Efficacy of intensive hand physical therapy in patients with systemic sclerosis

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ABSTRACT

Objective. *To evaluate the efficacy of a three-week period of complex and intensive hand physical therapy on hand function in patients with systemic sclerosis (SSc).*

Methods. *Thirty-one patients with SSc were treated. Hand stretching exercises, ergotherapy supplemented with thermal and mud baths, whirlpool therapy and soft tissue massage were daily used during a three-week period. The control SSc group (n=22) received similar management for their large joints leaving out their hands. The primary outcomes of this study were the Health Assessment Questionnaire (HAQ) and the Disabilities of the Arm, Shoulder and Hand (DASH). Hand Anatomic Index (HAI), Cochin Hand Function (CHFT) and clinical characteristics were also assessed before starting the therapy and six months afterwards.*

Results. *Six months after the investigation period, only the group receiving hand physical therapy showed improvement in HAQ and DASH scores compared to the baseline values ($p<0.05$). The improvement in median HAQ value (25%-75% quartiles) reached the clinical meaningful rate (baseline 1.125 /0.625-1.625/ versus 0.75 /0.25-1.5/ at six months). Visual analogue scales of global pain ($p<0.01$) and Raynaud's phenomenon ($p<0.05$) also had better results than at baseline. HAI, gripping strength and CHFT also showed some improvement, but did not reach the significance level of change by the end of the six-month observation period.*

Conclusions. *The complex physical therapy caused favourable changes in both the HAQ and the DASH indicating that this particular program had some long-term beneficial effect on hand function in patients with SSc.*

Introduction

Systemic sclerosis (SSc) is a connective tissue disorder characterised by vascular damage, fibrosis, and immunologic abnormalities. The end stage disease causes obliterative vasculopathy and tissue atrophy. Besides the internal organ involvement (1) affecting the lungs, heart, kidneys and gastrointestinal tract, musculoskeletal symptoms are also often present and significantly contribute to the impaired quality of life in SSc. Musculoskeletal manifestations affecting the hand function include arthralgia/arthritis, tendon friction rubs, joint contractures, digital tuft resorption, subcutaneous calcinosis, and decreased muscle grip strength. Hands are affected in up to 31% to 97% of SSc patients (2-4), leading to significant disability in both the limited cutaneous (lcSSc) and diffuse cutaneous (dcSSc) subsets of SSc (5, 6). One of the potential management options is to treat the synovitis to preserve hand function (6, 7). However, in a significant proportion of the cases further factors also contribute to hand function impairment including periarticular tissue fibrosis and hypoxia, skin tethering and atrophy, tendon injury ending up with contractures or osteolysis (8-10). There is a definite need for the efficient treatment of these particular hand damages because of its significant impact on range of motion and loss of function (3, 5, 6). Until now, only a few small case studies are available on the efficiency of musculoskeletal rehabilitation techniques in SSc including hand range of motion (ROM) exercises, paraffin wax bath, connective tissue massage, manual lymph drainage and patient education (11-18). Mugii *et al.* demonstrated that a daily simple stretching exercises for joints of the hand not only improved the total passive joint motion after one month following the interven-

tion, but the improvement was maintained after one year (11). Pils *et al*. (12) combined exercise therapy with paraffin wax treatment, and found that after 12 days of paraffin treatment, participants self-reported an improvement in skin stiffness, hand mobility, pain, and symptoms of Raynaud's phenomenon, although within three months after discontinuing the treatment the investigated parameters returned to baseline values. Sandqvist (13), Mancuso (14) and their co-workers confirmed the advantage of combining hand exercises with paraffin bath treatment. The improvement of hand function was independent of skin score and disease duration; however, neither of them conducted a follow-up longer than two months on the effects of their therapy. Bongi *et al*. (15) combined range of motion exercises with connective tissue massage and McMennell joint manipulation. This complex nine-week therapy twice per week improved fist closure, joint motion of the hands, hand function and quality of life, while the control group merely engaged in a range of self-administered motion exercises at home, which only improved fist closure. Bongi *et al*. reported that manual lymph drainage treatment for nine weeks was also effective (16). In a subsequent investigation (17), the authors used rehabilitation techniques combined and tailored for each SSc patient, also for a nine-week period. In each study the groups were reinvestigated after a nine-week follow-up period. Both complex physical therapies improved HRQoL and hand function, which was partially maintained for the next nine-week follow-up period. In another recent Italian controlled study (18) the efficacy of a three-month long period of occupational therapy with a self-administrated stretching programme was investigated after one and three months. At the end of the study period the occupational group showed a greater improvement concerning some hand function tests compared to the control group. As in previous studies (11-18), using a combined hand-exercise therapy in conjunction with paraffin baths, mud or soft tissue massage, a significant improvement was detected in the hand movement and function, but only for a two to three-month follow-up time.

Our earlier study (19) clearly indicated that a simple regular stretching home exercise was not effective enough to improve the hand function even if a repeated educational process was included for the patients. Based on this experience we decided to perform an intensive and combined physical therapy to clarify whether this approach was efficient for improving the hand function of SSc patients.

The aim of this study was to investigate whether a complex three-week long intensive hand-focused physical therapy intervention improved hand function for a long period in patients with SSc, who did not have significant inflammatory symptoms. The intensive stretching hand-exercise therapy in this study was completed by ergotherapy, mud-bath and thermal hydrotherapy.

Patients and methods

Study design

This particular prospective non-randomised, controlled, follow-up study consisted of a three-week intensive treatment period and the primary outcomes were namely the changes in Health Assessment Questionnaire Disability Index (HAQ-DI) (20) and in values of the Disabilities of the Arm, Shoulder and Hand (DASH) questionnaire (21), which were evaluated 24 weeks post-treatment. HAQ-DI predominantly represents the hand function (20), and DASH reflects the function of the upper limb.

The study protocol followed the Principles of the Declaration of Helsinki, 1964 and later amendments. The National Ethics Committee of Hungary gave their approval for conducting the study (5485- 2/2013/ EKU (338/2013.). All patients gave their written consent to the study, and did not participate in other physical treatment during the follow-up time.

Recruitment and participants

Patients between 18–75 years of age with joint contractures of the hand were eligible into the study. All patients were treated in our tertiary care centre of the Department of Rheumatology and Immunology of the University of Pécs, Hungary. Criteria of inclusion consisted of the presence of at least two hand joint-contractures as a manifestation of SSc based on the judgement of the rheumatologist (CV, TM), the ability to participate in a three-week physical therapy in another city 150 km away from the University, and the ability to be reassessed six months later. Limitation in ROM of a joint greater than 25% of the normal range was considered of threshold value for contractures.

Criteria of exclusion included (Fig. 1):

- patients with significant systemic inflammation (level of serum C-reactive protein (CRP) > 10 mg/l) and/or sign of synovitis, or disease activity (22)
- patients suffering from severe interstitial lung disease (FVC<50%) or pulmonary hypertension (WHO class III/IV (1, 23),
- cardiac diseases (LVEF<50%),
- patients with active skin ulcers,
- patients with severe urine incontinence,
- patients attending other physiotherapy within three months prior to the initiation of the study, and during the follow-up period.

In the intervention group the patients took part in a complex physiotherapy program including thermal baths (37-38°C, 30 minutes/day), mud baths of the hands (42°C, 20 minutes/day), whirlpool therapy of the extremities (37-38°C mineral water, 20 minutes/day), medical massages of the trunk and the upper extremities (mechanical, stimulating and vasodilator techniques in 30 minutes/occasion), exercise (30 minutes/day, isometric, isotonic and stretching hand exercises; 15 minutes isotonic and stretching exercises for the spine, hip and knee); ergotherapy (30-60 minutes; muscle facilitation, proprioceptive training, improvement of coordination and fine motor skills, practicing self-care tasks if needed).

The control group received similar therapy to their large joints, and the spine without being a subject to any treatment to the hands. The therapy of the control patients consisted of thermal baths (37-38°C, 30 minutes/day), mud

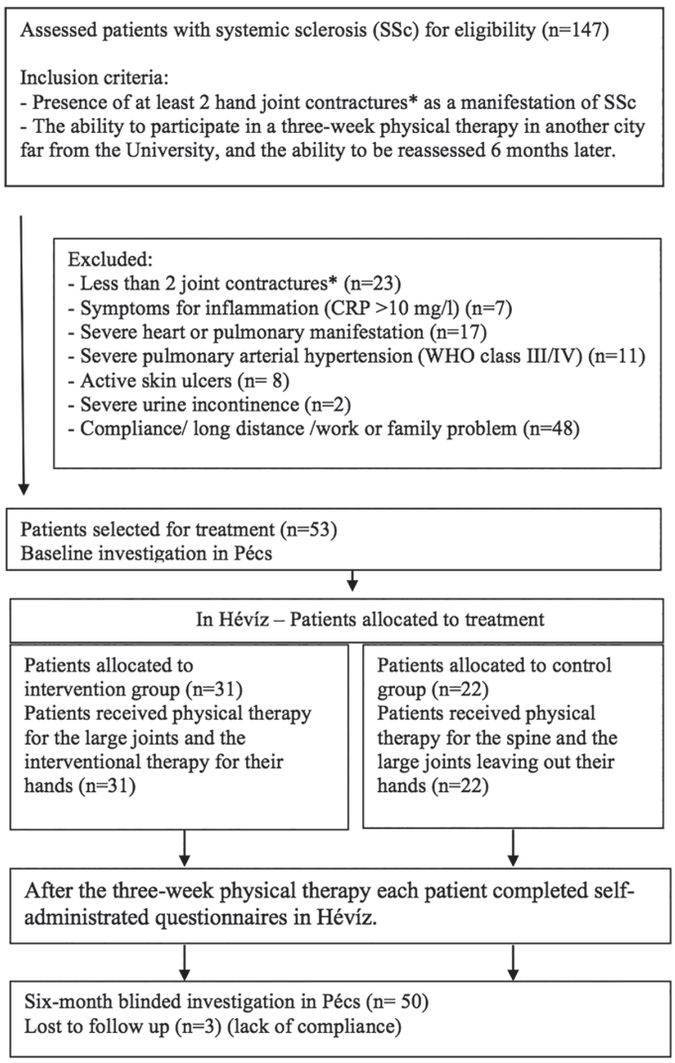

Fig. 1. Flow chart of participants through the study.

*Range of motion < 75% of the normal

pack for the spine (42°C, 20 minutes/day) medical massages of the trunk, exercise (30 minutes/day, isotonic and stretching exercises for the spine, hip and knee; breathing exercises – but not for the hands)

During the three-week long treatment, all therapy-types were given 5 days a week, except for mud baths (every other day) and thermal baths (daily).

Because of the nature of this particular complex therapy, the randomisation would be very difficult, as patients can communicate with each other during the study, therefore we decided that the test group and the patient control group is participating in the investigation in a different time framework, *i.e.* either members of the test group or the control SSc group were present at the site of the investigation. Otherwise we selected both hand exercise group patients and control SSc group in a consecutive order. The first 25-30 consecutive patients fulfilling the inclusion criteria were planned to contribute as an intervention group, and the next consecutive 25-30 patients were sorted into the control group. Baseline and 6 months evaluation of each case was blindly performed by independent physical therapists (ZBK, DK) in Pécs. The baseline assessment was performed 1–4 weeks before starting the study.

In Hévíz, at the end of the three-week physical therapy period, the local physical therapists (ES, AD, RPT) collected self-administered questionnaire data from each patients of both groups.

Outcomes of the study

The primary outcomes of this study were the changes of the HAQ-DI (20, 24, 25) and the DASH questionnaires (21). Both measures had been cross-cultural adapted into Hungarian and investigated as to their validities in patients with SSc (21, 26). Although HAQ is an overall assessment for disability, the 65% of its questions focus on hand function. In patients with SSc, global disability can be measured with the HAQ, and a good sensibility, reliability, validity, and responsiveness in measuring activities of the daily living, as well as the hand function (20, 24, 27).

The DASH questionnaire with 32 items has been shown to be reliable and valid in many upper limb surgical diseases and in rheumatoid arthritis or psoriatic arthritis, and also in SSc (21, 28).

In addition to these measures, the visual analogue scales (VASs) of the Scleroderma-HAQ (20, 25, 29-31) and other self-administered questionnaires, the Cochin Hand Function Test (CHFT) with 18 items (32, 33) and the Short Form Health Survey (SF-36) questionnaire, a measure of health related to quality of life had to be completed by all patients (20, 34). Hand anatomy index (HAI) (35) and delta Palm-fingertip Distance (delta FTP) were also calculated (36). HAI is calculated from the distances of open hand span minus closed hand span divided by lateral hand height. Delta FTP is the difference of the distance measured between the third fingertip and the distal palmar crease with fingers fully extended and fully flexed position. The grip strength of the hands was evaluated by using a JAMAR Hand Dynamometer (Lafayette Instrument, USA).

Statistical analysis

The distribution of values was assessed by using Kolmogorov–Smirnov test. Comparisons of the values of clinical data between groups were performed by using Mann-Whitney U test for non-parametric data or Student-T test for continuous variables and by Chi-square test for categorical variables. Wilcoxon signed-rank test was used for analysing the differences of test-results between baseline and at different time points of the study. Statistical analyses were made with IBM SPSS Statistics v 20.0 software package (IBM's Corporate, New York, USA).

Results

Fifty-three patients with SSc were eligible and consented to participate. Figure 1 shows the flow of participants through the study.

The general features of the subjects are reported in Table I. At baseline, there were no significant differences between the two groups in any of the clinical measures shown by the test analyses (Table I).

By the end of the three-week intensive physical therapy, the health related functional parameters showed a significant global improvement as compared to the starting values at the place of the therapy in both groups. However, the values of the HAQ got better significantly ($p<0.05$) exclusively in the group receiving physical therapy for the hands (Table II). At the onset the median value /25th to75th percentile/ of the HAQ was 1.125 /0.63-1.63/ and after three weeks treatment 0.937 /0.5-1.5/, though in the control group the baseline value was 1.0 /0.6-1.5/, and at the end of the three weeks 1.18 /0.75-1.5/, respectively. Results of the DASH, VAS-Pain, VAS-Raynaud's phenomenon, VAS-Pulmonary problems VAS-Globally problems showed better results in both groups immediately after the end of the three-week physiotherapy compared to the starting values of the test-results (Table II). The Physical Summary Index of SF36 improved significantly only in the control group after the three-week therapy ($p<0.05$) (Table II). The Cochin test showed some improvement in the interventional group, but the changes were not statistically significant in either of the groups (Table II).

After six months, only the hand-focused physiotherapy group showed a sustained improvement in HAQ, DASH, VAS-pain and VAS-Raynaud ($p<0.05$) (Table II). The average difference (improvement) of the HAQ result was 0.206, CI: 0.041 - 0.371, which was significant and higher than value of earlier defined "clinically meaningful change", 0.14 (34, 35). The power of the test was 0.74. When delta changes of measures from the baseline to the six-month follow-up were compared between the groups, a tendency of a larger improvement were observed in all values, favourable to the group receiving hand physical therapy, although these differences did not reach the statistical significance (Table III).

In the hand interventional group, comparing the changes of patients with better functional condition (HAQ<1, n=20) to the subjects with more severe condition (HAQ>1, n=11), a larger improvement ($p<0.05$) was found in HAQ, VAS-pain, VAS-Raynaud, and delta-FTP parameters in favour of the more severely-affected patients (data not shown).

Also in the hand treated group, patients whose improvement in HAQ reached the value of "minimally important difference for improvement" (HAQ score decreased more than 0.14 unit) (n=15) were significantly younger (average age was 56±12 years) ($p<0.05$), than those who did not improve to that extent (n=16, average age: 63±16 years). There was no significant difference between disease duration time at the baseline comparing these two groups.

Between the lcSSc and the dcSSc groups no significant differences in any the results of tests could be observed at the end of the three-week investigation period as well as after 6 months follow-up, both in the interventional and the control group (data not shown).

The hand focused three-week treatment modified neither the Physical nor the Mental Summary Indices of SF36, as measured at the six- month follow-up (Table II).

Generally the patients tolerated the treatment well. No serious adverse events happened with the 53 patients during the three-week physical therapy program. In both groups transitory mild hypertension occurred, usually on the first two days. In two cases a mild upper respiratory infection were evolved. Neither of the patients developed digital ulcer during the treatment. Three patients could not come to the six-month reinvestigation because of their family or work program.

Table I. Demographic and clinical data of 53 patients with systemic sclerosis (SSc).

	Hand-physical therapy group*	Control group*	*p*-values**
Number of patients	31	22	
Diffuse cutaneous SSc	18 (58%)	11 (50%)	0.561
Females	29 (93%)	20 (91%)	0.720
Average age	59.7 (14.5)	62.1 (8.4)	0.540
Disease duration (years)	11.1 (6.7)	13.0 (7.6)	0.342
Lung fibrosis on HRCT	16 (52%)	15 (68%)	0.228
FVC < 80%	11 (36%)	7 (32%)	0.781
Cardiac involvement[1]	13 (42%)	14 (64%)	0.119
Subcutaneous calcinosis	2 (7%)	4 (18%)	0.184
Anti-topoisomerase antibody	17 (55%)	12 (57%)	0.870
Anti-centromere antibody	6 (19%)	7 (33%)	0.253
Anti-RNA-polymerase-III	2 (7%)	0	0.225
Baseline measures:			
ESR mm/h	18.7 (9.3)	17.4 (9.6)	0.665
CRP mg/l	3.6 (2.4)	2.7 (2.1)	0.246
HAQ	1.125 /0.6-1.6/	0.875 /0.40-1.2/	0.155
DASH	34.2 /18.0-55.0/	37.5 /26.5-46.7/	0.802
CHFT	11.0 /3.0-26.0/	9.0 /5.75-28.0/	0.459
Hand anatomic index R	2.25 /1.6-3.2/	2.7 /1.6-3.37/	0.439
Delta-FTP R	29.8 /27.7/	29.8 /23.8/	0.885
VAS-Global	27.3 /22.4/	34.0 /18.7/	0.159

*Values are the number (%) of subjects or average (standard deviation) or median /25th to 75th percentile/, as appropriate; **Calculated by Pearson's chi squared test or Mann-Whitney U test; FVC: Forced vital capacity of the lungs; [1]Cardiac involvement means conduction defects, arrhythmias, pericarditis, myocardial ischaemia on ECG; HAQ: Health Assessment Questionnaire; DASH: Disabilities of the Arm, Shoulder and Hand; CHFT: Cochin Hand Function Test; Delta-FTP: Delta Finger To Palm Distance of the right hand (R); VAS-Global: Visual analogue scale 0-100 mm measure for overall problem with SSc.

Discussion

In this particular study, a complex three-week intensive hand-focused physical therapy intervention was performed, which improved hand function

Table II. Longitudinal evaluations of functional tests in 53 patients with SSc.

Interventional group (n=31)					
	Baseline (T0)	Three weeks (T1)	Six months (T6)	*p* (T0-T1)	*p* (T0-T6)
SF-36 Phys	36.5/28.3-41.9/	36.0/30.4-41.1/	35.3/28.0-48.8/	0.989	0.820
SF Mental	51.6 /38.5-56.8/	49.4 /39.7-58.5/	51.1 /37.5-59.5/	0.269	0.241
HAQ	1.125 /0.6-1.6/	0.937 /0.5-1.5/	0.75 /0.25-1.5/	**0.042***	**0.017***
DASH	34.2 /18.3-55.0/	29.3 /12.9-49.1/	32.5 /10.0-45.8/	**0.001***	**0.023***
Cochin	11 /3.0-26.0/	9.5 /4.0-23.5/	9 /2.0-20.0/	0.406	0.238
VAS pain	27.3 /22.4/	22.0 /20.9/	18.9 /20.0/	**0.002***	**0.030***
VAS RP	37.2 /28.0/	20.5 /22.3/	25.5 /22.8/	**0.011***	**0.046***
VAS DU	18.6 /30.9/	13.8 /24.1/	8.9 /20.9/	0.097	0.078
VASGI	19.6 /24.6/	11.1 /18.6/	15.6 /25.9/	**0.007***	0.779
VAS Pulm	16.9 /19.8/	14.1 /21.1/	14.6 /20.7/	**0.047***	0.433
VAS overall	29.8 /27.7/	18.9 /23.1/	21.6 /21.7/	**0.001***	0.082
dFTP R	70.0 /58.0-80.0/		76.0 /58.0-91.0/		0.217
dFTP L	77.0 /61.0-86.0/		78.0 /60.0-91.0/		0.939
HAI R	2.25 /1.6-3.15/		2.8 /1.9-3.3/		0.309
HAI L	2.73 /1.6-3.2/		2.97 /1.9-3.3/		0.308
Gripping R	137.6 /67.0/		152.8 /71.2/		0.075
Gripping L	132.0 /69.3/		143.8 /70.5/		0.187
Control group (n=22)					
	Baseline (T0)	Three weeks (T1)	Six months (T6)	*p* (T0-T1)	*p* (T0-T6)
SF-36 Physical	30.0 /25.8-36.6/	32.9 /28.9-38.9/	31.5 /27.0-43.4/	**0.020***	0.679
SF Mental	50.5 /43.1-60.0/	49.4 /39.7-58.5/	49.5 /37.0-57.1/	0.179	0.126
HAQ	0.875 /0.4-1.2/	1.18 /0.7-1.5/	0.875 /0.4-1.4/	0.378	0.442
DASH	37.5 /26.5-46.7/	38.7 /20.2-49.3/	37.8 /18.6-52.9/	**0.007***	0.948
Cochin	9 /5.75-28.0/	14.5 /6.5-33.2/	11.5 /5.3-29.3/	0.117	0.690
VAS pain	34.0 /18.7/	24.2 /22.8/	34.7 /24.6/	**0.007***	0.459
VAS RP	35.8 /27.9/	19.25 /20.0/	36.3 /35.1/	**0.003***	0.485
VAS DU	14.9 /21.3/	14.9 /20.8/	18.9 /34.3/	0.652	0.859
VASGI	17.2 /24.6/	17.6 /27.5/	20.2 /29.1/	0.432	0.182
VAS Pulm	23.7 /25.9/	17.4 /22.5/	25.8 /30.4/	**0.009***	0.727
VAS overall	29.8 /23.8/	21.9 /22.3/	33.3 /27.8/	**0.006***	0.586
dFTP R	74.0 /57.5-82.5/		78.0 /58.3-87.0/		0.420
dFTP L	79.0 /57.0-85.0/		78.5 /59.0-85.8/		0.840
HAI R	2.7 /1.6-3.37/		2.44 /1.6-3.0/		0.602
HAI L	2.69 /1.9-3.6/		2.78 /2.2-3.5/		0.896
Gripping R	133.4 /74.8/		139.7 /79.9/		0.711
Gripping L	127.7 /95.0/		127.2 /80.3/		0.571

Values are average (standard deviation) or median /25th to75th percentile/.
SF-36: Short Form Health Survey; Ph: Physical Component Scale and M: Mental Component Scale; HAQ: Health Assessment Questionnaire; DASH: Disabilities of the Arm, Shoulder and Hand; CHFT: Cochin Hand Function Test; VAS: Visual analogue scale 0-100 mm measure for Pain: pain of the body; RP: Raynaud's phenomenon; DU: digital skin ulcers; GI: gastrointestinal; Pulm: pulmonary; Glob: overall problems with SSc; dFTP: Finger To Palm Distance; R: Right; L: Left; HAI: Hand Anatomic Index; Gripping: Grip strength (in Newton).

for a six-month period in patients with SSc. The control SSc group, which received similar physical therapy only for their spine and the large joints leaving out their hands, could not reach improvement in hand function after six months compared to the baseline status. However, comparing the results between the patients and the control groups, there were no significant difference in the values of the functional tests, both at baseline and at six-month follow-up.

Most patients with SSc have significant musculoskeletal damage and loss of function predominantly, but not exclusively, affecting the hands (2-6). There is an essential need for certifying the effectiveness of the currently used physiotherapy and also for improving its fine technics to prevent or at least stop the progression of joint contractures and the impairment of hand function. Regular hand exercise combined with thermal therapy may be an appropriate approach for preventing the development of contractures or at least stop their progression. As in previous studies (11-18), using a combined hand-exercise therapy in conjunction with paraffin baths, mud or soft tissue massage, similarly to our investigation, a significant improvement was detected in the hand movement and function by the end of the treatment.

Our previous three years long follow-up study indicated that a regular education of patients to perform home exercises was not efficient enough to improve hand function (19).

In our present study mud and the whirlpool therapy were supplemented with medical massage to improve microcirculation. Ergotherapy (muscle facilitation, proprioceptive training and exercise for coordination) was also an integral part of the daily training program. In earlier studies the follow-up period took usually nine weeks (15-17). Our aim was to investigate whether a three-week long intensive, complex treatment would be an appropriate approach for achieving a lasting improvement in hand function.

Not surprisingly, the intensive program including regular exercise, ergotherapy with hydrotherapy led to significant transitory improvement in both the interventional and control groups in almost all of the anatomical, functional and global health-related parameters by the end of the three-week treatment period.

At the end of the six-month follow-up, hand-function improvement was maintained only in the hand-exercise group. In the hand-treated group of patients, several parameters, including HAQ, DASH, VAS-pain and VAS-Raynaud showed a long-term improvement. However, several additional parameters including dFTP, HAI, gripping strength and some VAS parameters showed only a transitory three-week improvement without any longstanding effect.

The most important finding was the six-month improvement in the primary outcome of HAQ values and furthermore in the DASH index as well. HAQ predominantly represents the hand function in SSc (19, 26-28), while DASH reflects the functionality of upper extremities including the hands, elbows and shoulders (19, 21). These

Table III. Measured values, changes from baseline and statistical data of groups after six months follow-up.

	Change assessment from baseline to six-month follow-up		
	Hand treated Mean 95%CI	Control Mean 95%CI	*p*
HAQ-DI	-0.206 -0.37 to -0.04	0.007 -0.36 to 0.38	0.217
DASH	-5.2 -9.3 to -1.1	-2.29 -8.5 to 4.0	0.414
CHFT	-2.0 -4.8 to 0.8	-0.5 -2.9 to 2.0	0.452
VAS Pain	-8.3 -15.8 to -0.7	0.44 -12.9 to 13.9	0.208
VAS RP	-12.8 -25.4 to -0.1	-0.94 -14.5 to 12.6	0.210
VAS DU	-11.1 -20.6 to -1.6	2.8 -13.2 to 18.8	0.105
VAS GI	-2.25 -11.2 to 6.7	5.94 -4.9 to 16.8	0.236
VAS Pulm	-1.6 -9.2 to 5.9	0.72 -15.8 to 17.2	0.762
VAS Global	-9.21 -18.8 to 0.5	1.83 -9.8 to 13.4	0.140
HAI R	0.12 -0.14 to -0.4	-0.18 -0.6 to 0.3	0.098
dFTP R	-0.52 -5.5 to 4.4	4.94 -3.7 to 13.6	0,833
Gripping R	15.3 -1.4 to 32.0	0.71 -24.3 to 25.8	0.263

HAQ: Health Assessment Questionnaire; DASH: Disabilities of the Arm, Shoulder and Hand; CHFT: Cochin Hand Function Test; VAS: Visual analogue scale 0-100 mm measure for Pain: pain of the body; RP: Raynaud's phenomenon; DU: digital skin ulcers; GI: gastrointestinal; Pulm: pulmonary; Glob: overall problems with SSc; dFTP: Finger To Palm Distance; R: Right; L: Left; HAI: Hand Anatomic Index; Gripping: Grip strength (in Newton).

two particular indices showed a parallel improvement in the investigated group of patients, and no change was observed in the patient control group (Table II). Control patients received physiotherapy leaving out the hands. It is reasonable to conclude that the improvement measured by these two particular instruments is explained almost exclusively by the improved hand function, and this is an indicator of the effectiveness of the treatment. Furthermore, the median six-month changes in HAQ results reached the value of "minimally important difference for improvement" (MID) (37, 38) in the hand-treated group, based on the HAQ MID estimated in previous clinical trials (-0.022 - -0.14 units).

In the intervention group though, the six-month average improvement of HAQ exceeded the MID value, the delta HAQ changes comparing to the control group, did not reach the statistical significance (Table III). At 80% power the number of patients would have been treated was 301 cases, in both the intervention and control groups, which was not feasible for this particular study.

The improvement in the HAQ and DASH were more pronounced in the more severely affected patients, compared to those showing less damage at baseline. It was also observed that patients exhibiting more improvement in HAQ were significantly younger compared to cases with no changes in HAQ values. Younger patients may have less degenerative joint damage as a comorbidity.

Results of VAS of Raynaud's phenomenon and global pain of the interventional group showed a significant improvement at the six-month follow-up. As also indicated in previous studies (29, 30, 39, 40) the repetitive transitory ischaemia caused by Raynaud's phenomenon is an important cofactor in the deterioration of hand function. Clinical studies examining the effectiveness of non-pharmacological therapy and rehabilitation for the management of Raynaud's phenomenon secondary to SSc are infrequent and they reveal contradictory results, partly due to inconsistent study designs (41). Investigations similarly focused on combined thermal and exercise-based therapy have also been proven to improve the symptoms of Raynaud's phenomenon over their limited follow-ups (12, 13). Even though our study is not randomised, it is controlled, and additionally, it goes beyond the aforementioned studies by incorporating a long follow-up period with an extensive patient base.

Interestingly, the Cochin hand function index did not improve significantly during the six-month period in either group, thus the hand-treated group showed a trend for some improvement that did not reach statistical significance (Table II). During the original validation of the Cochin test for SSc (32) consecutive patients were investigated including some with joint inflammation and with worse results of the average value (± SD) of the Cochin index /21.1±19.2/, as opposed to our study. In this study however, only cases with no signs of joint synovitis or systemic inflammation were included, and the overall Cochin index was much lower during the whole follow-up (the median values were: 9-11.5) (Table II). The different patient population may explain the differences, as the Cochin index may not be sensitive enough to changes in SSc patients without signs of joint inflammation. Further investigation is required to clarify the controversial findings.

The main limitation of our study is the low number of eligible SSc patients, which prevented us from dividing participants into subsets and subgroups

according to disease phase. Regarding eligibility, one-third of the patients were not suitable for the three-week treatment because of their cardiorespiratory or skin problems, and one-third of the patients could not participate because of having other occupational or family activities. The strength of our study is the fact that we were able to follow a relatively large number of patients for an extended period of time (six months) with a very low drop-out rate.

In conclusion, the three-week intensive exercise and ergotherapy for the hands supplemented with mud and hydrotherapy in warm water caused a long-term improvement in the hand function.

Key message

A complex hand-focused exercise program with ergotherapy, mud and hydrotherapy in warm water can lead to a long improvement of the hand function in patients with systemic sclerosis. Further investigations with larger participants are required to confirm the potential beneficial effect of the three-week intensive physical hand therapy in patients with systemic sclerosis.

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