

TMD in Patients with Primary Sjögren Syndrome: A Comparison with Temporomandibular Clinic Cases and Controls

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Aims: *The aim of this study was to investigate the prevalence of temporomandibular disorders (TMD) in patients with primary Sjögren syndrome (1°SS), analyze the impact of the disease on mandibular function, and assess psychosocial distress.*

Methods: *Sixty-three subjects, 60 women and 3 men, participated in the study; 21 1°SS patients were compared with age-matched and gender-matched groups of TMD subjects and controls. Patients were examined according to the Research Diagnostic Criteria for Temporomandibular Disorders. Results:* *Results showed that the subjective, clinical, and radiographic signs of TMD are not more common in patients with 1°SS than in controls. The impact of the autoimmune disease on mandibular function, eg, speech and chewing ability, revealed limitations in oral functioning similar to those in patients with TMD pain. Conclusion:* *Both 1°SS and chronic TMD may be associated with appreciable physical discomfort and psychosocial dysfunction. However, the underlying mechanisms of the oral dysfunction of 1°SS and TMD are quite different and essentially unrelated.*

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Primary Sjögren syndrome (1°SS) is an autoimmune disease that is characterized by lymphocyte infiltration of the salivary and lacrimal glands^{1,2} in particular, but pathologic involvement of the lungs, the kidneys, and the nervous system is common. Arthralgia, myalgia, and inflammatory myopathy are also frequent findings.³⁻⁵ Sjögren syndrome (SS) may exist as a primary condition or in association with other connective diseases such as systemic lupus erythematosus or rheumatoid arthritis, where it is known as secondary SS. Primary Sjögren syndrome exhibits a wide range of signs and symptoms that are related to the autoimmune nature of the disease. The most common complaints—keratoconjunctivitis sicca, xerostomia, fatigue, and myalgia/arthralgia—have been reported to have a profound impact on the quality of life.⁶

The masticatory muscles and the temporomandibular joint (TMJ) have often been found to be involved in connective tissue diseases such as rheumatoid arthritis,⁷⁻⁹ systemic lupus erythematosus,¹⁰ ankylosing spondylitis,^{11,12} psoriatic arthritis,^{13,14} and Reiter disease.¹⁵ Since xerostomia always and myalgia often are present in

patients with 1°SS,^{16,17} which involves and affects the physiologic mechanisms of the masticatory system, it is of interest to investigate the extent to which SS patients also exhibit temporomandibular disorders (TMD). To the authors' knowledge such a specific investigation into the presence of TMD in patients with 1°SS has not been performed previously.

The Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD), which were developed to aid in the diagnosis of TMD,¹⁸ use a dual axis system that allows a physical diagnosis based on pathophysiology to be placed on one axis (Axis I), and TMD-related parafunctional behaviors, psychologic status, and level of psychosocial function to be assessed on a second axis (Axis II). The instrument has been found to give acceptable reliability¹⁹ and to be valuable in the cross-cultural classification of patients.²⁰

The aims of the present study, using the RDC/TMD, were: (1) to investigate the prevalence of TMD in patients with 1°SS, and (2) to analyze the impact of the disease on mandibular function and assess psychosocial distress.

Materials and Methods

Patients

Sixty-three participants were enrolled in the study and divided into 3 groups. The first group (1°SS) consisted of 20 women and 1 man with a mean age of 65 years (range 44 to 78 years) who had previously been diagnosed with 1°SS.²¹ All met the Copenhagen²² and San Diego²³ criteria for SS and the criteria proposed by the European Community study group,²⁴ which consisted of specialists in oral medicine, ophthalmology, and rheumatology. The 21 patients belonged to a group of 34 consecutive diagnosed cases of 1°SS in Östergötland County, Sweden. The patients who volunteered to participate in this investigation—as well as in an intervention study that was presented previously²⁵—did not differ from the remaining known 1°SS patients with regard to age and gender distribution or in baseline secretion rates for unstimulated saliva.^{25,26} The second group (TMD) consisted of 21 TMD patients (RDC diagnosis) from the TMD Clinical Center in Linköping, Östergötland County, Sweden. The third group, which was selected as a control group (C), consisted of 21 general dentistry recall patients from a public dental clinic in Linköping. The patients in the C and TMD groups were matched according to gender and age with 1°SS patients. Thus, each group

consisted of 20 women and 1 man, with mean age of 65 ± 6.3 years for the 1°SS group, 66.0 ± 7.6 years for the TMD group, and 65.0 ± 9.1 years for the C group. There were no additional inclusion or exclusion criteria for the C group. Radiographs and the clinical examination were provided free of charge to all study subjects as compensation for participating in the study. The patients consented to participate in the study after being informed about it and its aims according to the guidelines of the Declaration of Helsinki.

Procedure

All patients underwent a clinical TMD examination and completed the RDC/TMD questionnaire according to the RDC/TMD specifications.¹⁸ Some additional questions were included; they concerned mandibular function, limitations in daily activities, and general physical symptoms. The senior author gathered all clinical data after calibration and assessment for reliability in the use of the RDC/TMD at the University of Washington's Department of Oral Medicine.

Clinical Examination

The RDC/TMD clinical examination (Axis I) involved the clinical assessment of the following TMD signs and symptoms.

Pain Site. Present pain was assessed as ipsilateral or contralateral to pain that was provoked by clinical examination of the masticatory muscles and by tests of jaw function.

Mandibular Range of Motion and Associated Pain. Jaw-opening patterns, vertical range of motion (extent of unassisted opening without pain, maximum unassisted opening, and maximum assisted opening), and extent of mandibular excursive movements (extent of lateral and protrusive jaw excursions) were assessed.

TMJ Sounds. Clicking, grating, and/or crepitus joint sounds were palpated for assessment during vertical, lateral, and protrusive excursions.

Muscle and Joint Palpation. Assessment of extraoral and intraoral masticatory and related muscles (20 muscle sites) was performed by means of bilateral palpation for pain and tenderness. The TMJ (4 joint sites) was also assessed by means of bilateral palpation.

A standardized examination protocol was used to record the pain in 16 extraoral sites and 4 intraoral myofascial sites. A summary score of these myofascial pain sites was calculated for each individual; it ranged from 0 to 20 points.²⁷

TMJ Imaging. The imaging of the TMJ consisted of 3 stages to minimize the radiation burden. The first stage was a panoramic x-ray examination (Siemens Zonarc) of the teeth and jaws including the TMJs for all of the participants. The second stage consisted of zonographic x-ray assessment (Siemens Zonarc) specifically designed for the TMJs in both lateral and frontal views to rule out possible deficiencies in the stage-1 examination, suspected osteoarthritis, or clinical signs of crepitus from the TMJ. When the image quality of the zonographic views was inferior, the third stage consisted of conventional tomography (CGR Stratomatic) in the lateral view.

The tomographic specifications for a positive diagnosis of osteoarthritis were that tomograms show 1 or more of the following: (1) erosion of normal cortical delineation, (2) sclerosis of parts or all of the condyle and articular eminence, (3) flattening of joint surfaces, and/or (4) osteophyte formation. All 1°SS patients and 20 of the 21 C patients were radiographically examined. Since the TMD group was investigated for a concurrent ongoing study, radiographic examination was performed only when clinically indicated; radiographs were taken for 14 of the 21 TMD patients. All radiographs were interpreted by the same person.

RDC/TMD Classification

The RDC/TMD classification system groups the most common forms of TMD into 3 diagnostic categories and allows multiple diagnoses to be made for a given patient. The RDC/TMD diagnostic groups are as follows.

- Group I—Muscle Disorders: (1) myofascial pain; and (2) myofascial pain with limited opening
- Group II—Disc Displacements: (1) disc displacement with reduction; (2) disc displacement without reduction, with limited opening; and (3) disc displacement without reduction, without limited opening
- Group III—Arthralgia, Arthritis, Arthrosis: (1) arthralgia; (2) osteoarthritis of the TMJ; and (3) osteoarthritis of the TMJ

RDC/TMD Axis II History Questionnaire

Questions about age and gender described the study population. Pain intensity was assessed with visual analogue scales (range 0 to 10). Temporal patterns of TMD-related pain and symptoms—pain localization, joint sounds, locking and catching of the TMJ,

tiredness and stiffness of the jaw, parafunctional habits, generalized musculoskeletal pain, and the perception of intraoral mouth pain—were reported on dichotomous scales.

Psychologic status was assessed by the depression score and nonspecific physical symptoms score with subscales of the Revised Symptom Checklist-90 (SCL-90-R).²⁸

Psychosocial functioning was assessed by means of the Graded Chronic Pain Scale, which yields a score of 0 to IV (0 = no pain, IV = severe psychosocial dysfunction). The scale reflects the severity of the impact of TMD on psychosocial functioning at home, work, or school. It also incorporates disability days that were lost because of TMD pain.²⁹

Additional items included 2 questions that assessed the degree of reduction in speech and chewing and provided a global estimate of the overall reduction in daily activities scored on a 0 to 10 scale (0 = “activity without any pain/discomfort at all,” 10 = “activity impossible due to discomfort/pain”).³⁰

Statistical Methods

Analysis of variance was used to test the significance of the differences among groups for continuous measures. A corresponding nonparametric statistical method, Kruskal-Wallis 1-way analysis of variance, was used for variables that were measured on an ordinal scale. This analysis was followed by nonparametric multiple comparison testing. When data consisted of frequencies in discrete categories, the Chi-square test was used to determine the significance of differences among independent groups. In cases in which the expected frequency in any individual cell was less than 5, Fisher's exact test was used for 2 × 2 tables. All statistical tests were 2-tailed and at the $P < 0.05$ significance level.

Results

The distribution of the subjective symptoms that were reported by the patients is shown in Table 1. The 1°SS group showed statistically significantly higher rates, compared to TMD and C groups, for the presence of widespread myofascial pain as well as intraoral pain. Painful or swollen joints other than the TMJ, as well as TMJ clicking, were reported more frequently for the 1°SS group compared to the other groups, but these differences did not reach statistical significance. In contrast, the 1°SS and C groups reported similar rates of occurrence for pain in the face, jaws, or TMJ as well as tiredness and stiffness of the jaws, while the distributions

Table 1 Frequency of Symptoms Reported by Patients in 1°SS, TMD, and C Groups

Symptom	% frequency			P value
	1°SS group	TMD group	C group	
Pain in the face, jaws, or TMJ	28 ^a	95 ^b	33 ^a	< 0.001
TMJ locking/catching	10 ^{a,b}	33 ^a	0 ^b	0.007
Clicking	57	33	33	0.240
Crepitus	19 ^{a,b}	52 ^a	14 ^b	0.018
Tiredness/stiffness in the jaw	24 ^a	71 ^b	10 ^a	< 0.001
Grinding/clenching	33	29	24	0.940
Intraoral pain	62 ^a	19 ^b	5 ^b	< 0.001
Myofascial pain in other parts of the body	71 ^a	24 ^b	24 ^b	0.001
Painful or swollen joints besides the TMJ	57	28	38	0.200

^{a,b} = Frequencies with the same letter are not significantly different at the level of $P < 0.05$ for a given variable (ie, in the first row, 1°SS and C had similar values, which did not differ significantly, while TMD had a higher value that differed significantly from the other 2 groups).

Table 2 Vertical Range of Motion (Mean \pm SD) for 1°SS, TMD, and C Groups

Activity	Range of motion (mm)			P value
	1°SS group	TMD group	C group	
Unassisted opening without pain	47.7 \pm 8.1 ^a	38.2 \pm 9.7 ^b	46.0 \pm 5.9 ^a	< 0.001
Maximum unassisted opening	51.2 \pm 4.8 ^a	43.3 \pm 7.7 ^b	49.7 \pm 5.6 ^a	< 0.001
Maximum assisted opening	52.1 \pm 4.8 ^a	45.2 \pm 6.7 ^b	50.6 \pm 5.9 ^a	< 0.001

^{a,b} = Frequencies with the same letter are not significantly different at the level of $P < 0.05$ for a given variable (ie, in the first row, 1°SS and C had similar values, which did not differ significantly, while TMD had a significantly lower value compared with those of the other 2 groups).

for these symptoms were markedly lower in these groups compared to the TMD group.

Measurements of vertical range of motion of the mandible—unassisted opening without pain, maximum unassisted opening, and maximum assisted opening—are shown in Table 2. The 1°SS and C groups were comparable for these mean range of motion measurements, each of which was significantly smaller for the TMD group.

No significant differences were found among the 3 groups for reports of parafunctional habits or extent of painful or swollen joints other than the TMJ. All 3 groups were comparable in the degree to which TMJ clicking sounds were present, but

the 1°SS group reported significantly fewer crepitus sounds than did the other 2 groups. No significant differences were found between the groups for extent of osteoarthritis as determined radiographically, although the sample sizes may have been too small to allow meaningful statistical comparisons.

Both the 1°SS group and the C group yielded significantly lower numbers of masticatory muscles that were tender upon palpation compared to the TMD group ($P \leq 0.001$). The mean values \pm standard deviation of myofascial pain scores were 6.3 \pm 6.3 for the 1°SS group, 5.8 \pm 4.7 for the C group, and 10.3 \pm 5.3 for the TMD group (Fig 1).

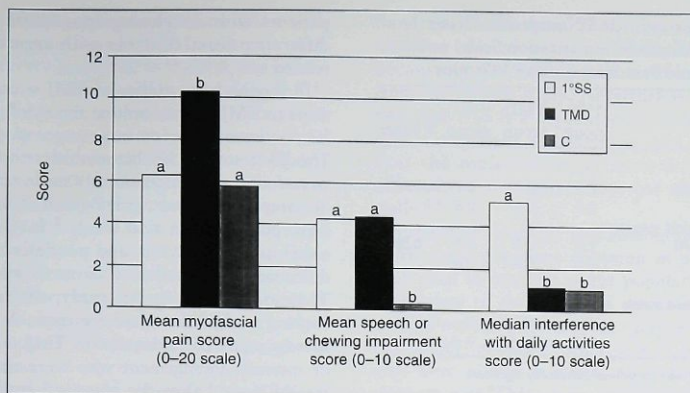


Fig 1 Comparison of 1°SS, TMD, and C groups. a,b = Mean or median of same letter within each group are not significantly different at $P < 0.05$ (ie, for the mean myofascial pain score, 1°SS and C had similar values, which did not differ significantly, while TMD had a higher value that differed significantly from the other 2 groups).

Table 3 Distribution of Diagnoses According to RDC/TMD Criteria for 1°SS, TMD, and C Groups

RDC/TMD diagnosis	% distribution			P value
	1°SS group	TMD group	C group	
Group I (myofascial pain)	29 ^a	91 ^b	33 ^a	< 0.001
Group II (disc displacement)	38	29	33	0.81
Group III (arthralgia, arthritis, arthrosis)	29 ^a	71 ^b	24 ^a	0.003

^{a,b} = Frequencies with the same letter are not significantly different at the level of $P < 0.05$ for a given variable (ie, in the first row, 1°SS and C had similar values, which did not differ significantly, while TMD had a higher value that differed significantly from the other 2 groups).

The distribution of diagnoses according to the RDC/TMD criteria is shown in Table 3. The 1°SS and C groups were comparable with regard to distribution of RDC/TMD Axis I diagnoses, while the TMD group showed much higher prevalence rates for Group I and Group III RDC/TMD diagnoses. No significant difference was found among the groups in rates of occurrence for Group II (disc displacement) TMDs.

The 1°SS and TMD groups were comparable with regard to subjective report of the level of mandibular functioning that is related to speech and chewing ability, while both groups reported significantly higher impairment of mandibular

function compared to the C group (Fig 1). In contrast, the 1°SS group showed significantly greater interference with daily activities as a result of their condition than was reported by the TMD and C groups (Fig 1).

The RDC/TMD Axis II measures of depression and presence of nonspecific physical symptoms, ie, somatization, were comparable across all groups, as shown in Table 4. In contrast, the TMD group showed significantly more psychosocial dysfunction than did either of the other groups.

Table 4 Depression and Nonspecific Physical Symptoms (SCL-90-R Somatization Scale) and Graded Chronic Pain Status in the 3 Groups

Measures	1 st SS group	TMD group	C group	P value
Depression				
Mean (SCL-90-R)	0.9	0.7	0.7	0.41
SD	0.7	0.5	0.5	
Nonspecific physical symptoms				
Mean (SCL-90-R)	1.0	0.9	0.7	0.39
SD	0.9	0.8	0.6	
Graded chronic pain status				
Median	0	2	0	< 0.05
Range	0-2	1-4	0-4	

A value of $P < 0.05$ was considered statistically significant.

Discussion

The 1stSS group consisted of a nonrandomly selected group of patients that comprised 62% of consecutively registered 1stSS cases in their geographic region. The demographic characteristics of this group, ie, mean age and gender distribution, are in agreement with other population-based studies of the region.^{2,21,31} Since 1stSS is a rheumatic disease with potential for causing changes in the TMJ and masticatory muscles that might affect jaw function, we decided to compare 1stSS patients with 2 groups of patients that were age-matched and gender-matched to the 1stSS patients: (1) a group of TMD clinic cases who were known to have pain and dysfunction in the masticatory muscles and/or the TMJ, and (2) a control group of patients who were relatively free of TMD-related symptoms and who were not seeking treatment for signs or symptoms of TMD.

The RDC/TMD has been shown to have acceptable reliability in adult¹⁹ and adolescent populations,²⁶ and it has been shown to be a valid instrument for cross-cultural studies²⁰ of TMD. In the present study, approximately 1 in 4 1stSS and C group subjects reported TMD-related pain, while, not surprisingly, all but 1 of the TMD subjects reported such pain. In studies of other autoimmune diseases that are related to secondary SS, such as rheumatoid arthritis⁷ and psoriatic arthritis,¹³ pain at rest or during jaw function was found to be significantly more common than in disease-free controls. Approximately 1/2 of patients with systemic lupus erythematosus reported pain and/or jaw dysfunction in the masticatory system,¹⁰ while

patients with ankylosing spondylitis showed no difference from controls with regard to TMD-related symptoms.¹¹

In longitudinal studies of TMJ sounds, fluctuations in TMJ sounds among the elderly were similar to those reported in younger populations.³² The TMJ sounds in rheumatoid arthritis patients were found to be significantly more common than in controls; however, grating sounds and clicking were combined in that study.⁸ In patients with ankylosing spondylitis and psoriatic arthritis, no difference compared with controls was found for TMJ sounds.^{12,14} In our study, the frequency of clicking was similar in all 3 groups. A 30-year follow-up study of patients with TMJ osteoarthritis or internal derangement who were conservatively treated found that the signs generally subsided over time.³³ The general conclusion from these studies is that there is no increased risk of developing signs of progressive TMD with increasing age.³²

In the present study, symptoms of fatigue/stiffness in the jaws or cheeks were more commonly reported in the TMD group, which is in agreement with other studies.^{34,35} These symptoms could be interpreted as a result of hyperactivity of the masticatory muscles because of bruxism or other parafunctional habits. In the present study, however, self-reports of bruxism did not differ among the groups; this finding fails to provide supporting evidence for the hypothesis that muscle hyperactivity is etiologic for TMD.

The 1stSS and C groups both showed vertical ranges of motion that were significantly greater than those observed in the TMD group; this is comparable to related differences between TMD groups and controls that have been reported in epidemiologic studies.³⁶ With regard to autoimmune diseases other than 1stSS, patients with rheumatoid arthritis,⁸ psoriatic arthritis,¹⁴ and ankylosing spondylitis¹² showed a significant reduction in vertical mouth opening compared with controls. The myofascial pain score was higher for the TMD group compared to the 1stSS and C groups. Similarly, the 1stSS group resembled controls but differed significantly from the TMD group with regard to masticatory capability.

In population-based studies, burning sensations in the mouth have been found in approximately 3% of the subjects³⁷; in TMD clinical studies, burning has been found in approximately 5% of the patients.³⁵ In our study, the 1stSS group exhibited a high frequency of burning mouth sensations, which is in agreement with other studies of 1stSS patients.²¹ The etiology of burning sensations in

the mouth and tongue seems to be complex, with several interacting factors.^{38,39} In patients with decreased salivary secretion, as in 1°SS, the intraoral mouth pain is often related to candidosis.^{21,40,41}

Generalized muscle and joint pain throughout the body has been reported to be common in 1°SS patients⁶; this coincides with the subjective reports in our study and with findings in a separate investigation of generalized disorders in this patient group (Lundström and Lindström, unpublished data). In a majority of patients with 1°SS, inflammatory myositis or inflammatory perivascular infiltrates have been seen in biopsies.³ In that study the neurophysiologic and immunologic investigation found frequent involvement of the peripheral nervous system.

Bjerrum and Prause⁴² reported that 50% of patients with 1°SS had reported psychologic problems, and 35% requested professional help from a psychologist or psychiatrist. A significant increase in the frequency of depressive symptoms and somatization was reported for 1°SS patients compared with controls and cancer patients.^{43,44} In our study, no significant difference was found between the groups in depression score or prevalence of nonspecific physical symptoms (somatization).

Chronic conditions of any kind may have a devastating impact on a patient's quality of life. The Graded Chronic Pain Scale, which integrates pain intensity, interference in daily activities, and pain-related disability days, was developed to quantify the level of pain-related psychosocial functioning²⁹ for people who were experiencing chronic pain conditions. In our study, the scale revealed that many patients in the TMD group reported an appreciably negative impact on levels of psychosocial functioning much more than did the 1°SS group (or the C group, who of course did not report appreciable levels of chronic pain). In an analysis of the more specific consequences of TMD pain or discomfort on limitation of mandibular functioning, eg, speech and chewing ability, no differences were found between the TMD and 1°SS groups, although both groups showed elevated mandibular dysfunction relative to the C group. However, an evaluation of the global estimate of limitation in daily activities showed that the 1°SS group was much more affected than the TMD or C groups. The implication of this finding is that the widespread changes, including important oral function changes—principally, extreme xerostomia associated with 1°SS—have a much more debilitating overall effect on the quality of life of patients who suffer from 1°SS than masticatory pain has for many of

those who suffer from TMD. This interpretation must be offered with much caution, however, because of the limited nature of relevant data from the present study. Nevertheless, the speculation that 1°SS is a psychosocially disabling condition is consistent with other reports that indicate that the majority of patients with 1°SS are affected not only physically, but also psychosocially.^{6,42}

Subjective, clinical, and radiographic signs of TMD were not more common in patients with 1°SS than in controls in the populations studied. Assessment of the impact of the autoimmune disease on mandibular function, eg, speech and chewing ability, revealed limitations in oral function that were similar in extent to those reported by patients with TMD pain. Both 1°SS and chronic TMD may therefore be associated with appreciable physical discomfort and psychosocial dysfunction. However, the underlying mechanisms of the oral dysfunction of 1°SS and TMD are quite different and essentially unrelated.

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