Journal of Dentomaxillofacial Radiology, Pathology and Surgery

Spring 2021, Volume 10, Number 2

Case Report: The Importance of Radiographic and Histologic Correlation in Diagnosis of Intra-Osseous Mucoepidermoid Carcinoma

Ali Lotfi[1], Mohammad Mehdizadeh[2], Sepideh Mokhtari[3], Saede Atarbashi-Moghadam[*4]

[1]*Assistant Professor, Department of Oral and Maxillofacial Pathology, school of dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.*
[2]*Associate professor, department of oral and maxillofacial Surgery, School of dentistry, Qom University of Medical Sciences, Qom.*
[3] *Education Development Office, School of dentistry, Tehran University of Medical Sciences, Tehran, Iran.*
[4] *Associate Professor, Department of Oral and Maxillofacial Pathology, school of dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.*

Use your device to scan and read the article online

Citation: Lotfi A, Mehdizadeh M, Mokhtari S, Atarbashi Moghadam S. The Importance of Radiographic and Histologic Correlation in Diagnosis of Intra-Osseous Mucoepidermoid Carcinoma. Journal of Dentomaxillofacial Radiology, Pathology and Surgery. 2021; 10(2):24-27. http://dx.doi.org/

http://3dj.gums.ac.ir

Article info:
Received: 2021/4/05
Accepted: 2021/4/17

Keywords:
Central Mucoepidermoid Carcinoma, Glandular Odontogenic Cyst, Salivary gland tumors

ABSTRACT

Central mucoepidermoid carcinoma (MEC) of the jaws is rare and it comprises 2–3% of all MECs reported in the literature. It may be similar to a glandular odontogenic cyst (GOC), mainly in incisional biopsies. The precise diagnosis of these lesions is essential since they have different treatment modalities and prognosis. This paper presented a 71-year-old male patient presented with a unilocular radiolucent lesion in the left posterior mandible. The lesion had some evidence of cortical perforation and soft tissue extension in radiographic features. Incisional biopsy of the lesion was performed and the specimen was diagnosed as the GOC. However, the excisional biopsy revealed proliferation of epidermoid and mucous cells in the cyst wall and the lesion was diagnosed as central low-grade MEC of mandible. Central mucoepidermoid carcinoma (CMEC) may resemble a GOC in the incisional biopsy. Therefore, we discuss the importance of radiographic and histopathologic correlation in the diagnosis of CMEC in this article.

*** Corresponding Author:**
Saede Atarbashi-Moghadam.
Address: Department of Oral and Maxillofacial Pathology, Dental School Shahid Beheshti University of Medical Sciences, Velenjak Street, Tehran, IR Iran.
Tel: +98-9122385589
E-mail: dr.atarbashi@gmail.com

Journal of Dentomaxillofacial Radiology, Pathology and Surgery

Case report

A 71-year-old man was referred to an oral and maxillofacial pathology center for evaluation of painless non-tender mild swelling in the left posterior mandible with unknown duration (Figure 1).

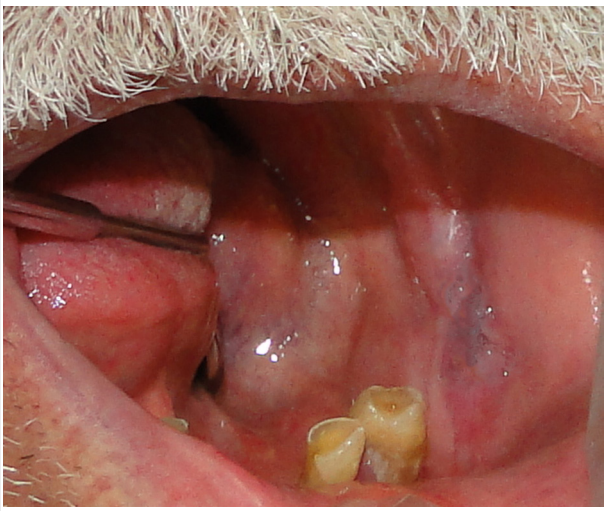

Figure 1: Intraoral photograph shows a mild swelling in the left posterior of the mandible.

On panoramic radiograph, a well-defined, unilocular radiolucent lesion with sclerotic borders, extending from the posterior body of the mandible into the ascending ramus was seen. CT scan revealed a destructive lesion in the left posterior body and ramus of the mandible causing cortical destruction (Figure2).

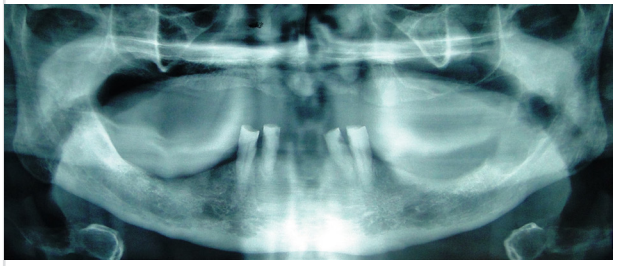

Figure 2: Radiograph shows a lesion in the left posterior body and ramus of mandible.

Soft tissue extension of the lesion was present as a mass in the oral space adjacent to the pterygoid plate. No septation or bone expansion was visible. Since the buccal fats were clear and the intraosseous lesion was well-defined, the possibility of peripheral MEC with an invasion of the underlying bone was excluded (Figure 3).

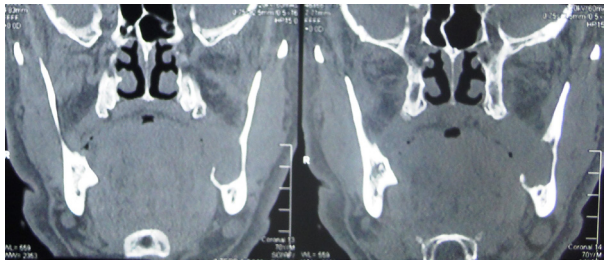

Figure 3: CT scan reveals cortical destruction and soft tissue extension.

On extraoral examination, no palpable lymph node was detected in the neck region. The aspiration of the lesion showed a serous-like fluid. His past medical history was unremarkable. Due to radiographic features and fluid aspiration, a cystic lesion such as odontogenic keratocyst and residual cyst were considered in the differential diagnosis. Therefore, an incisional biopsy was performed under local anesthesia. The histopathologic sections demonstrated a cystic lesion lined by stratified squamous epithelium, exhibiting small microcysts, clusters of mucous cells and some hobnail changes. Many cholesterol clefts and associated giant cells were also seen. There was no neoplastic island in the connective tissue (Figure 4).

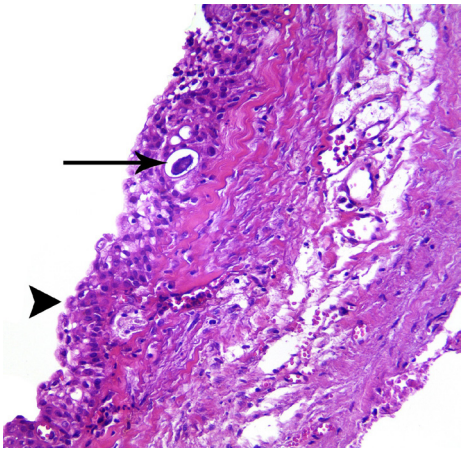

Figure 4: The microscopic sections demonstrate a cystic lesion lined by stratified squamous epithelium, exhibiting small microcyst (black arrow) and some hobnail changes (black arrowhead) without any neoplastic islands in the fibrous wall (H& E staining ×200).

Lotfi A, et al. The Importance of Radiographic and Histologic Correlation in Diagnosis of Intra-Osseous Mucoepidermoid Carcinoma. Journal of Dentomaxillofacial Radiology, Pathology and Surgery. 2021; 10(2):24-27. http://dx.doi.org/

According to these microscopic features, the diagnosis of GOC was considered. The lesion was completely excised. The excisional biopsy showed cystic spaces with nests and islands of epidermoid and mucous cells within the fibrous wall (Figure 5).

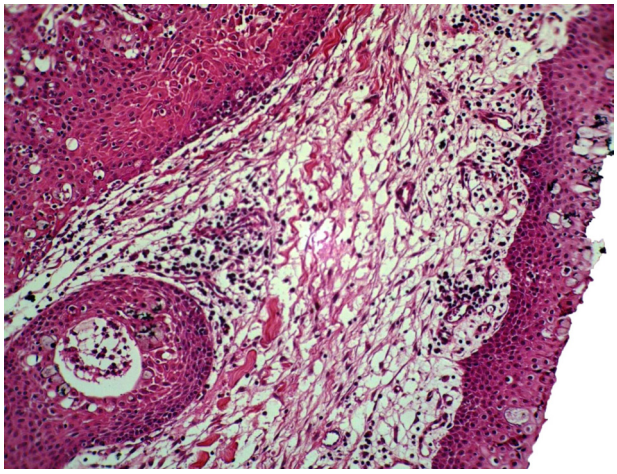

Figure 5: Microscopic section reveals a cystic lesion lined by stratified squamous epithelium containing mucus cells and microcysts accompanying tumoral islands. (H& E staining ×100).

Therefore, the final diagnosis was changed to low-grade CMEC. In further evaluation, no evidence of metastasis was found. The patient did not accept additional surgery and only underwent close follow-up.

Discussion:

MEC as the most common salivary gland tumor is composed of a mixture of mucus-producing cells, squamous (epidermoid) cells and intermediate cells (1). Intra-osseous or CMEC is most common in the posterior mandible. The frequent presenting symptom of this entity is cortical expansion. However, pain, trismus, paresthesia, tooth displacement and root resorption may be present (2). CMECs are histologically low-grade tumors that demonstrate prominent cyst formation, minimal cellular atypia and a relatively great percentage of mucous cells. Due to the low-grade nature of these lesions, wide local excision is the treatment of choice in most cases (3). The main microscopic differential diagnosis of CMEC is a GOC and some reports have presented this possible misdiagnosis (2, 4-6). The GOC has an unpredictable and potentially aggressive behavior with the strong predilection for the anterior region (4). Painless swelling is the most prevalent presentation. It is often exhibited as a well-defined, unilocular or multilocular radiolucency and cortical perforation may occur. Enucleation, curettage and local block excision have been designated as the treatment modalities for this lesion (5).

In the present case, a prominent cystic space was evident. GOC is lined by squamous epithelium of varying thickness. The superficial epithelial cells tend to be cuboidal to columnar, resulting in a hobnail and sometimes papillary surface. Duct-like spaces lined by cuboidal cells and mucin-producing goblet cells, cilia and focal spherical nodules may be seen (6). The differentiation of low-grade CMEC from GOC is challenging and there is significant overlap between these two entities.

The most frequent radiographic feature of CMECs is a well-defined unilocular or multilocular radiolucency. Chan et al (3) reported that CMECs have commonly well-defined sclerotic borders, internal amorphous sclerotic bone and many small loculations. However, in the present case, CT images clearly showed an extension of the lesion into the soft tissue reaching pterygoid plates. Cortical perforation has been reported in about 26% of GOCs (7) but soft tissue extension is unlikely. In the cases like this, the diagnosis should not be based on a single entity such as incisional biopsy and further investigation with a correlation of clinical, radiographic and histopathologic findings is necessary.

Histologically, GOC is frequently confused with CMEC. Differentiation of CMEC from GOC should be based on exact histologic examination of the lesion. The presence of the major criteria of GOC is very important in diagnosis. It should be kept in mind that superficial epithelial cells in the cyst line of GOC tend to be cuboidal to columnar. In the absence of this major criterion, the diagnosis of a cystic lesion as a GOC is very questionable. Additionally, the mere pres-

Journal of Dentomaxillofacial Radiology, Pathology and Surgery

ence of ciliated or goblet cells does not support the diagnosis of GOC. The other histologic point is that in problematic cases, especially in the posterior mandible, more sections of the tissue should be prepared and histologically evaluated to rule out CMEC. However, cellular atypia and solid epithelial proliferation is never present in GOC (5). On the other hand, the presence of superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcysts or duct-like structures is suggestive of GOC (8).

Immunohistochemistry may be helpful in the diagnosis of some problematic cases. Expressions of CK18 in CMEC and CK19 in GOC have been reported in the literature (4). In addition, in cases with GOC diagnosis particularly in incisional samples, a serial section of the lesion is highly recommended (2).

A practical and diagnostic application in problematic cases is the evaluation of MAML2 rearrangement or CRTC1-MAML2 fusion in MECs with fluorescence in situ hybridization (FISH) or RT-PCR which is negative in GOCs (9-11). Interestingly Nagasaki et al (11) reported a case of CMEC arising from previous GOC using MAML2 rearrangement.

Conclusion:

Correlation of radiographic and histopathologic features is necessary in the diagnosis of intraosseous lesions and any clue of malignant behavior of these lesions should be taken into consideration. Furthermore, CMEC may show a predominantly cystic pattern in gross and might reveal a large amount of fluid in aspiration. Therefore, attention to all clinical, radiographic and histopathologic features should be considered in the differentiation of CMEC and GOC.

Conflict of interest:

None declared

References:

1. Mokhtari S, Mokhtari S. Clinical features and differential diagnoses in laryngeal mucoepidermoid carcinoma. Clin Med Insights Pathol. 2012;5:1-6.https://doi.org/10.4137/CPath.S8435

2. Atarbashi Moghadam S, Atarbashi Moghadam F. Intraosseous mucoepidermoid carcinoma: report of two cases. J Dent (Shiraz). 2014; 15: 86-90.

3. Chan KC, Pharoah M, Lee L, Weinreb I, Perez-Ordonez B. Intraosseous mucoepidermoid carcinoma: a review of the diagnostic imaging features of four jaw cases. Dentomaxillofac Radiol 2013; 42: 20110162. https://doi.org/10.1259/dmfr.20110162

4. Mascitti M, Santarelli A, Sabatucci A, Procaccini M, Lo Muzio L, Zizzi A, et al. Glandular odontogenic cyst: review of literature and report of a new case with cytokeratin-19 expression. Open Dent J. 2014 ;8: 1-12. https://doi.org/10.2174/1874210601408010001

5. Prabhu S, Rekha K, Kumar G. Glandular odontogenic cyst mimicking central mucoepidermoid carcinoma. J Oral Maxillofac Pathol 2010; 14: 12-15.https://doi.org/10.4103/0973-029X.64303

6. Fowler CB, Brannon RB, Kessler HP, Castle JT, KahnMA. Glandular odontogenic cyst: analysis of 46 cases with special emphasis on microscopic criteria for diagnosis. Head Neck Pathol 2011; 5: 364-375.https://doi.org/10.1007/s12105-011-0298-3

7. Chrcanovic BR, Gomez RS. Glandular odontogenic cyst: An updated analysis of 169 cases reported in the literature. Oral Dis. 2018;24(5):717-724.https://doi.org/10.1111/odi.12719

8. Shah AA, Sangle A, Bussari S, Koshy AV. Glandular odontogenic cyst: A diagnostic dilemma. Indian J Dent. 2016;7(1):38-43.https://doi.org/10.4103/0975-962X.179371

9. Bell D , Lewis C, El-Naggar AK, Weber RS. Primary intraosseous mucoepidermoid carcinoma of the jaw: Reappraisal of the MD Anderson Cancer Center experience. Head Neck. 2016; 38 Suppl 1:E1312-7.https://doi.org/10.1002/hed.24219

10. Bishop JA, Yonescu R, Batista D, Warnock GR, Westra WH. Glandular odontogenic cysts (GOCs) lack MAML2 rearrangements: finding to discredit the putative nature of GOC as a precursor to central mucoepidermoid carcinoma. Head Neck Pathol. 2014;8: 287-90. https://doi.org/10.1007/s12105-014-0534-8

11. Nagasaki A, Ogawa I, Sato Y, Takeuchi K, Kitagawa M, Ando T, et al. Central mucoepidermoid carcinoma arising from glandular odontogenic cyst confirmed by analysis of MAML2 rearrangement: A case report. Pathol Int. 2018; 68: 31-35.https://doi.org/10.1111/pin.12609