoid carcinoma associated with dentigerous cyst

Sayed Mohammad Razavi<sup>1</sup>, Roya Yahyaabadi<sup>2</sup>, Saeedeh Khalesi<sup>3</sup>

<sup>1</sup>Dental Implant Research Center and Department of Oral and Maxillofacial Pathology, School of Dentistry, Isfahan University of Medical Sciences, <sup>2</sup>Department of Oral and Maxillofacial Pathology, School of Dentistry, Isfahan University of Medical Sciences, <sup>3</sup>Dental Materials Research Center and Department of Oral and Maxillofacial Pathology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

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Address for correspondence:  
Dr. Saeedeh Khalesi,  
Department of Oral and  
Maxillofacial Pathology,  
Dental Materials Research  
Center, School of Dentistry,  
Isfahan University of Medical  
Sciences, Isfahan, Iran.  
E-mail: s\_khalesi@dnt.mui.  
ac.ir

## ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland neoplasm. Central MEC (CMEC) is a rare primary intraosseous bony lesion with an incidence of 2%–4.3% of all MECs reported. In this article, we present a rare case of a CMEC in the anterior region of maxilla at a 43-year-old female patient that was arising from a dentigerous cyst. CMECs are extremely rare tumor. They are usually low-grade lesions with favorable prognosis. Odontogenic cysts are one of the origins of this lesion. Treatment of impacted tooth is necessary in the early stage for prevention of this neoplasm.

**Key Words:** Dentigerous cyst, malignant, mucoepidermoid carcinoma

## INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland neoplasm, that is, 2.8%–15% of all salivary gland tumors.<sup>[1]</sup> This tumor is affecting commonly the parotid gland (89.6%), submandibular gland (8.4%), and sublingual gland (0.4%).<sup>[2]</sup> There are three main cell types in histopathological features of MEC containing mucin-producing, epidermoid, and intermediate cells.<sup>[3,4]</sup> Central MEC (CMEC) is a rare primary intraosseous bony lesion with an incidence of 2%–4.3% of all MECs reported.<sup>[5]</sup> The pathogenesis of CMEC is still unknown. However, an origin from ectopic salivary glands, metaplastic transformation of odontogenic epithelium or neoplastic transformation of epithelial lining of odontogenic cyst have been suggested.<sup>[6,7]</sup> This lesion is more frequent in mandible than in the maxilla and 4<sup>th</sup> and 5<sup>th</sup> decades of life.<sup>[2,3]</sup>

It affects in a higher ratio females than males.<sup>[3]</sup> Given that several of these lesions have been reported, but a few of the lesion is seen in the anterior maxilla.<sup>[4,8]</sup> In this article, we present a case of a CMEC in the anterior region of maxilla in a 43-year-old female patient developing from a previously dentigerous cyst.

## CASE REPORT

A 43-year-old female patient presented to the Department of Oral and Maxillofacial Surgery at Dental School of Isfahan University of Medical Sciences with a chief complaint of pain and swelling in the anterior portion of maxilla since 2 months. There was no significant medical or familial history and tobacco use. On extraoral examination, the face

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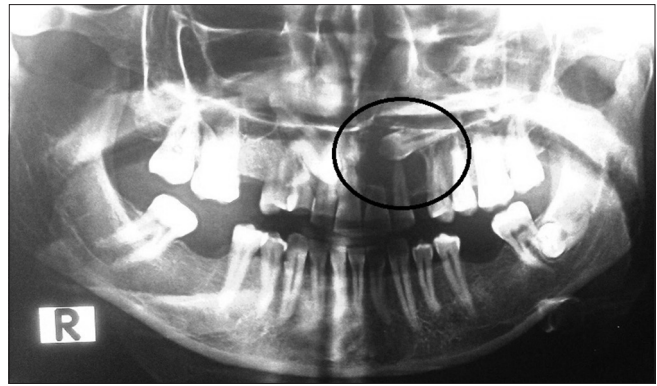


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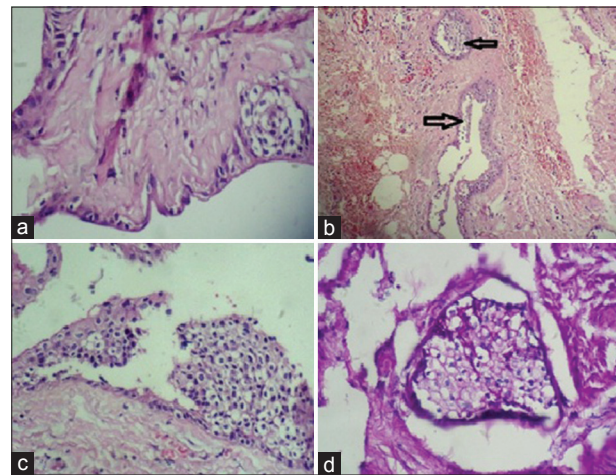
was normal. The submental and submandibular lymph nodes were not palpable or tender. In intraoral examination, swelling was smooth, firm and tender on palpation involving the anterior region of maxilla from the left lateral incisor to the left first premolar with the expansion of buccal cortical plate with intact overlying mucosa. The left canine tooth was absent. Panoramic radiograph showed well-defined unilocular radiolucency in the anterior left maxillary bone with an unerupted maxillary left canine tooth [Figure 1]. Based on the clinical and radiographical features, dentigerous cyst, keratocystic odontogenic tumor (KCOT), and adenomatoid odontogenic tumor (AOT) were considered in the differential diagnosis. An excisional biopsy was taken under local anesthesia. Histopathological examination showed a thin odontogenic epithelium 3–5 cell layers thick with nonkeratinized squamous cells and a few mucous cells in the superficial layer. The connective tissue wall was made up of loosely arranged collagen fibers and fibroblasts that suggested a dentigerous cyst [Figure 2a]. Connective tissue wall had numerous cystic spaces with foci of mucous, epidermoid, and clear cells [Figure 2b and c] which these cells stained by periodic-acid–Schiff (PAS) [Figure 2d]. Based on these findings, a final diagnosis of central low-grade MEC arising from a dentigerous cyst was made. She was referred to oral and maxillofacial surgery and oncologist for further treatment. After consultation with the oncologist, due to complete removal of the lesion in previous surgery and early detection, complementary therapies were not necessary and just be patient followed up. After 1 year, there was no recurrence of the lesion.

## DISCUSSION

CMEC is a rare malignant tumor in the jaw bone. The most cases of CMEC have been reported the posterior region of the mandible, but a few of this is seen in the maxilla.<sup>[7,9]</sup> Females are twice more affected than males with high incidence in the fourth and fifth decades.<sup>[9]</sup> Similar to other reported cases of CMEC, we presented a CMEC in a female patient with 43-year-old but unlike most other studies, this lesion involved maxillary bone. The most common clinical symptoms of CMEC include swelling of the involved area, pain, trismus, numbness or paresthesia and tooth mobility that was depended to extent affected adjacent structures.<sup>[3,7]</sup> Radiographic finding resemble to other odontogenic cysts or tumors show uni- or



**Figure 1:** Radiographic feature showing unilocular radiolucent lesion in around the crown of the left impacted canine tooth.



**Figure 2:** (a) A nonkeratinizing squamous cystic lining epithelium with a few mucous cells in the superficial layer and a connective tissue wall (H and E,  $\times 400$ ), (b) numerous cystic spaces with foci of mucous, epidermoid, and clear cells in connective tissue wall ( $\times 100$ ), (c) epidermoid and clear cells ( $\times 400$ ), (d) positive result for Periodic-acid–Schiff staining ( $\times 400$ ).

multi-lacunar radiolucencies.<sup>[4]</sup> Our case has pain and swelling in anterior region of maxilla with unilocular radiolucent lesion around the crown of canine tooth. The differential diagnosis is dentigerous cyst, KCOT, and AOT. Although in histopathological features, we have not observed the keratinized stratified squamous epithelium lining of cystic lesion we described the cystic lesion with nonkeratinized squamous epithelium with foci of mucous cells, epidermoid and intermediate cells set in a background of fibrous stroma.

The suggested origin of CMEC are as follows: (1) neoplastic change of entrapment of salivary glands within the mandible, (2) embryonic remnants of the submandibular gland within the mandible, (3) neoplastic transformation of mucus-secreting cells of epithelial lining of dentigerous cysts associated

with impacted tooth, and (4) neoplastic change and invasion from the epithelial lining of the maxillary sinus.<sup>[2,7]</sup> According to most studies, 50% of CMEC are associated with odontogenic cysts and impacted teeth, resemble to present case.<sup>[7]</sup> For maxillary CMEC, detection of the origin as the palate mucosa, gingival mucosa or maxillary sinus mucosa is different in comparison CMEC in the mandible.<sup>[10]</sup> However, the origin of maxillary CMEC in our case was identified, because the lesion was dentigerous cyst around the crown of impacted canine. Some criteria for the definitive diagnosis explained by Alexander that include: (a) presence of an intact cortical plate, (b) radiographic feature of destruction bone, (c) histopathologically confirmation, (d) observation of mucin production (positive PAS staining or mucicarmine staining), (e) absence primary lesions in salivary glands, and (f) exclusion of metastatic lesion or an odontogenic tumor.<sup>[11]</sup> All these criteria have been detected in the present case for diagnosis CMEC. Brookstone and Huvos had explained a staging system for CMEC. The lesions with intact cortical plates without evidence of bony expansion indicate Stage I disease. Stage II disease is surrounded by intact cortical bone that has some degree of expansion. Any instance of cortical perforation, breakdown of the overlying periosteum or nodal spread is Stage III disease.<sup>[12]</sup> The presented case has expanded bone, but buccal and lingual cortices were intact during surgery process and could be imputed as Stage II disease.

The main treatment of CMEC is surgery including conservative methods such as curettage, enucleation, marsupialization, local excision, and radical methods.<sup>[2,13]</sup> Recurrence rate of the cases with conservative surgical methods was 40%. According to studies, the radical methods of treatment such as segmental resection with or without adjacent therapy have recurrence rate of 4%.<sup>[13]</sup> Although the reported of CMECs were low-grade tumors in histopathological examination with favorable prognosis, Metastasis is reported in 9% of cases into regional lymph nodes, ipsilateral clavicle, lung, and brain.<sup>[14]</sup> Then, neck dissection and adjacent therapy is recommended for high-grade tumors.<sup>[9]</sup> Furthermore, maxillary CMECs have worse prognosis due to the possibility of local extensions into vital structures.<sup>[7]</sup>

## CONCLUSION

CMECs are extremely rare tumor. They are usually low-grade lesions and have favorable prognosis.

Odontogenic cyst is one of the origins of this lesion. Treatment of impacted tooth is necessary in early stage for prevention of this neoplasm.

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## Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

## REFERENCES

1. Chiu GA, Woodward RT, Benatar B, Hall R. Mandibular central mucoepidermoid carcinoma with distant metastasis. *Int J Oral Maxillofac Surg* 2012;41:361-3.
2. 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.
3. Kechagias N, Ntomouchtsis A, Mavrodi A, Christoforidou B, Tsekos A, Vahsevanos K, *et al.* Central mucoepidermoid carcinoma of the anterior region of the mandible: Report of an unusual case and review of the literature. *Oral Maxillofac Surg* 2015;19:309-13.
4. Takano M, Kasahara K, Matsui S, Ogawa C, Matsuzaka K. A case of mucoepidermoid carcinoma associated with maxillary cyst. *Bull Tokyo Dent Coll* 2012;53:119-25.
5. Raut D, Khedkar S. Primary intraosseous mucoepidermoid carcinoma of the maxilla: A case report and review of literature. *Dentomaxillofac Radiol* 2009;38:163-8.
6. Khan HA, Loya A, Azhar R, Din NU, Bell D. Central mucoepidermoid carcinoma, a case report with molecular analysis of the TORC1/MAML2 gene fusion. *Head Neck Pathol* 2010;4:261-4.
7. Spoorthi BR, Rao RS, Rajashekaraiah PB, Patil S, Venkatesiah SS, Purushothama P, *et al.* Predominantly cystic central mucoepidermoid carcinoma developing from a previously diagnosed dentigerous cyst: Case report and review of the literature. *Clin Pract* 2013;3:e19.
8. Purohit S, Manjunatha BS, Dagrur K, Harsh A. Intraosseous mucoepidermoid carcinoma of maxilla: A rare entity. *J Exp Ther Oncol* 2016;11:207-11.
9. Chundru NS, Prasanth T, Nandan SR, Rajesh A. Central mucoepidermoid carcinoma. *J Cancer Res Ther* 2015;11:657.
10. Li Y, Li LJ, Huang J, Han B, Pan J. Central malignant salivary gland tumors of the jaw: Retrospective clinical analysis of 22 cases. *J Oral Maxillofac Surg* 2008;66:2247-53.
11. Aggarwal P, Saxena S. Aggressive growth and neoplastic potential of dentigerous cysts with particular reference to central mucoepidermoid carcinoma. *Br J Oral Maxillofac Surg* 2011;49:e36-9.
12. 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;50:229-36.
13. Tucci R, Matizonkas-Antonio LF, de Carvalhosa AA, Castro PH, Nunes FD, Pinto DD Jr., *et al.* Central mucoepidermoid

carcinoma: Report of a case with 11 years' evolution and peculiar macroscopical and clinical characteristics. *Med Oral Patol Oral Cir Bucal* 2009;14:E283-6.

14. Zhou CX, Chen XM, Li TJ. Central mucoepidermoid carcinoma: A clinicopathologic and immunohistochemical study of 39 Chinese patients. *Am J Surg Pathol* 2012;36:18-26.