Subacute Necrotizing Sialadenitis
A Form of Necrotizing Sialometaplasia?

Tommaso Lombardi, MD, Dr Med Dent; Jacky Samson, MD; Roger Küffer, MD

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.


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.
Necrotizing sialometaplasia presents a striking anal-
ogy with SANS: a similar patient profile, with involve-
ment at a rather younger age in SANS; the same site dis-
tribution on the posterior lateral hard palate, with the
same spontaneous healing. From the reported cases, it
seems that the average delay preceding diagnosis is shorter
for SANS (about 1 week) than for NS (about 3 weeks).
Usually, NS presents as a large and/or deep ulcer, not or
slightly painful, often mimicking clinically a malignant
tumor, while in SANS the lesion is more often smaller,
painful, and rarely ulcerated. Healing time is longer, up
to 12 weeks for NS, whereas most SANS cases have healed
within 2 or 3 weeks. The main difference between NS and
SANS is the presence on the biopsy specimen of promi-
ient squamous metaplasia in the regenerating necrotic
acini and ducts, a classic problem of differential diagno-
sis with invasive squamous cell carcinoma. It should be
stressed that early cases of NS show only necrosis, and
that squamous metaplasia appears in a more advanced
stage of the disease.

Necrotizing sialometaplasia is thought to be the out-
come of the transient ischemia following vascular occlu-
sion resulting from the vasoconstriction effect of local
anesthesia, trauma from periodontal surgery, tooth in-
fecions, ill-fitting removable prostheses, or adjacent tu-
ors.\(^6,6\) It has also been observed in patients with eating
disorders.\(^7\) Interestingly, 1 of our patients with SANS also
had anorexia nervosa and underwent self-induced vom-
iting episodes. Anorexia might be another cause of is-
chemia: this disease has been associated with distal vas-
culopathy, giving rise in cases of associated trauma to
tissue necrosis of the extremities.\(^8,9\)

According to Fowler and Brannon,\(^2\) an early evalu-
ation of a biopsy specimen may show distinguishing fea-
tures: in SANS, inflammation would appear to be the ear-
eliest event before the acinar necrosis; on the contrary, in
NS, ischemia occurs first followed by the inflammatory
response. Necrotizing sialometaplasia is characterized by
mainly lobular acinar necrosis, whereas in SANS, only
focal necrosis is usually observed. Of greatest import-
ance is acinar or ductal squamous metaplasia, a key di-
agnostic criterion of NS that is not present in SANS. The
inflammatory infiltrate in SANS is of a mixed type (sub-
acute), composed of mononuclear cells (lymphocytes and
plasma cells) with somewhat numerous neutrophils and
sometimes eosinophils.

There have been only 3 published articles dealing with
SANS, which suggests that this lesion might be rela-
tively rare.\(^1,3\) On the basis of clinical and histopatho-
logic features, some authors consider SANS to be an
unusual subtype or early stage of NS,\(^3\) while others con-
sider it a distinct specific entity of infectious or allergic
cause.\(^1,2\)

Clinical features that support an infectious (possi-
bly 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 cur-
tent or recent upper respiratory tract infection. How-
ever, 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 presents a striking anal-
ogy with SANS: a similar patient profile, with involve-
ment at a rather younger age in SANS; the same site dis-
tribution on the posterior lateral hard palate, with the
exception that NS can also affect major salivary glands,
nasal cavity, maxillary sinus, and larynx; the same ne-
crosis of mucous acini of minor salivary glands; and the
same spontaneous healing. From the reported cases, it
seems that the average delay preceding diagnosis is shorter
for SANS (about 1 week) than for NS (about 3 weeks).
Usually, NS presents as a large and/or deep ulcer, not or
slightly painful, often mimicking clinically a malignant
tumor, while in SANS the lesion is more often smaller,
painful, and rarely ulcerated. Healing time is longer, up
to 12 weeks for NS, whereas most SANS cases have healed
within 2 or 3 weeks. The main difference between NS and
SANS is the presence on the biopsy specimen of promi-
ient squamous metaplasia in the regenerating necrotic
acini and ducts, a classic problem of differential diagno-
sis with invasive squamous cell carcinoma. It should be
stressed that early cases of NS show only necrosis, and
that squamous metaplasia appears in a more advanced
stage of the disease.

Necrotizing sialometaplasia is thought to be the out-
come of the transient ischemia following vascular occlu-
sion resulting from the vasoconstriction effect of local
anesthesia, trauma from periodontal surgery, tooth in-
fecions, ill-fitting removable prostheses, or adjacent tu-
ors.\(^6,6\) It has also been observed in patients with eating
disorders.\(^7\) Interestingly, 1 of our patients with SANS also
had anorexia nervosa and underwent self-induced vom-
iting episodes. Anorexia might be another cause of is-
chemia: this disease has been associated with distal vas-
culopathy, giving rise in cases of associated trauma to
tissue necrosis of the extremities.\(^8,9\)

According to Fowler and Brannon,\(^2\) an early evalu-
ation of a biopsy specimen may show distinguishing fea-
tures: in SANS, inflammation would appear to be the ear-
eliest event before the acinar necrosis; on the contrary, in
NS, ischemia occurs first followed by the inflammatory
response. Necrotizing sialometaplasia is characterized by
mainly lobular acinar necrosis, whereas in SANS, only
focal necrosis is usually observed. Of greatest import-
ance is acinar or ductal squamous metaplasia, a key di-
agnostic criterion of NS that is not present in SANS. The
inflammatory infiltrate in SANS is of a mixed type (sub-
acute), composed of mononuclear cells (lymphocytes and
plasma cells) with somewhat numerous neutrophils and
sometimes eosinophils.

Subacute necrotizing sialadenitis is an inflammatory nec-
rotizing 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 le-
esion intensity and importance of ischemia. In this re-
spect, SANS might represent an early stage or a minor
variant of NS rather than a distinct entity. The evalua-
tion 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.
REFERENCES


Call for Photographs

With the January 2001 issue, the ARCHIVES OF OTOLARYNGOLOGY introduced nonmedical photographs as cover art for the journal. We are bombarded with medical and technical information every minute of every day and this is our way of offering you, our readers, a moment to reflect, smile, breathe a little more deeply, maybe even escape for just a second and relax a bit. Do you have a scenic photograph you have taken that you think would make a great cover shot? We’d love to see it! Submissions should be from our readers, reviewers, authors, or anyone affiliated with the journal, and MUST be formatted horizontally. They can be black and white or color and at least 3.5 × 5 in but no larger than 8 × 10 in. If you wish to submit a digital photograph, please see our digital art submission guidelines located on our Web site: www.archoto.com. Due to legal concerns, no recognizable people should appear in the picture, and please include details about where the picture was taken, how you happened to be there, and anything else you think is interesting about the image. We need the photographer’s complete name, highest academic degree, city and state of residence, and a statement explaining how he or she is affiliated with the journal. Send submissions to ARCHIVES OF OTOLARYNGOLOGY, 1440 Clifton Rd NE, Suite 400, Atlanta, GA 30322. If you would like your photo returned, please enclose a self-addressed, stamped envelope. Cover photos will be chosen at the discretion of the ARCHIVES editorial staff.

Michael M. E. Johns, MD
Editor