

# A Mixed Method Study to Explore the Feasibility and Patient Satisfaction of Two Different Exercise Programs in Systemic Sclerosis Associated Microstomia

Ellinor Sydow (✉ [ellinorsydow@yahoo.com](mailto:ellinorsydow@yahoo.com))

Division of Rheumatology, University Hospitals Leuven, Leuven, Belgium <https://orcid.org/0000-0002-0161-9353>

**Sabien Severi**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

**Kristien Van der Elst**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

**Patrick Verschueren**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

**René Westhovens**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

**Jan Lenaerts**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

**Ellen De Langhe**

UZ Leuven: Katholieke Universiteit Leuven Universitaire Ziekenhuizen Leuven

---

## Research article

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

**Posted Date:** November 2nd, 2020

**DOI:** <https://doi.org/10.21203/rs.3.rs-100072/v1>

**License:**  This work is licensed under a Creative Commons Attribution 4.0 International License.

[Read Full License](#)

---

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

3  
4 Ellinor Sydow M.Sc.<sup>1</sup>, Sabien Severi B.Sc.<sup>1</sup>, Kristien Van der Elst M. Sc, PhD<sup>1</sup>, Patrick  
5 Verschueren MD, PhD<sup>1</sup>, René Westhovens MD, PhD<sup>1</sup>, Jan Lenaerts MD<sup>1</sup>, Ellen De Langhe MD,  
6 PhD<sup>1,2</sup>

7  
8 <sup>1</sup> Division of Rheumatology, University Hospitals Leuven, Leuven, Belgium

9 <sup>2</sup> Laboratory of Tissue Homeostasis and Disease, Department of Development and  
10 Regeneration, KU Leuven, Leuven, Belgium

11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44

45 **Abstract**

46

47 **Background**

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

54

55 **Methods**

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

65

66

67

68 **Results**

69 We included 6 women and 3 men, median age 60 years (range 40-75) and median disease  
70 duration 8 years (range 3-22). At 6 months, all patients in group A (n=4) and 4 in group B (n=5)  
71 improved with a median of 9mm (range 2-10) and 7mm (range 4-11), respectively. The  
72 proportion of executed to the planned number of exercises ranged between 63.7% and 98,9%  
73 in group A and between 48.5% and 97,4% in group B. Maintenance of the increase in oral  
74 aperture was noted in patients that continued to exercise daily. All 9 patients attended the  
75 interview that revealed three themes: drivers, challenges and perceived improvement.

76

77 **Conclusion**

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

81

82 **Keywords:** Systemic sclerosis, Microstomia, Occupational therapy, Exercise therapy,  
83 Therabite®  
84

85

86

87

88

89

90

91

92

93 **Background**

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

110  
111 In this pilot study, we aim to explore feasibility, patient satisfaction and effectiveness of two  
112 different exercise programs, Therabite® and orofacial exercises, in SSc-associated  
113 microstomia.

114

115

116

|

117 **Methods**

118 **Study design**

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

121

122 **Patient selection and randomization**

123 We addressed all adult SSc patients (>18 years) with microstomia (maximal oral aperture  
124 <40mm), fulfilling the ACR/EULAR 2013 criteria (12), that presented during 1 year at an  
125 outpatient visit at the Division of Rheumatology of the University Hospitals Leuven. Patients  
126 were classified according to criteria by LeRoy et al (13). Patients with a history of maxillary  
127 or mandibular fractures, infection, osteomalacia or osteoradionecrosis were excluded.

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

131

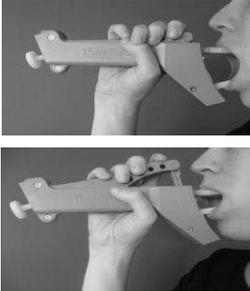
132 **Intervention**

133 Group A exercised with a passive jaw motion device (Therabite®) and Group B performed  
134 manual mouth-stretching exercises. Both groups were asked to exercise for 10 minutes, 3  
135 times/day for 3 months. Our department provided the device Therabite® free of charge to  
136 the participants for the duration of the study, without sponsorship. At the start of the  
137 intervention, the participants practiced their exercises together with the occupational  
138 therapist (ES) and written instructions (including photos) were provided. Detailed  
139 description of the exercises of both groups can be found in Figure 1.

140

Formatted: Right: 0.25"

141  
142  
143  
144  
145  
146  
147  
148  
149  
150  
151  
152  
153  
154  
155  
156  
157  
158  
159  
160  
161  
162  
163  
164  
165  
166  
167  
168  
169  
170  
  
171  
172  
173  
174  
175  
176  
177  
178  
179



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



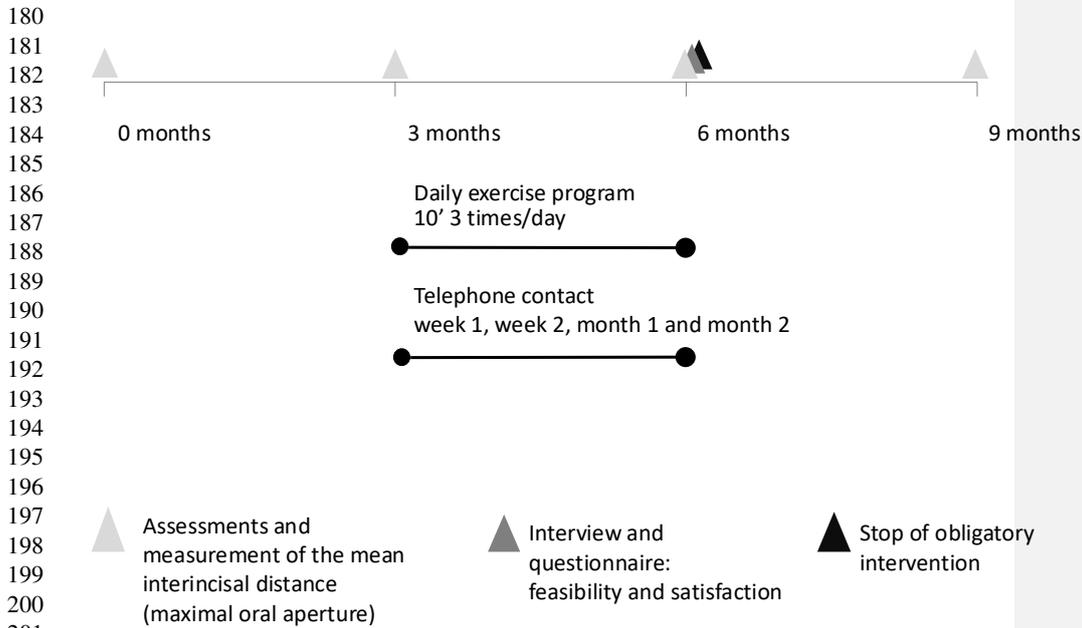
4 different stretching exercises  
The patient performs each stretching exercise 4 times for a duration of 30 seconds with 10 seconds rest.  
3 times/day

**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.

Formatted: Right: 0.25"



**Fig. 2. Study timeline**

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 (%).

Formatted: Right: 0.25"

216  
217

**Table 1. Interview guide follow-up calls.**

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?

218  
219

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

231  
232  
233  
234  
235

Formatted: Right: 0.25"

236 **Table 2. Interview guide.**

<b>Satisfaction</b>
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?
<b>Feasibility</b>
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?

237  
238

239 **Results**

240 **Patient recruitment**

241 During the 1-year recruitment period, 34 patients were considered eligible for the study. 9  
 242 patients consented to participate. The reasons to decline participation were as follows:  
 243 absence of subjective complaints (n = 8), participation not deemed feasible due to the travel  
 244 time (n=6), other physical complaints that were of higher priority to the patient (n=6), full-  
 245 time occupation (n=3), jaw complaints (n=1) and lack of motivation to practice (n=1).

246  
247

Formatted: Right: 0,25"

248 **Baseline patient data**

249 6 women and 3 men were included, with a median age of 60 years (range 40-75) and median  
250 disease duration of 8 years (range 3-22) (Table 3). 4 patients were allocated to the  
251 Therabite® group (group A), 5 to the manual group (group B). The two interventional groups  
252 were similar in terms of gender and occupational status, but differences were noted in age  
253 (group A = median 51,5 years and group B = median 62 years) and disease duration (group  
254 A= median 4 years and group B= median 15 years).

255

256 **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

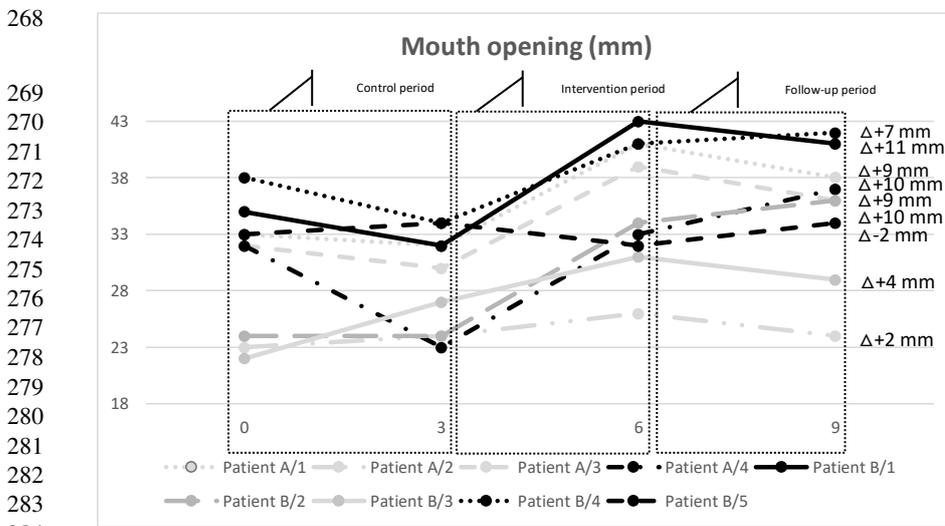
257 DcSSc = Diffuse Systemic Sclerosis LcSSc=Limited Systemic Sclerosis M = male F= female  
258

259 **Efficacy and compliance**

260 At time point 6 months, oral aperture improved in all patients in group A (n=4) and 4 patients  
261 in group B (n=5) with a median of 9mm (range 2-10) and 7mm (range 4-11), respectively (Fig.  
262 3). In one patient of group B, maximal oral aperture decreased 2mm over time. Compliance

Formatted: Right: 0.25"

263 ranged from 63.7% to 98,9% in group A and 48.5% to 97,4% in group B. During the follow-up  
 264 period there was always a decrease in oral aperture among the participants who stopped  
 265 practicing; there was also a decrease among the participants who had practiced at the start  
 266 of the follow-up period but discontinued later on. There was lasting improvement if they had  
 267 continued exercising 3 times/week, 1 time/day and maximal improvement at 2 times/day.



286 **Fig. 3. Maximal oral aperture (mm).**

289 **Semi-structured interviews**

290 All 9 participants of the exercise program participated in the interview that lasted 30-60 min.

291 Three main themes emerged from the data: drivers, challenges and perceived improvement

292 (Table 4).

293  
 294  
 295  
 296  
 297  
 298

299 **Table 4. Main themes and subthemes.**

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

300

301 **Theme 1: Drivers**

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

304

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

307

308 *“Yes, I have to. Yes, I should do it. Because it was a deal, right, then I should stick to it. That’s  
 309 the kind of person I am – we agree on it and then I go through with it, then I have to do it”*

310 (Male, 56, Therabite®)

311

312 *“I hoped there would be some improvement, but if it wasn’t for me, I hoped it could help  
 313 other people in the future”* (Female, 61, manual exercises)

314

Formatted: Right: 0.25"

315

316 **Motivated by functional disability.** All participants experienced functional disabilities due to  
317 microstomia. All patients were concerned that their microstomia would worsen and this  
318 motivated them to exercise.

319

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

323

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

330

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

333

#### 334 **Theme 2: Challenges.**

335 Participants mentioned several challenges related to the feasibility of the home-based  
336 exercises.

337

|

338 **Time investment.** In both groups, participants experienced a substantial time investment to  
339 complete the exercises on a daily base. Before initiating the exercise program, they expected  
340 that performing the exercises for 10 minutes three times during the day would be easy to  
341 complete.

342  
343 *“The duration, it’s no time at all, it doesn’t take long, but still it needs a while”* (Male, 56,  
344 Therabite®)

345  
346 *“3 times a day is REALLY... yes, it is really tough that way”* (Female, 40, Therabite®)

347  
348 All participants experienced that exercising three times a day was not feasible to implement  
349 into their daily routine. Each of them mentioned individual preference regarding the  
350 frequency of the exercises.

351  
352 *“3 times a week minimum but if I have to do it once a day, it would also be feasible”* (Female,  
353 61, manual exercises)

354  
355 *“Certainly not 3 times/day. 2 Times/day but reluctantly, I’d rather not really. Tough to always  
356 remember to do it. Keeping it up is difficult. But I do want to keep doing it in the mornings”*  
357 (Male, 60, manual exercises)

358  
359  
360 **Mental struggle.** All participants mentioned that it was hard to keep up their motivation to  
361 keep doing the exercises.

362

363 *"It's not something you do for fun, I'm telling you, the amount of effort is underestimated"*

364 (Female, 61, manual exercises)

365

366 ***Need of routines.***

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

370

371 *"In the mornings, first a drink, something to eat and then practice"* (Female, 47, Therabite®)

372

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

377

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

380

381 The participants that did not experience an effect on their daily routines were patients that  
382 spent the majority of their time at home and had already a lot of routines in place.

383

Formatted: Right: 0.25"

384 *"I can arrange my exercises however I like, for people who have to work I think it's far more*  
385 *difficult. If you need to work you can't just get it done at 12 o'clock"* (Male, 60, manual  
386 exercises)

387

388

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

392

393 *"Yes, at start, during the first week I was thinking 'do not do that, do not do that', if I have to*  
394 *be honest, I did not feel good, I had pain there, pain here, the first week was a real*  
395 *challenge"* (Female, 47, Therabite®)

396

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

401

402

#### 403 **Technical limitations of the Therabite®**

404 Most of the participants of the Therabite®group indicated limitations related to the device.

405 For half of them the possibilities were not challenging enough, and in the end, they could

406 open the mouth further than the device itself. 1 participant also missed horizontal exercises.

407

|

408

409 *"...at a certain point when I put the Therabite® into my mouth, then my teeth could actually*  
410 *come off of those pads"* (Male, 56, Therabite®)

411

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

414

415 *"But it was large, that was my husband's first reaction, 'but can't they make a small version,*  
416 *why does it have to be so big?'"* (Female, 40, Therabite®)

417

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

424

425

### 426 **3.3 Theme 3: Perceived improvement**

427 Participants also had several different perceptions about their improvement.

428

429 Experiencing **progress**.

430 Several participants experienced functional improvement in their daily living.

431

|

432 *“When I used to talk my mouth got tired, yes, when I talk, for me it’s much better now”*

433 (Female, 47, Therabite®)

434

435

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

437

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

440

441 *“I have not felt much difference myself. That is why I am pleasantly surprised. I had not*

442 *noticed it myself”* (Female, 71, manual exercises)

443

444 The participants had an overall positive experience:

445

446 *“If there was anyone else who was required to do it, I would immediately tell them ‘you*

447 *should try it, you won’t have any disadvantages”* (Male, 60, Therabite®)

448

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

452

453 *“I do hope though, that the three months I’ve done, that that wasn’t wasted effort, imagine*

454 *that”* (Female, 61, manual exercises)

455

Formatted: Right: 0.25"

456 **Necessity for continued training.** Everyone indicates that it is probably going to be necessary  
457 to continue practicing, but they all hope that it can be less intensive.

458

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

461

## 462 **Discussion**

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

473

474 The observed improvement in the maximum mouth opening suggests that the applied  
475 regime (frequency, duration and number of repetitions) is effective. The 30 second stretch  
476 duration and the 10 minute duration was based on the comments in the Maddali-Bongi  
477 study(5), which stated that to achieve an improvement in the maximum mouth opening of 4-  
478 5 mm, a stretch duration of 30 seconds, 50-60 times a day, a total of 30 minutes of exercise  
479 per day was required. Concerning type of exercise, we opted only to use exercises without

|

480 involvement of fingers in the mouth, because of sensitivity and wounds on the fingers, and  
481 this contrasts with other studies. Furthermore, our manual exercises had a longer stretch  
482 duration. The improvement gives an indication that manual exercises without using the  
483 fingers in the mouth can be efficient. Previous studies investigating the effects of the  
484 Therabite® device on other conditions have shown that the Therabite® device offers more  
485 comfort for the patient than using tongue depressors (11).

486  
487 In our study, the frequency (3 times/day) was a stumbling block for our professionally active  
488 patients. A number of patients were interested to participate but ultimately refused  
489 recruitment because the study seemed practically unfeasible. Only patients that experienced  
490 functional disability as a consequence of microstomia consented to participate. In clinical  
491 practice this will be a very important aspect to consider. This can be seen as selection bias  
492 and could have influenced the high adherence rate that was shown in this study. In previous  
493 studies it is clear that few participants persist in practicing (16). It is conceivable that a 3-  
494 month exercise period could increase patient compliance compared to a 6-month period.

495  
496 During the follow-up period there was always a decrease in oral aperture in the participants  
497 who stopped practicing. But there was also a decrease among the participants who had  
498 practiced at the beginning of the follow-up period but discontinued later on. There was  
499 improvement if they had continued exercising 3 times/week, 1 time/day and the most  
500 improvement at 2 times/day. This does mean that most of them did profit. It is unknown  
501 whether the improvement will disappear later on, to what extent and at which speed.  
502 Further research for long-term follow up is necessary.

503

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

512  
513 Various challenges also emerged from this study. The time investment and the mental  
514 struggle to keep up exercising threatened the feasibility of continuing to practice 3  
515 times/day in the long term. All participants considered 1 time/day to be feasible. In clinical  
516 practice health professionals could recommend adults with microstomia to exercise  
517 intensively (3 times/day) for 3 months to obtain an improvement and to maintain this  
518 improvement by exercising once a day. It is also important to try to do the exercises  
519 together with a routine daily activity. It was hardest to exercise at mid-day and all  
520 participants preferred the morning or the evening. The follow-up telephone call after 1  
521 week is crucial to be able to offer support and to prevent physical complaints such as pain in  
522 the corners of the mouth or cramps in the neck. It is crucial to downsize the instructions  
523 'keep the mouth as large as possible': is has to be as large as possible without getting pain  
524 during or after the exercise. Patient underestimated that they had to keep the same position  
525 for 30 seconds.

526

Formatted: Right: 0.25"

527 Furthermore, while deciding which intervention, it is important to consider the various  
528 advantages and disadvantages. The Therabite® has a purchase price of around 600 euros  
529 and is not reimbursed in Belgium while there are no costs associated with the manual  
530 exercises. With the Therabite®, the exercises are only vertical, while the manual exercises  
531 include both vertical and horizontal exercises. Participants also experienced that the  
532 Therabite exercises were not challenging enough and that they could open the mouth  
533 further than the device itself. Everyone in the manual group had an exercise they did with  
534 less pleasure, as therapist it will be important to coach the patient and to adapt the  
535 exercises according to the needs of the patient.

536

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

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

546 The strengths of this study are the availability of qualitative interview results for all patients,  
547 providing insight into the psychosocial aspects associated with feasibility of the proposed  
548 exercise programs. Limitations are the low number of participants precluding statements on

Formatted: Right: 0.25"

549 the effectiveness of either of the interventions and the lack of blinding of the health  
550 professional.

551 This study shows that exercises can improve the maximal oral aperture if the frequency,  
552 duration and the number of repetitions is sufficiently intensive. In conclusion, patients with  
553 SSc need to be aware of the benefit of physical exercises to improve microstomia. As a  
554 therapist, it is crucial to educate patients on this topic and our study can serve as a guidance  
555 of attention points to take in account. It is crucial to consider the frequency of exercises for  
556 the patients to determine if it is feasible to continue exercising or not.

557 Further studies are needed in order to define exercise programs that are feasible for active  
558 people (e.g. professionally active) and can be sustained in the long term. It should therefore  
559 be considered whether the same exercises with the same duration and number of  
560 repetitions would also have an effect with a lower frequency. To increase sample size, a  
561 multi-center study may be necessary in this rare disease.

562

### 563 **Acknowledgements**

564 Not applicable.

565

### 566 **Ethics approval consent to participate**

567 All research subjects participating in this study provided written informed consent. Consent  
568 was obtained from all participants by principal investigator. Documentation for the informed  
569 consent process as well as the signed consent forms is maintained in study binders at the  
570 Department of Rheumatology, University Hospital of Leuven in Belgium. All informed  
571 consent forms were reviewed and approved by the Ethical Committee of the University

Formatted: Right: 0.25"

572 Hospital of Leuven (S59817, 23 December, 2016). All subjects were also provided with copies  
573 of their signed informed consent forms to be kept in their own records. Copies of the  
574 informed consent forms are available for review if necessary.

575

#### 576 **Abbreviations**

577	SSc	Systemic sclerosis
578	QUAGOL	Qualitative Analysis Guide of Leuven
579	DcSSc	Diffuse cutaneous SSc
580	LcSSc	Limited cutaneous SSc
581	M	Male
582	F	Female

583

#### 584 **Authors' contributions**

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

588

#### 589 **Funding**

590 Not applicable.

591

#### 592 **Availability of data and materials**

593 All the data are maintained in the Department of Rheumatology, University Hospital of  
594 Leuven, Belgium.

Formatted: Right: 0.25"

595 **Consent for publication**

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

602

603 **Competing interests**

604 The authors declare that they have no competing interests.

605

606

Formatted: Right: 0.25"

607 **References**

- 608 1. Agarwal SK, Reveille JD. The genetics of scleroderma (systemic sclerosis). Vol. 22,  
609 Current Opinion in Rheumatology. 2010. p. 133–8.
- 610 2. Alantar A, Cabane J, Hachulla E, Princ G, Ginisty D, Hassin M, et al. Recommendations  
611 for the care of oral involvement in patients with systemic sclerosis. Vol. 63, Arthritis  
612 Care and Research. 2011. p. 1126–33.
- 613 3. WP. N. Oral management of the scleroderma patient. *J Am Assoc.* 1982;105(8):814–7.
- 614 4. Paquette DL, Falanga V. Cutaneous concerns of scleroderma patients. *J Dermatol.*  
615 2003;30(6):438–43.
- 616 5. Maddali-Bongi S, Landi G, Galluccio F, Del Rosso A, Miniati I, Conforti ML, et al. The  
617 rehabilitation of facial involvement in systemic sclerosis: Efficacy of the combination  
618 of connective tissue massage, Kabat’s technique and kinesitherapy: A randomized  
619 controlled trial. *Rheumatol Int.* 2011;31(7):895–901.
- 620 6. Albilal JB, Lam DK. Small Mouths ... Big Problems? A Review of Scleroderma and its  
621 Oral Health Implications. *J Can Dent Assoc [Internet].* 2007; Available from: [www.cda-  
623 adc.ca/jcda](http://www.cda-<br/>622 adc.ca/jcda)
- 623 7. Naylor WP, Douglass CW ME. The nonsurgical treatment of microstomia in  
624 scleroderma: a pilot study. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 1984;57:508–  
625 11.
- 626 8. Pizzo G, Scardina GA, Messina P. Effects of a nonsurgical exercise program on the  
627 decreased mouth opening in patients with systemic scleroderma. *Clin Oral Investig.*  
628 2003;7(3):175–8.
- 629 9. Pirrello R, Verro B, Grasso G, Ruscitti P, Cordova A, Giacomelli R, et al. Hyaluronic acid  
630 and platelet-rich plasma, a new therapeutic alternative for scleroderma patients: A  
631 prospective open-label study. *Arthritis Res Ther.* 2019;21(1):1–8.
- 632 10. Yuen HK, Weng Y, Bandyopadhyay D, Reed SG. Effect of a Multi-Faceted Intervention  
633 on Gingival Health Among Adults with Systemic Sclerosis NIH Public Access(2 Suppl  
634 65): S26-S32. \$watermark-text \$watermark-text \$watermark-text. Vol. 29, *Clin Exp*  
635 *Rheumatol.* 2011.
- 636 11. Pauli N, Andréll P, Johansson M, Fagerberg-Mohlin B, Finizia C. Treating trismus: A  
637 prospective study on effect and compliance to jaw exercise therapy in head and neck  
638 cancer. *J Sci Spec head neck.* 2015 Dec 1;37(12):1738–44.
- 639 12. Van Den Hoogen F, Khanna D, Fransen J, Johnson SR, Baron M, Tyndall A, et al. 2013  
640 classification criteria for systemic sclerosis: An american college of  
641 rheumatology/European league against rheumatism collaborative initiative. *Arthritis*  
642 *Rheum.* 2013 Nov;65(11):2737–47.
- 643 13. LeRoy EC, Medsger J. Criteria for the classification of early systemic sclerosis. *J*  
644 *Rheumatol.* 2001;28(7):1573–6.
- 645 14. Wood GD; Branco JA. A comparison of three methods of measuring maximal opening  
646 of the mouth. *J oral Maxillofac Surg.* 1979;37:175–7.
- 647 15. Dierckx de Casterle B, Gastmans C, Bryon E, Denier Y. QUAGOL: A guide for qualitative  
648 data analysis. *Int J Nurs Stud.* 2012 Mar;49(3):360–71.
- 649 16. Yuen HK, Marlow NM, Reed SG, Mahoney S, Summerlin LM, Leite R, et al. Effect of  
650 orofacial exercises on oral aperture in adults with systemic sclerosis. *Disabil Rehabil.*  
651 2012;34(1):84–9.
- 652 17. Poole J, Conte C, Brewer C, Good CC, Perella D, Rossie KM, et al. Oral hygiene in

653 scleroderma: The effectiveness of a multi-disciplinary intervention program. Disabil  
654 Rehabil. 2010;32(5):379-84.  
655

Formatted: Right: 0.25"

## Figures



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



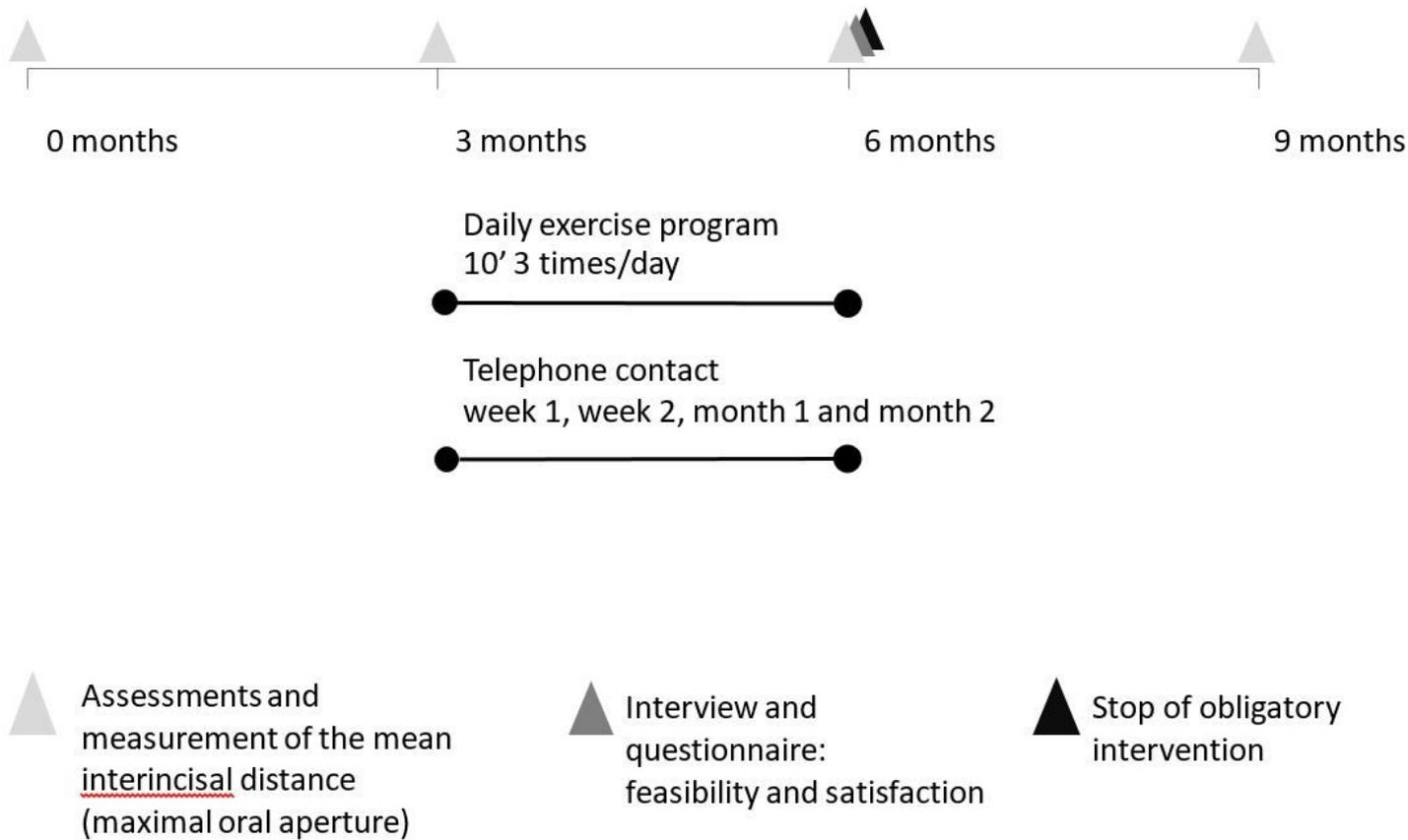
4 different stretching exercises

The patient performs each stretching exercise 4 times for a duration of 30 seconds with 10 seconds rest.

3 times/day

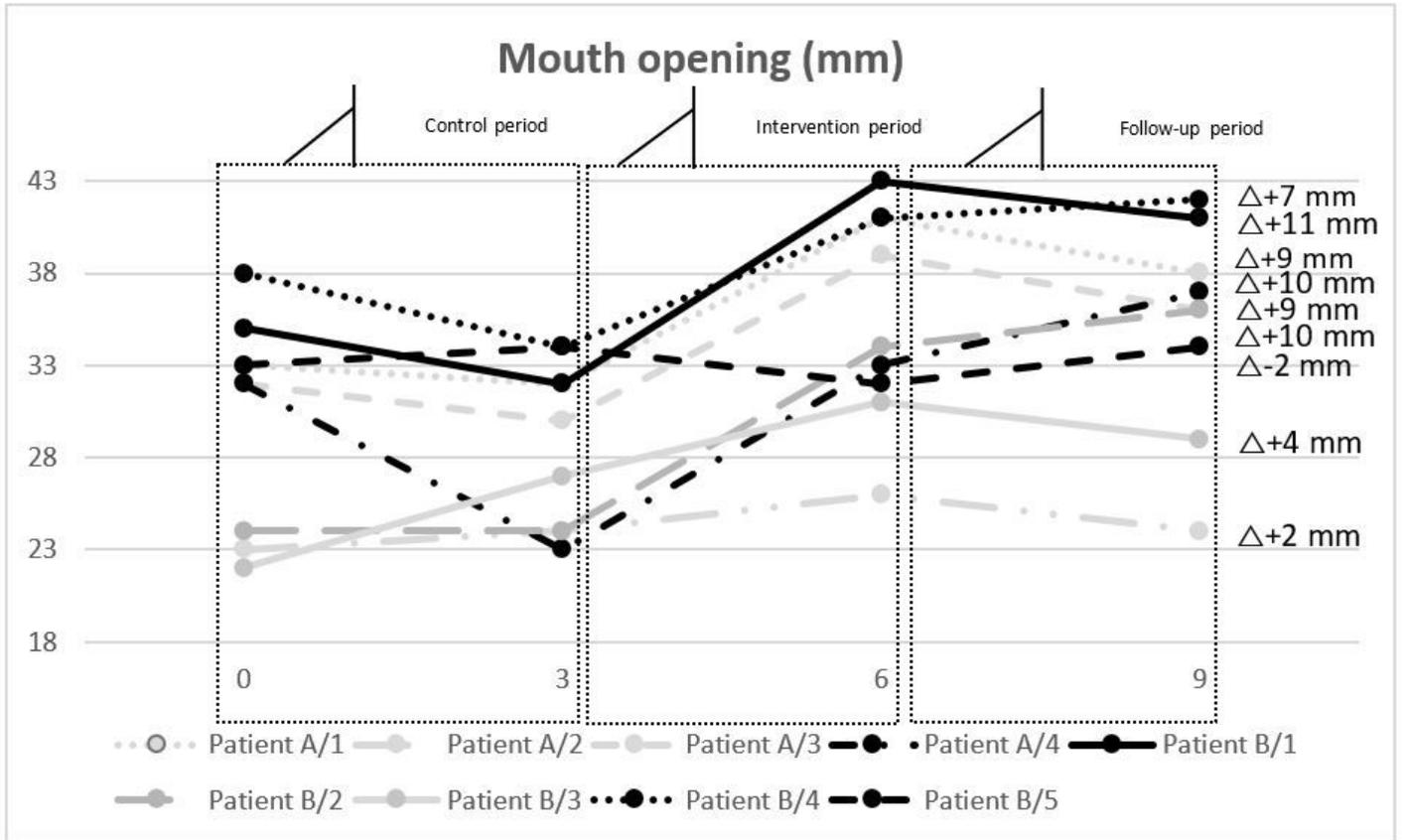
Figure 1

Therabite® versus manual exercises.



**Figure 2**

Study timeline



**Figure 3**

Maximal oral aperture (mm).