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Case Report

Extensive Necrotizing Sialometaplasia of Palate: A Case Report

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ABSTRACT

Necrotizing sialometaplasia (NS) is an uncommon, benign self-limiting lesion, locally destructive and an inflammatory condition that largely affects minor salivary glands and the major salivary glands in up to 10% cases. [1] A 19 year old male patient with bilateral painful ulcerative lesions over soft palate extending to the hard palate was presented to our department. Histopathological examination of punch biopsy revealed Pseudoepitheliomatous hyperplasia with replacement of the seromucinous lobules by aggregates of squamous cells and associated dense acute inflammation was observed. The pathogenesis of NS is unknown but is believed to be due to ischemia of vasculature supplying the salivary gland lobules. A simple biopsy is required to confirm the histological diagnosis and to rule out more serious disease processes. It is a self-limiting disease process and requires no specific treatment.

Keywords: Inflammation, Ischemia, necrotizing sialometaplasia, pseudoepitheliomatous hyperplasia, self-limiting disease.

INTRODUCTION

Necrotizing sialometaplasia (Salivary gland infarction) is a benign, self-healing lesion, affecting especially the minor salivary glands of the palate. It was first described by Abrams et al. in 1973. It is a relatively rare with incidence of <1% of biopsied oral lesions. It can occur at any age between 17 and 80 years with male predominance of 2:1. Classically, it involves the mucoserous glands of hard palate (80%), with two thirds of the cases being unilateral lesions. [2] It can also be seen in other sites like soft palate, lip, retromolar area, tonsillar fossa, gingiva, tongue, cheek, sinuses, nasal cavity, larynx and trachea, where the salivary gland tissue is located. The importance of NS is that it mimics malignant lesion, such as mucoepidermoid carcinoma and invasive squamous cell carcinoma. Although the underlying process is generally considered to be ischemic, [3] in many cases predisposing factors cannot be identified.

CASE REPORT

A 19 year old male came with complaints of throat pain and non-healing ulcer over soft palate for the past one month, with pain during swallowing. Patient also gave H/O usage of gutkha and tobacco chewing since 5 years.

On clinical examination, there was an ulcerative lesion over both sides of soft palate involving about 6*5 cm extending from bilateral anterior pillars, base of uvula and posterior half of the hard palate with slough over the lesion (Fig.1). Left submandibular lymph node was enlarged measuring about 3*2 cm and was not mobile, non-tender.

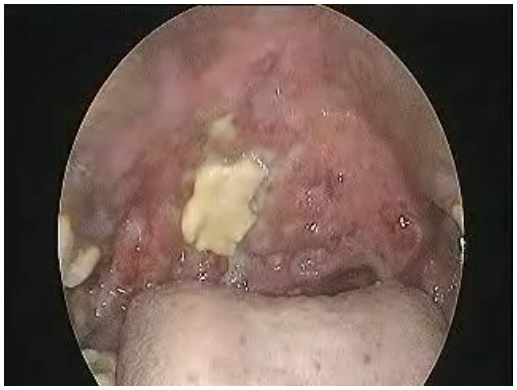

Fig.1: Intraoral view revealing the lesion on the both sides of the soft palate extending to the hard palate.

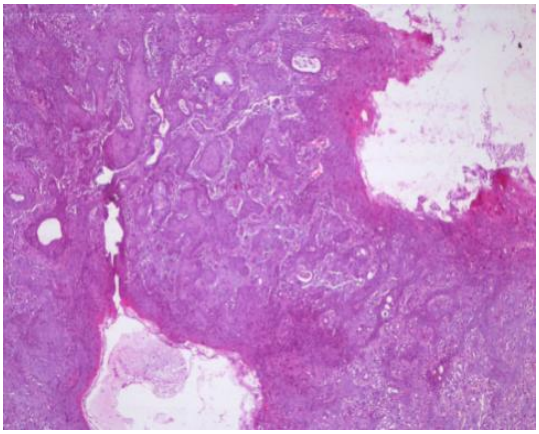

Fig.2: Pseudoepitheliomatous hyperplasia of overlying epithelium

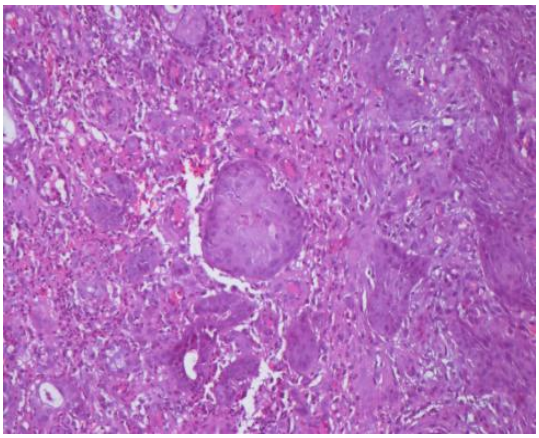

Fig.3: Higher magnification shows replacement of the seromucinous lobules by aggregates of squamous cells with associated dense acute inflammation.

A punch biopsy of the lesion histologically showed pseudoepitheliomatous hyperplasia, with elongated rete ridges deep into subepithelial tissue. Subepithelial minor salivary gland tissue showed extensive squamous metaplasia and stroma was composed of mixed inflammatory cells infiltrate predominantly comprising of neutrophils along with lymphocytes and occasional eosinophils and plasma cells. Occasional clusters of squamous cells showed nuclear atypia and scatter cells showed atypical mitosis (Fig.2,3). Patient was reassured and supportive treatment was given. He was followed up after 2 months and the ulcerative lesion had healed completely with fibrosis (Fig.4,5).

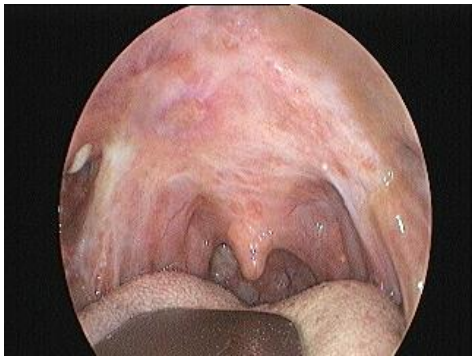

Fig.4: The ulcerative lesion had healed completely after period of 5 weeks.

Fig.5:Intraoral view showing complete regression of the lesion after a period of 2 months

DISCUSSION

NS is a self-limiting, variably ulcerated, benign process affecting the minor salivary glands. Although the etiopathogenesis of NS remains unknown there is general consensus that an ischemic event in the salivary gland precedes the development of the lesion. [4] Etiological factors for ischemia can be direct trauma, administration of local anaesthetic, ill-fitting dentures, alcohol, smoking, cocaine use, radiation, intubation, surgical procedures and upper respiratory tract infections [3] NS shows a male predilection with a male to female ratio of 2:1. Average age of presentation is around 46 years. In some cases, it may be seen in younger individuals

also. [5] It usually occurs spontaneously and the initial symptoms may be fever, chills, pain, malaise. Most common site involved is posterior part of hard palate at the junction of soft and hard palate. According to Anneroth and Hansen, five stages can be assumed in the pathogenesis of NS; infection, sequestration, ulceration, reparation and healing. [6] The different stages may occur simultaneously in different areas and the severity and extension of the damage depends on the healing capacity of the host tissues. The histological features of NS as described by Abrams *et al*. include lobular infarction or necrosis with simultaneous metaplasia of ducts and mucous acini, bland-appearing nuclear morphology of the squamous cells, prominent granulation tissue with inflammatory components and maintenance of the general lobular morphology. The preservation of normal lobular architecture of salivary glands seems to be a key feature that distinguishes NS from neoplasia. [2] The presence of residual lumina in some metaplastic nests is characteristic of NS and these features are not found in squamous cell carcinoma or mucoepidermoid carcinoma. [7] Brannon *et al*. described microscopic findings that include coagulation necrosis of the salivary gland acini during the early stage and squamous metaplasia of the ducts and reactive fibrosis during the late stage. [3] An extensive infarct leads to sequestration of the necrotic acini, resulting in ulceration. During the healing process of ulceration pseudoepitheliomatous hyperplasia is seen. However, if the infarct is limited in its extent, then sequestration and ulceration need not occur and necrosis may involve in a small area. [8]

Rizkalla and Toner investigated the use of immunohistochemistry to distinguish NS from squamous cell and mucoepidermoid carcinoma by identifying myoepithelial cells and the cytokeratin expression. They used several immunohistochemical markers such as p63, cytokeratin 5, cytokeratin 6, cytokeratin 7, S100, calponin, smooth muscle actin, CAM5.2. Their results showed that residual myoepithelial cells were identified at the periphery of the epithelial islands in all case of NS, in contrast to mucoepidermoid and squamous cell carcinoma. [9]

The management of NS includes symptomatic treatment and the lesions undergo spontaneous healing within 2-3 months with secondary intention. The duration of the healing process is usually related to the size of the lesion. The recurrence rate of NS is low. Even a full-thickness palatal lesion heals completely in 6 months.

CONCLUSION

In summary, NS is a self-limiting disorder of salivary glands mostly affecting the hard palate. Recognition of necrotizing sialometaplasia is essential because it may mimic malignancy both clinically and histologically. An adequate biopsy and an awareness of the disease entity are important to avoid unnecessary surgical intervention. The exact aetiology though cannot be determined in this case, the role of long term use of addictive substance (gutkha) as a predisposing factor needs to be assessed.

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