CASE REPORT
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A Case of Necrotizing Sialometaplasia After Laryngeal Mask Airway Ventilation

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ABSTRACT

Necrotizing sialometaplasia (NS) is a rare, benign, self-limiting inflammatory disease of the salivary glands that can clinically and histopathologically mimic malignancy. It most commonly presents as a painful ulcer of the hard palate. The lesion is believed to be the result of ischemia of the salivary gland lobules with subsequent repair and metaplasia. Necrotizing sialometaplasia heals spontaneously and requires no treatment. This report demonstrates a case of NS in a 33-year-old female patient who presented with a bilateral, painful swelling of the hard palate that evolved into an ulcer a few days after surgery with laryngeal mask airway ventilation. Hyperbaric oxygen therapy was attempted to enhance healing.

Keywords: Ischemia, laryngeal mask, necrotizing sialometaplasia, palatal ulcer, salivary glands

Introduction

Necrotizing sialometaplasia (NS) is a rare, benign inflammatory condition primarily affecting salivary gland tissue.[1] Initially described by Abrams in 1973, only around 200 cases have since been documented in medical literature.[2,3] The significance of NS lies in its ability to mimic malignant lesions both clinically and histologically, often leading to unwarranted, aggressive treatments.[3]

While the exact pathogenesis remains unclear, NS is generally believed to result from ischemia of minor salivary glands, followed by subsequent tissue repair and metaplasia.[3] A variety of ischemic triggers have been identified, including direct trauma (compression, occlusion, burns), local anesthetic injections, smoking, alcohol use, cocaine use, ill-fitting dentures, bulimia, radiation, intubation, and surgical procedures. Additionally, long-term use of salbutamol may cause mucosal dehydration, predisposing the tissue to injury.[4]

Necrotizing sialometaplasia typically affects individuals with an average age of 45.9 years and shows a male predominance (male-to-female ratio of 1.9 : 1).[3,5] It predominantly involves the minor salivary glands of the palate, with 80% of cases occurring in the posterior hard palate or at the junction of the hard and soft palates. Although most palatal lesions are unilateral, bilateral and midline cases have been reported.[5] Other, less frequent locations include the major salivary glands, lip, retromolar area, tongue, cheek, nasal cavity, trachea, and larynx.[3,5]

Case Presentation

A 33-year-old female presented to the otolaryngology clinic with complaints of pain, swelling of the hard palate, and a fever reaching 39°C, less than 24 hours following general anesthesia with laryngeal mask airway (LMA) ventilation for gynecologic surgery. Her medical history was unremarkable, with no chronic medication use. She had a history of smoking but quit 10 years prior.

The anesthesia protocol included the intravenous administration of propofol (200 mg), ketamine (25 mg), lidocaine (100 mg), and ibuprofen (600 mg). Laryngeal mask placement was performed without difficulty, although manual ventilation was initially impossible due to high airway resistance. Relaxation attempts with additional propofol and a low dose of rocuronium were unsuccessful, leading to a brief period of desaturation that required mask repositioning. The surgery

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lasted approximately 20 minutes, and the immediate postoperative period was uneventful. The following day, the patient contacted the anesthesia department with complaints of headache and hypoesthesia of the palate. Suspecting uvular swelling or a localized injury, she was referred to the ENT department for further evaluation.

Examination revealed mild oedema of the hard palate, and the patient was started on amoxicillin–clavulanic acid and analgesics. During a telephone consultation on the third postoperative day, angioedema was considered a possibility, prompting the addition of tranexamic acid and methylprednisolone to the treatment regimen. By the fourth day, the patient developed firm, bilateral swelling of the posterior hard palate, characterised by a purplish discolouration and central mucosal erosion with draining inflammatory exudate (Figure 1). A disinfecting mouthwash was prescribed. A computed tomography scan performed on the ninth day revealed a sharply defined, irregular hypodense lesion on the palate, measuring 3.1 × 1.2 × 4.4 cm, consistent with a liquefied hematoma. There was no bony erosion or lymphadenopathy (Figure 4). The leading diagnosis at this stage was a pressure ulcer or hematoma related to the laryngeal mask, but the lesion's progression, including the expanding central ulceration, was not typical for a pressure-related injury (Figure 2).

The clinical course aligned with the diagnosis of NS, and the patient was referred to a tertiary clinic, where hyperbaric oxygen therapy (HBOT) was suggested to improve healing (Figure 2). Over the course of 3 months, systematic follow-up showed complete resolution of the lesion and a full recovery from dysesthesia (Figure 3).

Discussion

Necrotizing sialometaplasia typically presents as an ulcerated lesion, though it can also appear as a non-ulcerating mass or swelling.[6] Lesion size can range from 0.7 to 5.0 cm,[5] and while ulceration usually remains superficial, there has been one case of full-thickness necrosis affecting the palate.[3] Symptoms such as fever, chills, or malaise may accompany NS in some cases, although this is less common.[2] The majority of lesions are painful, with pain occurring twice as often as asymptomatic cases. Some patients also experience paresthesia or anesthesia; this is thought to be caused by the involvement of the vasa nervorum of the greater palatine nerve in the vasculitis-like etiological process.[5]

Necrotizing sialometaplasia has a broad differential diagnosis due to its clinical similarities to other ulcerative or malignant lesions. In particular, it is frequently misdiagnosed as squamous cell carcinoma, mucoepidermoid carcinoma, adenocarcinoma, or other oncocytic malignancies. Additionally, NS may be confused with ulcerative conditions like necrotizing infection with species like *Streptococcus pyogenes*, traumatic ulcers, major aphthous ulcers, syphilis, tuberculosis, deep fungal infections, or agranulocytosis.[4] In some cases, where ulceration is absent, NS may present as a fluctuating swelling, resembling an abscess.[6] A less common variant, termed sub-acute necrotizing sialometaplasia (SANS), is suspected to have a viral or allergic origin and typically presents as an erythematous swelling of the palate with abrupt pain but without ulceration.[7,8]

Diagnosis of NS remains challenging and requires both clinical and histopathological evaluation. Incisional biopsy from the ulcer's base and its indurated edges yields the most

Figure 1. Picture of initial lesion on day 4. There is a firm, bilateral swelling of the posterior hard palate, characterized by a purplish discolouration and central mucosal erosion.

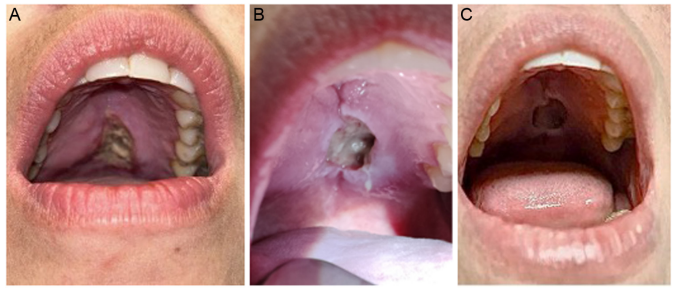

Figure 2. A-C. Pictures showing the evolution of the lesion on days 9, 15, and 24, with progressive ulceration.

Main Points

- Necrotizing sialometaplasia (NS) mimics malignancy, leading to potential misdiagnosis and unnecessary treatment.
- Ischemia of the salivary glands is the main cause of NS, with various triggers including trauma, intubation, and local anesthesia.
- Necrotizing sialometaplasia typically heals spontaneously with conservative management; biopsy and close follow-up are crucial for diagnosis and monitoring.

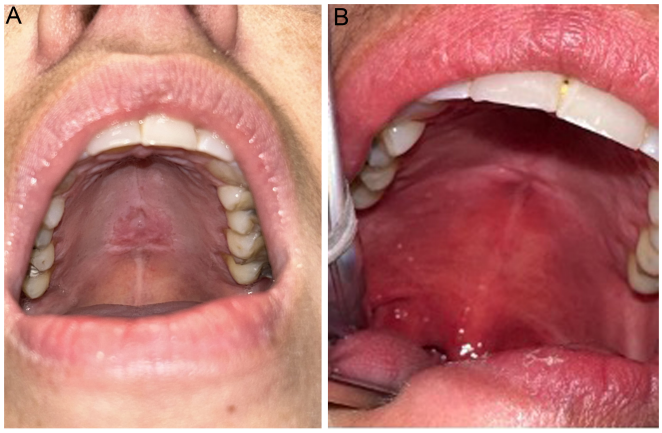

Figure 3. A, B. Pictures demonstrating spontaneous resolution and eventually complete healing of the lesion on days 59 and 227.

representative tissue samples.[4] Histopathological examination reveals 5 hallmark features: pseudoepitheliomatous hyperplasia, squamous metaplasia of ducts and acini, preservation of lobular architecture, lobular infarction (with or without mucin spillage), and secondary inflammation due to mucin extravasation.[6] The pseudoepitheliomatous hyperplasia and deeply seated islands of metaplastic squamous epithelium can lead to misdiagnosis as squamous cell carcinoma. Similarly, squamous metaplasia of the ducts juxtaposed with residual mucous cells may be mistaken for mucoepidermoid carcinoma. However, the preservation of the lobular structure, necrotic areas, and mixed inflammatory background are key distinguishing factors for NS.[4] Immunohistochemistry can support diagnosis, but hematoxylin–eosin staining remains the gold standard.[4,6]

Radiologically, NS may appear as saucerization of the underlying bone or a faint radiolucency.[3] In this case, biopsy was not performed due to the clear temporal relationship between the onset of symptoms and the LMA ventilation, and the absence of clinical or radiological signs suggestive of malignancy, although this would have given more certainty about the diagnosis.

Management of NS is typically conservative, with spontaneous healing occurring over a period of 2-12 weeks. Even in severe cases, such as full-thickness palatal necrosis, healing has been documented within 6 months.[4] If lesions fail to heal within a few months, repeat biopsies are advised to rule out malignancy.[4] Intra-lesional steroids have not been shown to reduce recovery time or alleviate associated anesthesia.[9] In this case, HBOT was proposed to the patient, since it promotes angiogenesis and improves wound healing, though its specific effect on recovery in NS remains unclear.[10] Further investigation into HBOT as a treatment option for NS may be warranted.

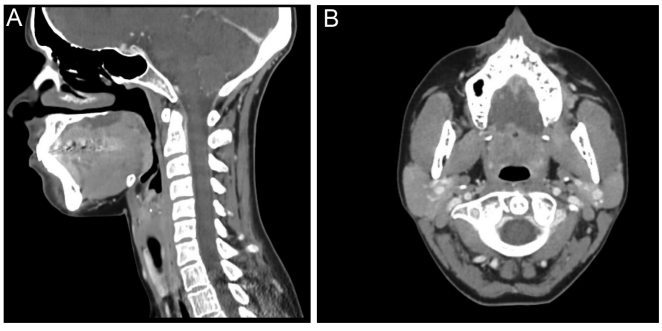

Figure 4. Sagittal (A) and axial (B) CT images show a sharply delineated, highly irregular hypodense zone in the palate.

Necrotizing sialometaplasia is a rare, benign inflammatory condition primarily affecting the salivary glands, most commonly in the hard palate. Despite its self-limiting nature, NS is often misdiagnosed as a malignant neoplasm due to its clinical and histological similarities to more aggressive conditions such as squamous cell carcinoma and mucoepidermoid carcinoma. Accurate diagnosis requires a thorough clinical evaluation combined with histopathological analysis to avoid unnecessary and potentially harmful treatments. While NS typically resolves without active intervention, close follow-up is essential to ensure complete healing. In cases of delayed recovery, repeat biopsies may be needed. Hyperbaric oxygen therapy has potential as a therapeutic adjunct, though further studies are required to confirm its efficacy in promoting faster healing.

Data Availability Statement: The data that support the findings of this study are available on request from the corresponding author.

Ethics Committee Approval: N/A.

Informed Consent: Written informed consent was obtained from the patient who agreed to take part in the study.

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References

1. Abrams AM, Melrose RJ, Howell FV. Necrotizing sialometaplasia. A disease simulating malignancy. *Cancer*. 1973;32(1):130-135. [CrossRef]
2. Krishna S, Bk R. Necrotizing sialometaplasia of palate: a case report. *Imaging Sci Dent*. 2011;41(1):35-38. [CrossRef]
3. Daudia A, Murty GE. First case of full-thickness palatal necrotizing sialometaplasia. *J Laryngol Otol*. 2002;116(3):219-220. [CrossRef]
4. Joshi SA, Halli R, Koranne V, Singh S. Necrotizing sialometaplasia: a diagnostic dilemma! *J Oral Maxillofac Pathol JOMFP*. 2014;18(3):420-422. [CrossRef]
5. Brannon RB, Fowler CB, Hartman KS. Necrotizing sialometaplasia. A clinicopathologic study of sixty-nine cases and review of the literature. *Oral Surg Oral Med Oral Pathol*. 1991;72(3):317-325. [CrossRef]
6. Carlson DL. Necrotizing sialometaplasia: a practical approach to the diagnosis. *Arch Pathol Lab Med*. 2009;133(5):692-698. [CrossRef]

7. van der Wal JE, van der Waal I. Necrotizing sialometaplasia: report of 12 new cases. *Br J Oral Maxillofac Surg*. 1990;28(5):326-328. [CrossRef]
8. Fowler CB, Brannon RB. Subacute necrotizing sialadenitis: report of 7 cases and a review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2000;89(5):600-609. [CrossRef]
9. Keogh PV, O'Regan E, Toner M, Flint S. Necrotizing sialometaplasia: an unusual bilateral presentation associated with antecedent anaesthesia and lack of response to intralesional steroids. Case report and review of the literature. *Br Dent J*. 2004;196(2):79-81. [CrossRef]
10. Sen S, Sen S. Therapeutic effects of hyperbaric oxygen: integrated review. *Med Gas Res*. 2021;11(1):30-33. [CrossRef]