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# Mucoepidermoid carcinoma of the retromolar region: case report

## *Carcinoma mucoepidermoide em região retromolar: relato de caso*

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## ABSTRACT

The present study aimed to report a case of mucoepidermoid carcinoma (MEC), focusing on its clinical-pathological characteristics. At intraoral clinical examination, a nodular lesion was observed in the right pterygomandibular raphe region, with three years of evolution. An incisional biopsy was performed, and the diagnostic hypotheses of salivary gland injury and sialadenitis were considered. Histopathologically, a malignant neoplastic process characterized by the proliferation of epidermoid, intermediate and mucosal cells was observed. The histopathological diagnosis of MEC was emitted. The present case praises the importance of early diagnosis and correct management of this disease, providing a better prognosis for these patients.

**Key words:** diagnosis; minor salivary glands; mucoepidermoid carcinoma.

## RESUMO

*Relatamos um caso de carcinoma mucoepidermoide (CME) com enfoque em suas características clinicopatológicas. No exame clínico intraoral, observou-se lesão de aspecto nodular em região de rafe pterigomandibular direita, com tempo de evolução de três anos. Biópsia incisional foi realizada, e as hipóteses diagnósticas de lesão de glândula salivar e sialadenite foram consideradas. Histopatologicamente, observou-se um processo neoplásico maligno caracterizado pela proliferação de células epidermoides, intermediárias e mucosas. O diagnóstico histopatológico de CME foi emitido. O presente caso enaltece a importância do diagnóstico precoce e do correto manejo dessa patologia, proporcionado um melhor prognóstico para os pacientes portadores de CME.*

***Unitermos:*** *diagnóstico; glândulas salivares menores; carcinoma mucoepidermoide.*

## RESUMEN

*Reportamos un caso de carcinoma mucoepidermoide (CME) con enfoque en sus rasgos clinicopatológicos. En la exploración clínica intraoral, se observó una lesión de aspecto nodular en región del rafe pterigomandibular derecho, con tiempo de evolución de tres años. Se realizó una biopsia por incisión, considerándose las hipótesis diagnósticas de lesión de glándula salival y sialadenitis. Histopatológicamente, se observó un proceso neoplásico maligno caracterizado por la proliferación de células epidermoides, intermedias y mucosas. El diagnóstico histopatológico fue de CME. El presente caso destaca la importancia del diagnóstico temprano y del manejo correcto de esa enfermedad, ofreciendo un mejor pronóstico para los pacientes portadores de CME.*

***Palabras clave:*** *diagnóstico; glándulas salivales menores; carcinoma mucoepidermoide.*

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## INTRODUCTION

Malignant neoplasms of the salivary glands encompass 3%-5% of all malignant tumors of the head and neck region; they may affect major or minor salivary glands[1, 2].

The mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy[3, 4]. Around 50% of the cases occur in major salivary glands: 80% in the parotid, 8%-23% in the submandibular, and 2%-4% in the sublingual. When considering intraoral involvement, the most frequently affected sites are palate and oral mucosa. Patients' mean age is approximately 45 years, with a slight predilection for women, in a 3:2 female/male ratio. Nevertheless, this women predominance is more pronounced, mainly, in tumors of the tongue and retromolar area[4, 5].

The clinical behavior of MEC is widely variable, ranging from indolent tumor growth to highly aggressive metastatic dissemination[6]. It is usually manifested as a painless mass, variably fixed, rubbery or soft in consistency. Owing to its location, normally superficial, an intraoral tumor can appear as a swelling of bluish-red color, mimicking a mucocele or a vascular tumor[2, 7].

At histopathology, MEC is a malignant neoplasm of the glandular epithelium, characterized by mucous, intermediate and epidermoid cells, with columnar aspect, and the possible presence of clear and oncocytoid cells[1, 2]. The tumors present solid, cystic or microcystic growth and, although not encapsulated and asymmetrical, they are often lobulated and somewhat circumscribed. Patterns such as irregular, stellate or pointed invasive silhouettes are less common. MECs arising in the minor salivary glands are located in the submucosa, where they are partially surrounded by salivary lobules; they can involve main ducts, and, in different ways, extend to the lamina propria. Perineural invasion can be demonstrated; however, necrotic areas are not common[7].

MEC can be classified as: low grade, intermediate grade, or high grade, according to its cytological features, invasion pattern, and cellular type[8, 9]. This grading is based on a set of characteristics, including necrosis, nuclear atypia, and size of the cystic component[10].

Treatment depends on location, clinical aspects, and histopathological grade[9]. The standard treatment for the main cancer types of the salivary gland is surgical resection combined with adjuvant therapy, in order to reduce failure rates. Although the role of adjuvant chemotherapy has not been confirmed, it has been used both to treat distant metastases and non-excisable disease and to reduce possible local/regional recurrences[5]. Prognosis will considerably depend on the clinical stage of the lesion, its anatomic location, its histopathologic grade, and the adopted treatment.

The present paper is aimed at reporting a case of MEC focusing on its clinical and histopathological characteristics.

## CASE REPORT

A female patient, aged 33 years, white, presented at the Stomatology Clinic of the Department of Dentistry of Universidade Federal do Rio Grande do Norte (UFRN), Natal, Brazil, reporting a painful lesion in the mouth, with duration of around three years. At intraoral clinical examination, a lesion of nodular aspect was observed in the right pterygomandibular raphe region. It was similar to the mucosa in color, had firm consistency, sessile implantation, and slow growth, measuring approximately 1 cm (**Figure 1**).

A panoramic radiograph was ordered to evaluate the possible lesion-associated bone loss; however, radiographic changes were not identified (**Figure 2**). Preoperative tests were also requested for the conduction of the excisional biopsy of the lesion. The diagnostic hypotheses were salivary gland lesion and sialadenitis.

FIGURE 1 – *Nodular lesion in the right pterygomandibular raphe region*

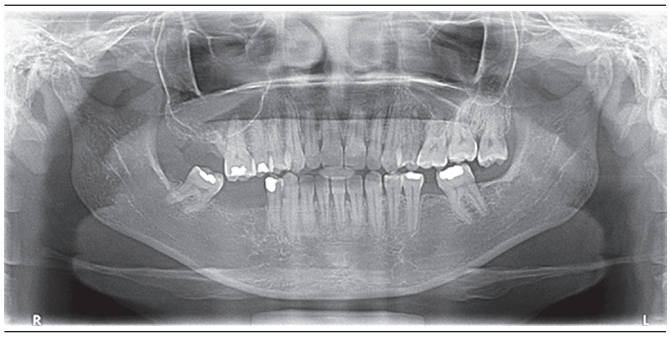

FIGURE 2 – *Panoramic radiograph with absence of radiographic changes in the region of the lesion*

At histopathologic examination, a lesion characterized by proliferation of epidermoid, intermediate and mucous cells was observed, surrounded by a stroma of fibrous connective tissue of varying density. Those cells were arranged in some regions, sometimes in a solid pattern, sometimes forming cystic structures of variable size, with the presence of an amorphous eosinophilic material compatible with mucus inside them, besides the formation of mucin pools. Cellular atypia was not seen; just some neoplastic cells with slight pleomorphism and hyperchromatism (**Figure 3**).

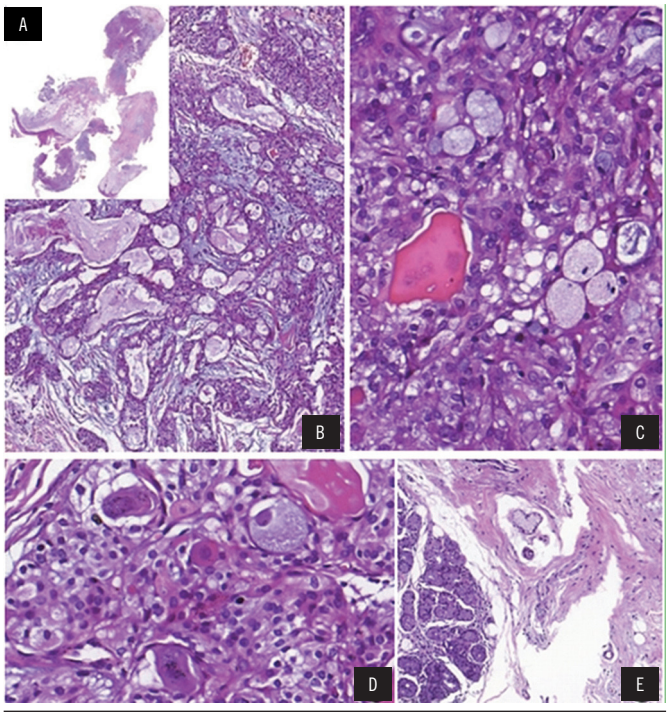


FIGURE 3 – *A) low-magnification photomicrograph revealing fragments of the neoplastic process richly cellularized and non-delimited; B) cystic and luminal spaces filled with eosinophilic material and solid areas; C) detail of morphology: mucous and epidermoid cells; D) high-magnification photomicrograph revealing detail of mucous, epidermoid and intermediate cells; E) glandular parenchyma completes the examined picture*

The histopathological diagnosis of CME was confirmed by microscopic analysis. The patient was referred to a reference hospital in the region to receive adequate treatment; she went through an extensive surgical excision of the neoplastic process. She has been under follow-up care, presenting no clinical or radiographic sign of recurrence 12 months after surgery.

## DISCUSSION

The oral cavity can be affected by a variety of lesions of different clinical and pathologic characteristics and biological behavior[11, 12]. This paper reports a case of MEC in a female patient, yet, so far, no consensus exists about gender predilection of this neoplasia. Some authors believe there is no preference, while others state that MEC is more prevalent among women (60.2%)[5, 12].

The benign clinical appearance of MEC often leads to a diagnosis of pleomorphic adenoma or mucous retention cyst, hemangioma, pigmented nevus and cystic processes[13, 14]. The clinical diagnosis proved imprecise in our report, making the conduction of an incisional biopsy fundamental for analysis and definite diagnosis of the case.

Our patient presented painful symptoms, although neither lymphadenopathy nor bone changes were observed in the radiographic examination, what made clinical diagnosis difficult. The absence of symptoms can delay diagnosis, making treatment less effective[14]. Besides, pain is not always present in the cases of MEC. At the same time, there are reports of symptomatic cases associated with lymphadenopathy, ulceration, or bone involvement[13, 14].

The MEC in this case was in a site with low degree of involvement by the neoplastic process. Major salivary glands are the most commonly affected, often represented by the parotid. Kolude *et al*. (2001)[15] analyzed 34 patients with MEC and verified that just 25% of the lesions affected minor salivary glands, most of them in the palate. Other oral affected areas in descending order are: buccal mucosa, alveolar mucosa, tongue, retromolar region, floor of the mouth, and lips.

Moreira *et al*. (2009)[16] observed that the parotid gland is the most frequent site of malignant lesions, followed by submandibular glands and minor salivary glands distributed throughout the oral cavity. In contrast, Ledesma-Montes and Graces-Ortiz (2002)[17] reported that the palate was the most common location.

Isolated tumors in the retromolar trigone are unusual. In general, most neoplastic processes diagnosed in that anatomic

site are squamous cell carcinomas, with rare exceptions. Lesions often extend to tonsils, anterior pillar, and soft palate[11, 14].

In our study, the excisional biopsy specimen was sent for anatomopathological analysis. This procedure promotes prompt diagnosis, enhances prognosis, and increases the chances of a successful treatment[12, 13]. MEC, as well as any other lesion of the maxillomandibular complex, must be diagnosed as soon as possible, because this improves outcomes. Therefore, the diagnosis of a malignant lesion in the initial stage favors treatment[5].

Lesion stage at the moment of diagnosis is important because the most advanced cases demand more complex treatments and have poor prognosis. Since the oral cavity permits easy access and inspection, late diagnoses cannot be justified[11]. The dental surgeon capacity for a careful examination of the stomatognathic system is fundamental to establish early diagnosis and avoid oral lesions, demanding, as a consequence, less invasive surgeries that result in better quality of life for the patient[5, 4].

Given the presented case, we highlight how important it is for dental surgeons to understand the clinical pathological features of MEC aiming at early detection and adequate treatment. This disease must be considered a diagnostic hypothesis in oral lesions of proliferative aspect, even when its clinical appearance does not suggest malignancy; the conduction of an incisional biopsy is essential for correct diagnosis and subsequent management of the patient.

## REFERENCES


1. Byrd SA, Spector ME, Carey TE, Bradford CR, McHugh JB. Predictors of recurrence and survival for head and neck mucoepidermoid carcinoma. Otolaryngol Head Neck Surg. 2013; 149(3): 402-8. PubMed PMID: 23695589.

2. Ellis MA, Graboyes EM, Day TA, Neskey DM. Prognostic factors and occult nodal disease in mucoepidermoid carcinoma of the oral cavity and oropharynx: an analysis of the National Cancer Database. Oral Oncol. 2017; 72: 174-8. PubMed PMID: 28797455.

3. Granic M, Suton P, Mueller D, Cvrljevic I, Luksic I. Prognostic factors in head and neck mucoepidermoid carcinoma: experience at a single institution based on 64 consecutive patients over a 28-year period. Int J Oral Maxillofac Surg. 2018; 47(3): 283-8. PubMed PMID: 28969884.

4. Barnes L, Eveson JW, Sidransky D, Reichart P. World Health Organization classification of tumors: pathology and genetics of head and neck tumors. IARC Press: Lyon; 2005.

5. Janet-Ofelia GC, Rafael MV, Guillermo GA, et al. Mucoepidermoid carcinoma of the salivary glands: survival and prognostic factors. J Maxillofac Oral Surg. 2017;16(4): 431-437. PubMed PMID: 29038625.

6. Brandwein MS, Ivanov K, Wallace DI, et al. Mucoepidermoid carcinoma: a clinicopathologic study of 80 patients with special reference to histological grading. Am J Surg Pathol. 2001; 25(7): 835-45. PubMed PMID: 11420454.

7. Coca-Pelaz A, Rodrigo JP, Triantafyllou A, et al. Salivary mucoepidermoid carcinoma revisited. Eur Arch Otorhinolaryngol. 2015; 272(4): 799-819. PubMed PMID: 24771140.

8. Luna MA. Salivary mucoepidermoid carcinoma: revisited. Adv Anat Pathol. 2006; 13(6): 293-307. PubMed PMID: 17075295.

9. McHugh CH, Roberts DB, El-Naggar AK, et al. Prognostic factors in mucoepidermoid carcinoma of the salivary glands. Cancer. 2012; 118(16): 3928-36. PubMed PMID: 22180391.

10. Seethala RR. An update on grading of salivary gland carcinomas. Head Neck Pathol. 2009; 3(1): 69-77. PubMed PMID: 20596994.

11. Goel AN, Badran KW, Braun APG, Garrett AM, Long JL. Minor salivary gland carcinoma of the oropharynx: a population-based analysis of 1426 patients. Otolaryngol Head Neck Surg. 2018; 158(2): 287-94. PubMed PMID: 28971745.

12. Honjo K, Hiraki T, Higashi M, et al. Immunohistochemical expression profiles of mucin antigens in salivary gland mucoepidermoid carcinoma: MUC4- and MUC6-negative expression predicts a shortened survival in the early postoperative phase. Histol Histopathol. 2018; 33(2): 201-13. PubMed PMID: 28649694.

13. Vander Poorten V, Hunt J, Bradley PJ, et al. Recent trends in the management of minor salivary gland carcinoma. Head Neck. 2014; 36: 444-55. PubMed PMID: 23559518.

14. Badlani J, Gupta R, Balasubramanian D, Smith J, Luk P, Clark J. Primary salivary gland malignancies: a review of clinicopathological evolution, molecular mechanisms and management. ANZ J Surg. 2018; 88(3): 152-7. PubMed PMID: 28982213.

15. Kolude B, Lawoyin JO, Akang EE. Mucoepidermoid carcinoma of the oral cavity. J Nat Med Assoc. 2001; 93: 178-84. PubMed PMID: 11405595.

16. Moreira ARO, Oliveira CDM, Figueirêdo EP, Silva RR, Lopes FF, Bastos EG. Levantamento epidemiológico das enfermidades das glândulas salivares em São Luís-MA: casuística de vinte anos. RFO. 2009; 14(2): 105-10. Available at: http://files.bvs.br/upload/S/1413-4012/2009/v14n2/a105-110.pdf.

17. Ledesma-Montes C, Garces-Ortiz M. Salivary gland tumours in a Mexican sample: a retrospective study. Med Oral. 2002; 7: 324-30. PubMed PMID: 12415215.



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