CASE REPORT

Necrotizing sialometaplasia of the palate

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Summary Necrotizing sialometaplasia is an uncommon inflammatory condition that affects salivary glands. A 38-year-old man with bilateral ulcerative painful lesions at the junction of the hard and soft palate was presented. An incisional biopsy was performed. Histopathologically, pseudoepithelomatous hyperplasia, lobular necrosis with through the maintenance of the architecture of salivary glands and squamous metaplasia of residual acinar and ductal elements with a bland appearance were observed. The complete self-healing of the lesions occurred in 3 weeks. Since this entity presents clinical and histopathological findings resembling either mucoepidermoid carcinoma or squamous carcinoma diagnostic failure may culminate in unnecessary mutilating surgery.

KEYWORDS
Inflammatory gland diseases; Self-healing disease; Palatal ulcer; Differential diagnosis

Introduction

Necrotizing sialometaplasia (NS) is an uncommon benign lesion and locally destructive, inflammatory condition that affects salivary glands. It was first described by Abrams et al. in 1973, and is predominantly found in the posterior hard palatal mucosa, with two thirds of the cases being unilateral lesions.1 Other locations include soft palate, lip, retromolar area, tongue, mucobuccal fold, tonsillar fossa, parotid, sublingual, submandibular, nasal cavity, incisive canal, maxillary sinus, and larynx. NS is more frequent in males with average age of 45.9. Clinical features vary from swelling to crater-like lesions which may be painful or with a complete lack of symptoms. Local anesthesia, heavy smoking, alcoholic abuse, traumatic injury, surgical procedure, upper respiratory infection, associated lesions, and allergies have been pointed out as etiological agents. These factors may affect the vascular system causing an ischemia in the salivary glands that may result in necrosis.2 Self-healing occurs in 4 days to 3 months, with no need for any additional treatment besides support procedures.3 Nevertheless, it is necessary to reach a conclusive diagnosis defined by microscopic exam as the typical histopathological appearance of pseudoepithelomatous hyperplasia can become a factor of confusion in the diagnosis of this condition.1,4,5 In this report, we demonstrate a new case of necrotizing sialometaplasia, emphasizing the potential diagnostic pitfall between
squamous carcinoma and mucoepidermoid carcinoma. Thus, failure in differentiating this lesion from a malignant process on a clinical and histopathological basis may culminate in unnecessary mutilating approach.2–9

Clinical summary

A 38-year-old, white male, self-referred patient presented, at the Stomatology Clinic of Odilon Behrens Hospital (Belo Horizonte, Brazil), bilateral ulcerative lesions at the junction of the hard and soft palate, adjacent to the midline. The patient reported that the symptoms had begun 7 days prior and was complaining of pain, with swallow difficulty, nausea and loss of weight (7 kg). In addition, the patient was a heavy smoker and admitted to alcohol consumption. The medical history and extra oral exam were not relevant. History of trauma or infection was not reported.

Intra-oral examination revealed two crater-like ulcers with irregular, raised, and soft borders, erythematous margins and necrotic center, with draining of inflammatory exudates, measuring: right 15 mm and left 20 mm in diameter (Fig. 1(A)).

The main diagnosis proposed was necrotizing sialometaplasia while differential diagnosis included mucoepidermoid carcinoma, squamous cell carcinoma, secondary syphilis or other ulcerated neoplasm of the minor salivary glands.

Routine hematological values were within the normal range and Venereal Disease Research Laboratory (VDRL) was not reactive. Thus, an incisional biopsy of the left-side lesion was performed. The specimen was sent to the Oral Pathology Laboratory of Federal University of Minas Gerais (Belo Horizonte, MG), and a diagnosis of necrotizing sialometaplasia was performed. The patient went through full followed up until the complete self-healing of the lesions

Figure 1  Clinical findings: (A) Two crater-like ulcers with irregular, raised, and soft borders, erythematous margins and necrotic center, with draining of inflammatory exudate, measuring right 15 mm and left 20 mm in diameter were observed. (B) The complete self-healing of the lesions, in approximately 3 weeks. Histopathological findings: (C) Maintenance of the architecture of salivary glands (H&E, X200). (D) Necrotic lobules are represented by small acinar-sized pools of mucin surrounded by thin fibrous septa ringed by a dense and diffuse inflammatory infiltration composed of neutrophils, lymphocytes, plasma cells and a great amount of eosinophils, in detail (H&E, X200). (E) Squamous metaplasia of residual acinar and ductal elements. Some nests show evidence of residual lumina (H&E, X200). (F) The metaplasic squamous islands and nests revealed a bland appearance, varying sizes and shapes, smooth and regular borders (H&E, X200).
occurred, which took approximately 3 weeks, as expected (Fig. 1(B)).

Pathological findings

Hematoxylin and eosin-stained paraffin sections revealed a mucosal fragment covered with thickened, acanthotic stratified squamous epithelium with parakeratosis. Areas of pseudoepithelialomatous hyperplasia and an area of ulceration could be visualized. Subjacent to the ulcerated area, lobular necrosis was noted with through the maintenance of the architecture of salivary glands (Fig. 1(C)). Necrotic lobules are represented by small acinar-sized pools of mucin surrounded by thin fibrous septa ringed by a dense and diffuse inflammatory infiltration composed of neutrophils, lymphocytes, plasma cells and a great amount of eosinophils in some regions (Fig. 1(D)). In the submucosa, squamous metaplasia of residual acinar and ductal elements was found. Some nests showed evidence of residual lumina (Fig. 1(E)). The metaplastic squamous islands and nests revealed a bland appearance, various sizes and shapes, smooth and regular borders (Fig. 1(F)). The component cells of these nests presented a bulky nucleus with occasional normal mitoses, without cellular atypia.

Discussion

According to Anneroth and Hansen, five stages can be assumed in the pathogenesis of NS: infarction, sequestration, ulceration, reparation and healing. These different stages may occur simultaneously in different areas and the severity and extension of the damage depend on the healing capacity of the host tissues. If the injury is extensive, the sequestration of the necrotic tissues results in ulcer formation with dense inflammatory response, as observed in our patient.²

Typical histopathological features of NS include: preservation of the lobular architecture of salivary glands, mixed inflammatory reaction, pseudoepithelialomatous hyperplasia of the covering epithelium, and squamous metaplasia of ducts and acini with cells exhibiting uniform nucleus with occasional normal mitosis without cellular atypia.³ The presences of residual lumina in some metaplastic nests are characteristic of NS. These latter features are not found in squamous cell carcinoma or in mucoepidermoid carcinoma.⁹

In the present case the first diagnostic hypothesis was necrotizing sialometaplasia although mucoepidermoid carcinoma, squamous carcinoma and other ulcerated neoplasm of minor salivary glands were included in the differential diagnosis. Secondary syphils was also considered since its oral lesions may show pleomorphic features. However, it was excluded by the serological exam (VDRL).

The patient in this report exhibits several aspects of the common clinical and microscopic features related to necrotizing sialometaplasia in the posterior palatal region. An uncommon histopathological finding was the local amount of eosinophilic granulocytes in the inflammatory process. This feature may indicate an immunologic or allergic mechanism in the pathogenesis of his disease, as emphasized by some authors.⁴ However, in our patient, no other inflammatory condition was observed or reported. Although many authors have reported the lesions to be painless, we agree with those who state that pain appears to be a chief complaint, mainly at the onset of the disease.⁴

Our patient was both a heavy smoker and a drinker of alcohol, two of the possible predisposed factors reported in the literature. These habits may induce arteriosclerosis of the small vessels, but the exact mechanism remains obscure.¹⁰

In conclusion, considering the self-healing behavior of this pseudotumoral condition, the correct diagnosis is required in order to avoid mismanagement of affected patients with an inadequate or unnecessary approach.

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References