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 acinar 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.


UBACUTE 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 entity1,2 and by others to be part of the spectrum of necrotizing sialometaplasia (NS).3

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 erythematous, 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.
Adjacent lobules or glands were sometimes normal (Figure 5) or showed an intermediary state with dedifferentiation of acini and ducts with an atrophic cell wall (Figure 6). There was no squamous metaplasia in any of the 3 cases.

**COMMENT**

There have been only 3 published articles dealing with SANS, which suggests that this lesion might be relatively rare. On the basis of clinical and histopathologic features, some authors consider SANS to be an unusual subtype or early stage of NS, while others consider it a distinct specific entity of infectious or allergic cause.

Clinical features that support an infectious (possibly viral) origin for SANS include sudden onset, several simultaneous or nearly successive cases in close living quarters, and in some patients an association with a current or recent upper respiratory tract infection. However, viral particles have never been found in samples of the affected glands. The presence of a few occasional eosinophils and spontaneous healing character of the lesions are not sufficient to prove an allergic origin.

Necrotizing sialometaplasia is characterized by mainly lobular acinar necrosis, whereas in SANS, only focal necrosis is usually observed. Of greatest importance is acinar or ductal squamous metaplasia, a key diagnostic criterion of NS that is not present in SANS. The inflammatory infiltrate in SANS is of a mixed type (subacute), composed of mononuclear cells (lymphocytes and plasma cells) with somewhat numerous neutrophils and sometimes eosinophils.

**CONCLUSIONS**

Subacute necrotizing sialadenitis is an inflammatory necrotizing lesion found in minor mucous salivary glands of the palate. The hard palate is also the preferential site of occurrence of NS, and in both diseases, the etiologic factors still remain obscure.

Differences between SANS and NS might rest on lesion intensity and importance of ischemia. In this respect, SANS might represent an early stage or a minor variant of NS rather than a distinct entity. The evaluation of more cases is needed, including the performance of viral, serologic, and molecular studies together with biopsy analysis, to provide new insights into the cause of SANS and its relation to NS.
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REFERENCES


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