

Necrotizing Sialometaplasia: A Diagnostic Dilemma

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Abstract

Necrotizing sialometaplasia is a benign self-limiting lesion of both major and minor salivary glands although more commonly the latter. The clinical and histological similarity between this entity and a malignant lesion implies a risk of unnecessary radical treatment. We report a case of necrotizing sialometaplasia in middle aged women presenting as a non-ulcerated swelling. This case illustrates the need for careful analysis of a biopsy specimen for a correct diagnosis.

Key words: Minor salivary glands, Necrotizing sialometaplasia, Oral ulcer, Palate

Introduction

Necrotizing Sialometaplasia (NS) is a benign condition that may be found at any site in the body that contains elements of the salivary gland. NS was defined by Abrams et al. in 1973 as a reactive necrotizing inflammatory process involving minor salivary glands of the hard palate [1].

It represents less than 1% of biopsied oral lesions [2]. Classically, it involves the mucoserous glands of the hard palate, but all sites with salivary tissue can be involved, including upper and lower lip, maxillary sinus, floor of mouth, tongue, retro molar area, oral mucosa, tonsillar fossa, major salivary glands, nasal cavity, larynx, soft palate and soft-hard palate junction [3,4]. Up to 10% of cases have been reported in major salivary glands.

Recognizing NS is important because this lesion mimics the appearance of malignant disease, both clinically and microscopically. Failure to recognize may result in unnecessary radical surgery. We report the clinical and histopathological features of a case of necrotizing sialometaplasia presenting as a non ulcerated nodule.

Case Report

A 28-year-old female presented with a smooth surfaced dome-shaped non ulcerated swelling at the junction of hard and soft palate on the left side of 6 months duration. The size of the lesion was 2×3 cm and the overlying mucosa had normal appearance with no pain or erosive changes (*Figure 1*).

Her past medical history revealed extraction of the

maxillary left first molar one year back under local anesthesia. Occlusal radiograph showed no bone involvement. A wide excisional biopsy was done. Microscopic examination revealed pseudoepitheliomatous hyperplasia of the surface epithelium (*Figure 2*). The submucosa showed focal necrosis of acini with preservation of lobular architecture and squamous ductal metaplasia (*Figures 3-5*). Mild nuclear pleomorphism

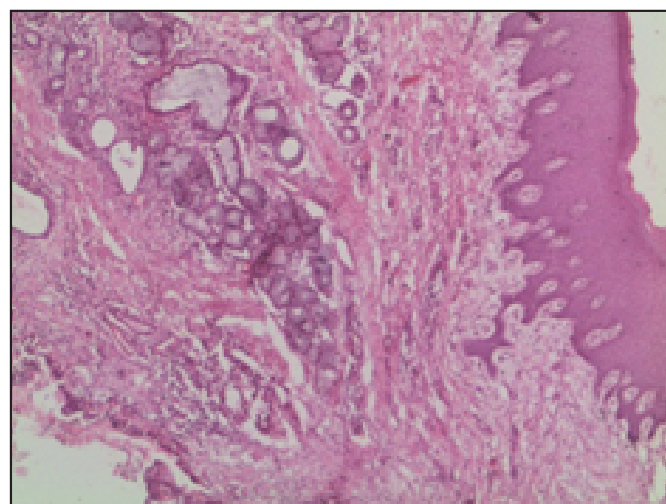


Figure 2. Pseudoepitheliomatous hyperplasia mimicking squamous cell carcinoma (H & E Original magnification X 40).

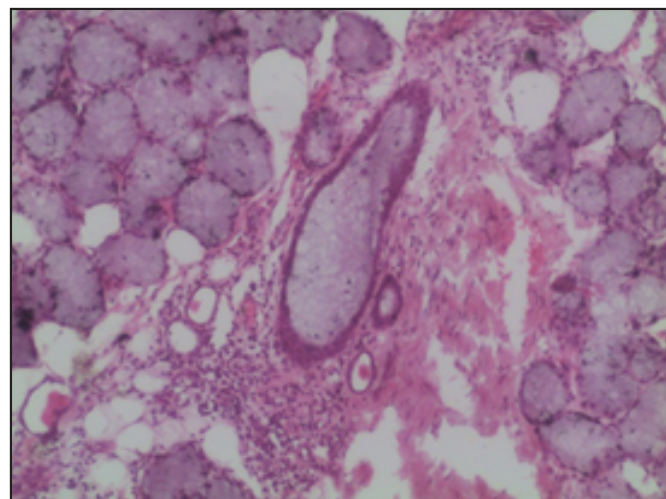


Figure 3. Focal acinar necrosis with dilated atrophic duct (H & E Original magnification X 100).

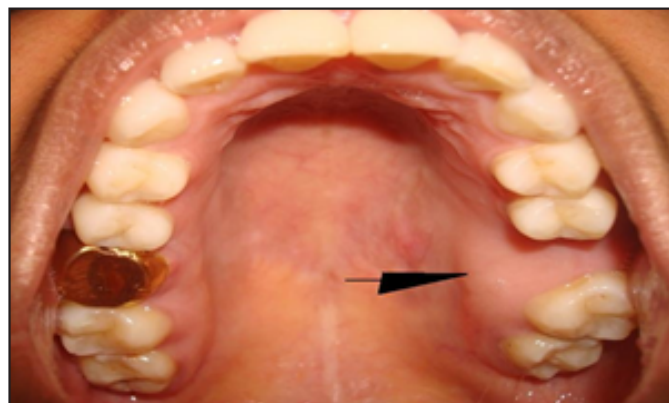


Figure 1. Clinical Photograph of non-ulcerated swelling (black arrow).

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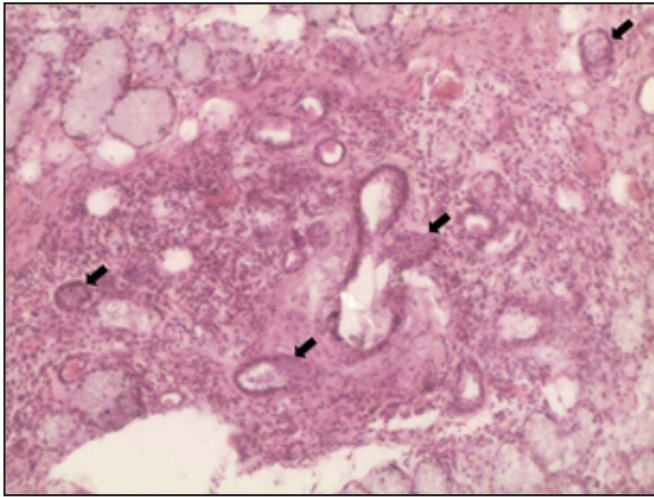


Figure 4. NS was composed of chronic inflammation, metaplastic squamous epithelium (black arrows) that mimicked well differentiated squamous cell carcinoma (H & E Original magnification X 40).

and chronic inflammatory cell infiltrate were also seen. All of these histopathological findings were consistent with the diagnosis of NS.

Discussion

The pathogenesis of NS is unknown, but ischemia involving the glandular tissue is probably the underlying cause of NS. However, the nature and pathophysiology of the ischemia remain unclear in many cases. Possible predisposing factors suggested are direct trauma, administration of local anesthesia, ill-fitting dentures, alcohol, smoking and cocaine use, radiation, surgical procedures, upper respiratory infections and chronic vomiting [5]. NS has also been reported in a patient with Buerger's disease and Raynaud's phenomenon supporting the role of ischemia as an important etiological factor [6]. In the present case application of local anesthesia to the hard palate resulting in vasoconstriction could be the etiological factor as the patient gave a history of tooth extraction in that region. In 1996, Shigematsu et al. found a relationship between the repeated application of local anesthesia into rat palate and histological changes similar to those observed in NS [7].

There is a wide range in the age at presentation (1.5–83 years), although most patients are older than 40 years (mean age 46) [4]. The incidence appears to be 2 to 3 times greater in males than females [6]. However, Kaplan et al. reported an apparently higher incidence in female patients [8]. Furthermore, Imai [9] et al. reported NS associated with eating disorder in female patients. Clinically, NS most often presents as a deep-seated ulcer, however, few cases manifest as non-ulcerated swelling or mass, as in the present case [10]. Generally, the lesions are unilateral but bilateral lesions are seen in around 12% of cases.

Abrams [1] et al. proposed the following histopathological criteria: necrosis of acinar cells of seromucinous glands; squamous metaplasia of salivary ductal epithelial and acini; pseudoepitheliomatous hyperplasia of the epithelium lining the gland; mucous pooling and granulomatous inflammatory response in or around the glands; and histologically benign nuclear morphology, although normal mitoses can sometimes



Figure 5. Complete resolution of the lesion after 10 weeks.

be observed. Brannon [4] et al. described the predominance of coagulative necrosis of acini in early lesions and of squamous metaplasia and reactive fibrosis in later lesions.

Anneroth and Hansen proposed five histologic stages in the development of necrotizing sialometaplasia: infarction, sequestration, ulceration, repair, and healing. They emphasized that these stages could overlap and would be dependent upon the extent and severity of damage [11]. Extensive infarct leads to sequestration of the necrotic acini, which results in the formation of the ulcer. However if the infarct is limited sequestration and ulceration do not occur, which could be the reason for the non ulcerative lesion in the present case. The characteristic squamous metaplasia of ductal elements may be misinterpreted as squamous cell carcinoma as has occurred in the present case. The presence of this metaplasia in residual viable salivary glands may be mistaken for mucoepidermoid carcinoma. These histological changes mimic carcinoma are due to the underlying reparative process but not due to the inherent nature of the lesion. However, the possibility of coexistence of a malignancy cannot be ruled out which warrants adequate biopsy and awareness of this disease entity [12].

Histological criteria to distinguish NS from a malignancy are:

1. Preserved general lobular morphology.
 2. Bland appearance of squamous islands or nests with no cytological evidence of malignancy and
 3. No findings of residual ductal lumina in any nest.
- Reactive atypia is occasionally noted but the general lobular appearance and the presence of intraepithelial inflammation in the squamous nests helps in coming to a diagnosis [4].

The differential diagnosis of NS should consider traumatic, inflammatory or infectious lesions like traumatic ulcer, major apthae, tuberculosis, tertiary syphilis or deep fungal infection in patients with AIDS or under immunosuppressive treatment and those of cancerous origin. The possibility of a subacute necrotizing sialadenitis should also be taken into account, although some authors consider it to lie within the spectrum of NS. According to Fowler et al., it is a non-specific acute inflammatory condition of unknown origin, histologically characterized by focal acinar necrosis (secondary to the inflammation) and atrophy of duct cells, without ductal metaplasia, pseudoepitheliomatous hyperplasia or fibrosis

[13]. It manifests as non-ulcerated erythematous nodular lesions on the palate accompanied by acute pain and has been reported in young people living in groups. The suggested etiology are infectious (viral) or allergy. The main differences from NS are smaller size of lesion, scarcity of ulceration, and absence of squamous metaplasia. Usually, no treatment is required for NS and the lesion heals by secondary intention within 4 to 10 weeks (average 5.2 weeks). Even a full-thickness palatal lesion communicating with nasal Cavity

resolved completely in six months [14].

Conclusion

In summary, recognition of necrotizing sialometaplasia is essential because this lesion may mimic malignancy such as squamous cell carcinoma or mucoepidermoid carcinoma both clinically and histologically. An adequate biopsy and an awareness of this disease entity are important to avoid inappropriate and unnecessary surgical resection.

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