A mixed method study to explore the feasibility and patient satisfaction of two different exercise programs in systemic sclerosis associated microstomia.

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Abstract

Background
Systemic sclerosis (SSc) is a severe autoimmune disease often leading to fibrotic cutaneous involvement of the face. Reduced oral aperture is associated with impaired food intake, oral hygiene and secondary dental problems. Stretching and oral augmentation exercises can increase oral aperture but are often hampered by low adherence rates. The aim of this mixed method study was to explore feasibility, patient satisfaction and effectiveness of two exercise programs in SSc-associated microstomia.

Methods
Adult patients (<18 years) suffering from systemic sclerosis (fulfilling the ACR/EULAR 2013 criteria) and microstomia (maximal oral aperture <40mm) were randomized to two groups. Group A exercised with a passive jaw motion device (Therabite®), and Group B performed mouth-stretching exercises. Patients were expected to exercise for 10 minutes, 3 times/day for 3 months, completed an exercise diary and were contacted 4 times by telephone. Patients were evaluated at baseline, 3 months (period without intervention), 6 months (after 3 months of intervention) and at 9 months (post-intervention visit). At month 6 semi-structured one-to-one interviews were conducted, recorded, transcribed verbatim and analyzed using Qualitative Analysis Guide of Leuven (QUAGOL).
Results

We included 6 women and 3 men, median age 60 years (range 40-75) and median disease duration 8 years (range 3-22). At 6 months, all patients in group A (n=4) and 4 in group B (n=5) improved with a median of 9mm (range 2-10) and 7mm (range 4-11), respectively. The proportion of executed to the planned number of exercises ranged between 63.7% and 98.9% in group A and between 48.5% and 97.4% in group B. Maintenance of the increase in oral aperture was noted in patients that continued to exercise daily. All 9 patients attended the interview that revealed three themes: drivers, challenges and perceived improvement.

Conclusion

Both interventions improve maximal oral aperture. The adherence to therapy was high but none of the patients considered it feasible to continue practicing 3 times/day. Future studies are needed in order to define feasible long-term exercise programs.

Keywords: Systemic sclerosis, Microstomia, Occupational therapy, Exercise therapy, Therabite®
**Background**

Systemic sclerosis (SSc) is a severe autoimmune disease and fibrotic cutaneous involvement of hands and face is a typical disease feature (1). Oral involvement with reduced oral aperture is frequent and associated with impaired food intake, reduced oral hygiene and secondary dental problems (2). Microstomia is defined as an interincisal distance smaller than 40mm (3). In SSc, microstomia is primarily caused by submucosal collagen deposits in perioral tissue (4). Various interventions have been examined. Exercises (5–8) as well as injection therapy (9) have been suggested to restore or maintain mouth opening and freedom of lip movement to improve patients’ quality of life. Several studies have shown that stretching (placing the thumbs in opposite corners of the mouth and pulling outward) and oral augmentation exercises (training with tongue depressors) can increase oral aperture in patients with SSc (5–8). In a study by Yuen (10) the authors could not show a significant improvement and highlighted the low exercise adherence rate. The passive jaw motion therapeutic device Therabite® is effective in increasing the range of motion in patients with temporomandibular joint and muscle disorders, but data in SSc-associated microstomia are lacking (2) (11). Furthermore, there are no studies investigating the feasibility of these exercises.

In this pilot study, we aim to explore feasibility, patient satisfaction and effectiveness of two different exercise programs, Therabite® and orofacial exercises, in SSc-associated microstomia.
Methods

Study design
A descriptive explorative convergent mixed method study was conducted from January 2017 to June 2018.

Patient selection and randomization
We addressed all adult SSc patients (>18 years) with microstomia (maximal oral aperture <40mm), fulfilling the ACR/EULAR 2013 criteria (12), that presented during 1 year at an outpatient visit at the Division of Rheumatology of the University Hospitals Leuven. Patients were classified according to criteria by LeRoy et al (13). Patients with a history of maxillary or mandibular fractures, infection, osteomalacia or osteoradionecrosis were excluded.

Research ethics committee approval was obtained from our local Institutional Review Board. Patients provided written informed consent and were randomized to two groups in a 1:1 ratio by means of a randomization list.

Intervention
Group A exercised with a passive jaw motion device (Therabite®) and Group B performed manual mouth-stretching exercises. Both groups were asked to exercise for 10 minutes, 3 times/day for 3 months. Our department provided the device Therabite® free of charge to the participants for the duration of the study, without sponsorship. At the start of the intervention, the participants practiced their exercises together with the occupational therapist (ES) and written instructions (including photos) were provided. Detailed description of the exercises of both groups can be found in Figure 1.
5 times 30 seconds of passive stretching with 30 seconds of rest in-between (= keep the mouth as far open as possible for 30 seconds) and

5 times 30 seconds active practice with 30 seconds rest in-between (= open the mouth gradually for 30 seconds)

3 times/day

4 different stretching exercises
The patient performs each stretching exercise 4 times for a duration of 30 seconds with 10 seconds rest.
3 times/day

**Fig. 1** Therabite® versus manual exercises.

**Study timeline and assessments**

A detailed study timeline can be found in Figure 2. Baseline visit was followed by a 3-month non-interventional observation period. The interventional phase of the study lasted for 3 months (from 3 to 6 months), followed by a post-interventional observation period of 3 months (from 6 to 9 months). Patients were assessed at baseline, 3, 6 and 9 months documenting oral aperture, skin thickness and patient reported outcomes.
Oral aperture was defined as the vertical distance from the bottom of the maxillary incisor to the top of the mandibular incisor with the mouth opened. All measurements were performed by the same assessor (ES) using a digital caliper. Three consecutive measurements were performed with a 5 seconds rest interval and averages were calculated (14).

During the interventional period, patients were contacted 4 times by telephone to address encountered problems and provide guidance (Table 1). These follow-up calls lasted 5-20 minutes. The subjects completed an exercise diary to document compliance. Compliance was defined as the proportion of executed exercises relative to the planned number of exercises and expressed in percentage (%).
Table 1. Interview guide follow-up calls.

| Question                                                                 |  |
|-------------------------------------------------------------------------|---|
| How did you experience your first week of mouth exercises?             |  |
| Can you perform all exercises or are there exercises that you have adjusted? |  |
| Are the exercises clear and easy to implement?                        |  |
| Did you ever have pain during or after exercise?                      |  |
| Can you sustain the duration of 10 minutes per session?                |  |
| Did you manage to practice 3 times/day?                                |  |
| How often did you not exercise during the previous week?              |  |
| What were the reasons for not exercising?                              |  |
| At what time of the day is it hardest to keep practicing?              |  |
| Do you have specific questions or concerns that you want to share with me? |  |

The interventional period ended at time point 6 months. When completing the interventional phase, patients were at liberty to continue exercising at their own pace, continuing diary recording but without follow-up telephone calls. The end of the interventional period (6 months) included a one-to-one interview, performed by ES, using a semi-structured interview guide (Table 2). Interviews were recorded, transcribed verbatim, anonymized and systematically analyzed using QUAGOL (15). ES read and reread all transcripts, and important units of meaning were systematically extracted, and grouped into natural subthemes and then overarching themes. A subset was independently analyzed by EDL and this was followed by a team discussion of combined findings together with our rheumatologists, a nurse and an occupational therapist. Based on these discussions the final themes and subthemes were refined.
Table 2. Interview guide.

Satisfaction
- You have been practicing for 3 months now, how have you experienced the last 3 months in terms of oral training?
- What expectations did you have in advance?
- To what extent have your expectations been met?
- What has happened differently than you expected?
- What benefit did the practice have for you?
- What improvements do you experience since the start of the oral training?
- What is getting worse/harder since the start of the oral training?
- What feeling do you get when you now hear the word 'Therabite®/mouth exercises'?
- What are the strengths/possibilities of the 'Therabite®/mouth exercises according to you?
- What are the weaknesses/downsides/pitfalls of the 'Therabite®/mouth exercises according to you?
- Which exercises did you not like and why?
- Which exercises did you prefer?
- Which aspects of the 'Therabite®/mouth exercises did you find less pleasant?

Feasibility
- How did exercising influence your daily routines?
- How do you see the feasibility of the training in the long term?
- How did you manage to continue to do the exercises daily?
- What influenced whether or not to carry out the exercises?
- What provided support/help?
- What was annoying/disruptive?
- If you could decide yourself how to continue the exercises, how would you do this?
- Is there anything else you want to tell or share about your experiences with mouth training?
- Do you have any questions or additions?
- Is there something that was overlooked?

Results

Patient recruitment

During the 1-year recruitment period, 34 patients were considered eligible for the study. 9 patients consented to participate. The reasons to decline participation were as follows:

- absence of subjective complaints (n = 8), participation not deemed feasible due to the travel time (n=6), other physical complaints that were of higher priority to the patient (n=6), full-time occupation (n=3), jaw complaints (n=1) and lack of motivation to practice (n=1).
Baseline patient data

6 women and 3 men were included, with a median age of 60 years (range 40-75) and median disease duration of 8 years (range 3-22) (Table 3). 4 patients were allocated to the Therabite® group (group A), 5 to the manual group (group B). The two interventional groups were similar in terms of gender and occupational status, but differences were noted in age (group A = median 51,5 years and group B = median 62 years) and disease duration (group A= median 4 years and group B= median 15 years).

Table 3. Baseline patient data.

| Age (years) | Gender | Diagnosis | Disease duration (years) | Dental status | Working status |
|-------------|--------|-----------|--------------------------|---------------|---------------|
| 56          | M      | DcSSc     | 5                        | Regular teeth | Sick leave    |
| 61          | F      | LcSSc     | 15                       | Dentures      | Housewife     |
| 40          | F      | DcSSc     | 3                        | Regular teeth | Housewife     |
| 71          | F      | LcSSc     | 15                       | Dental prosthesis upper teeth, lower teeth | Retired |
| 60          | M      | LcSSc     | 8                        | Dental prosthesis partly both above and below (5 regular teeth above and 3 below) | Sick leave |
| 47          | F      | LcSSc     | 3                        | Regular upper teeth, dental prosthesis lower teeth | Employee (50%) |
| 55          | F      | DcSSc     | 8                        | Regular teeth | Volunteer     |
| 61          | M      | LcSSc     | 22                       | Regular teeth | Retired       |
| 75          | F      | LcSSc     | 6                        | Dentures      | Retired       |

DcSSc = Diffuse Systemic Sclerosis LcSSc=Limited Systemic Sclerosis M = male F= female

Efficacy and compliance

At time point 6 months, oral aperture improved in all patients in group A (n=4) and 4 patients in group B (n=5) with a median of 9mm (range 2-10) and 7mm (range 4-11), respectively (Fig. 3). In one patient of group B, maximal oral aperture decreased 2mm over time. Compliance
ranged from 63.7% to 98.9% in group A and 48.5% to 97.4% in group B. During the follow-up period there was always a decrease in oral aperture among the participants who stopped practicing; there was also a decrease among the participants who had practiced at the start of the follow-up period but discontinued later on. There was lasting improvement if they had continued exercising 3 times/week, 1 time/day and maximal improvement at 2 times/day.

Fig. 3. Maximal oral aperture (mm).

Semi-structured interviews

All 9 participants of the exercise program participated in the interview that lasted 30-60 min. Three main themes emerged from the data: drivers, challenges and perceived improvement (Table 4).
Table 4. Main themes and subthemes.

| Main themes     | Subthemes                          |
|-----------------|------------------------------------|
| **Drivers**     | Motivated by the study             |
|                 | Motivated by functional disability |
|                 | Supportive factors                 |
| **Challenges**  | Time investment                    |
|                 | Mental struggle                    |
|                 | Need of routines                   |
|                 | Physical consequences              |
|                 | Technical limitations of the Therabite® |
|                 | Less enjoyable exercises           |
| **Perceived improvement** | Experiencing progress          |
|                 | Hope to retain progress            |
|                 | Necessity for continued training   |

Theme 1: Drivers

Participants highlighted several drivers that motivated them to perform the exercises at home.

Motivated by participating in a study. In both exercise groups, several participants were additionally motivated because of their participation in a study.

“Yes, I have to. Yes, I should do it. Because it was a deal, right, then I should stick to it. That’s the kind of person I am – we agree on it and then I go through with it, then I have to do it” (Male, 56, Therabite®)

“I hoped there would be some improvement, but if it wasn’t for me, I hoped it could help other people in the future” (Female, 61, manual exercises)
Motivated by functional disability. All participants experienced functional disabilities due to microstomia. All patients were concerned that their microstomia would worsen and this motivated them to exercise.

“Well, I don’t have lips anymore, when I drink a cup of coffee... I always need to clean up with a napkin, otherwise there’s always droplets and you do feel like that’s a handicap” (Male, 56, Therabite®)

Supportive factors. All participants of the Therabite® group and one patient of the manual exercise group described extrinsic motivational factors. They were primarily motivated by individuals in their environment, including their general practitioner, physiotherapist or partner. For some participants, the follow-up calls by the occupational therapist kept them motivated. Most of the participants placed their device or their instruction papers on a visible location, functioning as a memory aid.

“It was always before mealtimes, that she said ‘hey, have you done your exercises? Of course, yes, I need to do that first” (Male, 56, Therabite®)

Theme 2: Challenges.

Participants mentioned several challenges related to the feasibility of the home-based exercises.
Time investment. In both groups, participants experienced a substantial time investment to complete the exercises on a daily base. Before initiating the exercise program, they expected that performing the exercises for 10 minutes three times during the day would be easy to complete.

“The duration, it’s no time at all, it doesn’t take long, but still it needs a while” (Male, 56, Therabite®)

“All participants experienced that exercising three times a day was not feasible to implement into their daily routine. Each of them mentioned individual preference regarding the frequency of the exercises.

“All participants mentioned that it was hard to keep up their motivation to keep doing the exercises.”
“It’s not something you do for fun, I’m telling you, the amount of effort is underestimated”
(Female, 61, manual exercises)

Need of routines.
Participants highlighted how important it was to develop a routine, otherwise it was hard to persist and to remember to perform the exercises. To support daily routine, most of them combined their exercises with another activity that they were doing on a daily base.

“In the mornings, first a drink, something to eat and then practice” (Female, 47, Therabite®)

The mid-day period was challenging as many participants were not at home at this time of the day and mid-day planning was less structured compared to mornings or evenings. Other reasons were not able to do them in the presence of other people, travel days during the holiday and forgetfulness.

“For my daily routine, I scheduled everything around the oral exercises. Well, not always, but you do need to take it into account all the time” (Female, 40, Therabite®)

The participants that did not experience an effect on their daily routines were patients that spent the majority of their time at home and had already a lot of routines in place.
“I can arrange my exercises however I like, for people who have to work I think it’s far more difficult. If you need to work you can’t just get it done at 12 o’clock” (Male, 60, manual exercises)

Physical consequences. Most of the participants experienced physical consequences, especially in the first week. They felt a lot of soreness and experienced that their mouth/face needed to adapt to the exercises. This requested perseverance.

“Yes, at start, during the first week I was thinking ‘do not do that, do not do that’, if I have to be honest, I did not feel good, I had pain there, pain here, the first week was a real challenge” (Female, 47, Therabite®)

For some participants the physical consequences persisted after the first week, but the type and seriousness varied. Two participants adjusted the frequency of the exercises because of pain at the corners of the mouth (manual group). Other participants suffered from cramps at the neck, obliging one participant to adjust the exercise frequency (both groups).

Technical limitations of the Therabite®

Most of the participants of the Therabite® group indicated limitations related to the device. For half of them the possibilities were not challenging enough, and in the end, they could open the mouth further than the device itself. 1 participant also missed horizontal exercises.
“...at a certain point when I put the Therabite® into my mouth, then my teeth could actually come off of those pads” (Male, 56, Therabite®)

Two participants found that the device was too large, so they did not like to take the device along when leaving the house.

“But it was large, that was my husband’s first reaction, ‘but can’t they make a small version, why does it have to be so big?’” (Female, 40, Therabite®)

Less enjoyable exercises. Everyone in the manual exercise group had an exercise they did with less pleasure. In the Therabite® group, the participants had no comments about the exercises. Three out of five participants of the manual group found the exercise where they had to open the mouth without showing teeth heavy and less pleasant. Two participants found the exercise where they had to smile without showing teeth difficult. One participant suffered from mouth tears during the exercises where the mouth had to be wide open.

3.3 Theme 3: Perceived improvement

Participants also had several different perceptions about their improvement.

Experiencing progress.

Several participants experienced functional improvement in their daily living.
“When I used to talk my mouth got tired, yes, when I talk, for me it’s much better now”

(Female, 47, Therabite®)

“Yes, with eating and articulating. Eating is easier” (Female, 55, manual exercises)

Other participants did not experience subjective improvement but were surprised by the objective measurements.

“I have not felt much difference myself. That is why I am pleasantly surprised. I had not noticed it myself” (Female, 71, manual exercises)

The participants had an overall positive experience:

“If there was anyone else who was required to do it, I would immediately tell them ‘you should try it, you won’t have any disadvantages’” (Male, 60, Therabite®)

**Hope to retain progress.** Most of the participants were hoping that they could keep their improvement. They were curious if this could be sustained without further practice, but they were willing to continue practicing if necessary, but at a lower frequency.

“I do hope though, that the three months I’ve done, that that wasn’t wasted effort, imagine that” (Female, 61, manual exercises)
Necessity for continued training. Everyone indicates that it is probably going to be necessary to continue practicing, but they all hope that it can be less intensive.

“I think there will be a limit, to how far you can get, but I suspect that you have to keep it up regularly, otherwise it may worsen again” (Female, 71, manual exercises)

Discussion

The present study is the first to explore the feasibility and satisfaction of mouth exercises and the use of the Therabite® device for microstomia in patients suffering from systemic sclerosis. Previous studies have shown that oral augmentation exercises increase oral aperture (5–8), but did not include the experience of the participants. The result of the present study suggests that both interventions can improve the maximum oral aperture. The improvement in the observed maximum mouth opening (11 + -2mm) is comparable to previously published work by Pizzo et al (8) (10.7 + -2.06). In that study, participants were monitored every two weeks, while in the present study more self-discipline was expected from the participants. Other studies reported smaller improvements, namely 4.88 (5), 2.8 mm (16) or no improvement at all (17).

The observed improvement in the maximum mouth opening suggests that the applied regime (frequency, duration and number of repetitions) is effective. The 30 second stretch duration and the 10 minute duration was based on the comments in the Maddali-Bongi study (5), which stated that to achieve an improvement in the maximum mouth opening of 4-5 mm, a stretch duration of 30 seconds, 50-60 times a day, a total of 30 minutes of exercise per day was required. Concerning type of exercise, we opted only to use exercises without...
involvement of fingers in the mouth, because of sensitivity and wounds on the fingers, and this contrasts with other studies. Furthermore, our manual exercises had a longer stretch duration. The improvement gives an indication that manual exercises without using the fingers in the mouth can be efficient. Previous studies investigating the effects of the Therabite® device on other conditions have shown that the Therabite® device offers more comfort for the patient than using tongue depressors (11).

In our study, the frequency (3 times/day) was a stumbling block for our professionally active patients. A number of patients were interested to participate but ultimately refused recruitment because the study seemed practically unfeasible. Only patients that experienced functional disability as a consequence of microstomia consented to participate. In clinical practice this will be a very important aspect to consider. This can be seen as selection bias and could have influenced the high adherence rate that was shown in this study. In previous studies it is clear that few participants persist in practicing (16). It is conceivable that a 3-month exercise period could increase patient compliance compared to a 6-month period.

During the follow-up period there was always a decrease in oral aperture in the participants who stopped practicing. But there was also a decrease among the participants who had practiced at the beginning of the follow-up period but discontinued later on. There was improvement if they had continued exercising 3 times/week, 1 time/day and the most improvement at 2 times/day. This does mean that most of them did profit. It is unknown whether the improvement will disappear later on, to what extent and at which speed.

Further research for long-term follow up is necessary.
From the interviews, facilitating factors have been identified that motivate patients and increase feasibility of the exercises. It is important to take into account that different participants maintained a high adherence rate solely because of their participation in a trial. Supportive factors are also an attentive partner and/or involved health professionals. To continue exercising, the device or the papers with the exercises were always placed within reach, as a memory aid. In the future it would also be possible to work with a mobile application that gives a notification. If there are few supporting factors in patients, it may be important to provide more support via telephone monitoring.

Various challenges also emerged from this study. The time investment and the mental struggle to keep up exercising threatened the feasibility of continuing to practice 3 times/day in the long term. All participants considered 1 time/day to be feasible. In clinical practice health professionals could recommend adults with microstomia to exercise intensively (3 times/day) for 3 months to obtain an improvement and to maintain this improvement by exercising once a day. It is also important to try to do the exercises together with a routine daily activity. It was hardest to exercise at mid-day and all participants preferred the morning or the evening. The follow-up telephone call after 1 week is crucial to be able to offer support and to prevent physical complaints such as pain in the corners of the mouth or cramps in the neck. It is crucial to downsize the instructions ‘keep the mouth as large as possible’: is has to be as large as possible without getting pain during or after the exercise. Patient underestimated that they had to keep the same position for 30 seconds.
Furthermore, while deciding which intervention, it is important to consider the various advantages and disadvantages. The Therabite® has a purchase price of around 600 euros and is not reimbursed in Belgium while there are no costs associated with the manual exercises. With the Therabite®, the exercises are only vertical, while the manual exercises include both vertical and horizontal exercises. Participants also experienced that the Therabite exercises were not challenging enough and that they could open the mouth further than the device itself. Everyone in the manual group had an exercise they did with less pleasure, as therapist it will be important to coach the patient and to adapt the exercises according to the needs of the patient.

Most of the participants felt improvement in daily activities. The participants hope to retain progress without exercising but are willing to continue if they feel that their obtained results are diminishing. They all believe that practicing should be part of their lives, but they hope it can be less intensive. Further research is needed into feasible exercise programs with a lower frequency.

A recent study (9) suggests that a treatment with injections (hyaluronic acid and platelet-rich plasma) also improve both maximal oral aperture as quality of life. It is noteworthy that these injections require general anesthesia and are invasive procedures. Exercises are accessible for everyone and can be done without substantial costs.

The strengths of this study are the availability of qualitative interview results for all patients, providing insight into the psychosocial aspects associated with feasibility of the proposed exercise programs. Limitations are the low number of participants precluding statements on
the effectiveness of either of the interventions and the lack of blinding of the health professional.

This study shows that exercises can improve the maximal oral aperture if the frequency, duration and the number of repetitions is sufficiently intensive. In conclusion, patients with SSc need to be aware of the benefit of physical exercises to improve microstomia. As a therapist, it is crucial to educate patients on this topic and our study can serve as a guidance of attention points to take in account. It is crucial to consider the frequency of exercises for the patients to determine if it is feasible to continue exercising or not. Further studies are needed in order to define exercise programs that are feasible for active people (e.g. professionally active) and can be sustained in the long term. It should therefore be considered whether the same exercises with the same duration and number of repetitions would also have an effect with a lower frequency. To increase sample size, a multi-center study may be necessary in this rare disease.

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Ethics approval consent to participate

All research subjects participating in this study provided written informed consent. Consent was obtained from all participants by principal investigator. Documentation for the informed consent process as well as the signed consent forms is maintained in study binders at the Department of Rheumatology, University Hospital of Leuven in Belgium. All informed consent forms were reviewed and approved by the Ethical Committee of the University.
Hospital of Leuven (S59817, 23 December, 2016). All subjects were also provided with copies of their signed informed consent forms to be kept in their own records. Copies of the informed consent forms are available for review if necessary.

Abbreviations

| Abbreviation | Description |
|--------------|-------------|
| SSc          | Systemic sclerosis |
| QUAGOL       | Qualitative Analysis Guide of Leuven |
| DcSSc        | Diffuse cutaneous SSc |
| LcSSc        | Limited cutaneous SSc |
| M            | Male |
| F            | Female |

Authors' contributions

ES, EDL and RW designed the study. SS and ES were responsible for patient recruitment. ES performed the intervention and wrote the manuscript. All authors critically revised the manuscript for content and approved the final version for submission.

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Availability of data and materials

All the data are maintained in the Department of Rheumatology, University Hospital of Leuven, Belgium.
Consent for publication

All research subjects participating in this study provided written informed consent, including permission for their data to be utilized in publications. No names will be utilized in publications in order to maintain confidentiality. Consent was obtained from all participants by the principal investigator. Documentation for the informed consent process as well as the signed consent forms is maintained in study binders in the Department of Rheumatology at University Hospital of Leuven in Belgium.

Competing interests

The authors declare that they have no competing interests.
References

1. Agarwal SK, Reveille JD. The genetics of scleroderma (systemic sclerosis). Vol. 22, Current Opinion in Rheumatology. 2010. p. 133–8.
2. Alantar A, Cabane J, Hachulla E, Princ G, Ginisty D, Hassin M, et al. Recommendations for the care of oral involvement in patients with systemic sclerosis. Vol. 63, Arthritis Care and Research. 2011. p. 1126–33.
3. WP. N. Oral management of the scleroderma patient. J Am Assoc. 1982;105(8):814–7.
4. Paquette DL, Falanga V. Cutaneous concerns of scleroderma patients. J Dermatol. 2003;30(6):438–43.
5. Maddali-Bongi S, Landi G, Galluccio F, Del Rosso A, Miniati I, Conforti ML, et al. The rehabilitation of facial involvement in systemic sclerosis: Efficacy of the combination of connective tissue massage, Kabat’s technique and kinesitherapy: A randomized controlled trial. Rheumatol Int. 2011;31(7):895–901.
6. Albilia JB, Lam DK. Small Mouths ... Big Problems? A Review of Scleroderma and its Oral Health Implications. J Can Dent Assoc [Internet]. 2007; Available from: www.cda-adc.ca/jcda
7. Naylor WP, Douglass CW ME. The nonsurgical treatment of microstomia in sclerodermia: a pilot study. Oral Surg Oral Med Oral Pathol Oral Radiol. 1984;57:508–11.
8. Pizzo G, Scardina GA, Messina P. Effects of a nonsurgical exercise program on the decreased mouth opening in patients with systemic scleroderma. Clin Oral Investig. 2003;7(3):175–8.
9. Pirrello R, Verro B, Grasso G, Ruscitti P, Cordova A, Giacomelli R, et al. Hyaluronic acid and platelet-rich plasma, a new therapeutic alternative for scleroderma patients: A prospective open-label study. Arthritis Res Ther. 2019;21(1):1–8.
10. Yuen HK, Weng Y, Bandyopadhyay D, Reed SG. Effect of a Multi-Faceted Intervention on Gingival Health Among Adults with Systemic Sclerosis NIH Public Access(2 Suppl 65): S26-S32. $watermark-text $watermark-text $watermark-text. Vol. 29, Clin Exp Rheumatol. 2011.
11. Pauli N, Andréll P, Johansson M, Fagerberg-Mohlin B, Finizia C. Treating trismus: A prospective study on effect and compliance to jaw exercise therapy in head and neck cancer. J Sci Spec head neck. 2015 Dec 1;37(12):1738–44.
12. Van Den Hoogen F, Khanna D, Fransen J, Johnson SR, Baron M, Tyndall A, et al. 2013 classification criteria for systemic sclerosis: An american college of rheumatology/European league against rheumatism collaborative initiative. Arthritis Rheum. 2013 Nov;65(11):2737–47.
13. LeRoy EC, Medsger J. Criteria for the classification of early systemic sclerosis. J Rheumatol. 2001;28(7):1573–6.
14. Wood GD; Branco JA. A comparison of three methods of measuring maximal opening of the mouth. J oral Maxillofac Surg. 1979;37:175–7.
15. Diercks de Casterle B, Gastmans C, Bryon E, Denier Y. QUAGOL: A guide for qualitative data analysis. Int J Nurs Stud. 2012 Mar;49(3):360–71.
16. Yuen HK, Marlow NM, Reed SG, Mahoney S, Summerlin LM, Leite R, et al. Effect of orofacial exercises on oral aperture in adults with systemic sclerosis. Disabil Rehabil. 2012;34(1):84–9.
17. Poole J, Conte C, Brewer C, Good CC, Perella D, Rossie KM, et al. Oral hygiene in...
scleroderma: The effectiveness of a multi-disciplinary intervention program. Disabil Rehabil. 2010;32(5):379–84.