
Subacute necrotizing sialadenitis: a clinicopathological study

Lakshmanan Suresh, BDS, MS^{a,b} and Alfredo Aguirre, DDS, MS,^b Buffalo, NY
STATE UNIVERSITY OF NEW YORK AT BUFFALO

Objective. To report cases of extra-palatal subacute necrotizing sialadenitis (SANS), an uncommon condition that usually affects palatal minor salivary glands, and to characterize the etiopathogenesis, clinical features, and histology of this lesion.

Study design. Retrospective reviews of records for patients with SANS diagnosed between 1999 and 2005; only cases with complete clinical history and histology were included in the study.

Results. Five cases (3 women, 2 men) were identified. The majority of patients presented with painful 1.0 to 1.5cm swellings, with sudden and rapid increase in size. Two cases occurred in the buccal mucosa, 2 on the ventral surface of tongue, and 1 on the upper lip. Histology showed acinar necrosis surrounded by a dense polymorphous inflammatory infiltrate with focal exuberant tissue eosinophilia. Ductal atrophy was seen with minimal squamous metaplasia. In all the cases, healing occurred without any further treatment in 3 weeks. No recurrence was observed.

Conclusion. SANS is an uncommon, inflammatory condition of unknown etiology affecting minor salivary glands. SANS appears to be a self-limiting process that has distinct characteristic clinical and histologic features. Nevertheless, SANS shares some of the histologic features of early necrotizing sialometaplasia (NS), suggesting a possible relationship between the 2 conditions. Additional reporting of SANS would be helpful in better defining the condition and its delineation from NS. (*Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;104:385-90)

Subacute necrotizing sialadenitis (SANS) is a self-limiting, inflammatory condition of the minor salivary glands of unknown etiology. The lesion typically presents as a localized, palatal swelling with abrupt onset of pain.¹ SANS was first described by Werning et al.² in 1990 and since then to the best of our knowledge, there have been 26 cases reported in the English language literature.¹⁻⁵ All of the cases except 1 have been reported in the palate or tonsillar area.¹ We report 5 additional cases of SANS affecting extrapalatal minor salivary glands.

MATERIALS AND METHODS

A retrospective study of 5 biopsy-proven cases of SANS were obtained from the pathology records of the OMA pathology lab, School of Dentistry, State University of New York at Buffalo from January 2000 to October 2005. All cases were diagnosed as SANS as per the histopathological criteria described by Werning

et al.² Detailed clinical and histological characterization of all 5 cases was carried out, and a literature review was performed.

RESULTS

Clinical findings

A total of 5 cases were found. The ages of the patients ranged between 30 and 70 years with a mean age of 44.2 years. All patients were Caucasian and there was no sex predilection. Two cases occurred in buccal mucosa, 2 on the ventral surface of tongue, and 1 on the upper lip (Table I). In most cases the clinical presentation was a swelling with a recent, sudden rapid increase in size. The duration of the swelling ranged from 2 days to 1 year and pain was present in 4 of the 5 cases. In 1 case, the swelling had been present for more than a year, but the rapid increase in size occurred within a week. The size of the swellings ranged from 1 to 1.5 cm and there were no predisposing factors. All the cases were treated with excisional biopsy of the lesion, followed by a complete recovery within 3 weeks. Long-term follow-up was not available.

Histopathologic findings

All cases showed similar histologic characteristics (Table II). Intact surface mucosa (Fig. 1) and extensive areas of acinar necrosis were seen in all of the cases (Fig. 2). Minimal ductal atrophy and squamous metaplasia were also noted (Fig. 3). There was no evidence of any mucous spillage or fibrosis in any of the cases. A dense mixed inflammatory infiltrate of neutrophils, eosinophils,

^aSenior Resident, Advanced Oral and Maxillofacial Pathology Program, School of Dental Medicine, State University of New York at Buffalo; Currently, Assistant Lab Director, IMMCO Diagnostics, Inc, Buffalo, NY.

^bDirector, Advanced Oral and Maxillary Pathology Program; and Professor, Department of Oral Diagnostic Sciences, School of Dental Medicine, State University of New York at Buffalo.

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Table I. Demographics and clinical features of all cases of SANS reported in the English literature

Author/year	No. of cases	Age (y)	Sex	Race	Presentation	Location and duration (where reported)	Size, cm
Werning et al., 1990 ²	12	Range 18-29 (mean 20)	M	10 W 2 N/A	Firm painful nodules with erythematous and nonulcerated surface	8 hard palate 3 soft palate 1 anterior tonsillar pillar 1 d-4 wk	0.3-1.0
van der Wal et al., 1995 ³	3	17 20 15	M F M	N/A N/A N/A	Painful swelling No painful swelling Painful swelling of some days' duration	Hard palate, bilateral, 10 d R hard palate, N/A R hard palate, some days	0.8 N/A N/A
Fowler and Brannon, 2000 ¹	7	24 15 26 45 37 19 24	F F F M F M M	W W W W W A W	Nodule with recent pain Mildly tender nodule Swelling Nodule Firm tender mass Slightly painful Acutely painful mass	R hard palate, 6 y L, P, hard palate < 1 mo L, P, hard palate, 2 d R hard palate, 3 d R soft palate, 2 d L hard palate, 1 wk R hard palate, 2 d	0.5 × 1.2 0.8 N/A 1.5 2.0 0.5 N/A
Castro et al., 2002 ⁵	1	30	F	N/A	Painful submucosal nodule of 1 mo duration	L buccal mucosa	N/A
Lombardi et al., 2003 ⁴	3	23 22 40	F M M	N/A N/A N/A	Slightly painful nodule, bilateral Asymptomatic Unknown	P hard palate, 10 d P hard palate, 1 wk P hard palate	L 1 R < 1 0.8 0.8
Present cases	5						
1		51	M	W	Painful hard mobile lump with rapid increase in size	R buccal mucosa, 1 wk	1.5
2		30	M	W	Painful swelling with rapid increase in size	Ventral tongue, 5 d	1
3		33	F	W	Painful swelling with sudden increase in size	R upper lip, over 1 y	1
4		70	F	W	Painful swelling with rapid increase in size	R buccal mucosa, 2 d	1
5		37	F	W	Painless swelling with gradual increase in size	Ventral tongue, over 3 wk	1.5

SANS, Subacute necrotizing sialadenitis; M, male; F, female; N/A, not available; W, white; A, African American; R, right; L, left; P, posterior.

Table II. Summary of the histopathological findings of the present cases

Histological features	Case 1	Case 2	Case 3	Case 4	Case 5
Surface ulcer	Not present	Not present	Not present	Not present	Not present
Acinar necrosis	++	++	++	++	++
Atrophy of duct	+	+	+	+	+
Squamous metaplasia	-	Ductal	-	-	Ductal
Fibrosis	-	-	-	-	-
Mucous extravasation	-	-	-	-	-
Eosinophils	+++	+++	++	++	+++
Plasma cells	+	+	+	+	+
Neutrophils	+++	+	+	+	+
Lymphocytes	+	+	+	+	+
Charcot-Leyden crystals	-	-	-	-	-
Inflammation around nerves	Seen	Seen	None	None	Seen
Myositis	Yes	Yes	None	Yes	Yes
Other significant findings	Abscess next to a thick nerve fiber	Epi-myoepithelial islands	Papillary cystadenoma	Ductal ectasia	None

-, Negative; +, mild; ++, moderate; +++, severe.

lymphocytes, and plasma cells was present in the underlying connective tissue (Fig. 4). Strikingly, there was the presence of moderate to severe stromal eosinophilia in all

of the 5 cases (Fig. 5). Charcot-Leyden crystals, which are usually seen in extensive eosinophilia, were absent. Myositis was observed in 4 of the 5 cases.

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