

## Periapical mucoepidermoid carcinoma: a case report with insights for diagnosis and dental rehabilitation

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### Abstract:

Periapical radiolucent lesions may represent a challenge in daily clinical practice because many cystic lesions and tumours sometimes surround teeth with compromised pulp. In this paper we have showed an uncommon manifestation of mucoepidermoid carcinoma, firstly due to it intraosseous involvement and secondarily to involve the maxillary bone around the apex of a molar teeth without pulp vitality on clinical tests. An incisional biopsy was the goal of this diagnosis. The mucous and epidermoid cells lining a cystic cavity, and the marginal minor salivary glands also showing proliferative foci of these tumoral cells, led to the diagnosis of mucoepidermoid carcinoma on histopathology. A respective approach was made as primary treatment, which resulted in a bucconasal communication. An obliterating removable prosthesis was conducted over a in the office approach, significantly improving speech and swallowing as showed. The discussion pointed here highlights the case and the literature hot topics for diagnosis of periapical radiolucencies, objectifying to guide the clinicians reviewing the current state of art. The structured diagnostic approach used here and illustrations of the new case are opportune to achieve this objective.

**Keywords:** Mucoepidermoid Carcinoma, Periapical Diseases, Differential Diagnoses.

## **Carcinoma mucoepidermóide periapical: relato de caso com *insights* para diagnóstico e reabilitação oral**

### **Resumo:**

Lesões radiotransparentes periapicais podem representar um desafio na prática clínica diária porque muitas lesões císticas e tumores às vezes circundam dentes com polpa comprometida. Neste trabalho mostramos uma manifestação incomum de carcinoma mucoepidermóide, primeiro devido ao envolvimento intraósseo e, secundariamente, por envolver o osso maxilar ao redor do ápice de um dente molar sem vitalidade pulpar em testes clínicos. Uma biópsia incisional foi o objetivo deste diagnóstico. As células mucosas e epidermóides que revestem uma cavidade cística, e as glândulas salivares menores marginais também apresentando focos proliferativos dessas células tumorais, levaram ao diagnóstico de carcinoma mucoepidermóide na histopatologia. Foi feita a respectiva abordagem como tratamento primário, que resultou em comunicação buconasal. Uma prótese removível obliterante foi realizada em consultório, melhorando significativamente a fala e a deglutição, conforme mostrado. A discussão aqui apontada destaca o caso e os temas quentes da literatura para o diagnóstico de radiolucências periapicais, objetivando orientar os clínicos na revisão do estado da arte atual. A abordagem diagnóstica estruturada aqui utilizada e as ilustrações do novo caso são oportunas para atingir esse objetivo.

**Palavras-chave:** Carcinoma mucoepidermoide, doenças periapicais, diagnóstico diferencial.

## **Carcinoma mucoepidermoide periapical: caso clínico con *insights* para el diagnóstico y rehabilitación bucal**

### **Resumen:**

Las lesiones radiolúcidas periapicales pueden representar un desafío en la práctica clínica diaria porque muchas lesiones quísticas y tumores a veces rodean dientes con pulpa comprometida. En este trabajo mostramos una manifestación inusual del carcinoma mucoepidermoide, primero por afectación intraósea y, secundariamente, por afectación del hueso maxilar alrededor del ápice de un molar sin vitalidad pulpar en ensayos clínicos. Una biopsia incisional fue el objetivo de este diagnóstico. Las células mucosas y epidermoides que recubren una cavidad quística, y las glándulas salivales menores marginales que también muestran focos proliferativos de estas células tumorales, llevaron al diagnóstico de carcinoma mucoepidermoide mediante histopatología. Se utilizó como tratamiento primario el abordaje respectivo, que resultó en la comunicación buconasal. Se realizó una prótesis obliterante removible en el consultorio, lo que mejoró significativamente el habla y la deglución, como se muestra. La discusión destacada aquí resalta el caso y los temas candentes en la literatura para el diagnóstico de radiolucidez periapical, con el objetivo de guiar a los médicos en la revisión del estado actual del arte. El enfoque de diagnóstico estructurado utilizado aquí y las nuevas ilustraciones de casos son oportunos para lograr este objetivo.

**Palabras clave:** Carcinoma mucoepidermoide, enfermedades periapicales, diagnóstico diferencial.

## **INTRODUCTION**

Periapicopathies is a term used to describe the inflammatory periapical diseases associated with necrotic dental pulps, often leading to periapical radiolucencies attributed to the chronic periapical diseases termed dental granuloma and radicular inflammatory cyst (RIC) (KONTOGIANNIS *et al.*, 2014). On the other hand, some periapical radiolucencies or

diseases may not be related to an inflammatory disease from the teeth, highlighting the importance of an adequate use of semiotechnique and complementary exams, like tests of pulp vitality and radiographs/ computerized tomography (CT), respectively (PONTES *et al.*, 2015).

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Mucoepidermoid carcinoma (MEC), the topic of this paper, are generally of slow growing and with low cell atypia on histopathological examination, being described in the past as a benign tumor, until the 1991 World Health Organization classification (TAKANO *et al.*, 2012). This is the most common malignant salivary gland neoplasm, comprising about 5% to 10% of salivary gland tumors. Its prevalence is higher in women aged 40 to 50 years, although there are reports of patients affected between the first and seventh decades of life. The parotid gland is the most commonly involved site, followed by minor salivary glands of the palate, submandibular glands, and sublingual, consecutively. Additionally, other glands outside the stomatognathic system must be involved, like the breast gland, but these specific statistics were not of interest in this paper (MERNA *et al.*, 2018; BASARAN *et al.*, 2018; ABT *et al.*, 2019).

The intraosseous variant of MEC is a rare entity, seen in 2% to 3% of cases of MEC in the jaws. According to Abt and collaborators (2019), less than 200 cases have been reported since 1939 in the literature. These may mimic benign odontogenic cysts or tumors in both clinical and radiographic aspects, and thus the histopathological diagnosis is the decisive phase.

The etiopathogenesis of intraosseous MEC is uncertain. Several theories and speculations include: (1) entrapment of retromolar, submandibular, or sublingual mucous

glands in the mandible during the embryonic developmental stage; (2) mucosal metaplasia and neoplastic transformation of the epithelial lining of an odontogenic cyst; (3) iatrogenic entrapment of the minor salivary glands; (4) neoplastic transformation of the maxillary sinus epithelium; and (5) remnants of the dental lamina (LI *et al.*, 2018; SHERIN *et al.*, 2011; LI *et al.*, 2012).

The aim of this study is to report a case of a primary intraosseous CME involving the apex of a tooth that did not respond to pulp vitality tests, mimicking the conventional periapical pathosis (e.g., RIC, periapical granuloma). Moreover, is to attach a guided review to elucidate the important steps for diagnosis and a simple option of rehabilitative approach. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Ethics Committee of 5.580.762.

## METHODS

This study is a case report and a brief exploratory and integrative literature review to obtain the state of art for diagnosis of periapical MEC: first, an opened narrative review seeking to identify diagnostic criteria and differential diagnoses that can guide the clinical approach; secondarily, a brief systematized review looking for cases of CME in dentate patients that may provide data for differential diagnosis of CME in a location next to dental apex. For this last systematized step of review, the MEDLINE databases was assessed trough pubmed.com platform, using the research terms “Mucoepidermoid carcinoma” AND “intraosseous”, resulting in 57 papers; and “Mucoepidermoid carcinoma” AND “periapical”, resulting in 8 papers. Cases published in languages other than English and Intraosseous MEC in edentulous patients or without a context of tooth involvement were excluded, and them, 8 articles were selected to be included in the table of results aiming to obtain helpful data for diagnosis of MEC mimicking periapical diseases.

## RESULTS

### Case report

A 50-years-old female patient came to the dental clinics with the complaint: "I have do fix my teeth, and I have a blister on the roof of my mouth". She reported the appearance of the lesion 4 years ago, without other symptoms. Clinical examination revealed a floating swelling on palpation in the corresponding region of the palate (Figure 1a). The tooth had a negative response to the pulp vitality test and percussion, which reinforced the hypothesis that it was an inflammatory root cyst. Her medical history was not contributory. An aspiration puncture was performed, and it's content revealed a slight presence of bloody material, suggesting the possibility of a solid lesion. A CT scan showed a hypodense lesion involving the root of tooth 27, breaking through the cortical bone and infiltrating the palate (Figure 2).

**Figure 1.** Initial clinical aspect of lesion.



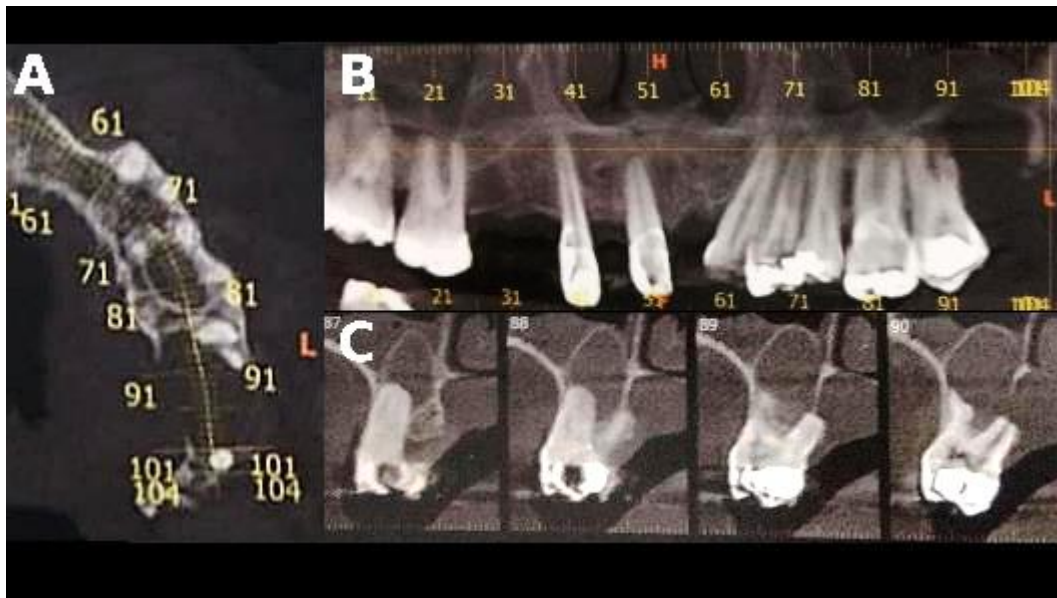
**Legend:** It shows the left palatine mucosa with a floating overgrowth next to the the molar teeth.  
**Source:** Files of authors.

Combined with the findings of the imaging exam, the differential diagnoses included periapical cyst and other more aggressive lesions like ameloblastoma, central giant cell

lesion, and a salivary gland tumor. An incisional biopsy was performed and histological analyses revealed a lesion composed by cystic spaces lined either by regions of single layer cells or amounts of cells with conspicuous cytoplasm sometimes eosinophilic, sometimes clear and broad, often with round nucleus with disperse chromatin. These morphologic characteristics were sufficient to conclude the diagnosis of a periapical MEC (Figure 3).

The patient was referred to a head and neck surgeon and followed for a total excision of the lesion with a safety margin, creating an oronasal communication as a sequel (Figure 4). The final diagnosis after remotion of lesion was Low-grade MEC stage T2N0M0, with a significant cystic component and "mural" growths present, including those nearby the minor palatine salivary glands. Because of the early diagnosis, there was no need for chemo and radiotherapy.

**Figura 2.** Cone bean CT Scan.

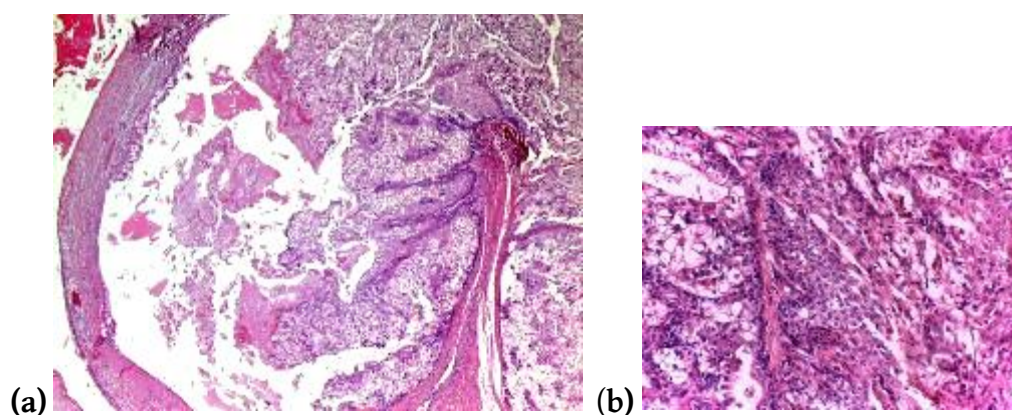


**Legenda:** (a) – axial view of lesion on right side with rupture of both vestibular and palatine cortical plates; (b) – panoramic reconstruction showing a periapical hypodense lesion involving the roots of tooth 27 extending near to tuber and maxillary sinus.; (c) parasagittal sections with presence of hypodense area associated with the root of tooth 27 and lifting the maxillary sinus.

**Fonte:** Files of authors.



**Figure 3.** Histopathologic view of case, stained with Hematoxylin-Eosin.



**Legend:** (a) – Lower power field view showing cystic space at left side and the solid proliferation of mucoepidermoid cells at right; (b) – high power field showing a solid area of tumor containing clear mucous cells with some intercalated epidermoid cells.

**Source:** Files of authors.

**Figure 4.** Clinical view of the surgical sequelae.



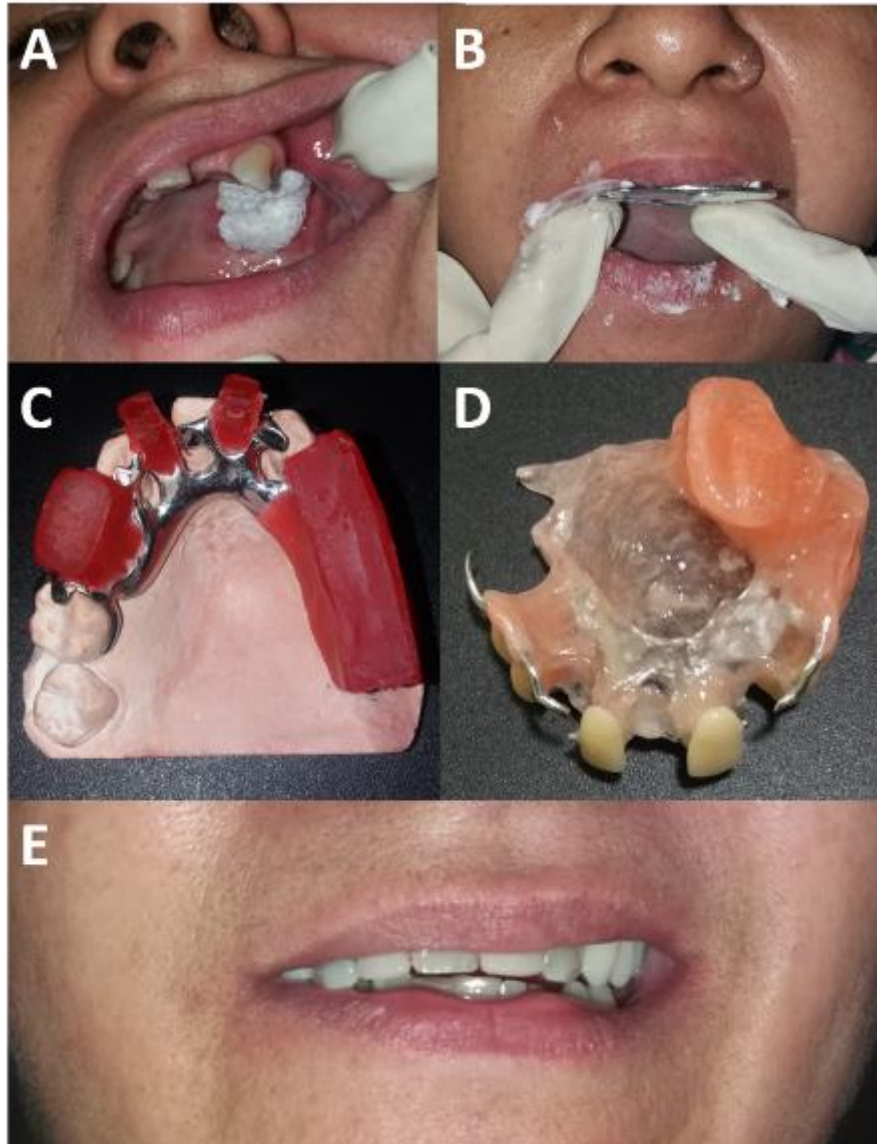
**Legend:** An opening in the palate was observed, communicating with the nasal cavity, after the en bloc removal of the region that involved by the tumor.

**Source:** Files of authors.

As rehabilitative treatment was made “in office” with an obturator partial removable prosthesis adapted to plug the region of the tumor resection on the palate (Figure 5). Using this obturator removable prosthesis, it was possible to create an hygienic and practical devise to sealing the nasal cavity, promoting adequate mastication and phonation (Annexes part 1 and 2, showing her speaking with and without the obturator prosthesis and the improvement

in phonation). A five-years follow-up was obtained without any recurrence, evaluated through biannual consultations that included dental evaluation and adjustments to the prosthesis such as relining and polishing, in addition to other restorative dental treatments.

**Figure 5.** Rehabilitation sequence with removable oblitative prosthesis of acrylic.



**Legend:** (a) – Cautious use of gauze to seal the communication before the molding procedure; (b) – Alginate molding; (c) Metal structure with staples and wax rollers for records; (d) Acrylic prosthesis, noting the nasal extension in acrylic that was created from relining with soft acrylic after the prosthesis was already made; (e) Patient with new possibility to smile, chew and talk.

**Source:** Files of authors.



### Literature review

Diagnosis should follow a systematized knowledge of diseases that may involve periapical location, therefore some "warning signs" need to be valued and open doors to new hypotheses beyond the trivial. The basic exam of tooth, evaluating the depth of cavities or restorations together the vitality tests, cares to be reinforced on general practice as protocol (with tetrafluoroethane or refrigerant gas), mainly on the presence of periapical bone lesions. The typical and atypical manifestations of inflammatory periapical lesions, as well as of other intraosseous lesions, is reviewed here for clinicians preparing for diagnostic challenges such a intraosseous MEC.

The table 1 summarized important data about cases reported in literature. The preferred location for lesions are the posterior regions of jaws, specially mandible. Some cases showed confounding factors for inflammatory periapical disease like previous endodontic treatment, pain on percussion, and the literature has not been concerned to describe results of tooth-vitality test. On radiography varies between uni and multilocular lesions; they may or may not present cortical perforation, bone expansion, root resorption or displacement. Some lesions suggested a cystic appearance, like in the present case reported, as an osteolytic unilocular lesion with well-defined and regular borders. There are also lesions with aspect of a benign tumour, showing multilocular appearance, slowly growth. And alert signs was also found, like fast growth, persistent pain, and invasive radiographic pattern (erosive osteolytic or ill-defined margins). Finally, tomography seems to reaffirm itself as the most appropriate exam to investigate details and raise suspicions of malignancy in a periapical lesion (e.g. degree of bone destruction and invasion of maxillary sinus, orbit, or even muscles through extraosseous expansion).

### DISCUSSION

Intraosseous mucoepidermoid carcinomas are mostly detected in the mandible, with higher prevalence in the posterior region and is twice as associated with women as men (BASARAN *et al.*, 2018). Suspicious symptomatology includes clinically identifiable swelling, pain, and may also manifest paresthesias, trismus and tooth mobility. Radiographic findings

show well circumscribed radiolucent areas, unilocular or multilocular, associated with an impacted tooth or cysts in 50% of cases.

The following aspects was rationally considered in the present case as evidence of the intraosseous origin of this CME: 1. the intraosseous epicenter of lesion, since it is rounded and well delimited, and shows that the medullary destruction has a larger diameter than the region of cortical rupture; 2. absence of pulpal vitality and significantly increased duration of symptoms to presume an origin from an RIC.

**Table 1** – Cases reported in literature involving CME in periapical regions, from the last five-years.

Reference	Sex*/age	Involved tooth / Symptoms	Other important findings	Imaging exams	Rehabilitation**
Basaran <i>et al.</i> , 2021	F*/ 49	46 / asymptomatic	No data of tooth-vitality test. Diagnosed by “a mass encountered” after remotion of this tooth 46.	large and multilocular from the angle, to the symphysis of mandible.	S: subtotal mandibulectomy reconstructed with osseo-septo-cutaneous free fibular flap. P: none.
ABT <i>et al.</i> , 2019	M/ 65	37 / pain on retromolar pad palpation and probing.	No data of tooth-vitality test. A minor radiolucency 2.5 years priorly without diagnosis.	retromolar ill-defined radiolucency in a vital tooth, mimicking periodontal pocket.	S: marginal mandibulectomy reconstructed with titanium plate and iliac crest bone graft. P: none.
LI <i>et al.</i> , 2018	F/ 33	37 / regional asymptomatic swollen.	No data of tooth-vitality test.	large and unilocular reabsorbing distal root of 37 and ascending the branch, without cortical erosion.	S: partial mandibular resection with placement of a microvascular fibula myocutaneous flap. P: none.
HARADA <i>et al.</i> , 2021	M/ 66	27 / pain for 4 years without other details. Pain on percussion.	No data of tooth-vitality test. Teeth 26 and 27 received endodontics and were not mobile.	Multilocular with multiple cortical erosions surrounding roots of 27 with partial resorption of the distal root.	S: partial osteotomy and palatoplasty with 10 mm margin. P: A dental-maxillary prosthesis was fabricated and placed.
HERNÁNDEZ-ARENAS <i>et al.</i> , 2019	M/ 13	Lower posterior left teeth / 2 years of asymptomatic enlargement in left mandible	No data of tooth-vitality test. Citric fluid obtained on puncture.	Large and multilocular expansive without cortical perforation, in the area of body and ramus near to molars roots without its reabsorption.	S: partial mandibular resection reconstructed titanium plate. P: none.
KHATANIAR <i>et al.</i> , 2022	F/ 51	Lower anterior teeth / Rapidly non-tender swelling for one month.	No data of tooth-vitality test. History of breast carcinoma.	large and multilocular without cortical perforation, in the chin region, covering the midline, covering apices of the anterior roots without resorption.	S: segmental resection reconstructed with titanium plate and osteomyocutaneous free fibular flap. P: none.
DOMINGUEZ-MEDINA <i>et al.</i> , 2021	M/ 54	36 / occasional pain for one year. Pain on percussion.	Tooth with endodontic treatment. Non-fluctuant, vestibular bulge in the gums adjacent to tooth 36.	Unilocular periapical well-defined radiolucency of approximately 15 mm	S: marginal mandibular body resection reconstructed with titanium plate. P: none.
MARUYAMA <i>et al.</i> , 2021	M/ 67	47 / asymptomatic	History of a cystic lesion related to tooth 48 without histopathological diagnosis 11 years earlier	Ill-defined radiolucency involving the region of 4, touching the distal part of tooth 37 and reabsorbing the lingual cortex.	S: partial mandibular resection. P: none.

\* M: male; F: female. \*\* P: prosthetic; S: surgical. # Of the three cases reported, this one involved a periradicular context, the others are apparently in edentulous patients or areas. The reference 35 were excluder from table because tumor involved only soft tissue, mimicking a sinus tract, but without relation with root apex of tooth

Source: BRAZÃO-SILVA *et al.*, 2024.

However, the histopathological aspect showed areas with minor salivary glands in close contact with tumor proliferation. The theory of its origin from the neoplastic transformation of the epithelial lining of odontogenic cysts is supported by data showing about 50-70% of intraosseous MEC associated with dental cysts, and classic authors writing that it may not be quite rare (MERNA *et al.*, 2018; WALDRON and MUSTOE, 1989). It is presumed to be arising from transformation of the mucus-producing cells commonly found in epithelial linings of these cysts. One should also take into account the fact that the posterior region of the mandible is the most frequent site for both odontogenic cysts and intraosseous mucoepidermoid carcinomas (SHERIN *et al.*, 2011; ZHOU *et al.*, 2012). On the other hand, neoplastic transformation and invasion from the lining of the maxillary sinus and neoplastic transformation of entrapped minor salivary glands within the maxilla is also considered (HERNÁNDEZ-ARENAS *et al.*, 2019).

Despite its epidemiology and characteristics of periapical MEC may not yet be well established, the review of cases showed a preference to the involvement of the posterior maxilla and mandible, and signs such extraosseous manifestation or clinically visible swelling in the absence of symptoms or with symptoms like paresthesia, tooth mobility, must be a kind or “red flags” for diseases mimicking tooth inflammatory diseases, requiring biopsy as the best procedure. Biopsy is mandatory for these cases. The initial cases without the lesion rupture of cortical bone makes difficult to deal with biopsies, making mandatory the need of opening a mucosal flap followed by a bone window with a dental surgical drills to collect the sample for histopathological diagnosis (BASARAN *et al.*, 2018; SHERIN *et al.*, 2011; LI *et al.*, 2018). The discussion from this point onwards included some other lesions not-rarely reported as periapical radiolucency, facing the present reported case. These lesions take in the differential diagnosis includes the RIC, ameloblastoma, central giant cell lesions, lateral periodontal cyst, odontogenic keratocyst.

The majority of periapical MEC cases were of low histological grade; thus, the data contribute to what was found by Merna and collaborators (2018). The histological aspects of MEC includes the mucinous, intermediate, and epidermoid cells found in variable proportions and the predominance of cystic spaces of varying sizes, lined with mucus-secreting cells and intermediate-type cells determines the low-grade of MEC (TAKANO *et al.*,

2012; LI *et al.*, 2012; MERNA *et al.*, 2018; BASARAN *et al.*, 2018; ABT *et al.*, 2019). The histologic differential diagnosis of MEC involves necrotizing sialometaplasia, pleomorphic adenoma, cystadenoma, squamous cell carcinoma, and clear cell tumors. Common features of this differential diagnosis are strands, sheets, and clusters of mucous, squamous, intermediate, and clear cells seen in mucoepidermoid carcinoma, and the immunohistochemical profile of keratins is helpful in differential diagnosis (All MECs expressed CKs 7, 8, and 18, but few expressed Cks 10/13 and 14) (LI *et al.*, 2012). Another dilemma of histological diagnosis is referred to Glandular Odontogenic Cyst (GOC). The expression of Ck7, 8 and 18, as well as mucicarmine favours the diagnosis of MEC, whereas expression of Ck19 and Ck13 favours GOC diagnosis (KAUR *et al.*, 2021). The rearrangement of the Mastermind like (MAML)-2 gene is reported to reveal the diagnosis of MEC, but recent findings suggested that both lesions may share this alteration representing sometimes different spectrum of the same disease (MARUYAMA *et al.*, 2021). Because of the similarity between intraosseous MEC and some of these mentioned benign diseases, the preoperative diagnosis becomes challenging, thus the association of biopsy results with additional microscopic and biomolecular findings must be critical, and clinical-imaging correlation is essentially requested.

The discussion from this point onwards included some other lesions not-rarely reported as a periapical radiolucency, facing the present reported case, with the aim of showing important aspects of clinical assessment and formulation of diagnostic hypotheses. These main lesions to be taken into the differential diagnosis includes the RIC, ameloblastoma, central giant cell lesions, lateral periodontal cyst, odontogenic keratocyst.

### **Radicular Inflammatory Cyst (RIC)**

The palatal swelling seen in the present case does not completely rule out the possibility of an inflammatory root cyst, since it can also rupture the cortical bone, although this is uncommon. In RIC this swelling is usually hard due to the expansion of the palatine bone, but if there is cortical perforation, it may also be soft on palpation. A bluish coloration in the palatal swelling is suspect for MEC, but may also appear in RIC (NABIL *et al.*, 2013; DESHMUKH *et al.*, 2014; NAKHAE *et al.*, 2016). The RIC occurs mainly in the maxillary region (60% of cases) involving the apex or a lateral side of the tooth root of the necrotic tooth if

there are accessory root canals, preferentially in males and between the third and sixth decades of life (NABIL *et al.*, 2013; DESHMUKH *et al.*, 2014). They are mostly asymptomatic, but can occur in the presence of pain and sensitivity to percussion, cause mobility, root resorption of the involved element and tooth displacement (NABIL *et al.*, 2013; DESHMUKH *et al.*, 2014).

Surgical removal of a lesion clinically and radiographically diagnoses as RIC is indicated in the following situations: inaccessibility to the apical third during endodontic treatment, periapical lesions that show no signs of regression on follow-up imaging examinations after therapy, lesions larger than 2cm, histopathological diagnosis in lesions whose origin is inconclusive for RIC (PAVASKAR *et al.*, 2013; COSTA *et al.*, 2020). Microscopic examination is almost always conclusive in pointing to a chronic inflammatory lesion with a pathological cavity lined partially or completely by non-keratinized stratified squamous epithelium (NABIL *et al.*, 2013; DESHMUKH *et al.*, 2014; NAKHAEI *et al.*, 2016).

### **Ameloblastoma**

If the periapical lesion is multilocular, suspicion must inevitably consider ameloblastoma as a differential diagnosis, configuring radiolucent images classically called "soap bubbles" or "honeycomb". However, in a large number of cases of unicystic ameloblastomas and in some solid lesions the characteristic aspect of well-defined unilocular radiolucent lesion is present, so that its development in the periapical region can simulate periapicopathies (AMORIM and FREITAS, 2003; BASSEY *et al.*, 2014). The literature points out ameloblastoma as one of the lesions that should enter into a differential diagnosis of periapical lesions in vitalized teeth or in teeth without vitality, but that do not regress to therapy. This benign tumor can also originate from the epithelial rests of Malassez located in the apical periodontal region. It requires surgical intervention that may include margin enlargement due to its aggressive and recurrent behavior, and it is in this sense that early diagnosis of smaller lesions may favour a lower therapeutic morbidity (SCIUBBA *et al.*, 2005; ODUKOYA and EFFIOM, 2008; DEVILLIERS *et al.*, 2011).



### **Central Giant Cell Lesion (CGCL)**

CGCL is defined as an intraosseous lesion consisting of fibrous cellular tissue that contains multiple foci of haemorrhage, aggregations of multinucleated giant cells, and occasionally trabeculae of bone tissue. It is a mildly aggressive, asymptomatic, slow-growing lesion that does not cause root resorption and has a low recurrence rate. It is highly prevalent in children and young adults and is more frequently found in the anterior region of the mandible. It is generally classified as a circumscribed, non-cortical, multilocular, radiolucent lesion. However, occasionally it can present as a well circumscribed, radiolucent, cortical lesion that can be found near the tooth apex, resembling apical periodontitis (CANDEIRO *et al.*, 2020).

### **Lateral Periodontal Cyst (LPC)**

LPC presents on radiographic appearance as a well-circumscribed, round or teardrop-shaped radiolucency with an opaque margin along the lateral surface of the tooth root, usually located between the roots of vital mandibular canines and premolars. On clinical examination CPL manifests as a small swollen soft tissue volume increase within or slightly inferior to the interdental papilla. Such disease is usually asymptomatic and is often discovered during routine radiographic examination. However, because of its location, it can easily be diagnosed as a lesion of endodontic origin (KEREZLOUDIS *et al.*, 2000).

### **Odontogenic Keratocyst (OK)**

Santos and collaborators (2013) reports having found 27 cases in English literature where keratocysts involved the periapical region and slight predominance in the maxilla, mainly in the anterior region (SANTOS *et al.*, 2013). Although commonly asymptomatic, their growth can lead to cortical bone expansion, displacement, and tooth resorption. Radiographically, they represent unilocular lesions with sclerotic edges in most, however the multilocular aspect also characterizes it, being or not associated with impacted teeth. Given the characteristics of periapical involvement, radiolucency and expansive growth of the

keratocyst, it should be considered among the diagnostic hypotheses raised in this case report (LUNAWAT *et al.*, 2020).

### Final considerations

Despite being considered a neoplasm with low malignancy potential, about 10% of patients affected by MEC die by tumor related causes. The MECs located in the submandibular gland and those with high histopathological grade are considered the most aggressive as a whole. For intraosseous MEC, it is also noteworthy that the intracystic pattern of proliferations correlates with better prognosis, being very useful in the clinical outcome (CAPODIFERRO *et al.*, 2020). Some clinic and radiographic aspects have been mentioned in another classification system for intraosseous jaws tumors: lesions with intact cortical bone and no bone expansion are classified as stage 1 and have a better prognosis; lesions that expand the bone but do not disrupt the cortex are stage 2; lesions that disrupt the integrity of the periosteum or cause cortical perforation with or without nodal involvement and masses with nodal involvement are stage 3, with the worse prognosis (BROOKSTONE and HUVOS, 1992). This is controversial in the present case, which perforated cortical bone and would be interpreted as a stage 3, but the histological aspect showed a low-grade pattern and the outcome was favourable.

The therapeutic approach is very much guided by the level of cell differentiation of the lesion, history of relapse, and signs such as perineural or lymphovascular involvement (MERNA *et al.*, 2018). In general, intraosseous mucoepidermoid carcinoma is treated by bloc surgical resection with safety margins, even if they are low-grade tumors. If present lymphovascular involvement, chemo and/or radiotherapy can be used as adjuvant treatment. Cases treated conservatively, such as curettage or enucleation, may recur in 40% - 45%, which is why these procedures are not recommended, while the recurrence rate in cases treated by bloc resection with clear margins drops to 13% (BASARAN *et al.*, 2018).

In the present paper was also shown the forms of rehabilitation obtained in the case reports reviewed (table 1). Many of them were included advanced surgical modalities of anatomic reconstruction. However, the simplest procedures like a removable prosthesis, could be sufficient to some patients and accessible to more dental professionals, promoting

a way of rehabilitation with low cost and able to promote a reasonable quality of masticatory function and phonation, like in the present case (additional file).

## CONCLUSION

Oral lesions may take different forms or be similar in appearance, even though they are of completely different etiologies. It is worth emphasizing the importance of the dental surgeon in early diagnosis, seeking to know all the possible causes of the lesion and adopting the best conduct for the correct diagnosis. In the present report, the absence of vitality in tooth 27 associated with the CT scan only, could have led to endodontic treatment and prosthodontic preservation of the case, without being aware of the severity of the lesion, which in turn would increase morbidity for the patient. MECs mimicking periapical disease is uncommon but it is found to be a real possibility in a dental office.

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