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Case Report

A case report on Necrotizing Sialometaplasia: Misleading ulcers of the oral cavity

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Abstract

Necrotizing sialometaplasia is a rare, self-limiting condition characterized by necrosis and squamous metaplasia of the salivary gland tissue. First described in 1973, it typically affects the minor salivary glands of the palate. The exact cause remains uncertain, though trauma, surgical procedures, and ischemia are considered possible triggers. Histopathologically, necrotizing sialometaplasia is characterized by coagulative necrosis, squamous metaplasia, and inflammatory changes. Management involves conservative treatment, most cases resolving spontaneously within weeks to months. Accurate diagnosis is crucial to avoid misdiagnosis and unnecessary treatment.

Keywords: Necrotizing sialometaplasia, Salivary gland disease, Mucoepidermoid Carcinoma, Squamous Cell Carcinoma, Self-limiting condition.

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1. Introduction

Necrotizing sialometaplasia (NS) is a benign, self-limiting inflammatory condition of the salivary glands that can closely resemble mucoepidermoid carcinoma or squamous cell carcinoma in both clinical presentation and histological appearance. This can lead to a diagnostic conundrum and unnecessary, aggressive surgery.[1] In 1973, Abrams et al. published the first report on this pathological lesion.[2] Necrotizing sialometaplasia shows a 2:1 male predominance and typically affects individuals between the ages of 17 and 80 years.[3]

It is known to affect the hard palate's minor salivary gland tissue. Nonetheless, there have been reports of involvement in other mucous glandular tissues, including the trachea, nasal cavity, and floor of the mouth.[4-7]

Necrotizing sialometaplasia typically begins as a localized swelling, progressing to a tender, erythematous nodule that subsequently ulcerates, forming a non-indurated necrotic base. It commonly presents on the hard palate as an ulcerative lesion or swelling, often associated with pain and discomfort.[3]

2. Case Report

A 50-year-old male patient who is a farmer by profession came to the Department of Oral Medicine and Radiology with a primary complaint of a painless ulcer on the right side of his hard palate for 2 weeks. The lesion began as a small, peanut-sized swelling that gradually increased and eventually ruptured to form an ulcer in that region. The patient had no relevant medical history or any known history of drug allergy. However, he gave a significant history of chronic tobacco use in the smoked form, smoking about 10 cigarettes per day for the past 30 years. His dental history consisted of an uneventful extraction of the tooth 45 one year ago. He maintained average oral hygiene, which included brushing his teeth once daily with a toothbrush and toothpaste.

On clinical examination, the patient was found to be conscious, oriented, moderately built, and nourished with stable vital signs. There were no signs of any systemic diseases or underlying infections, no TMJ abnormalities were detected, and there were no palpable lymph nodes. Intraoral examination revealed a solitary ulcer of size 1.5 x 1.0 cm on the posterior part of the hard palate concerning the second molar, 2.5 cm lateral to the midline and 1.5 cm away from the free gingival margin. The ulcer showed irregular margins

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with thick exudates and a yellowish slough covering the floor. Areas of erythema surrounded the lesion. On palpation, the ulcer was non-tender, with sloping edges and a base that felt firm in consistency. The remaining oral cavity was unobtrusive other than periodontal pockets concerning teeth-17,27 and 46, and significant calculus and staining.

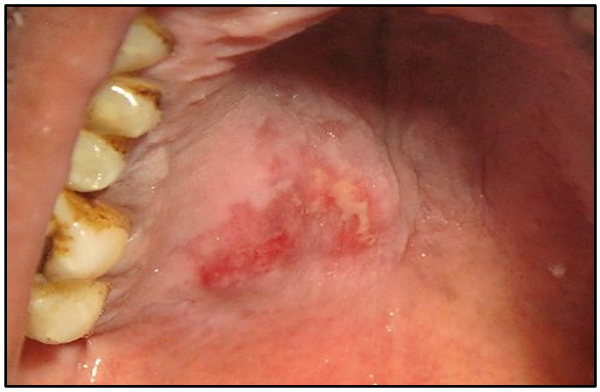


Figure 1: shows a solitary ulcer on the posterior hard palate

Based on the above observations, a provisional diagnosis of a malignant ulcer on the hard palate was made. The two important differential diagnoses made in this scenario were Mucoepidermoid Carcinoma and Squamous Cell Carcinoma. Clinical investigations included thermal vitality tests on the adjacent teeth (16, 17, and 26), which were found to be vital. Intraoral Periapical radiograph of the specified region showing no evidence of osseous involvement, and an incisional biopsy was performed for establishing a definitive diagnosis.

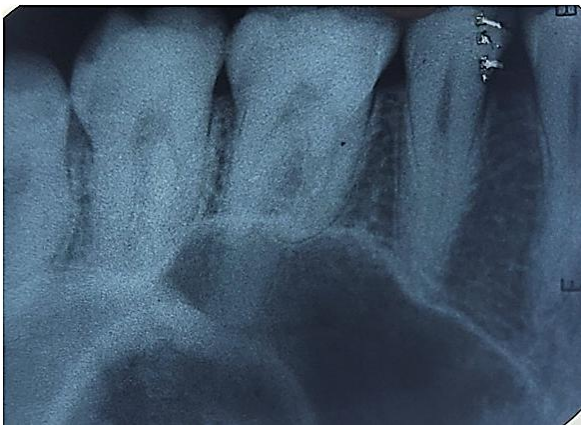
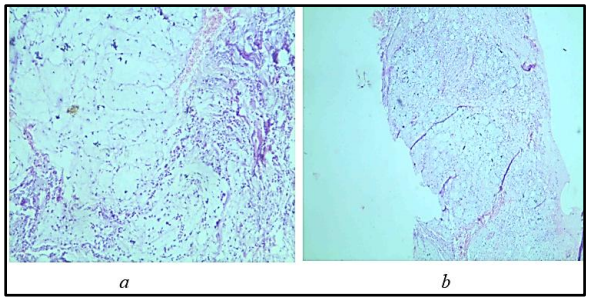


Figure 2: Intraoral periapical radiograph of the specified region shows no osseous involvement.

Histopathological examination revealed squamous metaplasia of the ductal epithelium, characterized by bland-appearing nuclei and an absence of cellular atypia, consistent with a diagnosis of necrotizing sialometaplasia involving the hard palate. The patient underwent an excisional biopsy. On reviewing after a week, the surgical site demonstrated good post-operative healing with no signs of recurrence. The prognosis remains favorable.



Figures 3: show eosin and hematoxylin sections showed squamous metaplasia of ductal epithelium with a bland appearance of the nuclei and no evidence of any cellular atypia.

3. Discussion

Necrotizing sialometaplasia (NS) is a rare, benign, and self-limiting inflammatory disorder that primarily involves the minor salivary glands, with a predilection for the hard palate. It poses a significant diagnostic challenge due to its striking clinical and histopathological similarity to malignant oral neoplasms, particularly mucoepidermoid carcinoma and squamous cell carcinoma.[2] The above-discussed case involving a 56-year-old male smoker with a solitary ulcer on the posterior hard palate highlights these diagnostic intricacies.

The etiopathogenesis of NS can be primarily attributed to ischemic necrosis of salivary gland tissue. Various predisposing factors have been associated with the onset of ischemia leading to necrotizing sialometaplasia, including local trauma, dental injections, ill-fitting dentures, smoking, upper respiratory tract infections, and surgical interventions within the oral cavity.[8] Trauma from the intubation procedure for general anesthesia can also result in NS; Bulimia Nervosa, an eating disorder with marked binge eating followed by purging,[9-10] has also been suggested recently to cause NS. Senapati S et al. reported its occurrence due to immune-mediated vasculitis such as Granulomatosis with polyangiitis, Wegener's disease.[11] Our patient had a long-standing history of smoking, which may have contributed to the local ischemic events in the specified region.

Clinically, NS most often manifests as a sudden-onset, painless ulceration of the posterior hard palate, although initial presentation may involve a nodular swelling that subsequently ulcerates within 2 to 3 weeks.[12] Although the hard palate is the most common site, necrotizing sialometaplasia can also involve other locations such as the retromolar pad, gingiva, lip, tongue, nasal cavity, sinuses, and larynx.[13] Approximately two-thirds of palatal NS cases are reported to be unilateral.[14] Early manifestations typically include submucosal swelling, which may or may not be accompanied by pain or paresthesia. Prodromal symptoms such as fever, chills, or general discomfort may precede the lesion.[3] Within 2–3 weeks of onset, the lesion often progresses to a crater-like ulcer with raised margins,

measuring between1 to 5 cm in diameter. At this stage, pain is usually absent.

In the above-mentioned case, the patient reported an initial swelling that was asymptomatic, which eventually ulcerated, consistent with the typical clinical progression of NS. The ulcer presented with irregular margins, sloping edges, a firm base with thick exudates, and yellowish slough covering the floor, and was tender on palpation- features that mimic malignancy and often result in diagnostic uncertainty.

Necrotizing sialometaplasia can pose a diagnostic challenge, as both squamous metaplasia and necrosis are also observed in various inflammatory and neoplastic salivary gland conditions.[15] According to Anneroth and Hansen, the histopathogenesis of NS progresses through five stages: infarction, sequestration, ulceration, repair, and healing. Histologically, the condition exhibits a spectrum of features, including ulceration, lobular necrosis, sequestration of necrotic acini, pseudo-epitheliomatous hyperplasia of the surrounding epithelium, squamous metaplasia of the ductal epithelium, and inflammatory changes.[12]

Biopsy of our case showed squamous metaplasia with bland nuclear features and absence of cellular atypia, supporting our diagnosis of Necrotizing Sialometaplasia. Importantly, pseudo-epitheliomatous hyperplasia of the surface epithelium, seen in some cases, can mimic invasive squamous cell carcinoma, underscoring the importance of histopathological expertise in avoiding misdiagnosis.[3] Immunohistochemical staining is useful in identifying residual myoepithelial cells in necrotizing sialometaplasia. This can be demonstrated through the expression of markers such as calponin, smooth muscle actin, and cytokeratin-7.[16]

Radiographically, intraoral periapical imaging generally reveals no osseous involvement, which is consistent with the benign nature of NS.[17] In our case, the Intraoral Periapical radiograph of the region confirmed the absence of any osseous involvement. Additional clinical investigations like thermal vitality testing of adjacent teeth revealed normal pulpal responses, ruling out any odontogenic infection as a source of ulceration.

Management of NS is largely conservative. As the lesion is self-limiting, treatment involves patient reassurance, maintenance of oral hygiene, and symptomatic care.[18] Leite M et al. advocated using low-level laser therapy or photo-biomodulation to treat necrotizing sialometaplasia.[19] Our patient in this case underwent an excisional biopsy for a definitive diagnosis, followed by observation. On follow-up, he showed satisfactory post-operative wound healing without any signs of recurrence, indicating the favorable prognosis typically associated with this condition.

4. Conclusion

Necrotizing sialometaplasia is a rare, benign lesion of the oral cavity that clinically and histopathologically resembles malignant conditions. It is typically self-limiting. A comprehensive understanding of its clinical features, along with a detailed patient history, is crucial for an accurate diagnosis. It often presents with deceptive clinical features that may result in undue concern as well as improper treatment planning. Clinicians must maintain a strong clinical suspicion, especially in high-risk individuals such as smokers and tobacco users, to ensure correct diagnosis and appropriate management. A multidisciplinary and careful diagnostic approach ensures optimal results and preserves the well-being of the patient.

5. Source of Funding

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6. Conflict of Interest

None.

References

1. Mesa ML, Gertler RS, Schneider LC. Necrotizing sialometaplasia: Frequency of histologic misdiagnosis. *Oral Surg Oral Med Oral Pathol.* 1984;57(1):71-3.
2. Abrams AM, Melrose RJ, Howell FV. Necrotizing sialometaplasia. A disease simulating malignancy. *Cancer*. 1973;32:130–5.
3. Brannon RB, Fowler CB, Hartman KS. Necrotizing sialometaplasia clinicopathologic study of sixty-nine cases and review of the literature. *Oral Surg Oral Med Oral Pathol.*1991;72(3):317-25.
4. Batsakis JG, Manning JT. Necrotizing sialometaplasia of major salivary glands. *J Laryngol Otol.* 1987;101(9):962–6.
5. Devine M, Sammut S, Conn B, Lopes V. Necrotising sialometaplasia in the floor of mouth. *Oral Maxillofac Surg*. 2014;18(1):119–21.
6. Maisel RH, Johnston WH, Anderson HA, Cantrell RW. Necrotizing sialometaplasia involving the nasal cavity. *Laryngoscope.* 1977;87(3):429–34.
7. Romagosa V, Bella MR, Truchero C, Moya J. Necrotizing sialometaplasia (adenometaplasia) of the trachea. *Histopathology.* 1992;21(3):280–2.
8. Fowler CB, Brannon RB. Necrotizing sialometaplasia: a clinicopathologic study of sixty-nine cases and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2000;89(5):577–85.
9. Wu YH, Kuo YS, Lin PY, Chiang CP. Necrotizing sialometaplasia - Case report. *J Dent Sci.* 2020;15(3):390–2.
10. Salvado F, Nobre MA, Gomes J, Maia P. Necrotizing sialometaplasia and bulimia: A case report. Medicina (Kaunas). 2020;56(4):188.
11. Senapati S, Samal SC, Kumar R, Patra S. Necrotizing sialometaplasia: Manifestation of a localized unclassified vasculitis. *Indian J Pathol Microb.* 2016;59(2):232-4.
12. Anneroth G, Hansen LS. Necrotizing sialometaplasia. *Int J Oral Surg.* 1982;11(5):283–91.
13. Maisel RH, Johnston WH, Anderson HA, Cantrell RW. Necrotizing sialometaplasia involving the nasal cavity. *Laryngoscope.* 1977;87(3):429-34.
14. Kandula S, Manjunatha BS, Tayee P, Astekar M. Bilateral necrotising sialometaplasia. *BMJ Case Rep*; 2016:bcr2015211348.
15. Albert MA, Raymond JM, Francis VH. Necrotizing sialometaplasia: A disease simulating malignancy. *Cancer.* 1973;32(1):130–5.
16. SA, Na HY, Choe JY, Hong SN, Lee H, Park S, Kim JE. Necrotizing sialometaplasia: A malignant masquerade but questionable precancerous lesion, report of four cases. *Shin BMC Oral Health*. 2020;20(1):206.

17. Ghandour L, Bahmad HF, Nasr M, Younes S. Necrotizing sialometaplasia of the palate: a case report and literature review. *Case Rep Dent*. 2021;2021:6676285.
18. Ariji Y, Kimura Y, Hayashi N. Imaging features of necrotizing sialometaplasia of the hard palate: report of a case. *Dentomaxillofac Radiol.* 2001;30(5):308–11.
19. Leite MG, Freitas ES, de Vasconcelos Ii AJ, Pinheiro TN. Low-level laser therapy approach to bilateral necrotizing sialometaplasia of the hard and soft palates. *J Lasers Med Sci.* 2020;11 (3):348-54.

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