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Necrotizing Sialometaplasia and the importance of considering it as a differential diagnosis of intraoral neoplasms

Sialometaplasia Necrotizante y la importancia de considerarla como un diagnóstico diferencial de neoplasias intraorales

Sialometaplasia Necrosante e a importância de considerá-la como diagnóstico diferencial de neoplasias intraorais

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Abstract

Necrotizing sialometaplasia is a benign and self-limiting pathological entity that occurs in minor salivary glands, most frequently located on the border between the hard and soft palate. It has an unclear etiology, being associated with physical, chemical or biological injury, which would produce necrosis due to ischemia of the glandular tissue and a subsequent attempt to repair, which histologically translates into tissue metaplasia. Clinically, it can present as an irregular ulcer, with raised edges and a necrotic base, giving it an appearance that is easily confused with a malignant oral neoplasm. Although they have a low prevalence, their knowledge can avoid unnecessary and mutilating clinical maneuvers. We present the case of an adult, who had a family history of malignant neoplasm of the head and neck, and his clinical evolution. Relevant aspects are discussed. Ulcerated lesions with a crater-like appearance, located on the palate, should evoke this lesion as part of their diagnostic hypotheses, their knowledge can avoid carrying out mutilating and unnecessary treatments.

Keywords: necrotizing sialometaplasia, minor salivary glands; mouth neoplasms.

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Resumen

La sialometaplasia necrotizante es una entidad patológica benigna y autolimitante que se presenta en glándulas salivales menores, ubicada con mayor frecuencia en el límite entre el paladar duro y blando. Presenta una etiología poco clara, asociándose a trauma físico, químico o biológico, que produciría necrosis por isquemia del tejido glandular y un posterior intento de reparación, que histológicamente se traduce en una metaplasia del tejido. Clínicamente puede presentarse como una úlcera irregular, de bordes sollevados y fondo necrótico, confiriéndole un aspecto fácilmente confundible con una neoplasia maligna oral. Si bien presentan una baja prevalencia, su conocimiento puede evitar maniobras clínicas innecesarias y mutilantes. Se presenta el caso de un adulto, quien contaba con antecedentes familiares de neoplasia maligna de cabeza y cuello, y su evolución clínica. Lesiones ulceradas de aspecto crateriforme, ubicadas en el paladar, deberían evocar a esta lesión como parte de sus hipótesis diagnósticas, su conocimiento puede evitar llevar a cabo tratamientos mutilantes e innecesarios.

Palabras clave: sialometaplasia necrotizante, glándulas salivales menores, neoplasias de la boca.

Resumo

A sialometaplasia necrosante é uma entidade patológica benigna e autolimitada que ocorre nas glândulas salivares menores, mais frequentemente localizadas na margem do palato duro e mole. Tem etiologia incerta, estando associada a uma lesão física, química ou biológica, que produziria necrose por isquemia do tecido glandular e posteriormente uma tentativa de reparação, que se traduz histologicamente em metaplasia tecidual. Clinicamente, pode apresentar-se como uma úlcera irregular, com bordas elevadas e base necrótica, o que lhe confere um aspecto facilmente confuso com uma neoplasia oral maligna. Embora tenham baixa prevalência, seu conhecimento pode evitar intervenções clínicas desnecessárias. Apresentamos o caso de um adulto, com histórico familiar de neoplasia maligna de cabeça e pescoço, e sua evolução clínica. Aspectos relevantes são discutidos. Lesões ulceradas com aspecto de cratera, localizadas no palato, devem evocar essa lesão como parte de suas hipóteses diagnósticas, seu conhecimento pode evitar a realização de tratamentos mutilantes e desnecessários.

Palavras-chave: sialometaplasia necrosante, glândulas salivares menores; neoplasias bucais.

Introduction

Necrotizing sialometaplasia is a benign and self-limiting reactive process that occurs most frequently in minor salivary glands.⁽¹⁾ Its etiology is unclear. It has been suggested that a physical, chemical, or biological trauma could cause ischemia and subsequent glandular tissue necrosis. Subsequently, the attempted repair would result in tissue metaplasia, causing a necrotic ulcer of poor appearance.⁽²⁾

This lesion is uncommon, with a prevalence of less than 1%.⁽¹⁾ However, it is important because its clinical and histopathological appearance can be easily confused with oral malignant neoplasms such as mucoepidermoid carcinoma or squamous cell carcinoma. Therefore, it is essential to consider its existence to give the lesion some time and avoid unnecessary clinical maneuvers and alarming the patient.⁽¹⁻³⁾ The following is a case of necrotizing sialometapla-

sia. A literature review is conducted, and clinical aspects are discussed.

Background

A 48-year-old male patient sought care at the dental emergency unit for hemimaxillary pain lasting one week, which increased when chewing. He also said that he had a bad taste in his mouth. The virtual medical history included arterial hypertension and type II diabetes mellitus—both under treatment—smoking five cigarettes/day and occasional social drinking. His family history included the death of his mother due to maxillofacial malignant neoplasm, without specifying the diagnosis.

Figure 1: There is a dirty-base and crater-shaped ulcer with defined borders of 10x10 mm in diameter on the palatine mucosa in relation to the left maxillary molars. Roots of teeth 2.6 and 2.7.



The intraoral clinical examination showed only the roots of teeth 2.6 and 2.7 and palatal volume increase. (Figure 1) There was a crater-shaped ulcer with a dirty base and defined borders measuring 10x10 mm in diameter. Root debris was removed, and 0.12% chlorhexidine mouthwash was administered twice daily for 7 days to improve local conditions and reduce inflammation. A follow-up appointment was scheduled one week later to assess the need for a biopsy and histopathological exam.

Figure 2: Checkup after one week. Less inflammation, homogeneous base with scar tissue.



In the checkup, the patient reported fewer symptoms and a clear regression of the ulcer (Figure 2), fibrin in the base, and decreased inflammation. We decided not to perform a biopsy because necrotizing sialometaplasia was posed as a hypothetical diagnosis. The patient attended checkups in the following four weeks, and after one month, complete tissue repair was observed (Figure 3).

Figure 3: Checkup after four weeks. Healthy tissues and total ulcer remission are observed.



Discussion

Necrotizing sialometaplasia is an entity of unclear pathophysiology. It is suggested that it could be produced by ischemia of minor salivary glands caused by several conditions: direct trauma, drinking, cocaine use, radiotherapy, local anesthesia with vasoconstrictors, surgical procedures, smoking, and bulimia. It has also been associated with chronic inflammation.^(4,5)

In this case, the patient reported smoking and occasional drinking and also presented a chronic infectious focus in the neighboring teeth, which might account for the etiology of this case. Drug use cannot be ruled out even if the patient does not report it.

This condition occurs mainly in men in their fifties, although it can appear at any time. It initially appears as a non-ulcerated enlargement, usually associated with pain and paresthesia. The necrotic tissue detaches after two to three weeks, clinically corresponding to a well-demarcated irregular ulcer with raised edges and a necrotic base. Its size varies between 0.5 and 3 cm.⁽¹⁻⁴⁾ It is most frequently located in the palatine mucosa, at the boundary between the hard and soft palate, and unilaterally, although it may appear in other sites such as the sinus mucosa,

larynx, and floor of the mouth. In most cases, it is described as a symptomatic and painful lesion.⁽³⁻⁴⁾ In this case, the ulcer presented clinical characteristics similar to those described in the literature: unilateral lesion on the boundary between the hard and the soft palate. One aspect that might cause confusion is the associated increase in volume, as this increase was located adjacent to the ulcer, probably caused by the oral infectious foci present when seeking care.

The first diagnostic impression suggested a neoplastic lesion of the minor salivary glands. However, it is always prudent to consider necrotizing sialometaplasia as a differential diagnosis, especially when clinical appearance is inconclusive: absence of necrosis due to the presence of tumor mass, absence of great vascularity as in oral cavity neoplasms, or absence of color change, which might suggest mucoepidermoid carcinoma of the minor salivary gland. Other differential diagnoses of necrotizing sialometaplasia include squamous cell carcinoma, subacute necrotizing sialoadenitis, lymphomas, histioplasmosis, and syphilis.^(4,5)

Some cases present with bulky lesions detected with imaging tests such as MRI and CT.⁽⁴⁻⁵⁾ For this reason, a biopsy is performed in most cases to confirm the diagnosis. In this case, we decided not to perform a biopsy because, after one week of follow-up, necrotizing sialometaplasia was diagnosed after the inflammation and symptoms caused by the odontogenic infection subsided. The patient was kept under observation until the ulcer completely subsided.

The histopathology of necrotizing sialometaplasia includes lobular necrosis of the salivary glands, pseudoepitheliomatous hyperplasia of the lining epithelium, and prominent squamous metaplasia of the salivary duct epithelium. In addition to histopathology, immunohistochemical tests may be helpful because they will be positive for cytokeratin-7, contrary to squamous cell carcinoma.⁽⁴⁾

As a benign and self-limited lesion, necrotizing sialometaplasia does not require special treat-

ment as it will fully heal after 3 to 12 weeks, depending on its extension. It has been suggested that topical corticosteroids and low-power lasers would shorten the recovery time. Recurrence is infrequent and has only been reported once in the literature.⁽⁷⁻⁹⁾

As mentioned above, the initial clinical aspect was relevant to raise suspicion of a malignant lesion. This led to patient follow-up and, finally, to conclude that the lesion was benign. Careful management of these lesions is essential, where clinical experience, medical history, physical examination, and a biopsy will lead to the correct treatment, as there have even been reports of mutilating treatments such as max-

illectomy with a final diagnosis of necrotizing sialometaplasia.⁽⁴⁻⁷⁾

Conclusions

Necrotizing sialometaplasia is a rare, benign, and self-limiting lesion that can mimic a malignant neoplasm and lead to radical and unnecessary treatment. A biopsy is required in most cases to rule out malignant neoplasms. Managing this lesion requires a good medical history of the case and relevant clinical expertise. Any ulcerated crater-like lesion on the palate should include this lesion as part of its diagnostic hypothesis.

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The authors have no conflict of interest regarding the publication of this paper.

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1. Conception and design of study
2. Acquisition of data
3. Data analysis
4. Discussion of results
5. Drafting of the manuscript
6. Approval of the final version of the manuscript

JdelaM has contributed in 1, 2, 3, 4, 5 y 6.

FD has contributed in 3, 4, 5 y 6.