Necrotizing sialometaplasia of the oral palate: a rare cause of painful mouth ulceration.

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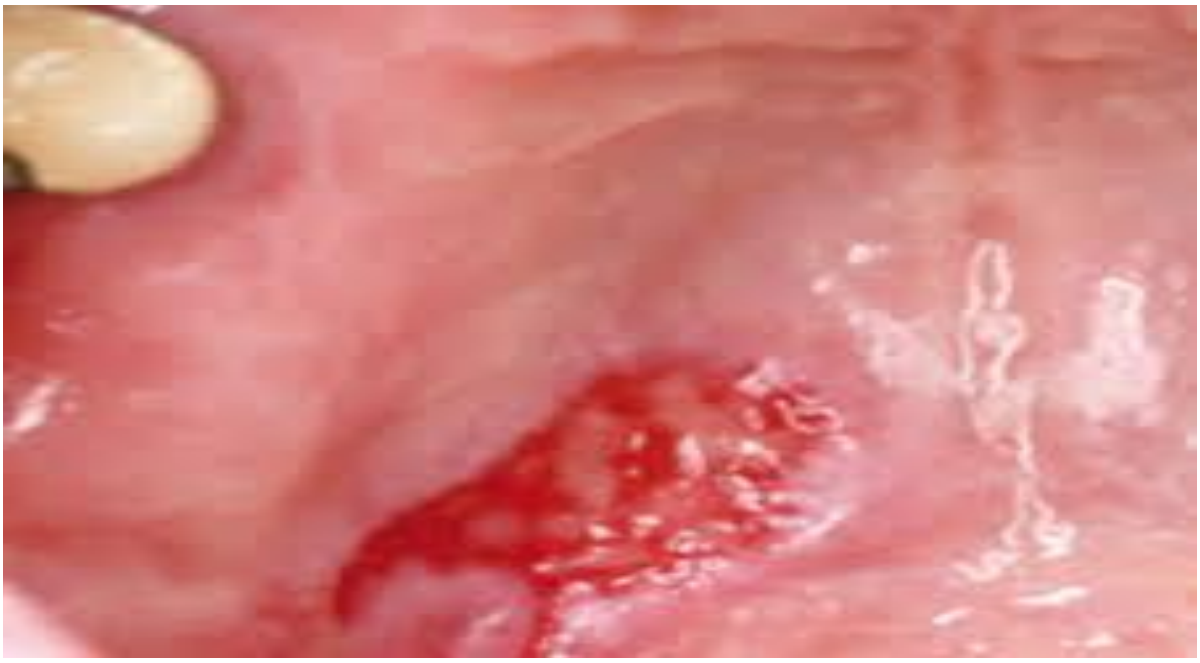
Introduction:

Necrotizing sialometaplasia (NMS) is a rare benign inflammatory condition affecting the salivary glands. This pathology mainly affects the accessory salivary glands of the palate. We present a patient who consults for painful bleeding ulcers.

Case report:

45-year-old patient with no alcohol or smoking history complaining of a painful lesion located in the oral palate that is bleeding upon food contact associated with dental paraesthesia for 2 months. The patient was advised by a medical practitioner to use local treatments based on oral care by mouthwashes and buccal miconazol but without clinical improvement.

Then the patient consulted our department where we did a biopsy that reveals a non-specific ulceration of the fleshy bud type. (figure 1).



In addition, TPHA-VDRL and HIV 1 and 2 serology were prescribed as well as complete blood cell count and protein electrophoresis which was normal. Thereafter, we performed a second biopsy which came back in favor of glandular necrosis associated with an epidermoid metaplasia of the acinus, the excretory ducts and mucus patches. The patient received a low dose of oral corticosteroid therapy with good improvement.

Discussion:

Necrotizing sialometaplasia (NMS) is a rare inflammatory disease of the salivary glands. The prevalence is 0.03% of the oral mucosa lesions [1.2]. In 1958, Saunders reported two clinical observations suggestive of palatal NMS. In 1973, it was identified for the first time after seven cases of palatal lesions by Abrams et al. The sex ratio of SMN is 2/1 [1.4]. The SMN has a wide age range (1.5-83 years), although the majority of patients are of more than 40 years old [5]. 88% of NMS cases affect the salivary glands accessories and 12% the main salivary glands.79% are localized on the palate [6] as our reported case.

In fact, there are many theories that have been suggested as causes:

- Tobacco smoking (MNS represents the terminal stage or ulcerative nicotinic stomatitis).

-Vitamin A deficiency [2, 10].

- An allergic mechanism (recognized by the presence of polymorphonuclear eosinophils in the inflammatory SMN infiltrate) [2.6].

- Infectious diseases such as HIV [2.6].

- Anorexia and bulimia [9], since several cases occurred in young women with these behaviors [2].

- Salivary lobule necrosis due to an ischemic event [9-10, 2, 9,6] either by a vascular pathology such as atherosclerosis, or by trauma (injections of local anesthetics [7], dental care chemical products use [7], ingestion of hot beverages [7], wearing dentures [7], locoregional interventions [6.7.10] or the presence of upper aerodigestive tract tumor.

Clinically, MNS may be manifested by a local throat pain. In some cases, it is associated with sensory disorders in the area concerned such as paraesthesia [11], hypoaesthesia or anesthesia [2]. Rarely associated with fever, migraines or lymphadenopathy. The differential diagnosis at the initial stage can be an abscess of dental origin or a tumor of the accessory salivary glands (pleomorphic adenoma).

Moreover, at the ulceration stage, we can think of a carcinoma, a canker sore (Sutton periadenitis), an infectious ulcer (tuberculosis, syphilis, systemic mycosis), agranulocytosis, non-Hodgkin's lymphoma or sarcoma [11, 14], Rarely palatal artery thrombosis [15]. The clinical evolution of MNS is usually good after 4 to 10 weeks without treatment [14]. Scarring occurs by second intention [11, 14]. Lesion extension and secondary infections have been reported.

Macroscopically, it is often a crateriform ulcer [1,11,12,13] of the palatal mucosa measuring from 5 to 20 mm in diameter that can be unilateral or bilateral with a whitish base covered by necrotic tissue and fibrino-leukocytic exudate, surrounded by an erythematous or whitish halo [2].

MNS can present in two forms [11,12]. First, the ulcerative form is the necrotizing sialometaplasia which correspond to the clinical case. Second, the tumor form. It is a subacute necrotizing sialadenitis that constitute the initial stage of MNS or a more localized form. The SMN preferentially affects the accessory salivary glands, in particular in the hard palate and the hard-soft palate junction [2, 9]. The main salivary glands such as the parotid and submandibular glands are rarely affected.

Histologically, MNS is characterized by five criteria [2, 7, 10 ,11]:

- Infarction or lobular necrosis.

- The persistence of phantom nuclei.

- An epidermoid metaplasia simultaneously involving canals and mucous acini.

- The presence of an inflammatory infiltrate and tissue granulation.

- Preservation of the lobular aspect although inflammatory and metaplastic alterations affect several glandular lobules.

In general, NMS is diagnosed at advanced necrotic or ulcerative stage. Management is based on:

- Symptomatic treatment for analgesic purposes [11].

- Oral hygiene measures to avoid secondary infection [9-10].

- At the stage of necrosis, we can proceed with the curetage of

necrotic tissue.

At the initial stage, systemic corticosteroid therapy can stop the evolution towards the necrotic stage, but its role is still under discussion. Regular follow-up is needed until recovery.

Conclusion :

Necrotizing Sialometaplasia is a rare pathology but can easily be recognized nowadays. Ignorance or late diagnosis may lead to the need of an invasive management. The histopathological aspect is not always clear. However, in that case, 1 or 2 weeks of close lesion observation might be needed to see its evolution and to be able to establish the diagnosis (SN, squamous cell carcinoma, muco-epidermoid carcinoma, etc.)

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