

Necrotizing Sialometaplasia and Gastric Disorders: A Rare Case Report

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Abstract

Case Report

Necrotizing sialometaplasia (NS) is an uncommon, self-limiting, benign inflammatory condition, which involves predominantly the minor salivary glands. Its prevalence is 0.03% of oral mucosa lesions biopsied. It is known to be related to local factors that may cause vascular obstruction, with consequent infarction and ischemic necrosis of the salivary glands nevertheless it can be the manifestation of a specific systemic condition. The aim of this case report is to describe the clinical and histopathological aspect of this lesion and to determine the potential association with gastric disorders (GD). We present a case of 45-year-old male patient with bilateral ulcerative lesion on the hard palate who suffered from gastric ulcer and gastroesophageal reflux disease (GORD). A biopsy was performed, and a histopathological diagnosis of NS was established. The possible contribution of gastrointestinal disorders to the pathogenesis of NS is discussed. Given the diagnostic difficulty and risk for aggressive management strategies, necrotizing sialometaplasia is an important diagnostic entity to consider in patients with oral ulcerative lesions. Reassurance and observation are the primary management strategy, as these lesions spontaneously resolve over time.

Keywords: Necrotizing sialometaplasia, ulcerative lesions of oral mucosa, salivary glands, gastroesophageal reflux disease (GORD), case report.

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INTRODUCTION

Necrotizing sialometaplasia (NS) is a benign, locally destructive, inflammatory condition that affects the salivary glands and most frequently the palatal minor salivary glands, first described by Adams *et al.*, in 1973. [1] Its uncommon and represent less than 0.03% of oral mucosa lesions biopsied.[2]

Clinically, necrotizing sialometaplasia manifests as an ulcerative lesion of the oral mucosa. A single lesion in the transition between the hard and soft palate is the most frequent presentation, although bilateral and even multiple lesions with random distribution on the hard palate may appear. The palatal location is not exclusive: other zone with ancillary salivary glands can be affected. However, more rarely, the major salivary glands may also be affected. Exceptionally, the condition may also affect the seromucosal glands of the upper airways; nasal cavities, sinus cavities and nasopharynx. [3]

This lesion is interesting because it may mimic malignant lesions such as squamous cell carcinoma (SCC) or, less commonly, mucoepidermoid carcinoma (MEC) both clinically and microscopically, this may lead to an unnecessary radical treatment in case of misdiagnosis.

Few case reports of NS associated to gastrointestinal disorders were published. We here in, present a rare case of bilateral necrotizing sialometaplasia at the posterior hard palate of a middle-aged male patient with a history of gastric ulcer and gastroesophageal reflux disease.

CASE HISTORY

A 45-year-old man, was referred to our department of periodontology by his general dental practitioner with a two-week history of bilateral palatal swellings that had developed into two deep ulcers after the first three days. [Figure 1A,1B] The patient was complaining of severe pain of the palate.

Amoxicillin/clavulanic acid antibiotics and chlorhexidine mouthwash were administered to the patient by his dentist for 7 days without clinical improvement.

His medical history revealed a gastric ulcer and gastroesophageal reflux disease (GORD) with no history of smoking, alcohol intake, trauma or recent surgery.

Oral examination was performed including assessment of : oral dryness, acid and bitter taste, glossalgia, halitosis, itching, burning and pharyngeal discomfort in relation to his GORD.

A complementary imaging exam, two retro alveolar radiography and cone beam computed tomography (CBCT) revealed no bone loss. [Figure 2A,2B]

Blood tests were demanded : fasting blood glucose, sedimentation rate (ESR), blood count (CBC), serology (HIV, hepatitis B and C, syphilis) and

tuberculin intradermal test (IDR). All of them were negative.

An incisional biopsy of the lesion was performed under local anaesthesia involving the edges, healthy tissue of the palate and a portion of tissue from the central area of the lesion. The histopathological findings showed lobular necrosis of the salivary glands, pseudoepitheliomatous hyperplasia of the covering epithelium and prominent squamous metaplasia of the epithelium of salivary ducts suggestive of a benign fibromatous lesion. The diagnosis of Necrotizing sialometaplasia was retained.

Analgesics were administrated to the patient: 2 g of paracetamol / day for 5 days Adequate oral hygiene (tooth brushing and chlorhexidine rinses without alcohol) was prescribed to prevent the superinfection of the ulcerated area. The patient was referred to his gastrologist for an adequate GORD management. The patient was regularly followed up. After two weeks, a partial healing of the lesions was observed. [Figure 3A, 3B] And after four weeks, the lesions were almost recovered. [Figure 4A,4B]



Figure 1A



Figure 1B

Figure 1A and 1B: Initial clinical aspect shows 2 irregular shaped ulcers in the posterior part of the palate with raised erythematous margins.

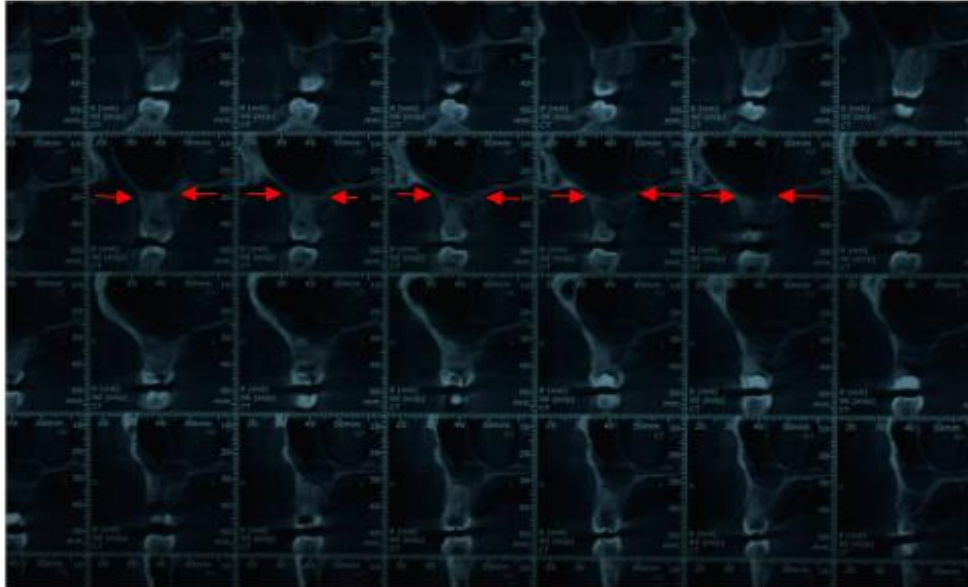


Figure 2A

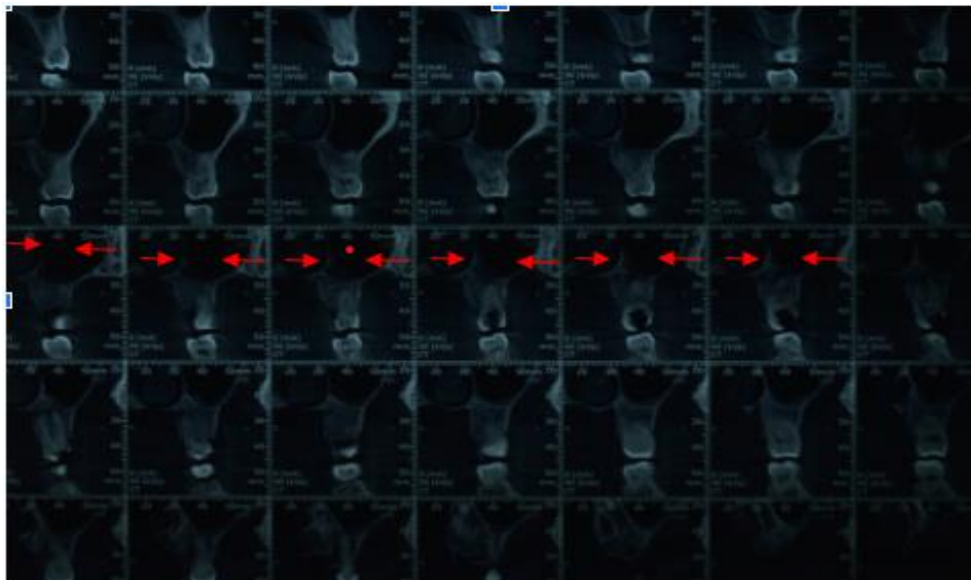


Figure 2B

Figure 2A and 2B: CBCT demonstrating bone integrity: no bone lysis caused by the lesions



Figure 3A



Figure 3B

Figure 3A and 3B: Two weeks-follow-up showing regression of bilateral lesions



Figure 4A



Figure 4B

Fig 4A and 4B: Four weeks-follow-up showing important remission and presence of a mild erythema area.

Table 1: Histologic features of necrotizing sialometaplasia

1	Pseudoepitheliomatous hyperplasia of the overlying squamous mucosa
2	The squamous metaplasia ducts and acini
3	Inflammation secondary to extravasion of mucin
4	Lobular infarction with or without mucin spillage
5	preservation of the normal lobular architecture of the salivary glands

DISCUSSION

The diagnosis of Necrotizing sialometaplasia can be a serious challenge for dentists, it can resemble other more concerning conditions. The main differential diagnoses include Malignant lesions (like squamous cell carcinoma or mucoepidermoid carcinoma), Granulomatosis, Inflammatory conditions (such as syphilis, tuberculosis, Behçet's disease, lupus erythematosus) and other salivary gland disorders (including sialadenitis, sialolithiasis, or benign tumors like pleomorphic adenoma). [4-6]

Given the potential similarity in clinical presentation, histopathological examination is crucial to differentiate necrotizing sialometaplasia from these conditions and to establish an accurate diagnosis. There are 5 key histopathologic features of NS as listed in [Table 1]. [4]

The etiopathogenesis of NS is somewhat controversial: Most authors believed it is related to ischemia which is caused mainly by traumatic injuries, such as accidental injury of maxillary, teeth extraction, trachea after intubation, denture wearing, local anesthetic injecting, alcohol and tobacco use, and cocaine abuse. [4,7]

Bulimia, have been implicated as potential predisposing factor., which is characterized by recurrent episodes of binge eating, followed by compensatory behaviors, including self-induced vomiting by a

mechanical gag reflex. [8] Sporadic case reports have been published that correlated vomiting with NS revealing that the mechanical irritation and acidic environment resulting from frequent vomiting might contribute to the development of necrotizing sialometaplasia in the oral cavity. [9, 10]

In this report we describe a case of NS associated to gastric ulcer and gastroesophageal reflux disease (GORD) which is defined as a gastrointestinal motility disorder that results from the reflux of stomach contents into the esophagus or oral cavity. The oral and maxillofacial manifestations of GORD may include dental erosion, xerostomia, mucositis, aphthous like ulcerations, sour taste, burning mouth, hyperesthesia, bruxism, and/or temporomandibular disorder (TMD). [11]

Clinically, oral lesions are frequently observed in patients with GORD, where gastric contents (pH 1–1.5) consisting of acids, pepsin, bile salts, and trypsin may reflux to the esophagus and reach the oral cavity, leading to high levels of dental erosion and sometimes caries. It may also cause damage to oral soft tissues that are not adapted to their harmful potential. [12]

In an experimental model of rat chronic acid reflux esophagitis, the researchers observed inflammatory cell infiltration in the mucosa of the back of the tongue proving that acid reflux can also lead to an inflammatory response in the oral mucosa. [12-16]

The regurgitation of gastric acids may also contribute to "overload" saliva secretion activity of the minor salivary glands located under the mucosa lining. This demand for additional secretion leads the acinar salivary gland cells to increase activity, manifested by extra production and release of secretory granules which result in hypertrophy of these cells. This condition may contribute, as a predisposing factor, to a locally ischemic event in the minor salivary glands. [9]

The palate is the first oral site to be in contact with the gastric acid content which may explain the predominant location of the ulcerative lesions described in the necrotizing sialometaplasia.

Previously, it was reported a significant decrease in mucosal pH levels in the hard and soft palate in bulimic patients as well patients with gastroesophageal disease leading to an inflammatory response in the oral mucosa and an ischemic necrosis of the salivary glands. Therefore, the exact relationship between necrotizing sialometaplasia and GORD requires further research to establish a definitive causal link, as the current understanding is based on limited observations and case reports.

Consulting with healthcare professionals and specialists is essential for accurate diagnosis and appropriate management of both conditions.

This article may be helpful to other clinicians to be aware of this rare entity in future: oral palatal ulcers more specifically bilateral lesions occur, a meticulous history questionnaire looking for gastrointestinal disorders as a possible etiological factor.

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