A mixed method study exploring patient satisfaction and feasibility of two exercise programs in systemic sclerosis associated microstomia

Ellinor Sydow¹, Kristien Van der Elst¹, Patrick Verschueren¹, Jan Lenaerts¹, René Westhovens¹, Ellen De Langhe¹,²

¹ Division of Rheumatology, University Hospitals Leuven, Leuven, Belgium
² Laboratory of Tissue Homeostasis and Disease, Department of Development and Regeneration, KU Leuven, Leuven, Belgium

Corresponding address:
Ellinor Sydow
Division of Rheumatology, University Hospitals Leuven, Herestraat 49, 3000 Leuven, Belgium
ellinor.sydow@uzleuven.be

ORCiD ID: https://orcid.org/0000-0002-0161-9353

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Abstract

Objectives. Systemic sclerosis (SSc) is often leading to fibrotic cutaneous involvement of the face and reduced oral aperture with impaired food intake and oral hygiene. Oral 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 suffering from SSc and microstomia (maximal oral aperture <40mm) were randomized to two groups. Group A exercised with a 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. Patients were evaluated at baseline, 3 months (period without intervention), 6 months (after 3 months of intervention) and at 9 months (post-intervention). At month 6 semi-structured one-to-one interviews were conducted.

Results. We included 6 women and 3 men, median age 60 years and median disease duration 8 years. At 6 months, all patients in group A (n=4) and 4 in group B (n=5) improved with a median of 9mm and 7mm, respectively. The adherence ranged between 63.7% and 98,9% in group A and between 48.5% and 97,4% in group B. 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®
Key messages:

- Exercises can improve the maximal oral aperture if the frequency, duration and the number of repetitions is sufficiently intensive.
- Continuing to exercise 3 times a day is not feasible for adults with systemic sclerosis.
- It is necessary to define feasible exercise programs for professionally active patients that can be sustained in the long term.

Introduction

Systemic sclerosis (SSc) is a severe autoimmune disease and fibrotic cutaneous involvement of hands and face is a typical disease feature (1). 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). 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. The subtype of SSc was 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. A description of the exercises of both groups can be found in Supplementary Figures S1 and S2, available at Rheumatology Advances in Practice online.

**Study timeline and assessments**

A detailed study timeline can be found in Figure 1. 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. These follow-up calls lasted 5-20 minutes. Interview guides can be found in Supplementary Tables S1 and S2, available at Rheumatology Advances in Practice online. The subjects completed an exercise diary to document adherence. Adherence was defined as the proportion of executed exercises to the planned number, expressed in percentage (%).
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. Interviews were recorded, transcribed verbatim, anonymized and systematically analyzed using QUAGOL (15). ES read and reread all transcripts and important units of 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. Based on these discussions the final themes and subthemes were refined. Detailed description of this process can be found in Supplementary Figure S3, available at Rheumatology Advances in Practice online.

Ethics

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.

Results

Patient recruitment
During the 1-year recruitment period, 34 patients were considered eligible for the study. 9 patients consented to participate. The recruitment process can be found in Figure 2.

**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) (Supplementary Table S3, available at *Rheumatology Advances in Practice* online.). 4 patients were allocated to the Therabite® group (group A), 5 to the manual group (group B). No significant difference was found between the groups (p>0.05 using the Mann-Whitney U test).

**Efficacy and adherence**

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 (Figure 3). In one patient of group B, maximal oral aperture decreased 2mm over time. Adherence 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 (Supplementary Table S4, available at *Rheumatology Advances in Practice* online.). There was lasting improvement if they had continued exercising 3 times/week, 1 time/day and maximal improvement at 2 times/day.

**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 (Table 1), challenges (Table 2) and perceived improvement (Table 3).
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.

*Motivated by functional disability.* All participants experienced functional disabilities due to microstomia and were concerned that their microstomia would worsen.

*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 or from the follow-up calls by the occupational therapist. Most of the participants placed their device or their instruction papers on a visible location, functioning as a memory aid.

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. All participants experienced that exercising three times a day was not feasible to implement into their daily routine.

**Mental struggle.** All participants mentioned that it was hard to keep up their motivation to keep doing the exercises.

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

**Physical consequences.** Most of the participants experienced physical consequences, especially in the first week. 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 or pain at the neck.

**Technical limitations of the Therabite®**

The participants of the Therabite® group indicated limitations related to the device. For half of them the possibilities were not challenging enough, and they also missed horizontal exercises. Two participants found that the device was too large, so they did not like to take the device along when leaving the house.
**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.

### 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. Other participants did not experience subjective improvement but were surprised by the objective measurements. The participants had an overall positive experience:

**Hope to retain progress.** Most of the participants were hoping that they could keep their improvement.

**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.

### 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 SSc. 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 oral aperture (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 oral aperture suggests that the applied regime (frequency, duration and number of repetitions) is effective. The 30 second stretch duration and the 10 minute duration 3 times a day was based on the comments in the Maddali-Bongi study (5). 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. 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. Also, 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 adherence 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. There was improvement if they had continued exercising 3 times/week, 1 time/day and the most improvement at 2 times/day. 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. 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 and all participants preferred to exercise in 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. 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. 6 of the 9 participants reported side effects. 2 participants from the Therabite® group and 1 participant from the manual group did not experience any side effects. The side effects varied in nature and severity. 2 participants exercising with manual exercises had pain at the corners of the mouth, so they had to adjust the frequency of the exercise. 1 participant from the Therabite® group had cramps under the chin and 3 participants suffered from neck cramps (2 from the Therabite® group and 1 from the manual group). In one of these participants, exercising with therabite®, these pains were so intense that the participants had to adjust the frequency to 2 times/day. Yawning was provoked in one participant during the manual exercises.

The Therabite® has a purchase price of 412.80 euros (exclusive VAT) 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.

The participants 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, there was nothing added to aim to reduce this bias.

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.

Further studies are needed in order to define exercise programs that are feasible for professionally active patients 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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ES, EDL and RW designed the study. ES was responsible for patient recruitment, 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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Conflicts of interest: The authors have declared no conflicts of interest.

Availability of data and materials: All the data are maintained in the Department of Rheumatology, University Hospital of Leuven, Belgium.
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Figure legends

Fig. 1. Study timeline

Fig. 2. The recruitment process

Fig. 3. Maximal oral aperture (mm)
| Main themes                  | Subthemes                        | Participant’s narratives                                                                 |
|-----------------------------|----------------------------------|------------------------------------------------------------------------------------------|
| Drivers                     | Motivated by the study           | “I made a promise, if I make a promise to someone, I will keep it” (Male, 60, Therabite) |
|                             |                                  | “If it doesn’t help me, maybe it will help somebody else. For the future, that others    |
|                             |                                  | are getting better of it” (Female, 61, manual)                                             |
|                             | Motivated by functional disability | “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               | “It was motivating that you called me. It was a good motivation. It was a short         |
|                             |                                  | conversation but still...” (Female, 55, manual)                                           |
Table 2. Theme of ‘challenges’, subthemes and some of the participants’ narratives

| Main themes               | Subthemes                        | Participant’s narratives                                                                                                                                                                                                                                                                                                                                                          |
|---------------------------|----------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Challenges                | Time investment                  | “The duration, it’s no time at all, it doesn’t take long, but still, it needs a while” (Male, 56, Therabite®)                                                                                                                                                                                                                   |
|                           |                                  | “3 times a day is REALLY... yes, it is really tough that way” (Female, 40, Therabite®)                                                                                                                                                                                                                                      |
|                           |                                  | “Certainly not 3 times/day. 2 Times/day but reluctantly, I’d rather not really. Keeping it up is difficult. But I do want to keep doing it in the mornings” (Male, 60, manual exercises)                                                                                                                                                                                                 |
|                           | Mental struggle                  | “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                 | “In the mornings, first a drink, something to eat and then practice” (Female, 47, Therabite®)                                                                                                                                                                                                                                 |
|                           |                                  | “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®)                                                                                                                                                         |
|                           | Physical consequences            | “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®)                                                                 |
|                           | Technical limitations of the Therabite® | “…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®)                                                                                                                                                                    |
|                           | Less enjoyable exercises.        | “The third exercise, laughing with a closed mouth, that was hard, I wanted to perform perfectly but I didn’t feel if I did it well” (Man, 61, Therabite)                                                                                                                                                                                  |
Table 3. Theme of ‘Perceived improvement’, subthemes and some of the participants’ narratives

| Main themes          | Subthemes                     | Participant’s narratives                                                                                                                                 |
|----------------------|-------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------|
| Perceived improvement| Experiencing progress         | “Yes, with eating and articulating. Eating is easier” (Female, 55, manual exercises)                                                                     |
|                      |                               | “I have not felt much difference myself. That is why I am pleasantly surprised. I had not noticed it myself” (Female, 71, manual exercises)          |
|                      |                               | “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       | “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. | “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) |
Figure 1. Study timeline

- 0 months
- 3 months
- 6 months
- 9 months

- Daily exercise program
  10’3 times/day

- Telephone contact
  week 1, week 2, month 1 and month 2

- Assessments and measurement of the mean interincisal distance (maximal oral aperture)

- Interview and questionnaire: feasibility and satisfaction

- Stop of obligatory intervention

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34 adult SSc patients (>18 years) with microstomia (maximal oral aperture <40mm), fulfilling the ACR/EULAR 2013 criteria

Excluded patients (1):
1. Jaw complaints

Declined participation (24):
8. Absence of subjective complaints
6. Participation not deemed feasible due to the travel time
6. Other physical/medical complaints that were of higher priority to the patient
3. Fulltime occupation
1. Lack of motivation to practice

4 patients
Group A: Therabite®

5 patients
Group B: Manual mouth-stretching

Figure 2. The recruitment process
Figure 3. Maximal oral aperture (mm)