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RESEARCH ARTICLE

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Temporomandibular joint range of motion measurements in systemic sclerosis patients with temporomandibular dysfunction and comparison with measurements in patients with only temporomandibular dysfunction

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ABSTRACT

Objectives: The main objective of this study was to determine the range of motion of the temporomandibular joint (TMJ) in systemic sclerosis (SSc) patients with temporomandibular dysfunction (TMD) and to compare these data with the measurement results of patients with only TMD.

Method: Patients who presented to the oral and dental clinic of a university hospital were retrospectively evaluated. Seventeen SSc patients with TMD were included in the study (mean age: 42.9 ± 9.4 years; age range: 21–53). The other group consisted of twenty patients who had only a diagnosis of TMD (mean age: 40.4 ± 8.6 years; age range: 29–58).

Results: We detected microstomia in 64.7% of SSc patients with TMD. In the SSc-with-TMD group, mouth opening measurements were lower compared to those with only TMD ($p < .05$). In the SSc-with-TMD group, left laterotrusion movement measurements were lower compared to those with only TMD ($p < .05$).

Conclusions: This study highlights the presence of TMD in patients with SSc and demonstrates impairments in TMJ movements.

KEY POINTS

- This study highlights the presence of TMD in patients with SSc and demonstrates impairments in TMJ movements.
- In our study group, SSc patients with TMD had more restricted mouth opening and left laterotrusion movements compared to patients with only TMD.
- According to our study data, xerostomia occurs in almost half of SSc patients with TMD.

KEYWORDS

Systemic sclerosis; temporomandibular dysfunction; temporomandibular joint; microstomia

Introduction

Systemic sclerosis (SSc) is a rare autoimmune disease characterized by widespread skin fibrosis, vascular abnormalities, and internal organ involvement [1,2]. The prevalence of the disease ranges from 2 to 10 cases per million individuals [3]. Skin involvement, musculoskeletal system, gastrointestinal system, heart, lungs, and kidneys may be observed in patients with SSc [1,4,5]. SSc is classified into two subtypes: limited cutaneous and diffuse cutaneous. Skin fibrosis is confined to the distal extremities, face, and neck in the limited subtype while in the diffuse subtype, the abdomen, thorax, and proximal regions of the elbows and knees are also affected [2,6].

Temporomandibular dysfunction (TMD) serves as a broad umbrella term for various conditions affecting the temporomandibular joint (TMJ) and its adjacent soft tissues, characterized primarily by pain, joint noises, malocclusion, and restricted mouth opening [7,8]. TMD is the most common cause of chronic pain in the stomatognathic system [9,10]. One study found that TMD is a significant public health problem affecting approximately 34% of the general population [11]. The same study estimated that by 2050, the global prevalence of TMD will reach 44%. TMD is diagnosed through a detailed clinical examination and radiological investigation [12,13]. It is reported that more than 90% of SSc patients have signs and symptoms of TMJ [4,14]. In the limited number of previous studies, patients with SSc were found to exhibit more TMD-related symptoms compared to healthy control subjects [15–17]. In SSc patients with TMD, functional limitation is much more frequently due to fibrosis of the facial skin, perioral

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tissues, and masticatory muscles. Joint abnormalities such as condyle resorption, erosive synovitis, and synovial fibrosis can contribute to TMD in these patients [18,19].

TMD encompasses a group of conditions affecting different structures of the stomatognathic system, including the masticatory muscles, articular disc, and TMJ [13,20]. A range of motion within normal limits is an important indicator of a joint's proper function. The most common signs and symptoms of TMD include limited movement in the TMJ [21]. An interincisal distance of less than 40 mm is defined as a microstomia [22]. Microstomia may lead to speech impairment, dental caries and chewing difficulties [21–23].

In SSc patients, due to the pathophysiology of the disease, fibrosis and microstomia in the craniofacial region negatively affect the mechanics of the TMD and the flexibility of the surrounding tissues. However, the number of scientific studies focusing on the development of TMD in SSc patients is still insufficient. A significant portion of the available data consists of limited case series with methodological limitations [18,24–27]. In particular, our literature search did not reveal any study that directly compared TMJ range of motion measurements in individuals diagnosed with SSc and TMD with those in patients with only isolated TMD. This situation leads to a lack of full understanding of the specific quantitative effect of SSc on TMJ range of motion. Based on these shortcomings, the main objective of this study is to determine the range of motion of the TMJ in SSc patients with TMD and to compare these data with the measurement results of patients with only TMD. Therefore, the aim is to investigate the specific effects of SSc on TMJ mobility and to contribute to clinical assessment and rehabilitation approaches for this patient group.

Materials and methods

Study design

In this cross-sectional study, the records of patients who applied to the dental faculty hospital of a university in Türkiye between January 1 2022, and December 31 2023, were retrospectively examined.

Participants

Based on a retrospective review of the hospital information system, 17 adult patients with SSc were identified; these patients met the 2013 American College of Rheumatology/European League Against Rheumatism (ACR/EULAR) classification criteria and were followed up at the Faculty of Dentistry with a diagnosis of TMD [28]. The TMD-only group consisted of 20 patients who applied to the dental faculty during the study period, whose appropriate records could be found in the hospital information system, and who were diagnosed with TMD but not SSc. The study included a total of 17 SSc patients with TMD (14 females, 3 males; mean age: 42.9 ± 9.4 years, range: 21–53) and 20 patients with only TMD (15 females, 5 males; mean age: 40.4 ± 8.6 years, range: 29–58). In our study, the Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) were used for the diagnosis of TMD [29]. Therefore, all participants fundamentally meet these diagnostic criteria. The exclusion criteria for the study were defined as a diagnosis of localized scleroderma, concomitant fibromyalgia, a history of head, neck, or oral cancer, a history of orthodontic treatment, the presence of fewer than 10 natural teeth, and a history of maxillofacial surgery.

Because this was a retrospective study that included all eligible SSc-and-TMD records available within the prespecified two-year period, no a priori sample size calculation determined recruitment. Maximum mouth opening was designated as the primary outcome. For an independent two-group comparison with a two-sided α of 0.05, 80% power, and a large standardized mean difference of $d = 0.90$ based on contemporary dental-research recommendations, 21 participants per group would be required [30]. The available sample (17 and 20 participants) therefore did not reach this benchmark. A sensitivity analysis indicated that these group sizes provided 80% power to detect $d \approx 0.95$. Accordingly, the study should be considered exploratory, and smaller but potentially relevant group differences may have been missed.

Study ethics

Ethical approval for the study was obtained from the ethics committee of a university hospital on June 7 2024, with approval number B.30.2.ATA.0.01.00/433. The principles of the Helsinki Declaration were adhered to throughout the study.

Outcome measurements

Data regarding age, sex, organ involvement–related symptoms, presence of Raynaud phenomenon, disease duration, age at symptom onset, presence of additional chronic diseases, smoking and alcohol use, autoantibody results, and TMJ measurements of patients were obtained from the hospital information systems. (In our clinic, preclinical maneuvers, including routine passive

stretching before measurements, are performed to differentiate joint contracture from transient muscle spasms.) The patients' subtypes of SSc were noted (limited cutaneous SSc and diffuse cutaneous SSc). Limited cutaneous SSc was defined as skin thickening distal to the elbows and knees [2,6]. Maximum interincisal opening, right and left laterotrusion movements and protrusion measurements were obtained from the hospital information system. Maximum mouth opening < 40 mm was considered reduced [22]. To diagnose xerostomia, the unstimulated whole saliva collection test, an objective indicator of hypofunction of the salivary glands, was used [31].

In patients with SSc, the severity of skin thickening is assessed using the Modified Rodnan Skin Score (MRSS). Patients' MRSS results were obtained through the hospital information system. For this score, 17 anatomical body areas are evaluated, including the face, chest, abdomen, upper arms, forearms, fingers, thighs, lower legs, and the dorsum of the hands and feet [32]. The patient's skin thickness is assessed by clinical palpation, and each area was scored on a scale of 0–3 (0 = normal skin; 1 = mild thickness; 2 = moderate thickness; 3 = severe thickness).

Statistical analysis

SPSS version 23.0 was used for statistical analysis. Descriptive statistics were calculated, and the distribution of numerical variables was assessed using the Shapiro–Wilk test. The independent-samples t-test, chi-square test, or Mann–Whitney U test was used for between-group comparisons, as appropriate. Categorical variables are presented as number (percentage), and numerical variables as mean ± standard deviation or median [interquartile range]. Between-group effects for numerical outcomes are reported as mean differences with 95% confidence intervals (CIs) and Cohen's d with 95% CIs. In accordance with dental-research-specific recommendations, absolute d values of 0.10, 0.40, and 0.90 were interpreted as small, medium, and large effects, respectively [30]. Confidence intervals were used to convey the magnitude and precision of the estimated effects [33]. A two-sided p value < 0.05 was considered statistically significant.

Results

Maximum mouth opening was lower in the SSc-with-TMD group than in the TMD-only group (mean difference, −6.60 mm; 95% CI, −11.60 to −1.60; $p = .012$; Cohen's d = 0.98; 95% CI, 0.30 to 1.67), representing a large effect according to dental-specific thresholds. Left laterotrusion was also lower in the SSc-with-TMD group (mean difference, −1.80 mm; 95% CI, −3.43 to −0.17; $p = .034$; Cohen's d = 0.79; 95% CI, 0.11 to 1.46), representing a medium effect. The other movement measurements did not differ significantly between groups, and their confidence intervals included the null value (Table 1).

The clinical characteristics of the SSc-with-TMD group are presented in Table 2.

Discussion

In our study, we detected microstomia in 64.7% of SSc patients with TMD. In our participant group, we found that SSc patients with TMD exhibited greater limitations in mouth opening and left lateral movement compared to patients with only TMD. We found a xerostomia prevalence of 47.1% in our SSc patients with TMD.

The most common signs and symptoms of TMD include limited movement in the TMJ [21]. Previous studies have reported that the prevalence of microstomia in patients with SSc ranges between 52% and 91.4% [4,34,35]. Similarly, in our study, we detected microstomia in 64.7% of SSc patients with TMD. Additionally, reported mouth opening measurements in patients with SSc range from

Table 1. Comparison of demographic and clinical characteristics of SSc-with-TMD group and TMD-only group.

	SSc-with-TMD group (n:17)	TMD-only group (n:20)	*p* value	Cohen's d (95% CI)	Mean difference (95% CI)
Age (years)	42,9 ± 9,4	40,4 ± 8,6	0,389**	0.28 (0.00 to 0.93)	2.50 (−3.57 to 8.57)
Sex (female/male)	14 (82,4)/ 3 (17,6)	15 (75,0)/ 5 (25,0)	1,000*	—	—
Smoking status (yes/no)	3 (17,6)/ 14 (82,4)	3 (15,0)/ 17 (85,0)	0,659*	—	—
Mouth opening (mm)	33,3 ± 9,5	39,9 ± 2,7	0,012**	0.98 (0.30 to 1.67)	−6.60 (−11.60 to −1.60)
Right laterotrusion (mm)	8,4 ± 4,2	8,9 ± 1,5	0,679**	0.16 (0.00 to 0.81)	−0.50 (−2.74 to 1.74)
Left laterotrusion (mm)	6,3 ± 2,9	8,1 ± 1,6	0,034**	0.79 (0.11 to 1.46)	−1.80 (−3.43 to −0.17)
Protrusion (mm)	4,3 ± 1,7	4,7 ± 1,0	0,344**	0.29 (0.00 to 0.94)	−0.40 (−1.37 to 0.57)

Data are presented as mean ± standard deviation or n (%). Mean differences were calculated as the SSc-with-TMD group minus the TMD-only group. Cohen's d values are presented as absolute values; values of 0.10, 0.40, and 0.90 indicate small, medium, and large effects, respectively. Cohen's d was not calculated for categorical variables. *Fisher's exact test; **Independent-samples t-test. CI: Confidence interval; SSc: Systemic sclerosis; TMD: Temporomandibular dysfunction.

Table 2. Demographic and clinical characteristics of the SSc-with-TMD group ($n = 17$).

Disease duration (years), median [interquartile range]	7,0 [10,0]
Age at disease onset (years), mean ± SD	35,1 ± 9,3
SSc subtype (limited/diffuse), n (%)	10 (58,8)/7 (41,2)
Additional chronic disease (yes/no), n (%)	6 (35,3)/11 (64,7)
Raynaud phenomenon (yes/no), n (%)	14 (82,4)/3 (17,6)
Gastrointestinal symptoms (yes/no), n (%)	12 (70,6)/5 (29,4)
Dyspnea (yes/no), n (%)	7 (41,2)/10 (58,8)
Xerostomia (yes/no), n (%)	8 (47,1)/ 9 (52,9)
Microstomia (yes/no), n (%)	11 (64,7)/6 (35,3)
Global MRSS, mean ± SD	17,1 ± 7,9
Antinuclear antibody positivity (positive/negative), n (%)	13 (76,5)/4 (23,5)
Anti-Scl-70 positivity (positive/negative), n (%)	3 (17,6)/14 (82,4)

SSc: Systemic Sclerosis, TMD: Temporomandibular dysfunction, MRSS: Modified Rodnan Skin Score.

33.2 to 40.1 mm in the literature [4,15,36–39]. In our study, mouth opening measurements (33.3 ± 9.5 mm) obtained in the SSc-with-TMD group were among the lowest measurements reported in the literature for patients with SSc. Microstomia is common in SSc patients and is also highly prevalent in SSc patients with TMD. This significant limitation in mandibular function and our finding of one of the lowest mouth opening values in the literature are related to the complex effects of SSc pathophysiology on the craniofacial region. In this context, although our study primarily examines TMD in SSc patients, the duration of the disease, the severity of skin fibrosis, the SSc subtype, and the possible effects of the applied immunosuppressive treatments on mandibular mobility should not be overlooked [15,36,40].

In our study, mouth opening movements were more restricted in SSc patients with TMD compared to patients with only TMD. Additionally, in our study, we found that left laterotrusion movement was lower in SSc patients with TMD compared to patients with only TMD. In SSc patients with TMD, not only mouth opening but also laterotrusion movements should be considered in treatment planning. In line with our study data, one study showed that SSc patients with TMD had more restricted measurements of maximum mouth opening, left laterotrusion, right laterotrusion, and protrusion movements compared to healthy controls [18].

In patients with SSc, salivary secretion rates may be reduced due to glandular fibrosis or secondary Sjögren's syndrome [36]. Xerostomia may lead to increased rates of dental caries and a higher frequency of Candida infections [41]. In one study, it was reported that xerostomia was more uncomfortable for patients than having a reduced mouth opening in SSc patients [42]. Although data are limited, the reported prevalence of xerostomia in patients with SSc ranges from 29.2% to 80% [4,34,35]. In line with the literature, we found a xerostomia prevalence of 47.1% in our SSc patient group with TMD. Nearly half of patients with SSc and TMD experience xerostomia, a finding that should be taken into consideration in clinical practice. Additionally, in SSc patients, secondary Sjögren's syndrome and medication use, which can contribute to xerostomia, should also be considered when interpreting these data.

The principal limitation is the small sample size, which reflects the rarity of SSc and the eligibility criteria. The available sample was smaller than the 21 participants per group required to detect a dental-research benchmark large effect ($d = 0.90$) with 80% power and was adequately powered only for effects of approximately $d \geq 0.95$. Consequently, the estimates are imprecise, as reflected by their wide 95% CIs, and null findings should not be interpreted as evidence of equivalence; Type II error remains possible. The sample size also precluded reliable relationship or regression analyses within the SSc-with-TMD group and multivariable adjustment for potential confounders. Other limitations include the retrospective design and possible missing data, lack of information on SSc treatments, possible measurement variability in retrospectively recorded TMJ mobility assessments, and possible selection bias in forming the comparison group. Therefore, the results should be regarded as exploratory. The strengths of our study include being, to our knowledge, the first study to directly compare the TMJ range of motion of SSc patients with TMD and the patients with only TMD, and providing a detailed characterization of a rarely studied clinical population.

This study highlights the presence of TMD in patients with SSc and demonstrates impairments in TMJ movements. Furthermore, according to our study data, in SSc patients with TMD, mouth opening and left laterotrusion movements are more restricted compared to patients with only TMD. In our study patient group, xerostomia was observed in almost half of the SSc patients with TMD. This study provided data that could form the basis for future studies on the

clinical characteristics of SSc patients with TMD. Larger, multicenter trials are necessary to better understand how TMD affects quality of life in this population and to establish optimal treatment protocols.

Author contributions

CRediT: **Köksal Sarihan MD, MSc, PhD:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing; **Doğan Durna DMD, MSc, PhD:** Conceptualization, Data curation, Investigation, Project administration, Resources, Validation, Writing – original draft, Writing – review & editing; **Ayhan Kul MD, MSc, PhD:** Conceptualization, Data curation, Methodology, Resources, Software, Supervision, Writing – review & editing; **Binali Çakur DMD, MSc, PhD:** Conceptualization, Data curation, Formal analysis, Methodology, Supervision, Writing – review & editing.

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Data availability statement

The datasets for this study are not publicly available due to patient confidentiality but are available from the corresponding author upon reasonable request.

Ethics approval

Ethical approval for the study was obtained from the ethics committee of a university hospital on June 7 2024, with approval number B.30.2.ATA.0.01.00/433. The study was conducted in accordance with the principles of the Declaration of Helsinki.

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