



Central Mucoepidermoid Carcinoma Arising in a Dentigerous Cyst: A Challenging Case Report

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Central mucoepidermoid carcinoma (MEC) is a rare neoplasm of the jaw which is more common in the posterior of mandible, and the most likely origin is odontogenic epithelium. The aim of the present report is to describe a case of maxillary MEC arising in a dentigerous cyst in a 57-year-old female. It is crucial for pathologists to aware of the histopathologic features of this rare neoplasm to have a correct diagnosis.

Key words: Dentigerous cyst, mucoepidermoid carcinoma, neoplasm, odontogenic cyst

INTRODUCTION

Central mucoepidermoid carcinomas (CMECs) of the jaws are rare, which may originate from ectopic salivary gland tissue, epithelium of odontogenic cysts, and maxillary sinus or submucosal salivary glands with intrabony extension.^{1,2} These are more common in the fourth and fifth decades of life with female propensity. Most of the cases are found in the posterior of the mandible.³ A variety of radiologic imaging has been reported which appears to be nondiagnostic, and biopsy is essential for a definitive diagnosis.⁴ Common clinical symptoms include swelling, pain, trismus, paresthesia, and tooth mobility.³ The treatment of choice is radical surgery. Adjuvant radiotherapy is recommended for high-grade cases.⁵ This report presents a 57-year-old woman who suffered from CMEC of the maxillary anterior region developing in a dentigerous cyst (DC).

CASE REPORT

A 57-year-old woman was referred to the Department of Oral and Maxillofacial Surgery, Shahid Beheshti University of Medical Sciences, Tehran, Iran, for evaluation of painless swelling of the left anterior maxillary vestibule with 1-month

duration. The patient had no history of previous trauma or medical problems. Intraoral examination revealed buccal cortical expansion in the area of left maxillary impacted canine. The overlying mucosa was intact. The panoramic radiograph showed a well-defined pericoronal corticated radiolucency of the left impacted canine measuring 1 cm × 1 cm [Figure 1]. On cone-beam computed tomography, buccal cortical destruction was evident [Figure 2a and b]. There was no cervical lymphadenopathy and laboratory blood tests were within normal limits. Aspiration of the lesion showed a serous-like fluid. Due to radiographic features and fluid aspiration, DC, odontogenic keratocyst, and calcifying odontogenic cyst were considered in the differential diagnosis. The lesion completely excised under local anesthesia. Grossly, the lesion consists of multiple pieces of cystic brown soft tissue with elastic consistency accompanied by a canine totally measuring 2.3 cm × 1.5 cm × 0.3 cm. One piece attached to the cement-enamel junction of tooth. Microscopic examination showed a cystic lesion lined by nonkeratinized stratified squamous epithelium with varying thickness and few mucous cells. The underlying fibrous connective tissue revealed infiltration of chronic inflammatory cells, cholesterol clefts, associated giant cells, and one epithelial island containing clear

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Figure 1: Periapical radiograph showing a unilocular radiolucent lesion around the crown of left maxillary impacted canine

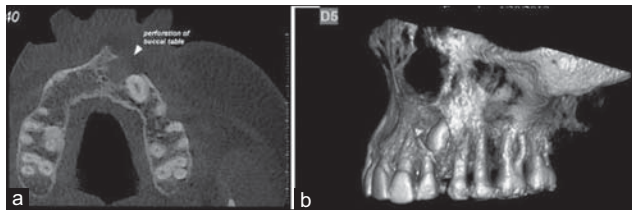


Figure 2: (a) Cone-beam computed tomography demonstrates buccal cortical destruction and (b) three-dimensional reconstruction

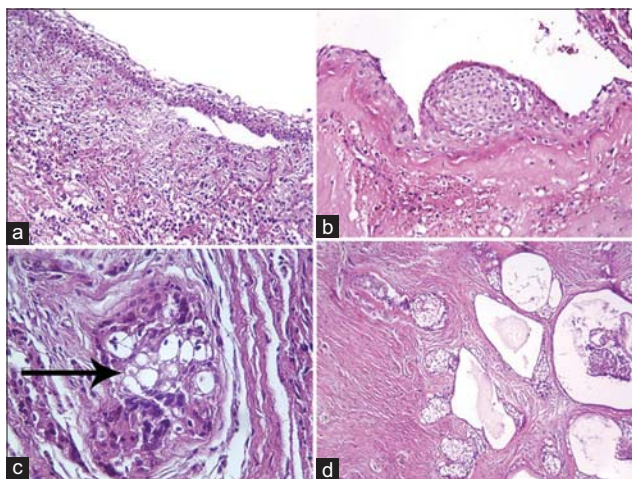


Figure 3: Histopathological sections showing (a) a cystic lesion lined by nonkeratinized stratified squamous epithelium with exocytosis and spongiosis (H and E, $\times 200$). (b) Epithelial lining showing nodular thickening (H and E, $\times 200$). (c) An epithelial island (black arrow) containing clear and squamous cells (H and E, $\times 400$). (d) Several islands of squamous and clear cells with microcystic formation (H and E, $\times 200$)

and squamous cells [Figure 3a-d]. Therefore, serial sections of the lesion were performed and additional sections showed several islands of squamous and clear cells with microcystic

formation. According to the aforementioned microscopic and radiographic features, the diagnosis of “MEC arising in DC” was confirmed. In spite of the fact that CMEC requires radical surgery, due to patient’s dissatisfaction to undergo second surgery, distance of tumoral islands to bony borders, and initial complete enucleating of the lesion, close clinical and radiographic follow-up was performed. The patient is free from disease for 15 months postoperatively.

DISCUSSION

Odontogenic epithelium surrounding impacted teeth has the potential to differentiate into a wide-ranging of jaw lesions, from cysts to neoplasms. Uncommon secondary development of tumors from DC including adenomatoid odontogenic tumor, ameloblastoma, MEC, and squamous cell carcinoma has been reported.⁶ CMECs are rare, forming 2%–3% of all MECs.¹ Some authors found that about half of the CMECs were associated with dental cysts or impacted teeth,⁷ whereas others^{1,8} showed no significant relationship between CMECs and impacted teeth. Long-term chronic inflammation may motivate neoplastic transformation in odontogenic cysts.⁶ Kalburge *et al.*⁶ indicated that tumoral transformation of odontogenic cysts tends to arise with increasing age. Our case was also in the sixth decade of life that implies the importance of precise evaluation of pericoronal lesions with increasing age. Most of the reported cases of CMECs presented in the posterior of the mandible,³ which disagreed with our case. The most clinical signs and symptoms reported include pain and swelling, but our case showed painless swelling similar to He *et al.* reports¹ and in contrast to others.^{3,9} Brookstone and Huvos⁸ introduced a staging system for CMEC that includes Stage I: Intact cortical plate without expansion, Stage II: Intact cortical plate with bony expansion, and Stage III: Any cortical perforation or breakdown of the overlying periosteum or nodal spread. In our patient, there was perforation in the buccal cortical plate; thus, it can be considered as Stage III. Histopathologically, MEC revealed that a neoplasm composes of nests and islands of epidermoid, mucous, and intermediate cells showing cystic spaces in a fibrous stroma.⁵ A significant number of CMEC have been stated to be largely low-grade cystic lesions.⁴ Glandular odontogenic cyst (GOC) may be microscopically mistaken with CMEC.^{10,11} GOC is lined by stratified squamous epithelium with varied thickness and surface cuboidal or columnar ciliated cells. Small microcysts and clusters of mucous cells are also described which may probably cause diagnostic challenges.¹⁰ Nevertheless, examination of multiple sections usually results in the differentiation of these lesions. Furthermore, CMECs demonstrate *MAML2* gene rearrangements, which often are not found in GOCs.¹⁰

Due to multi-potentiality of odontogenic epithelium of DC, Razavi *et al.*³ recommended removal of impacted teeth in early stage to prevent neoplastic transformation. The clinical behavior of MECs arising in odontogenic cysts or *de novo* has not been reported.⁹ Hence, most of the CMECs are low-grade malignancies, radical surgery is the treatment of choice, and adjuvant radiotherapy is recommended for high-grade ones. Recurrence rate after conservative surgery was about 40%.³ Metastasis is reported about 9%.¹⁰

Taken together, in DC cases with suspicious structure in the underlying connective tissue, serial sections of the lesion are highly recommended. Furthermore, clinical and radiographic follow-up of impacted tooth, especially in elder adults, is suggested. This issue highlights the importance of accurate examination of pericoronal lesions with increasing age.

Declaration of patient consent

All appropriate patient consent forms were obtained. In the form, the patient has given her consent for her 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 is no conflict of interest.

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