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Effect of orofacial exercises on oral aperture in adults with systemic sclerosis

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Abstract

Purpose—To examine the effect of a home orofacial exercise program on increasing oral aperture among adults with systemic sclerosis (SSc).

Method—Forty-eight adults with SSc were assigned randomly to the multi-faceted oral health intervention or usual dental care control group. Participants with an oral aperture of < 40 mm in the intervention group received an orofacial exercise program, which included daily manual mouth-stretching and oral augmentation exercises twice a day with a total of 6 minutes for 6 months. The outcome measure was oral aperture which was measured at baseline, 3-months, and 6-months intervals.

Results—A significantly larger increase in oral aperture for participants received the orofacial exercise program was found when compared to those in the usual care at 3 months (P=0.01), but not at 6 months evaluation. Participants' adherence rate to the exercise program was low (48.9%).

Conclusions—The orofacial exercise program intervention for adults with SSc and microstomia did not show significant improvement at 6 months. In addition to the low exercise adherence rate, insufficient frequencies, repetitions, and durations of the orofacial exercises may contribute to these results.

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Keywords

Scleroderma; microstomia; interincisal distance

Introduction

People with systemic sclerosis (SSc, scleroderma) frequently exhibit unique orofacial manifestations, such as microstomia or small mouth. Small mouth and mouth furrows are the two top orofacial concerns reported by adults with SSc.[1] Microstomia is defined as an interlabial distance less than 45 mm or an interincisal distance less than 40 mm.[2] In severe microstomia, the interincisal distance is less than 30 mm.[2] Microstomia in SSc is mainly caused by submucosal collagen deposits which contributes to fibrosis in perioral tissue.[1] Anywhere from 43% to 80% of adults with SSc display microstomia;[3–5] the mean interincisal distance for adults with SSc is about 33 mm.[5,6]

In addition to the psychological distress secondary to facial disfigurement, microstomia can result in multiple functional debilitating sequelae, such as drooling and difficulty with mastication and speech.[2,5,7] As a result, microstomia can have a profound negative impact on the relationships of patients with others (e.g., social interaction) and on their quality of life.[7] Reduction in mouth opening may also interfere with normal oral hygiene and dental treatment, and has been shown to be associated with dental caries in people with SSc.[8]

Preventive measures through mouth-stretching and oral augmentation exercises have been shown to reverse the progression of microstomia.[2,9,10] In their pioneering study, Naylor and associates[2] compared their newly designed mouth-stretching and oral augmentation exercises (experimental condition) to a set of traditional orofacial grimacing exercises (comparsion condition) in a randomized, controlled trial in nine patients with SSc (5 in the experimental group, 4 in the comparison group). Both groups showed an increase in maximal oral aperture after 3 months of exercise, but no statistically significant difference was found between the two groups, perhaps due to insufficient statistical power from such a small sample size.

Subsequent studies on the evaluation of mouth-stretching and oral augmentation exercises either employed a single group research design without a control group,[8,10] or compared a home orofacial exercise program (i.e., including mouth-stretching and oral augmentation exercsies) to physiotherapy on the orofacial area plus a home exercise program.[9] It is, therefore, unclear whether Naylor and associates' home exercise program is more effective than no prescribed orofacial exercise.

The purpose of this study was to investigate whether adults with SSc who received an orofacial exercise program will show a significant improvement in the size of oral aperture when compared to a usual care control group at intervals of 3- and 6-months post baseline. This study was part of a larger trial evaluating the effect of a multi-faceted intervention program on oral health status in people with SSc employing a single-blind, randomized, controlled design.

Methods

Participants

Study participants were recruited through the rheumatology clinic at a university facilitated by its local connective tissue disease database (CTDD). The CTDD contains medical

information on the majority of patients with SSc who received consultation and/or treatment at the university rheumatology clinic beginning in 2001. Participants eligible for the study were adults (aged > 18 years old) who were diagnosed with SSc at least 1 year prior to study baseline evaluation, and fulfilled the American College of Rheumatology preliminary classification criteria for SSc,[11] and were diagnosed with SSc at least 1 year prior to study baseline evaluation. Exclusion criteria were localized scleroderma (e.g. morphea, linear scleroderma, and en coup de sabre), less than 10 natural teeth, an upper and/or lower full denture, requirement for antibiotic therapy prior to dental examination, use of a rechargeable, oscillating-rotating-pulsating or sonic powered toothbrush, or an adapted flossing device similar to Reach® Access™ Flosser, performance of mouthstretching exercise on a regular (e.g., daily) basis, complaint of any major jaw joint problems (e.g., severe pain or dislocation), or currently receiving periodontal disease treatment.

Procedures

Informed consent was explained and completed at the university research dental clinic, and baseline mouth evaluation was conducted. Following the baseline evaluation, eligible participants were randomly assigned to one of two groups: a multi-faceted oral health intervention, or a usual dental care control group. Participants in the multi-faceted oral health intervention group were each given a a rechargeable, powered Oral-B® oscillating-rotating-pulsating toothbrush and a Reach® Access Flosser, and taught how to use them. Participants in the control group were each given a manual toothbrush (Oral-B® Complete Advantage Deep Clean toothbrush) and dental floss (Crest® glide shred guard floss).

A block randomization was used with a block size of seven and an allocation ratio of 4:3, which led to random assignment of 4 participants to the intervention group and 3 to the control group. Participants in both groups received an individual, face-to-face training session at baseline, and were instructed to implement the learned oral hygiene techniques twice a day at home for 6 months. The protocol was approved by the Institutional Review Board of the university where the study was conducted.

Orofacial exercises

Participants who were assigned to the intervention group with an oral aperture of less than 40 mm were taught to perform manual mouth-stretching and oral augmentation exercises by a trained research coordinator. Briefly, the manual mouth-stretching exercise involved placing the right thumb at the corner of the left side of the mouth and the left thumb at the corner of the right side of the mouth.[2,10] Participants were instructed to simultaneously stretch both sides of the mouth horizontally as far as they could and hold this position for 15–20s and then rest for 10s before repeating the stretching. The oral augmentation exercise involved inserting a wood stick (2cm × 1.5cm × 9.5cm; provided to participants) between the upper and lower teeth at one side of the mouth corner. Participants stretched the mouth opening by turning the stick on the corner and gently pushing the stick as far back towards the posterior teeth as possible.[10] The participant held the stick in this position for 15–20s and then removed the stick and rested for 10s before repeating the entire process on the opposite side of the mouth. Participants were instructed to repeat each exercise three times consecutively, and to perform each type of exercise two times a day (morning and evening). Handouts with pictures showing the exercises were given. The duration of stretching was based on the efficacy evidence of studies on stretching exercise to improve joint range of motion (ROM) in which 15s and 30s duration of static stretching are equally effective as long as the total daily duration of stretching is the same.[12,13] Prolonged stretching beyond 60s would not result in significant gain in ROM.[12,13]

Self-monitoring

Participants received monthly calendars to keep a record of their daily oral hygiene and were resupplied with these at the 3-month evaluation. On each day of the study follow-up, participants were asked to record whether they had brushed their teeth, flossed, and performed orofacial exercises (intervention group only) by marking “yes” or “no” on the calendar. At the end of each month, participants mailed the completed calendar back to the research coordinator in a self-addressed, stamped envelope. Telephone reminders were made to those participants who had not returned their calendar.

Maintenance phase

The maintenance phase consisted of three telephone calls (about 15–20 mins each) at 2-week, 2-month, and 5-month intervals post baseline evaluation. These monitoring phone calls aimed to encourage compliance and to answer any questions or issues that the participants had in relation to implementing the oral hygiene (and orofacial exercises) protocols.

Blind assessment

At 3- and 6-months post baseline, participants received another clinical mouth assessment. Two calibrated dental hygienists completed the oral exams and conducted mouth evaluations at both baseline and each of the two subsequent assessments.[14] Each assessment was exactly the same as the baseline evaluation. Efforts were made to have the same examiner stay with the same participant at each assessment. The oral examiners were blinded to the participants’ group assignment. Participants were blinded to the other type of intervention available in the study. To avoid bias in the collection of the outcome measures, the oral examiners were instructed not to ask participants about any treatment-related issues of the study. Based on the analysis of data from 21 adults with SSc who had their mouth evaluated simultaneously by the two dental hygienists, the intra-class correlation coefficient [ICC (2,3)] for the interrater reliability on the size of oral aperture exceeded 0.99, which indicates excellent agreement.

Outcome measure of the orofacial exercise

The primary outcome measure was the change in the maximum oral aperture from baseline to the 3- and 6-months post baseline evaluations. Maximum oral aperture was measured, using a small metal ruler, as the distance in mm between the upper and lower incisal edges of the right central incisors when the participants were requested to open their mouths as wide as possible.[2] If the right central incisors were absent, the left central, right lateral, or left lateral incisors were substituted in that order.[2] Three successive trials of maximum oral aperture measurement, with a 5s pause in between each measurement, were conducted and recorded.[15]

Data analysis

An average of the three successive trials for each oral aperture measurement was computed for comparisons between and within groups. We used intent to treat analysis to compare the mean oral aperture changes at baseline, 3, and 6 months between and within the intervention and control groups. Participants who did not complete the 3 or 6 months evaluation were considered not to have had any changes in oral aperture. In addition, for participants with less than 40mm oral aperture at baseline, we used an appropriate to treat analysis approach to compare the mean oral aperture changes at baseline, 3, and 6 months between those with orofacial exercises instruction given in the intervention group and no-exercise instructions in the control group, as well as comparisons within each group. Between group oral aperture changes at baseline, 3, and 6 months were as follows: change from baseline (B) to 3 months

(F1), i.e., first change = F1-B, change from 3 months (F1) to 6 months (F2), i.e., second change = F2-F1, and change from baseline (B) to 6 months (F*), i.e., overall change = F*-B (F*=F2, unless F2 was missing, in which case the value of F1 was imputed). For within group oral aperture measure, we evaluated the first change, second change, and overall change.

Since the oral aperture change measures did not meet the assumptions of normality, non-parametric statistical methods were used to analyze the data. Specifically, the Mann-Whitney U-test was used to evaluate significant increase (one-sided at $\alpha = 0.05$) in the size of oral aperture for the intervention or orofacial exercise group when compared to the control or no-exercise group, respectively. The Wilcoxon signed rank test was used to evaluate significant differences (two-sided at $\alpha = 0.05$) within groups per change measure.

Results

There were 48 participants eligible for the study, of whom 26 were assigned to the intervention group. Descriptive statistics for the study sample are listed in Table 1. The mean (SD) age of the participants was 50.7±13.0 years old, ranging from 22 to 76 years; 79.2% were female; 54.2% were African American. The mean (SD) disease duration based on the available date of diagnosis (first non-Raynaud phenomenon manifestation of SSc) was 7.6±6.1 years. The ratio of participants with diffuse cutaneous SSc to participants with limited cutaneous SSc was 7:9 (see Table 1)

Six participants (3 from intervention group) dropped out after completion of the baseline evaluation, and one participant (from control group) forgot to bring her maxillary partial denture at each of the follow-up evaluation (no oral aperture measurement was performed). As a result, there were 41 participants who had the 3-months oral aperture assessment data. An additional three participants (1 from intervention group) did not return for the 6-months assessment. Some of the known reasons for participant drop out included sickness, diagnosis of cancer, incarceration, complaint of sore throat after the dental cleaning in the intervention group; and hip replacement, deceased, military service, and unable to re-schedule the final visit before the termination of the study in the control group.

Among the 48 participants, 28 had a mean oral aperture size of less than 40mm at baseline and were considered for appropriate to treat analysis. Of these 28 participants, 13 received orofacial exercises instruction. Among participants included in the appropriate to treat analysis, their mean (SD) age was 51.3±12.3 years old, ranging from 23 to 76 years, with 78.6% of them being female, and 57.1% were African American. The mean (SD) disease duration based on the available date of diagnosis was 8.6±7.1 years. The ratio of participants with diffuse cutaneous SSc to participants with limited cutaneous SSc was 1:1. (see Table 1).

Adherence rates

Adherence rates were determined by computing the ratio of the number of exercise sessions performed by the participants to the number of sessions requested. Of the 13 participants who received the orofacial exercises instruction, two did not have the exercise record because they did not return the monthly chart. The mean (SD) adherence rate of the 11 participants was 48.9% ±32.6% with only 3 participants achieving 70% or higher adherence rate.

Intent to treat analysis for between and within group comparisons

Compared to the control group, the intervention group showed a significantly larger change (i.e., increase) in the size of oral aperture from baseline to 3 months (i.e., first change;

P=0.04), but not from baseline to 6 months (i.e., overall change; P=0.19) (see Table 2). There were significant differences in the overall change of the oral aperture size in both the intervention and control groups (see Table 2).

Appropriate to treat analysis for between and within group comparisons

Compared to the no-exercise group, the orofacial exercise group demonstrated a significantly larger increase in the size of the oral aperture on the first change (P=0.01), but not on the overall change (P=0.19) (see Table 3). There was a significant difference in the overall change of the oral aperture size in the orofacial exercise group but not the no-exercise group (see Table 3).

Discussion

The results from our study do not support the hypothesis that adults with SSc who receive the orofacial home exercise program show a significant increase in the size of oral aperture, when compared to the no-exercise/control group. Results of the analysis using the intent to treat approach agree with those using the appropriate to treat approach.

At 3 months evaluation, there was a significant increase in the size of the oral aperture within the orofacial exercise / intervention group and a slight decrease in the size of the oral aperture within the no-exercise / control group. Among participants in the orofacial exercise group, mean improvement in oral aperture was about 2.8mm from baseline to 3 months, which is much less than the improvement (4.8mm) reported by Maddali-Bongi et al.[9] Frequency, repetition, and duration of stretching of the orofacial exercises may contribute to this observed difference. In Maddali-Bongi et al's study,[9] patients with SSc in the control group were requested to perform 3 types of home orofacial exercise: mouth stretching for at least 5 min of continuous stretching, 3 times/day, two consecutive oral augmentation of at least 8 min each continuous stretching in duration per day, and a series of orofacial grimacing once a day for 9 weeks. A total of about 25–30 mins of orofacial exercise was performed on a daily basis. Whereas, participants in the present study, were requested to perform orofacial exercise for a total of about 6 min a day (1 min total, 2 times/day of mouth stretching exercise, and 2 min total, 2 times/day oral augmentation exercise). Poole et al. did not find any significant improvement in their participants' oral aperture after 6 months of a home orofacial exercise program.[8] This may have been due to insufficient stretch duration (five stretches with 3–5s each per day).

There was a significant increase in the oral aperture between 3 and 6 months in the no-exercise /control group, which was unexpected. Seven participants in the control group (including two with an oral aperture size of more than 40 mm) demonstrated an increase in oral aperture with an average of 6.6 mm (ranging from 4 mm to 9 mm) between 3- and 6-months evaluations. Two participants in this subgroup had changed their medications during the study period by starting immunosuppressive drugs. Two participants reported they practiced mouth opening exercise: one with an oral aperture of 42 mm at 3-months evaluation reported she opened her mouth as wide as she could; another, with an oral aperture of 36 mm at 3-months evaluation, bought a Neckline Slimmer after seeing a commercial on television advertising its ability to tone up the tissue under the chin two months before the 6-month evaluation. Her oral aperture increased by average of 7.7mm between 3- and 6-months evaluations. We could not locate the other three participants to ask them what they might have done that could have contributed to the substantial increase in their oral aperture. Thus, all of the reasons for the significant increase observed in oral aperture between 3 and 6 months in the no-exercise/control group are unknown.

On the other hand, a 43-year-old White female participant in the orofacial exercise group, with limited cutaneous scleroderma, demonstrated a dramatic reduction in the size of oral aperture by an average of 12.7mm between 3 and 6 months. She reported that her mouth opening got tighter but we were unable to locate the cause for this loss of movement.

Still, based on the findings from the present study, as well as previous orofacial exercise intervention studies,[2,9,10] low exercise adherence rate may also contribute to the lack of improvement in the size of oral aperture. The studies of both Naylor et al.[2] and Pizzo et al.[10] employed a very vigorous monitoring system which required the patients visit the clinic for oral aperture evaluation every two weeks during the entire study period. As a result, their adherence rate may be high. However, when the monitoring was withdrawn, adherence to the recommended orofacial exercise program decreased dramatically.[2] None of the previous studies monitored the adherence rates of the home orofacial exercises.[2,8–10] The adherence rate of the present study was less than 50%, considered to be fair to poor when compared to traditional home-based physical exercise studies, which, in general, exceed a 60% adherence rate.[19]

The majority of participants who received the orofacial exercise instruction in our study indicated that they discontinued the exercises after 6 months due to soreness on the lips or at the jaw joint, decreased pigmentation at the mouth corner, forgetfulness, or not having time. Only three participants reported continuing the orofacial exercises on a weekly basis, among whom, one just did the oral augmentative exercise, without stretching the mouth at the corners using her thumbs because the stretching aggravated her dry, chapped lips.

A simple mouth stretching exercise may be more feasible and effective in the long run when minimal or no monitoring can be implemented. This could include exercises such as patients opening their mouth as wide as possible (with or without the aid of their hands for additional sideways stretching) and simple orofacial grimacing (which can be done any time of the day and at any place without the requirement of special equipment, e.g. a wooden stick). Such interventions may be as effective as a structured orofacial exercise program. However, in order to achieve an improvement in mouth opening of average of 4–5 mm, as found in the study of Maddali-Bongi et al,[9] the stretched mouth positions would need be held for about 30s, 50–60 times a day (a total of 30 min orofacial exercise). Further research efforts should be conducted on how to increase and maintain participants' adherence with simple mouth stretching and orofacial grimacing exercises at adequate frequencies, repetitions, and duration, as well as incentives for incorporating these exercise regimens into their daily routines.

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Table 1

Characteristics of the Participants at Baseline

Intent to Treat Group	**Overall (N=48)**	**Intervention group (n_1=26)**	**Control group (n_2=22)**
Characteristic	**Mean±SD(Range) or N(%)**	**Mean±SD(Range) or n_1(%)**	**Mean±SD(Range) or n_2(%)**
Oral aperture at baseline (mm)	36.5±9.7 (10.0–56.7)	36.2±11.0 (10.0–56.7)	36.8±8.0 (22.7–50.3)
Age (years)	50.7±13.0 (22–76)	51.9±14.3 (22–76)	49.2±11.4 (23–73)
Disease duration (years)[‡1]	7.6±6.1 (1.0–24.7)	8.3±6.4 (1.5–24.7)	6.8±5.8 (1.0–17.8)
Female	38 (79.2%)	21 (80.8%)	17 (77.3%)
African American[§]	26 (54.2%)	13 (50.0%)	13 (59.1%)
Diffuse cutaneous subset	21 (43.8%)	13 (50.0%)	8 (36.4%)
Calcium channel blocker	26 (54.2%)	12 (46.2%)	14 (63.3%)
Immunosuppressant	11 (22.9%)	8 (30.8%)	3 (13.6%)

Appropriate to Treat Group	**Overall (N=28)**	**Orofacial Exercise group (N=13)**	**No-exercise group (N=15)**
Characteristic	**Mean±SD(Range) or N(%)**	**Mean±SD(Range) or n_1(%)**	**Mean±SD(Range)or n_2(%)**
Oral aperture at baseline (mm)	30.1±6.7 (10.0–38.3)	27.4±7.4 (10.0–35.0)	32.4±5.3 (22.7–38.3)[†]
Age (years)	51.3±12.3 (23–76)	51.76±14.09 (23–76)	50.87±10.93 (31–73)
Disease duration (years)[‡2]	8.6±7.1 (1.0–24.7)	11.3±7.7 (1.5–24.7)	6.2±5.9 (1.0–17.8)
Female	22 (78.6%)	11(84.6%)	11 (73.3%)
African American[§]	16 (57.1%)	7 (53.8%)	9 (60.0%)
Diffuse cutaneous subset	14 (50.0%)	8 (61.5%)	6 (40.0%)
Calcium channel blocker	15 (53.6%)	5 (38.5%)	10 (66.7%)
Immunosuppressant	7 (25.0%)	5 (38.5%)	2 (13.3%)

[†] P=0.049;

[‡1] N=42, due to missing data;

[‡2] N=25, due to missing data;

[§] Only African Americans and Caucasians.

Table 2

Changes in the Size of Oral Aperture using the Intent to Treat Analysis (N=48)

	Group												
	Intervention (n_1=26)						Control (n_2=22)						
Oral aperture Measure Variable	**Mean**	**SD**	**Min**	**Max**	**Med**	***P-value* for within group difference***	**Mean**	**SD**	**Min**	**Max**	**Med**	***P-value* for within group difference***	***P-value* for between group difference†**
1st change (F1-B)	1.44	2.83	−3.30	8.70	0.67	0.02	−0.09	3.16	−5.30	8.30	0.00	0.71	0.04
2nd change (F2-F1)	0.71	3.38	−12.70	5.00	1.33	0.04	2.35	3.53	−6.00	9.00	2.00	0.01	0.88
Overall change (F‡-B)	2.14	2.88	−4.00	11.00	2.00	0.001	2.26	4.28	−3.33	15.67	0.50	0.02	0.19

SD=standard deviation, Min=minimum, Max=maximum, Med=median, B=baseline, F1=3 months, F2=6 months.

*Two-sided P-value using Wilcoxon signed rank test for each change

†One-sided P-value for significantly greater changes among intervention group when compared to control group using Mann-Whitney U-test

‡6 months measure recorded (F2, if non-missing, F1, otherwise)

Table 3

Changes in the Size of Oral Aperture using Appropriate to Treat Analysis (N=28)

	Group													
	Orofacial Exercise							No-exercise						
Oral aperture Measure Variable	**n**	**Mean**	**SD**	**Min**	**Max**	**Med**	***P-value* for within group difference***	**n**	**Mean**	**SD**	**Min**	**Max**	**Med**	***P-value* for within group difference***	***P-value* for between group difference†**
1st change (F1-B)	12	2.81	3.25	−1	8.67	1.83	0.01	12	−0.61	3.83	−5.33	8.33	−1.67	0.41	0.01
2nd change (F2-F1)	11	−0.06	4.48	−12.67	3.33	1.33	0.27	10	3.53	4.44	−6	9	3.33	0.04	0.98
Overall change (F‡-B)	12	2.75	3.82	−4	11	2	0.02	12	2.33	5.43	−3.33	15.67	0.17	0.31	0.19

SD=standard deviation, Min=minimum, Max=maximum, Med=median, B=baseline, F1=3 months, F2=6 months.

*Two-sided P-value using Wilcoxon signed rank test for each change

†One-sided P-value for significantly greater changes among intervention group when compared to control group using Mann-Whitney U-test

‡6 months measure recorded (F2, if non-missing, F1, otherwise)