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Necrotizing sialometaplasia of palate- a clinical case

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Abstract: *Necrotizing sialometaplasia is an uncommon, benign, self-limiting inflammatory condition that manifests mostly as an ulcerative lesion on the palate. It is believed that the lesion is a result of vascular ischemia essentially induced by trauma. Until the present, the establishment of the diagnosis of necrotizing sialometaplasia endures to be a challenge. The significance of this disease lies in the fact that both its clinical and histologic characteristics may mimic those of a malignant neoplasm, especially mucoepidermoid carcinoma or squamous cell carcinoma. A combination of histopathological and clinical findings is usually beneficial in establishing the affirmative diagnosis. In this article we present a review of the literature and a report of a clinical case of necrotizing sialometaplasia of palate in a young man, heavy smoker.*
Key words: *Necrotizingsialometaplasia, palate*

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I. Aim

The aim of this report is to present a clinical case of necrotizing sialometaplasia of the hard palate and the to stress on the importance of confirming the diagnosis by a simple incisional biopsy.

II. Introduction

Necrotizing sialometaplasia was first described in 1973 by Abrams et al. [1] Necrotizing sialometaplasia is an extremely rare, benign, self-limiting lesion of salivary gland tissues. This condition can be found in any salivary gland, including the parotid and submandibular glands, although it is mostly encountered in minor salivary glands of the palate. [5,7,10] It can present in any age group, though most of the patients are older than 40 years. The occurrence comes to be approximately 2 times greater in males than in females. [6,7,10,11] It seems that there is no particular racial predilection. [14] Clinically, it manifests itself as a deep ulcer or erythematous nodule that is questionable for malignancy. [13]

III. Clinical case

We present a clinical case of a 32- years old male patient, a heavy smoker, that appeared in the Department of Oral and Maxillofacial surgery with a "crater-like" ulceration on the mucosa to the left of the midline of his posterior hard palate. The lesion had a soft necrotic central portion and elevated and indurated margins surrounded by an erythematous halo. (Fig. 1) He reported that at first he noted a nonulcerated erythematous swelling that over a relatively short period of time turned into an ulceration. The lesion had been resent for approximately 10 days. It had a dimension of 10 mm in diameter at the time of the visit. The ulcer was associated with no pain but rather paresthesia in the area. There were no present indications of regional lymphadenopathy. No radiolucency was noted on the radiographs. Differential diagnosis of necrotizing sialometaplasia of minor salivary gland of palate, a salivary gland neoplasm or squamous cell carcinoma were considered. An incisional biopsy was performed. The diagnosis of necrotizing sialometaplasia was established through history, clinical examination and histopathology using hematoxylin-eosin stain. (Fig.2) No treatment was needed. The patient was advised to maintain an excellent oral hygiene and to rinse with chlorhexidine solution 3-4 times a day for 2 weeks. The lesion healed spontaneously in about 10 weeks leaving no scar. (Fig.3)

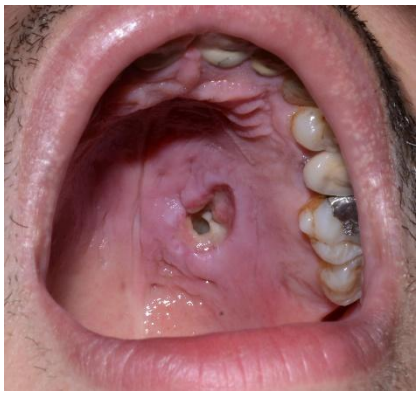

Fig.1:A clinical photograph at the day of the visit

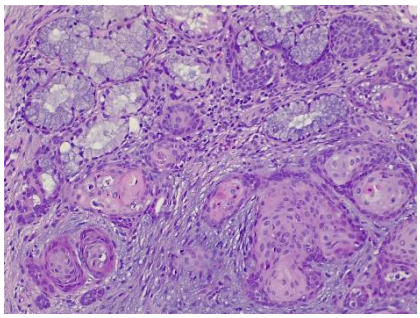

Fig.2: Histopathological image

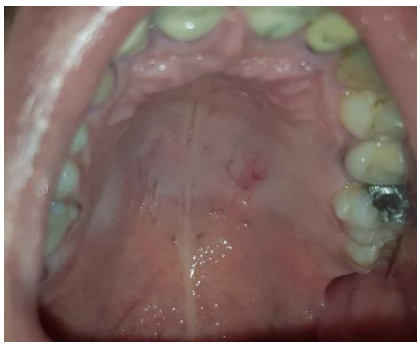

Fig.3:A clinical photograph 10 weeks after the appearance of necrotizing metaplasia of palate

IV. Discussion

Necrotizing sialometaplasia is a benign, self-limiting, necrotizing disorder that involves the mucus-secreting glands, mainly the minor salivary glands. [1,11,16] There are announcements of the development of necrotizing sialometaplasia for the whole of the upper aerodigestive tract where salivary gland tissue is present. It is generally found on the hard palate. [1,16] Regarding location, the majority of cases were reported to occur on the posterior of the hard palate, while the junction between the hard and soft palate was the second most common site. [6] Involvement of the underlying bone of the hard palate is infrequent, but this does not exclude the diagnosis of necrotizing sialometaplasia. [8] Necrotizing sialometaplasia has been reported in all regions where salivary gland tissue is located which counts the nasal cavity, maxillary sinuses, incisive canal, tongue, lower lip, buccal mucosa, retromolar pad, tonsillar fossa, soft palate, trachea and larynx. [8,16] There is existing data that the major salivary glands may be also involved and most of the reports find this condition when a surgical intervention on a major salivary gland for treatment of unrelated problems is performed. [5,8]

The etiology of this disease remains unknown. [12,13] Most authors support infarction as the cause of necrotizing sialometaplasia. It is believed that infarction is a result of compromised blood supply (ischemia) due to local tissue injury of different kinds, e.g., trauma, atherosclerosis, localized vasculitis. [1,7] Association of the lesion with factors like smoking, alcohol use, ill-fitting dentures, prior dental injection, recent surgery, infections and systemic disease have been described. [6,10,16] Predisposing factors for the development of necrotizing metaplasia may be also found in the setting of bulimia. [7] Self-induced vomiting is generally a result of finger insertion distally into the mouth and pharynx so that to initiate a gag reflex. The physical trauma caused by the self-induced vomiting and the food ingestion as well as the chemical trauma due to low pH of the stomach

content result in vascular injury and compromised blood supply to the regional salivary gland lobules. All that leads to later ischemic necrosis or infarction. [16] Necrotizing sialometaplasia has also been reported to be associated with other upper gastrointestinal disorders such as esophagitis, dysphagia, hiatal and umbilical hernia, gastric and duodenal ulcers, and vomiting after silastic ring vertical gastroplasy. [2,3] Romagosa et al. reported three cases of necrotizing sialometaplasia involving submucosal mucoserous glands in the trachea following prolonged translaryngeal intubation. [15] It has also been described in the larynx secondary to atheromatous embolization. [17]

Clinical symptoms of necrotizing sialometaplasia are variable. [6] Lesions generally emerge spontaneously and appear as exophytic palatal tumours, ulceration or nonulcerated erythematous swellings. [16] According to Brannon et al. who analyzed 69 cases of necrotizing sialometaplasia reported in the literature, most cases are of painful lesions. [6] There are some cases that were associated with no pain. [10, 16] The lesion usually presents as a clearly circumscribed ulceration, frequently 1 to 3 cm in diameter. [7] The margins of the ulcer are often raised, erythematous and indurated resembling a carcinoma. [7,10] Involvement of the palate mainly comes as a single, unilateral ulcer on the posterior hard palate or at the junction of the hard and soft palates. [7] Nevertheless, bilateral synchronous and metachronous lesions are reported. [9,10,13] Rarely, the necrotizing sialometaplasia may also occur in the midline. [10] Anesthesia or paresthesia of the palatal mucosa is described as an early indicator of the ulceration and might play a part in the clinical confusion of necrotizing sialometaplasia with a malignant neoplasm. [6,10] In some cases, the surface of the mucosa is intact and the lesion is raised and fluctuant, giving the wrong impression of an abscess formation. In addition, sometimes the clinical history reveals that a non-ulcerated swelling precedes the ulceration. [6,7]

As for the histological findings, microscopically, necrotizing sialometaplasia shows clearly noticeable similarity to mucoepidermoid carcinoma or squamous carcinoma. [1, 16] According to Abrams at al. the histological features which are most helpful in the differentiation of necrotizing sialometaplasia from the mentioned threatening diseases are „lobular infarction or necrosis; bland-appearing nuclear morphology of the squamous cells; simultaneous metaplasia of ducts and mucous acini; prominent granulation tissue and inflammatory components, and maintenance of the general lobular morphology in spite of the fairly extensive inflammatory and metaplastic changes often involving more than one lobule." [1] It is the preservation of lobular architecture that is the best histologic sign for the necrotizing sialometaplasia. [7] Histologic characteristics that exclude the diagnosis of necrotizing sialometaplasia and draw out a concern for a malignant neoplasm are: perineural invasion, presence of apparently neoplastic goblet cells or proliferation thereof; atypical mitoses; marked nuclear pleomorphism in conjunction with high nuclear to cytoplasmic ratios. [7] Anneroth and al. stated that the following 5 histologic stages in the evolution of the necrotizing sialomethaplasia could be seen: infarction, sequestration, ulceration, reparative and healed. They emphasized that these stages could overlap and would be dependent upon the extent and severity of damage. [4] The diagnosis can be additionally amplified by immunohistochemistry. Carlson et al. state that the "incorporation of an antibody including myoepithelial markers (smooth muscle antibody, p63, calponin), basement membrane markers (laminin, collagen type IV), E-cadherin, and various cytokeratins (CK5, CK6, CK7, CAM 5.2)" could be helpful for establishing a final diagnosis when in doubt. Yet, till today there is no definitive immunophenotype and this is the reason why a properly oriented hematoxylin-eosin section stays the gold standard for diagnosis. [7,10]

The differential diagnoses of greatest import include mucoepidermoid carcinoma and squamous cell carcinoma. [1,5,7,15] Non neoplastic conditions to be considered include rare granulomatous lesions such as syphilitic gumma, tuberculous ulcer, midline lethal granuloma. [13] It is important to keep in mind that healed necrotizing sialometaplasia might present the same histopathological characteristics as chronic sialadenitis or might show degenerative age changes. [4]

The lesion resolves spontaneously within 6 to 10 weeks and do not require any treatment. [1,4,8] An incisional biopsy is essential to confirm the diagnosis of necrotizing sialometaplasia and to rule out a malignancy. [16] The healing begins with a proliferation of the surface epithelium in order to cover the epithelial defect. [4] The healing process is rather slow in view of the considerable surface of exposed submucosa to heal by secondary intention. [1] The patient must be closely followed up until healing is complete. [8] As the lesion is self-limited, some patients may not seek treatment and consequently are not biopsied and diagnosed. [4, 8, 12]

V. Conclusion

Necrotizing sialometaplasia is a benign self-limiting disorder of salivary glands. Unfortunately, it has been clinically and microscopically confused with a malignant neoplasm, resulting in inappropriate treatment. A simple incisional biopsy is required to confirm the histologic diagnosis and to rule out more serious disease processes. Usually, no treatment is required and the lesion heals by secondary intention within up to 10 weeks.

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