Subacute Necrotizing Sialadenitis

A Form of Necrotizing Sialometaplasia?

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Objectives: To report our experience of subacute necrotizing sialadenitis (SANS), an unusual lesion of the minor salivary palatal glands, and to discuss its relationship with necrotizing sialometaplasia (NS).


Setting: Academic center, referral center, and an ambulatory care center.

Patients: Three patients (1 woman, 2 men), aged 22, 23, and 40 years at diagnosis.

Intervention: All 3 patients underwent incisional biopsy.

Main Outcome Measures: Clinical description of SANS, ability to make the diagnosis preoperatively, clinical evolution, histologic features, and comparison with the much more frequent NS.

Results: Three patients presented with a lateral palatal nodule (1 case bilateral, 1 case ulcerated) of 7 to 10 days’ duration, 0.8 to 1.0 cm in size, slightly or not painful. No patient was correctly diagnosed prior to undergoing a biopsy. In all 3 cases, the biopsy specimen showed acinic necrosis surrounded by a dense polymorphous inflammatory infiltrate with atrophy of ductal cells but no squamous metaplasia. Healing occurred without any further treatment in up to 3 weeks. No recurrence was observed in 2 cases; 1 patient was lost to follow-up.

Conclusions: SANS is a painful spontaneously resolving necrosis of the palatal salivary glands, easily misdiagnosed preoperatively. The main differences from NS are smaller size of lesion, scarcity of ulceration, and absence of squamous metaplasia. Although initially described as a new autonomous entity, SANS might be an early or minimal form of NS.


SUBACUTE NECROTIZING sialadenitis (SANS) is a self-limiting inflammatory lesion of the minor salivary glands of unknown cause. It is considered by some authors to be a distinct entity and by others to be part of the spectrum of necrotizing sialometaplasia (NS).

To the best of our knowledge, only 22 cases of SANS have been reported, mostly on salivary palatal glands. Most (77%) of the patients are young men who have spent time living in close quarters such as military barracks. The disease usually presents as a unilateral, erythematous, nonulcerated swelling, often painful, of the posterior hard palate (rarely the soft palate) that heals in a few days or weeks. Incisional biopsy shows a “subacute” inflammation of the affected salivary glands, represented by a mixed infiltrate of lymphocytes and neutrophils. Most of the glandular structures are lost or atrophic, and the remaining acini are necrotic. An infectious and an allergic origin have been suggested, based on the simultaneous occurrence in close-quarter communities and on the rapid spontaneous healing. We present 3 new cases of SANS of the palate occurring in 2 men and 1 woman, and we discuss the possible relationship of SANS with NS.

METHODS

We conducted a retrospective review of records of patients who had biopsy-proven SANS. Between 1996 and 2001, 3 patients treated in the Division of Stomatology underwent incisional biopsy of the palatal salivary glands affected by SANS. Clinical, surgical, and histopathologic records were reviewed for age at diagnosis, presenting symptoms, site of lesion, complications, and recurrences.
The Table summarizes clinical and histopathologic patient data. Patient 1 was a 23-year-old woman who was referred for a slightly painful bilateral symmetric swelling of the hard palate, adjacent to the first molars, of 10 days’ duration. Overlying mucosa was erythematous but not ulcerated (Figure 1). The patient was known to have anorexia nervosa, with occasional crises of bulimia. A biopsy was performed on the larger (about 1 cm) left-side nodule, which had a yellow spot in its center. The 2 lesions healed spontaneously in 3 weeks and did not recur during 2 years of follow-up.

Patient 2 was a 22-year-old man referred for a unilateral nonpainful submucosal nodule of 0.8 cm on the left side of the posterior palate, facing the second premolar, which had been present for about 7 days. The overlying mucosa was slightly erythematosus, and a small yellowish ulcer was present in the center (Figure 2). The lesion completely healed in 3 weeks and did not recur during 4 years of follow-up.

Patient 3 was a 40-year-old man with a 0.8-cm swelling of the lateral hard palate. No clinical details accompanied the biopsy findings. The lesion had almost completely disappeared 2 weeks later, but long-term follow-up was not available.

The 3 biopsy specimens showed similar histopathologic characteristics. The nodules consisted of a group of palatal minor mucous salivary glands showing large areas of necrosis with pyknosis or complete loss of cell nuclei and some homogenization of the cell cytoplasm (Figure 3). Necrotic areas were surrounded by a dense polymorphous inflammatory infiltrate (Figure 4) composed mainly of lymphocytes and plasma cells mixed with a variable amount of neutrophils and a few occasional eosinophils; eosinophils were more numerous in 1 case.
Necrotizing sialometaplasia presents a striking anatomic and histologic resemblance to invasive squamous cell carcinoma. It is a destructive lesion that involves minor salivary glands and presents as a destructive, painless mass involving the oral and nasal mucosa. The lesion is characterized by the replacement of normal salivary gland acinar and ductal epithelium with squamous metaplastic epithelium. The lesion is often associated with a history of smoking and is more common in middle-aged and older adults. The differential diagnosis includes invasive squamous cell carcinoma, chronic obstructive sialadenitis, and benign salivary gland tumors. The clinical and histologic features of necrotizing sialometaplasia are similar to those of invasive squamous cell carcinoma, but the latter typically involves major salivary glands and is associated with a more aggressive clinical course. Necrotizing sialometaplasia is a self-limiting lesion that may resolve spontaneously, but it may also recur if the underlying cause is not identified and treated. The pathogenesis of necrotizing sialometaplasia is unclear, but it may be caused by a combination of local irritants, immune factors, and genetic predispositions. The lesion is thought to result from the interplay between local factors, such as inflammation and ischemia, and systemic factors, such as immune reactions and genetic susceptibility. The treatment of necrotizing sialometaplasia is typically conservative, with surgical intervention reserved for patients who fail to respond to conservative management or have recurrent lesions.
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REFERENCES


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