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Case Report

Conflicting Diagnosis of a Palatal Lesion: A Case Report of Necrotizing Sialometaplasia

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ABSTRACT

Necrotizing sialometaplasia (NS) is a rare condition which can be easily confused with malignancy. Its presentation initially parallels mucoepidermoid carcinoma or squamous cell carcinoma, however, NS is a benign, self-limiting inflammatory disease of the mucus-secreting minor salivary glands. NS is thought to be caused by trauma which induces vascular ischemia of these minor salivary glands. Diagnosing NS continues to be difficult, and clinical history remains one of the pillars of ruling out cancer. This report details a case of NS in a 37-year-old male who presented to the Womack Army Medical Center Oral and Maxillofacial Surgery Clinic with an exophytic mass in his right-hand palate. He had a history of recent trauma induced to the area via incision and drainage to address what was believed to be a palatal space infection. Later, an incisional biopsy was performed after the exophytic mass persisted. The initial local pathology report favored low-grade mucoepidermoid carcinoma, but after review by the Joint Pathology Center at Walter Reed Military Medical Center, the consensus diagnosis was established as NS. The patient returned to the clinic five weeks later with the exophytic mass no longer present and the biopsy site healing well.

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Introduction

Necrotizing sialometaplasia (NS) is a rare, benign inflammatory reaction of both major and minor salivary glands. Its clinical and histologic presentation may imitate squamous cell carcinoma or mucoepidermoid carcinoma. Therefore, NS can be easily misdiagnosed and potentially treated with an unnecessary surgery. Abrams et al is credited as the first to report this reactive necrotizing inflammatory process – specifically involving the hard palate (2). It originates most commonly as the result of trauma, but other known causes include denture use, an adjacent tumor or cyst, injection of local anesthesia, and smoking. The incidence of NS is approximately 0.03% of oral biopsy specimens, making it extremely rare (11). NS occurs most commonly in Caucasians with males more affected than females. It encompasses any age group ranging from 17 to 80 years with a mean age of 50 years in men and 36 years in women. Minor salivary glands – specifically of the palate – comprise 80% of reported cases (4). However, NS can occur anywhere that salivary gland tissue exists, and cases have been reported in the

retromolar pad, gingiva, lip, tongue, cheek, nasal cavity, sinuses, and larynx. Reported cases in extra salivary sites include the lungs, breast, and skin (5-12).

NS initially presents as a non-ulcerative swelling with associated pain or paresthesia. Within a few weeks, necrotic tissue sloughs off, leaving a crater-like ulcer. While the pain has usually subsided at this point, this is when the lesion most closely resembles malignancy as a non-healing ulcer. Histopathologic evaluation reveals acinar necrosis of followed by squamous metaplasia of the salivary ducts. Although the glands are necrotic, they maintain their lobular architecture unlike the dysplasia seen in malignancy. And contrary to malignancy, NS is a self-limiting process that resolves over the course of 5-6 weeks. Surgical intervention is rarely required except for the diagnostic biopsy, and supportive care for treatment of symptoms is sufficient.

Case Report

A 37-year-old male underwent placement of a dental implant at

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edentulous site #3 in conjunction with an indirect sinus lift in 2013 at a different military installation. The patient had returned to that clinic shortly after implant placement complaining of pain and congestion. Within a month, the implant had lost stability. The implant was removed and the site curetted. The patient was lost to follow up due to multiple deployments. Upon the patient's arrival to Fort Bragg in 2016, he was evaluated by a periodontist for restorative therapy at edentulous site #3. A radiopaque mass was noted in the right maxillary sinus on pantomogram, and the patient was referred to the Womack Army Medical Center Oral and Maxillofacial Surgery clinic for evaluation. The patient was asymptomatic at the time of his initial presentation to the Oral Surgery clinic except for the complaint of occasional congestion. Due to the patient leaving the area for a month, conservative management was initially employed by prescribing Augmentin, Sudafed, and Afrin as well as obtaining a cone beam computed tomography image to assess for mucosal thickening in the right maxillary sinus. The patient returned to the clinic seven weeks later complaining of severe right sinus pain and a swelling on the roof of his mouth. A well-defined, fluctuant mass was noted on the right hard palate. All teeth in the right maxilla tested vital, and the palatal mass was determined to be a palatal space infection associated with a communication from the right maxillary sinus. An incision and drainage procedure were performed which yielded no frank pus. The patient was placed on antibiotics and appointed for close follow up.

At the one week follow up, the patient returned with the palatal swelling still present. At that time, a computerized tomography scan of the head was obtained, and the patient was appointed for an incisional biopsy of the lesion. Five days later, a nine by five-millimeter incisional biopsy was performed and submitted for histopathologic examination. The preliminary diagnosis from the hospital's general pathology department favored low-grade mucoepidermoid carcinoma. Therefore, the specimen was sent to the Joint Pathology Center at Walter Reed Military Medical Center for expert consultation. The case was reviewed independently by members of the Head, Neck, and Endocrine pathology sections and reviewed in conference. After comprehensive discussion, the Joint Pathology Center yielded a final diagnosis of NS. The patient was seen three weeks after the incisional biopsy for follow up and to provide pathology results. At that time, the palatal mass was no longer present, and the biopsy site was healing well.

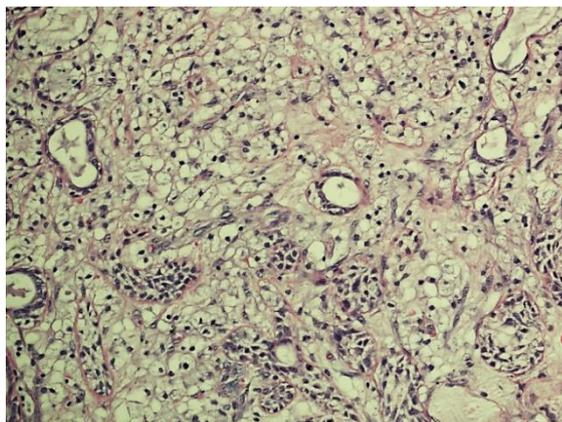


Figure 1.1: Sheets of clear cells intermixed with ductal structures are clearly evident. Muciphages can also be appreciated. Hematoxylin and eosin (H&E) stain x200.

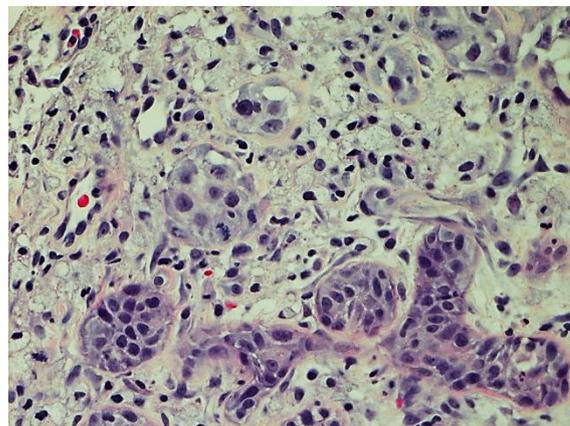


Figure 1.2: The specimen exhibits increased mitotic activity (arrows) in the glandular ductal epithelium, and squamous metaplasia. Hematoxylin and eosin (H&E) stain x400.

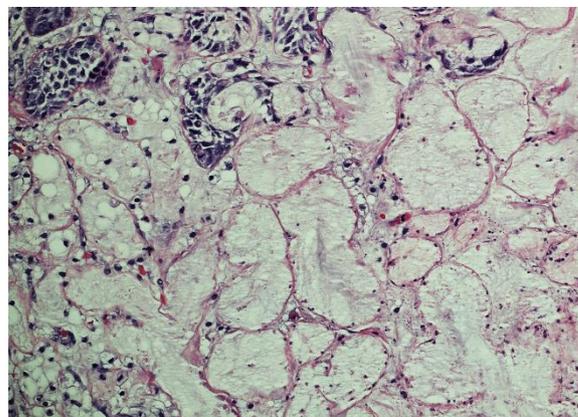


Figure 1.3: Necrotic mucous acini preserving the lobular architecture of minor salivary glands, typical of necrotizing sialometaplasia. Hematoxylin and eosin (H&E) stain x200.

Discussion

NS by definition involves metaplasia of the salivary gland ducts and acini – more specifically, squamous metaplasia from the normal cuboidal epithelium. Also, by definition, necrosis of the salivary gland lobules is involved. This ischemic type of necrosis occurs most frequently at the hard palate. The etiology of NS remains largely a mystery, but the most logical answer appears to be ischemia of vasculature supplying the salivary gland lobules. Direct trauma to the minor salivary glands remains the most likely etiological factor, but other factors that contribute to ischemia include administration of local anesthetic, ill-fitting dentures, alcohol, smoking, cocaine use, radiation, and surgical procedures. While our patient had no recent dental procedures at the time of his initial presentation with the palatal mass, he did have intermittent sinus congestion of the right maxillary sinus for three years following the failed indirect sinus lift and failed dental implant placement. It is very likely that the Schneiderian membrane was violated at the time of this procedure in 2013, and allogeneic bone graft material was introduced into the right maxillary sinus. This chronic nidus could have led to dehydration of the adjacent mucosa, thus making it more susceptible to local trauma. Also, the etiology for the initial palatal mass was skewed when the incision and drainage procedure was

performed eleven days prior to the incisional biopsy. It is possible that the specimen contained necrotic salivary gland tissue as well as early squamous metaplasia caused by the incision and drainage procedure, thus masking the etiology of the initial lesion. In any event, the lesion resolved spontaneously in a fashion very typical of NS.

Approximately two thirds of NS cases involving the palate are unilateral, as was the case for our patient (16,17). The lesion may also occur in the midline. The sizes range from 0.7cm to 5.0cm (average 1.8 cm) (15). NS commonly presents with an ulcer after the necrotic mucosa sloughs off. These ulcerative lesions of the palate simulate malignant processes. Our patient is unique in the fact that his NS lesion presented with pain as most NS lesions are usually painless. In fact, anesthesia of the palatal mucosa is reported as an early indicator of NS (18). The margins of the ulcer are often everted and indurated resembling a carcinoma. Radiographically, most NS lesions show no bony involvement except for a few cases showing saucerization of the underlying bone (3). The appearance and location of the lesion in our patient led our differential diagnosis prior to the incisional biopsy to include pleomorphic adenoma, B-cell lymphoma, and low-grade mucoepidermoid carcinoma. Other possibilities include primary adenocarcinoma of the palate, squamous cell carcinoma, subacute necrotizing sialadenitis, major aphthous ulcer, secondary syphilis, and tuberculous ulcer. The diagnosis of NS relies heavily on clinical history. While biopsy is necessary to confirm the diagnosis, biopsy alone may lead to more misdiagnoses of malignancy and therefore unnecessary surgery or radiotherapy. The diagnosis can be further supplemented via immunohistochemistry demonstrating focal to absent immunoreactivity for p53, low immunoreactivity for MIB1(Ki-67), and the presence of 4A4/p63 and calponin-positive myoepithelial cells. However, hematoxylineosin staining remains the gold standard (19). Anneroth and Hansen described the histopathogenesis of NS by proposing five histological stages: infarction, sequestration, ulceration, repair, and healing (10). Histological features exhibit a spectrum ranging from ulceration, lobular necrosis, sequestration of necrotic acini, pseudoepitheliomatous hyperplasia of adjacent epithelium, squamous metaplasia of ductal epithelium, and inflammatory changes (2). Our case showed pseudoepitheliomatous hyperplasia of epithelium with acinar cell necrosis and squamous metaplasia of ductal epithelium. Most importantly, the lobular architecture was maintained, thus helping the Joint Pathology Center to favor NS over mucoepidermoid carcinoma. Typically, no treatment is required, and the lesion heals by secondary intention within four to ten weeks (average five weeks). One case even reported a full-thickness palatal lesion communicating with nasal cavity that resolved in six months (3). In our case the lesion was almost completely healed at five weeks, and the patient was lost to follow up after that time. In conclusion, NS is a benign self-limiting process of salivary glands. Unfortunately, it can be misdiagnosed as a malignancy resulting in inappropriate treatment. A simple incisional biopsy is required to confirm the diagnosis. In our case, the exact etiology cannot be determined as the chronic sinusitis and the acute phase status post

incision and drainage overlap prior to the incisional biopsy. Fortunately, the lesion did resolve, thus ruling out malignancy and saving our patient from further surgical intervention.

Disclaimer

The views expressed herein are those of the authors and do not reflect the official policy of the Department of the Army, Department of Defense, or the U.S. government.

Disclosure

None of the authors reported any disclosures.

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