Necrotizing sialometaplasia associated with bulimia: case report and literature review

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Necrotizing sialometaplasia (NSM) is a self-limiting disorder affecting mainly the minor salivary glands. The significance of NSM resides in its clinical and histopathological resemblance to carcinoma. Few cases of NSM associated with eating disorders have been reported to date. We present here the clinical features and histomorphology of an additional case of bulimia-associated NSM closely mimicking an invasive carcinoma. A high index of suspicion and good communication between clinician and pathologist are essential in recognizing this entity and preventing unnecessary surgical therapy. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;103:e39-e42)**

Necrotizing sialometaplasia (NSM) was initially characterized by Abrams and colleagues in 1973 as a benign, self-limiting, necrotizing process involving mucous salivary glands on the hard palate.[1] Since then it has been described at other sites where minor salivary gland tissues are present, throughout the upper aerodigestive tract.[2] The significance of NSM is that it may histologically simulate a malignant process.[3] This condition is considered to be associated with an ischemic event, and is associated with clinical factors such as smoking, alcohol use, denture wearing, recent surgery, systemic disease, and trauma.[4]

In 1998, NSM of the hard palate was first associated with chronic self-induced vomiting in two bulimic patients.[5] An additional case was reported by Scully and Eveson in 2004.[6] Bulimia nervosa, an eating disorder seen mainly in young adult females, is characterized by the ingestion of large amounts of food, followed by self-induced vomiting or laxative abuse. Bulimia is often associated with borderline weight and nutritional status and patients are often reluctant to admit to the problem or to seek treatment.

We present here an additional case of NSM of the hard palate associated with bulimia that was histologically misinterpreted as carcinoma in the original biopsy by several experienced pathologists in 2 referral centers. Clinical correlation, histopathologic review of incisional and excisional material, and additional consultation resulted in a final diagnosis of NSM. This prevented further surgical intervention.

CASE REPORT

A 32-year-old white woman sought medical advice for a painful lesion of the hard palate. Her past medical history included chronic anxiety disorder and bulimia, for which she received both inpatient and outpatient treatment. Surgical history consisted of a tonsillectomy in 1977, suprapubic hernia repair in 1979, and a left oophorectomy in 1994 to remove a dermoid cyst. The patient had a history of cigarette smoking, and claimed to have recently quit.

The patient reported a 12-week history of a painful lesion on the left side of the hard palate that had began as an area of erythema. The lesion developed into a nodule, which "ruptured" and became an ulceration. She had self-treated with over-the-counter herbal remedies, golden seal and myrrh, which were ineffective. The patient was evaluated in a community hospital emergency room where amoxicillin was prescribed. Two weeks later she was referred to our institution. Extraoral examination revealed no enlargement of the parotid glands or evidence of xerostomia. There was no lacrimal swelling, keratoconjunctivitis sicca, or arthropathy. No cervical lymphadenopathy was noted. On intraoral examination the lingual tooth erosion characteristic of bulimia, and an irregular ulceration with raised borders,

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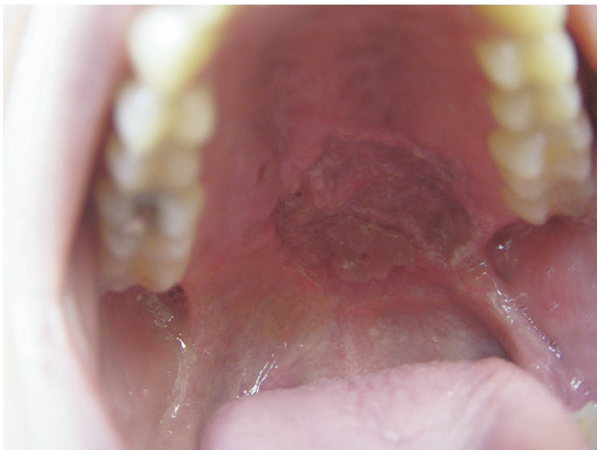

Fig. 1. Clinical photograph shows a broad, shallow, ulceration of the posterior left hard palate that crosses the midline. The mixed red and white lesion has ragged borders and measures approximately 1.5 × 2 cm.

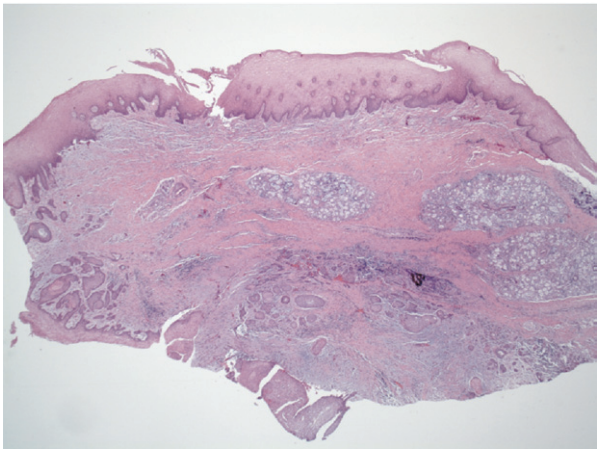

Fig. 2. Biopsy section. Pseudoepitheliomatous hyperplasia (left lower corner) and squamous metaplastic islands with an apparent infiltrative pattern. Note minimal inflammatory infiltrates (hematoxylin and eosin [H&E], original magnification ×4).

1.5 × 2.0 cm in diameter on the hard palate, were noted (Fig. 1). Maxillofacial computed tomography (CT) scan revealed no palatal bony erosion or maxillary sinus opacification. An excisional biopsy of the lesion was performed. The pathologic diagnosis was low-grade mucoepidermoid carcinoma. A second opinion on the biopsy specimen was requested from another referral center. The second diagnosis rendered was well-differentiated squamous cell carcinoma. A wide local resection of the lesion was performed, in which a partial thickness of the palatal bone was resected without creating an oro-antral fistula. There were negative surgical margins intraoperatively. No definitive residual tumor was identified in this specimen. The significant discrepancy between the patient's history of bulimia, and the physical and pathologic findings, raised a suspicion of NSM rather than carcinoma. Retrospective histopathologic examination of the excisional biopsy resulted in a revised diagnosis of NSM, rather than squamous cell or low-grade mucoepidermoid carcinoma. No further surgery was performed. Clinical follow-up at 6 months showed near complete healing of the palatal ulceration, but unfortunately the patient's chronic eating disorder persists despite intensive outpatient counseling.

Histology showed focal ulceration of the overlying epithelium and pseudoepitheliomatous hyperplasia. Despite a focally prominent infiltrative pattern (Fig. 2), the overall lobular architecture of the minor salivary glands was preserved and most epidermoid cells had a bland appearance with only slight cytologic atypia. The remote and recent fibrosis with focal myxoid appearance

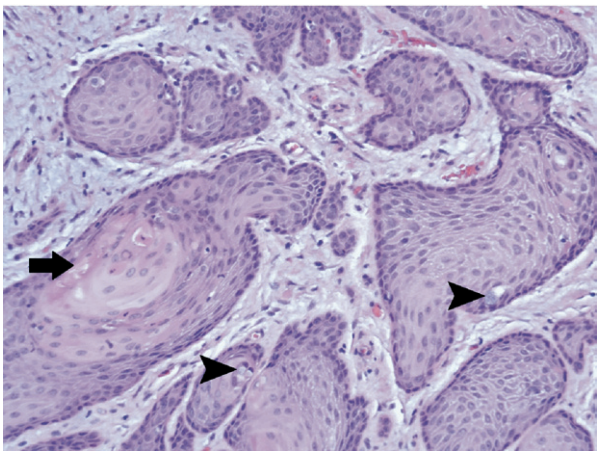

Fig. 3. Metaplastic islands with focal areas mimicking dyskeratosis (arrow) and residual mucous cells (arrowheads). Note the bland nuclear features, myxoid stroma, and lack of inflammation (H&E, original magnification ×20).

was originally interpreted as desmoplastic reaction. The interlobular connective tissue contained scant, focal chronic inflammatory infiltrate, which did not include eosinophils. There were multiple epidermoid nests and islands of squamous metaplasia identified in the ducts of minor salivary glands, some displaying an infiltrating pattern. Some of these nests showed evidence of partial ductal lumina and/or mucous cells (Fig. 3), and occasional mitotic figures were seen (Fig. 4). Collectively, these histological features support the final diagnosis of NSM.

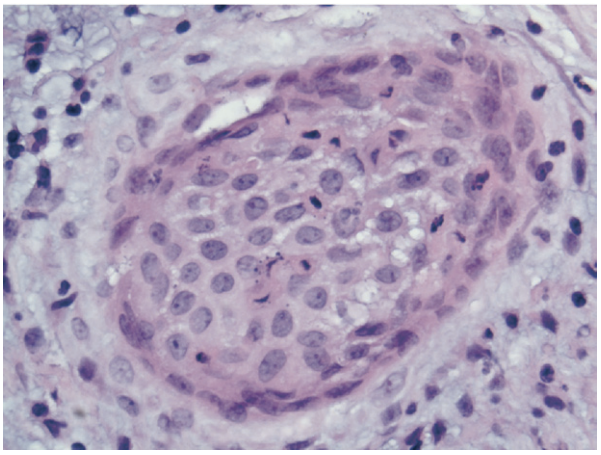

Fig. 4. A high-power view of a metaplastic epithelial island with mitotic figures (H&E, original magnification ×60).

DISCUSSION

NSM is a rare (0.03% of biopsied oral lesions[7]), benign, self-healing, reactive inflammatory process that most frequently affects the minor salivary glands.[8] The pathogenesis is considered to result from ischemia although it has been associated with various traumatic, iatrogenic, environmental, and infectious etiologies.[2] To our knowledge, including the present case, 4 bulimic patients who developed NSM of the hard palate have been reported[5,6] to date. All cases have occurred in Caucasian females, ages 29, 20, 21, and 32, with an average age of 25.5 years. Clinically these cases presented as ulcerations of the hard palate; 3 of the patients were smokers.

Bulimia and other conditions such as gastroesophageal reflux disease (GERD) and chronic alcoholism may cause regurgitation of stomach contents into the oral cavity. In some cases, patients experience 2 or more of these conditions concurrently. Often, affected patients are reluctant to admit current or past episodes of bulimia or alcoholism and a thorough medical, dental, and social history and clinical examination are required to discover the cause of chronic regurgitation. Clinical findings of the oral complications of bulimia include sialadenosis, xerostomia, and lingual tooth erosion.[9] Sialadenosis is a painless, noninflammatory enlargement of salivary glands that is usually associated with an underlying systemic disorder such as diabetes, alcoholism, malnutrition, anorexia nervosa, and bulimia. When secondary to bulimia, sialadenosis usually involves the parotid, either uni- or bilaterally, although involvement of palatal minor salivary glands has been reported.[10] Up to 29% of patients with bulimia may exhibit sialosis.[11] In our patient, there was no significant enlargement of the minor salivary glands adjacent to the lesion (Fig. 1). Patients with a long history of chronic bulimia may develop salivary hypofunction with resulting xerostomia.[6] A characteristic, progressive pattern of tooth erosion is observed, depending on the frequency and chronicity of exposure to gastric acids.[12] Lingual surfaces of the maxillary anterior teeth are severely eroded with demineralization of both enamel and dentin. The lingual and occlusal surfaces of premolars and molars become eroded so that occlusal portions of amalgam restorations are raised, and surrounding occlusal dentin is scooped out by a combination of chemical and mechanical wear. The pathogenesis of NSM is widely believed to be related to ischemic changes[2]; in bulimic patients these might be secondary to chronic mechanical injury of palatine mucosa by induced vomiting.[5] Binge eating and purging to maintain weight is estimated to occur in 15% of the adolescent population.[13] Approximately 1.0% of women and 0.3% of men meet the diagnostic criteria for bulimia nervosa.[14] The clinical scenario of a nonhealing palatal ulcer in an adolescent or young adult patient with clinical signs of bulimia should place NSM high on the differential diagnosis. Squamous cell carcinoma of the hard palate is rare, in addition it is rare in young persons.[15] Common histopathologic features of NSM are overall preservation of the lobular architecture of the minor salivary glands and ductal squamous metaplasia. In a small sample where the architecture cannot be assessed, NSM mimics squamous cell and/or mucoepidermoid carcinoma, and the distinction can be extremely challenging. Moreover, the hyperplastic and metaplastic changes can be florid and the local healing with subsequent fibrosis can simulate an infiltrative process. Histologic features of NSM may have some relation to the age of the lesion, with early lesions exhibiting coagulative necrosis, while older lesions show fibrosis and squamous metaplasia.[16] In the current case, in addition to all these confounding factors, the inflammation was mitigated likely by the antibiotic therapy administered prior to biopsy, depriving the examining pathologists of an important diagnostic clue. The major importance of NSM is the danger of misdiagnosis, because it may mimic carcinoma clinically and histologically. The addition of this case report to the literature strengthens the association of bulimia with NSM. The clinician's awareness of this condition, thorough history taking, and open communication with the pathologist, are imperative in preventing a histopathologic misdiagnosis leading to unnecessary and disfiguring surgery.

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