Necrotizing Sialometaplasia in a Patient With an Eating Disorder: Palatal Ulcer Accompanied by Dental Erosion Due to Binge-Purging

Tomoaki Imai, DDS, PhD,* and Masahiro Michizawa, DDS, PhD†

This report describes a case of necrotizing sialometaplasia (NS) accompanied by significant dental erosion of the maxillary teeth of the palatal surfaces owing to chronic self-induced vomiting. This observation contributed to the determination of an immediate and appropriate provisional diagnosis of NS in a patient with an eating disorder, which subsequently was confirmed histopathologically as NS. The diagnostic challenges presented by NS associated with eating disorders and its management are discussed.

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Eating disorders (EDs) are a group of psychopathologic conditions that affect the relation between a patient’s body and food consumption, leading to abnormal eating behavior and potential or overt medical complications.1,2 Mucosal, dental, and salivary manifestations have been reported in patients with EDs, and the management of their oral health remains challenging.1 In addition to these oral pathologies, necrotizing sialometaplasia (NS) is considered an uncommon but emerging associated complication.1,3

NS is a self-limiting inflammatory condition that commonly occurs as a deep ulcer in the posterior hard palate.4 This lesion also can be found in major salivary glands and at other sites that contain salivary glands, including the nasal cavity, trachea, buccal mucosa, maxillary sinus, tonsil, and tongue.3,5 The incidence appears higher in male patients, with a male-to-female ratio of approximately 2:1, and particularly is increased in men older than 40 years.4,6 However, Kaplan et al7 recently reported an apparently higher incidence in female patients. This lesion can be misdiagnosed histologically as a malignancy owing to the difficulty in the differential diagnosis from squamous cell carcinoma or mucoepidermoid carcinoma.6 Unlike squamous cell carcinoma, the lobular architecture and bland morphology of metaplastic squamous cells are preserved in NS without significant cellular atypia. NS also can be differentiated from mucoepidermoid carcinoma by the presence of incorporated luminal elements of remnant mucous cells and mucin pools into metaplastic islands, whereas mucoepidermoid carcinoma often exhibits features of cystic spaces lined by neoplastic cells.8 Although the etiology has not been established, it is generally accepted to involve ischemic circulation of the salivary gland lobules followed by an inflammatory response. Clinical examination shows that ischemia typically develops in the oral cavity as a consequence of smoking, local injury, blunt force trauma, denture wear, or surgical procedures.4,6 In other sites, NS can occur in the trachea after intubation and the larynx owing to atheromatous embolization.9,10 It also has been reported in patients with other conditions, including pregnancy, diabetes, sickle cell disease, Buerger disease, lymphoma, upper respiratory tract infections, cocaine abuse, bulimia, and chronic vomiting.4,11-14

Patients with EDs carry a negative mental image of their own body and a lack of self-esteem and, accordingly, are often defensive and unwilling to fully disclose their conditions when visiting a dental clinic with any oral symptom.1,2,15 A diagnosis of NS in these patients is thus largely dependent on the identification of an
objective diagnostic cue. This report describes a case of NS accompanied by significant dental erosion of the maxillary teeth of the palatal surfaces owing to chronic self-induced vomiting. This observation contributed to the determination of an immediate and appropriate provisional diagnosis of NS in a patient with an ED, which subsequently was confirmed histopathologically as NS. The diagnostic challenges presented by NS associated with EDs and its management are discussed.

Report of Case

A 26-year-old woman complaining of an ulcerative stomatitis on the posterior hard palate was referred to the authors’ center. The patient reported that a painful lesion had developed in this location 12 days previously, which was followed by submucosal fluctuation and rupture of the ulcer 4 days later. The patient’s height and weight were 160 cm and 40 kg, respectively (body mass index, 15.6 kg/m²). She was afebrile and had not experienced fatigue or nocturnal sweating. She was a nonsmoker with no apparent history of alcohol abuse, recent surgery, or trauma. Although she described the use of psychiatric services on the interview sheet, she refused to provide further details. She was not prescribed antipsychotic or antianxiety drugs at that time.

Intraoral examination disclosed a 12- × 13-mm ulcer 12 mm deep, with the necrotic surface located on the right side of the hard palate (Fig 1). The ulcer had thickened keratotic borders without tenderness or induration and did not penetrate the nasal cavity. Regional cervical lymphadenopathy was not detectable by palpation. The right side of the submandibular gland was slightly enlarged and tender, whereas the parotid and lacrimal glands were not swollen. Hyposalivation was observed, but keratoconjunctivitis sicca and arthralgia were not apparent. There was dental erosion involving the dentin on the palatal surfaces of the maxillary anterior and posterior teeth and the enamel on the occlusal surfaces of the maxillary molars. This erosive pattern indicated an association with intrinsic acid regurgitation or vomiting, likely an ED. The occlusal surfaces of the dentition displayed no facets created by attrition, which produces a similar amount of wear on the opposing teeth. Dental caries and periodontal disease were not apparent. Laboratory tests showed hypokalemia (potassium level, 2.7 mEq/L) and iron deficiency hypochromic anemia (hemoglobin, 11.5 g/dL; mean corpuscular volume, 74.4 fl; mean corpuscular hemoglobin, 22.7 pg; serum iron level, 24 μg/dL; unsaturated iron-binding capacity, 343 μg/dL). Hematologic tests did not show an inflammatory response. Hepatic and renal functions and serum levels of amylase and soluble interleukin-2 receptor were within normal limits. Serologic testing ruled out intraoral syphilis. Based on these findings, the authors provisionally diagnosed NS in the patient with an ED with binge-purging symptoms, without inquiring about the patient’s dietary habits to avoid losing her trust. The patient refused to consent to imaging procedures, including computed tomography and magnetic resonance imaging, but did approve

**FIGURE 1.** Mirror views of the ulcer in the hard palate and dental erosion. A, The base of the lesion is covered with necrotic debris at the initial visit. The occlusal surfaces of the posterior teeth have depressed cusps and edges, raising the level between the metal inlays and the adjacent tooth surface (arrowheads). Palatal surfaces show a polished cervical appearance of the wear facets involving dentin. In anterior untreated teeth, loss of palatal contour and exposure of dentin are apparent (arrows). B, The ulcer is almost completely healed after 3 weeks.

a histopathologic examination to confirm the inflammatory condition and to exclude neoplasms such as carcinoma or lymphoma.

During the patient’s initial visit, the authors performed an incisional biopsy of the anterior aspect of the ulcer, including the peripheral margin (Fig 2). The specimen featured a mucosal fragment lined with hyperplastic parakeratinized epithelium. Although the underlying lobular architecture of the palatal salivary gland tissue was maintained, some lobules were necrotic with stromal inflammatory infiltration secondary to extravasation of mucin. Squamous metaplasia of ducts was evident with a benign nuclear morphology. These findings were compatible with NS.

On her next visit, conducted in a comfortable setting, the patient was presented with the histopathologic diagnosis and the indication of a possible background of ED with binging-purging. The patient was relieved to learn that her lesion was not malignant and then admitted to binge eating, self-induced vomiting, occasional laxative use, and amenorrhea. The EDs had persisted as anorexia nervosa for more than 10 years and binging-purging developed in the past several years. These inappropriate events had become temporarily more frequent before the manifestation of the initial symptom of the lesion on her palate. Scarring over the dorsum of the patient’s hands (Russell sign) was not evident. During the stable period, vomiting could almost always be self-induced without the application of pressure with a finger. Upon worsening of binging-purging, she admitted to an increase in undue mechanical stress to the palate with food or instruments. The rapid development of the palatal ulcer caused the patient concern, which likely led to a stable level of abnormal eating behaviors by the time of her initial visit. The patient continued regular visits to her psychiatric doctor for further care. Although professional dental examinations had been discontinued for several years, the patient agreed to resume regular visits to her dentist. The palatal ulcer healed spontaneously within 4 weeks without complications.

**Discussion**

**DIFFERENTIAL DIAGNOSIS OF PALATAL ULCER**

The clinical differential diagnosis of a palatal ulcer generally includes traumatic, inflammatory, and infectious processes and malignant neoplasms and requires clinical information, including the course (acute or chronic), symptoms (painful or not), and a patient’s background. Self-inflicted injury by patients with EDs and depression based on a dislike of the body might contribute to the diagnosis. The relatively rapid appearance of an ulcer and the lack of induration at its borders are likely to indicate an inflammatory condition rather than invasive carcinoma or salivary gland tumors, although non-Hodgkin lymphoma, in particular extranodal natural killer/T-cell lymphoma, and the possibility of other sarcomas cannot be completely excluded. A necrotic deep ulcer emerging after possible causative mechanical stress and followed by mucous swelling that can be fluctuant is clinically consistent with NS. With regard to specific inflammatory diseases, serologic testing ruled out intraoral syphilis. An intraoral ulcer owing to tuberculosis is painful, but the patient did not report spontaneous pain or severe tenderness in the ulcer at her initial visit or exhibit the

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**FIGURE 2.** Microscopic appearance of a specimen from the incisional biopsy (hematoxylin and eosin stain). A, Necrotic mucous glands and squamous metaplastic islands show scattered stromal inflammatory infiltration (magnification, x100). B, Metaplastic cells show a benign nuclear morphology with minimal pleomorphism and no atypical mitotic figures (magnification, x400).

characteristic general symptoms of tuberculosis, including fatigue, slight fever, and cough. Intraoral invasive fungal infections such as aspergillosis, a rare condition, usually present as a necrotic ulcer in an immunocompromised host, such as chemotherapy-induced granulocytopenia or acquired immunodeficiency syndrome. Wegener granulomatosis, an autoimmune disease characterized by systemic vasculitis of small arteries and veins, typically presents in the head and neck as rhinitis and sinusitis, accompanied by cough and hemoptysis. The oral mucosa can be involved as granular erythematous gingiva and uncommonly as a palatal ulcer.

An accurate diagnosis is occasionally made only after several alternative tracks of thinking, and that of NS in patients with EDs is no exception. Previous cases have been misdiagnosed histopathologically as carcinoma at initial biopsy and surgically resected. In others, no ED-associated intraoral symptoms other than palatal NS were assessed or presented. The present case is unique in that dental erosion as an objective intraoral clue potentially shared an associated background with the ulcer and thus aided in providing an immediate and appropriate provisional diagnosis of NS, which was subsequently confirmed by histopathology (Table 1).

An incisional biopsy with an equivocal clinical diagnosis such as palatal ulcer might lead to a misdiagnosis without consideration of the differential diagnosis of NS based on histopathology. It is important to note that an accurate clinical diagnosis helps guide the pathologist. Further, the possibility that a malignancy is simultaneously accompanied by NS should be investigated by biopsy.

EATING DISORDERS WITH BINGING-PURGING SYMPTOMS

Anorexia nervosa and bulimia nervosa are the most commonly encountered EDs. Bulimia is characterized by recurrent episodes of binge eating, followed by compensatory behaviors, including self-induced vomiting, excessive exercise, and misuse of laxatives, emesis, and other medications. Individuals with bulimia are frequently of normal body weight or even slightly overweight. In contrast, patients with anorexia maintain a distorted body image, with a fear of gaining weight, and therefore maintain body weights less than 85% adjusted for age. Further, anorexics are more likely to develop microcytic anemia with iron deficiency, whereas bulimics usually are not anemic. Anorexia is further categorized into restricting or binge/purge subtypes. Individuals with the latter subtype regularly engage in binge eating and inappropriate compensatory behaviors to maintain a low body weight. Like bulimia, this subtype also can be associated with vomiting-associated oral manifestations, such as dental erosion.

In the present patient, whose body weight was approximately 75% of that expected for her age, it was concluded, based on her psychiatric consultation, that her condition was best described as anorexia of the binge-eating/purging type. In contrast, to the authors' knowledge, all previous cases of NS associated with EDs reported in the literature have been categorized as bulimia, although body weight and blood test results were not reported. The patients studied in these and the present report share binging-purging symptoms. Anorexia with these symptoms features highly diagnostic transitions with bulimia during the clinical course. As advocated by Frydrych et al., it would be reasonable to assume that the distinction in EDs between vomiting and nonvomiting subtypes is important with respect to the effects of habitual behavior on intraoral tissues.

ORAL MANIFESTATIONS IN EATING DISORDERS

Potential complications of EDs in the oral and maxillofacial regions include dental erosion, caries, decreased salivary flow, periodontal diseases, atrophic mucosa, and sialosis. During vomiting, the gastric contents initially confront the palatal surface of the maxillary dental arch. This surface also often contacts the tongue surface, which retains gastric fluid among its filiform papillae, resulting in the erosion of polished surfaces. Further, the palatal surface of the maxillary dental arch is unlikely to receive direct salivary protection because of its relatively long separation from the openings of the major salivary glands. Frequent vomiting thus leads to the predominantly lingual erosion of the maxillary anterior and posterior teeth. Eroded enamel exhibits uniform, spoon-like surfaces, rounded margins, loss of contour of untreated teeth, and notched incisal surfaces of the anterior teeth. Moreover, mechanical wear leads to abrasion characterized by a sharply cut, flat, and angled appearance. Extrinsic acid usually causes dental wear on the facial and occlusal surfaces as a result of its intraoral entrance from the anterior aspect.

Sialosis is characterized by noninfectious swelling of major salivary glands, particularly the bilateral parotids glands. This manifestation frequently can be found in patients who vomit or purge. In the present case, sialosis might have developed as unilateral swelling of the submandibular gland, although cases without parotid enlargement are an unusual pattern of sialosis.

Compensatory behaviors associated with EDs, such as frequent self-induced vomiting or misuse of laxatives, can alter the electrolyte balance and decrease saliva flow owing to dehydration. Although no serologic assays are available to confirm this diagnosis,
<table>
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<td>NR</td>
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<td>NR</td>
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<td>NR</td>
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<td>NR</td>
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<td>1st, MEC; 2nd, SCC</td>
<td>none</td>
<td>uncertain duration</td>
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<td>Carlson\textsuperscript{5} (2009)</td>
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<td>NR</td>
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<td>Kaplan et al\textsuperscript{7} (2012)</td>
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<td>NS</td>
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<td>unilateral submandibular gland</td>
<td>&gt;10 yr</td>
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Abbreviations: EDs, eating disorders; F, female; MEC, mucoepidermoid carcinoma; NR, not reported; NS, necrotizing sialometaplasia; SCC, squamous cell carcinoma.

hypokalemia may be a specific predictor of recent vomiting episodes.\textsuperscript{28} The degree of dental erosion may be related to the duration of binging-purging.\textsuperscript{29} In the present case, findings of dental erosion accompanied by serum hypokalemia were consistent with the clinical history of a relatively long history of an ED with binging-purging and an increased frequency of these episodes before the manifestation of NS.

Among the possible predisposing systemic conditions for NS, diabetes with atherosclerosis,\textsuperscript{14} autoimmune diseases with related vasculitis,\textsuperscript{5} and Buerger disease with Raynaud phenomenon\textsuperscript{15} can cause a narrowing of peripheral vessels, leading to a compromised blood supply.\textsuperscript{11} Intriguingly, peripheral vasculopathy also has been reported in patients with EDs.\textsuperscript{30,31} Sickle cell anemia predisposes to ischemia owing to an increase in nonfunctional erythrocytes and blood viscosity and has been hypothesized to be a potential underlying cause of NS.\textsuperscript{11}

Local injury with ischemia is considered an etiologic component of NS.\textsuperscript{4,5,32} Rye et al\textsuperscript{13} reported that ischemia likely has a greater impact in the posterior hard palate, which is supplied primarily by a branch of the greater palatine artery, whereas surrounding areas are supplied from multiple vessels. In patients who vomit/purge, undue pressure or microtrauma to the posterior palatal mucosa owing to rapid food ingestion or use of a finger or instrument to induce vomiting is considered a local predisposing factor for NS.\textsuperscript{21,22} When insufficiently protected by a decreased saliva output, the mucosa is likely to be fragile because of direct long-term gastric acid-induced chemical injury and potential nutritional deficiencies.\textsuperscript{1} Because the pain threshold is increased in patients with EDs, especially those with binging-purging,\textsuperscript{33} this characteristic also might contribute to the development of NS in EDs.

In the present case, temporary worsening of binging-purging before the development of NS would likely have led to an increase in traumatic stress and chemical exposure of the palate. Iron deficiency hypochromic anemia could compromise the steady supply of oxygen to the palate locally affected by frequent abnormal eating behaviors. These systemic and local factors might trigger irreversible ischemic changes in the palatal salivary gland tissues for a short time. Details of the mechanisms of NS against a background of EDs with binging-purging await the accumulation of additional experimental and clinical studies.

Burkhart et al\textsuperscript{15} presented a detailed and systematic protocol for approaching patients with suspected bulimia, which aimed to encourage them to agree to comprehensive oral health treatment. Using a secure and private location at an appropriate time and with particular awareness of a patient’s body language, the clinician should suggest possible causes of the oral symptoms, establish the patient’s associations with foods, and assess the patient’s body-image concepts and eating habits. The approach in the present case was consistent with these methods. A systematic approach to establishing a diagnosis of NS associated with EDs should aid clinicians in convincing the patient to consider resuming preventive dental care aimed at minimizing damage to the enamel and oral soft tissues.

The development of oral manifestations in EDs might potentially exacerbate its etiologic factors, such as a lack of self-esteem or a dislike of one’s body.\textsuperscript{1} Considering the gap between the dramatic progression and striking appearance of the ulcer and its self-limiting nature, the proper diagnosis and appropriate management of NS based on an appropriate evaluation of hard and soft oral tissues are challenging responsibilities for oral specialists. A deep palatal ulcer that occurs subsequent to a painful erythematous swelling in a young adult woman with a suspected ED with binging-purging should make NS a differential diagnostic priority.

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References