Case Report

Necrotizing Sialometaplasia of Palate with Belligerent Clinical Stance: A Case Report and Review

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Abstract

Necrotizing sialometaplasia (NS) is a benign condition affecting the salivary glands often misdiagnosed as neoplasia based on clinical findings alone. It is considered under the group of tumor-like lesions according to the WHO classification of salivary gland tumors. Here, we report a rare case of NS in the anterior palate which presented as an ulcer in a 70-year-old male. Complete remission was observed around 8 weeks. Adequate biopsy from the representative area should be done to recognize the histological picture among the five histological stages described by Anneroth to avoid misinterpretation and inappropriate treatment of the nonneoplastic inflammatory condition of the salivary glands.

Key words: Anterior palate, necrotizing sialometaplasia, minor salivary glands

Introduction

Necrotizing sialometaplasia (NS) is a rare self-limiting lesion which is locally destructive, inflammatory condition that affects the salivary glands. It was first described by Abrams *et al.* in 1973.[1] It is frequently misdiagnosed as a malignancy in spite of its relatively innocuous nature. This pitfall in diagnosis can be overcome if one is aware of the clinical and pathological features of this idiosyncratic entity.[2] This report presents a case of NS in a 70-year-old male with a chronic history tobacco and alcohol abuse.

Case Report

A 70-year-old male patient reported with an unhealing wound in the palate for 15 days. He gave history of total extraction due to loosening of the teeth in duration of 1 month. A swelling appeared 20 days later which then ulcerated. It was associated with intermittent pain of pricking type which radiated to the temporal region. In addition to this, the patient admitted to a 30 years history of alcohol abuse and even tobacco in both smokeless and smoked form for 5–6 times/day. No relevant medical history was observed.

Extraoral examination did not reveal any abnormality. Intraoral examination revealed a tender solitary ulcer on the right side of anterior hard palate measuring 3 cm × 4 cm with punched out margins exposing the superficial bone [Figure 1]. It was covered by necrotic slough. The ulcer extended posterior to the alveolar ridge mucosa behind maxillary incisors region up to 4 cm anterior to the junction of hard and soft palate anteroposteriorly. Mediolaterally, it extended from alveolar ridge adjacent to canine and premolar region crossing midline up to 1 cm. Based on these features, a provisional diagnosis of NS was proposed. Our differential diagnosis included deep-seated fungal infection, NS, mucoepidermoid carcinoma, and squamous cell carcinoma. These differential diagnoses were considered based on clinical appearance and anatomical location of lesion.

Occlusal radiograph [Figure 2] and orthopantomograph revealed mild surface erosion in the palate. Routine hematological values were within the normal range. Thus, an incisional biopsy was performed and was sent for histopathological examination. A diagnosis of necrotizing sialometaplasia was rendered based on the histopathological features. The patient was treated by debriding the necrotic slough and irrigated with betadine solution. He was advised

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to maintain good oral hygiene with saline rinse 3–4 times a day and also cessation of habits.

Pathological findings

Microscopic evaluation of hematoxylin- and eosin-stained section demonstrated parakeratinized stratified squamous epithelium of varying thickness with elongated rete ridges and an area of ulceration. Lesion tissue was composed of necrosed salivary glands with preservation of lobular architecture [Figure 3]. The necrotic acni showed pale acinar outline with hyperchromatic or absence of nuclei [Figure 4]. Metaplastic squamous islands of ductoacinar unit with residual lumina and inflammatory infiltrate were apparent [Figure 5]. No cytological atypia was observed in squamous islands [Figure 6].

Discussion

NS is benign condition affecting the salivary glands often misdiagnosed as a neoplasia based on clinical findings alone. It is considered under the group of tumor-like lesions according to the WHO classification of salivary gland tumors.[3]

Incidence of NS reported is 0.03%–0.063%.[4] Rajendran states most of the cases occur spontaneously while others may be associated with history of trauma, radiation therapy, or surgery.[1] The precise etiopathogenesis is not known. It has been considered that the ensuing events causing NS may be due to some kind of physical, chemical, or biological assault over the local blood vessels leading to ischemia followed by infarction, necrosis, and ductal proliferation of the salivary glands.[5] The ischemia may be due to trauma, administration of local anesthetics, infection, smoking, alcohol, intubation, Ill-fitting dentures, radiation, and surgical procedures.[3,6,7] Histopathogenesis of NS is classified into five histological stages by Anneroth and Hansen. These five stages follow a sequential order as it follows infarction, sequestration, ulceration, repair, and healing.[8]

NS is predominantly seen in palate but also noticed in other locations such as soft palate, lip, retromolar area, tongue, major salivary glands, mucobuccal fold, tonsillar fossa, nasal cavity, incisive canal, maxillary sinus, and larynx.[7] It most commonly

Figure 1: Ulcer on the anterior palate.

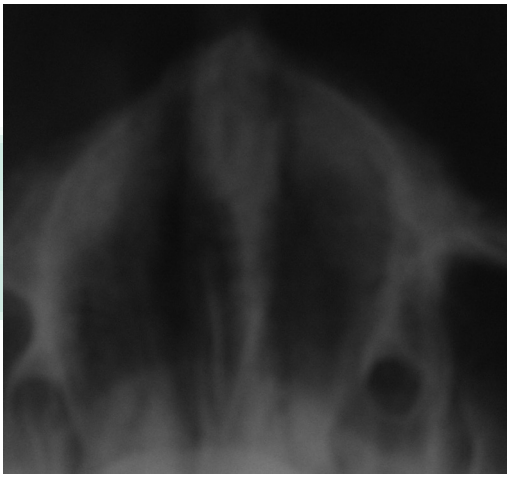

Figure 2: Occlusal radiograph reveals mild surface erosion in the palate.

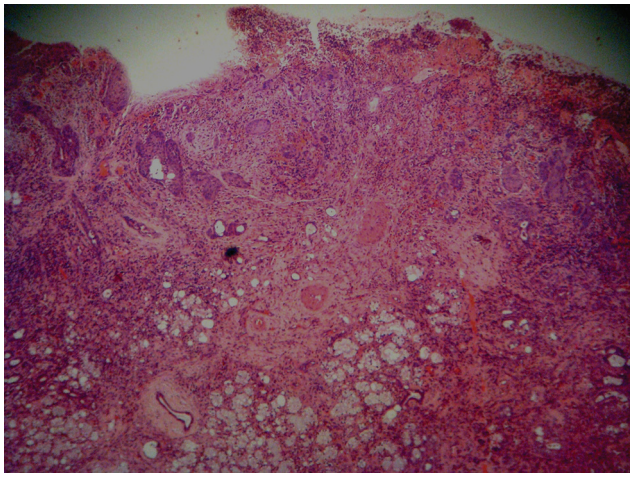

Figure 3: Photomicrograph shows ulcerated epithelium with preservation of lobular pattern of the preexisting salivary glands (×10).

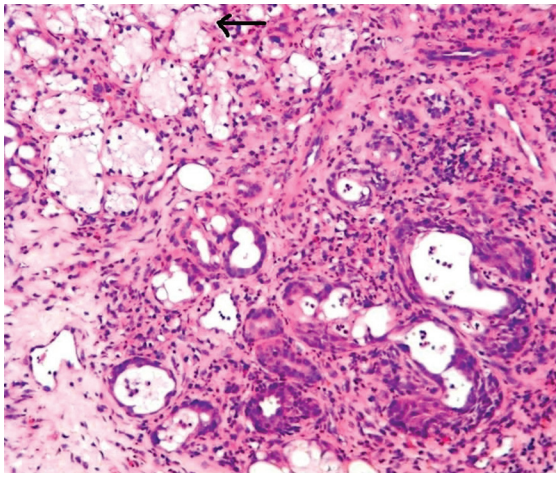

Figure 4: Photomicrograph shows acinar degeneration with pale acinar outline, and nuclei are hyperchromatic or absent (×40).

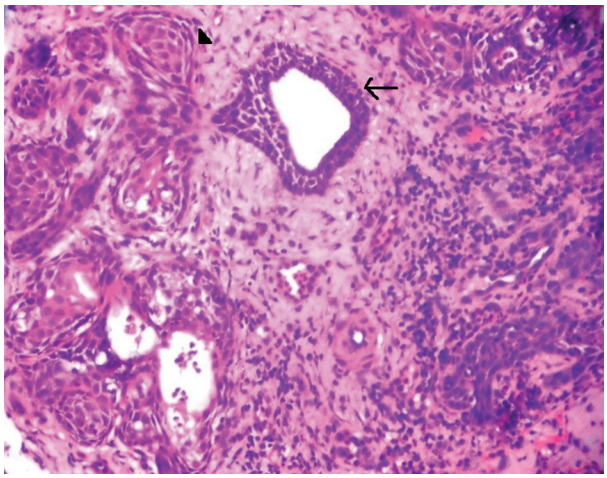

Figure 5: Photomicrograph shows squamous metaplasia of ducts (arrow) and squamous islands (arrowhead) with dense inflammatory infiltrate in stroma (×40).

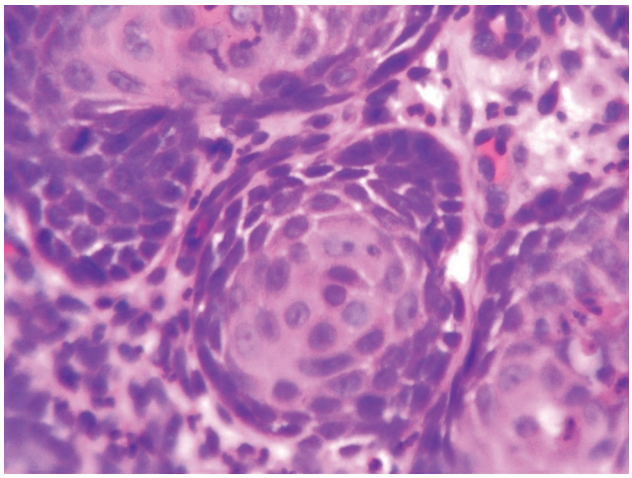

Figure 6: Photomicrograph shows squamous islands in lamina propria with bland cytological features (×40).

appears as deep crater-like ulcers ranging from 1 to 3 cm in size. They may also occur as submucosal swellings that may slough and leave characteristic crater-like ulcers. These ulcers are usually unilateral but may be bilateral and rarely metachronous. Average age of patients with NS falls in range of 17–80 years with male predominance of 2:1.[2] Radiographically, NS shows no bony involvement, but a small number of cases demonstrated saucerization of the underlying palatal bone.[7]

Our case is unique since it presented as an ulcer in the anterior hard palate crossing the midline. The possible hypothesis we thought was due to frequent local anesthetic injections for total extraction which might have caused vascular damage resulting in an ulcer that might have extended up to anterior palate. Synergetic effects of smoking and alcohol consumption might have aggravated the condition.

The main histopathological features are coagulative necrosis of glandular acini which is evident as degeneration of mucous acini with preservation of acinar outline and lobular architecture. It is associated with spaces filled with mucin and mixed inflammatory cell infiltrate. Another key finding is the presence of squamous metaplasia of ducts which are commonly noticed as solid squamous islands close to the elongated epithelial rete ridges. Cells lining the ducts undergo squamous metaplasia which later proliferates leading to lumen obliteration. These obliterated ducts appear as bland squamous islands with nonexistence of atypical features. The overlying mucosa will be hyperplastic demonstrating pseudoepitheliomatous hyperplasia.[1] Due to the presence of these squamous islands, it is most commonly misdiagnosed as squamous cell carcinoma. Other microscopic differential diagnosis is mucoepidermoid carcinoma, subacute necrotizing sialadenitis, and Sutton's disease.[5]

Treatment

NS is managed by simple observation until the healing phase is complete as it has been observed that the lesion resolves spontaneously. It usually heals within 5–6 weeks, but size of the lesion and the presence or absence of bony perforation also influence the healing period. Prognosis is good once it is diagnosed accurately.[1] In our case, the lesion showed signs of remission after 2 weeks of follow-up. Complete remission of the lesion was observed approximately after 8 weeks.

Conclusion

As the lesion resolves spontaneously, accurate analysis of the clinical appearance followed by adequate biopsy from the representative area should be done to recognize the histological picture among the five histological stages described by Anneroth to avoid misinterpretation and inappropriate treatment of the nonneoplastic inflammatory condition of the salivary glands.

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Conflicts of interest

There are no conflicts of interest.

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