ORIGINAL ARTICLE

Occupational Therapy Treatment to Improve Upper Extremity Function in Individuals with Early Systemic Sclerosis: A Pilot Study

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Objective. To determine the feasibility and preliminary effects of occupational therapy to improve upper extremity function in patients with early systemic sclerosis (SSc; scleroderma) who have upper extremity contractures.

Methods. A single-group pilot clinical rehabilitation trial was conducted at the University of Michigan Scleroderma Center. Patients with SSc and ≥ 1 upper extremity contracture (n = 21) participated in a total of 8 weekly in-person occupational therapy sessions. The therapy consisted of thermal modalities, tissue mobilization, and upper extremity mobility exercises. The participants were instructed to perform upper extremity exercises at home between sessions. Feasibility was measured by the percent enrollment as well as session attendance and session duration. The primary outcome measure was the Shortened Disabilities of the Arm, Shoulder and Hand measure (QuickDASH); secondary and exploratory outcomes included the Patient-Reported Outcomes Measurement Information System (PROMIS) physical function measures; objective measures of upper extremity mobility, strength, and coordination; and skin thickening. Linear mixed models were used to determine the effects of treatment on the primary and secondary outcomes.

Results. Fifty percent of potentially eligible subjects (24 of 48) were interested in participating. Twenty-one (88%) of the 24 subjects were enrolled, and 19 (91%) of these 21 subjects completed all sessions. The mean \pm SD age of the participants was 47.9 \pm 16.1 years; 100% had diffuse SSc, and the mean disease duration was 3.1 years. At 8 weeks, participants had statistically significant improvement in the QuickDASH and PROMIS physical function measure (*P* = 0.0012 and *P* = 0.004, respectively). Approximately one-half of participants in the sample achieved improvement in the QuickDASH and PROMIS measure that exceeded minimally important differences.

Conclusion. In-person treatment sessions were feasible in the patients with SSc and resulted in statistically significant and clinically meaningful improvements in upper extremity and physical function. In future studies, the effects of SSc should be compared with those in a control condition, and the durability of treatment effects should be examined.

INTRODUCTION

Systemic sclerosis (SSc; scleroderma) is a rare, debilitating disease of the connective tissue that not only affects the skin but also can cause severe damage to the internal organs. Despite gains in drug therapies to help control symptoms, patients with SSc face the significant challenge of managing a chronic disease that has a huge impact on daily life. Musculoskeletal complications of SSc can be

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¹Susan L. Murphy, ScD, OTR: University of Michigan and VA Ann Arbor Health Care System, Geriatric Research Education and Clinical Center, Ann Arbor, Michigan; ²Mary Whitehouse Barber, OTR, Carol Dodge, OTR/L, severe, especially in patients with early disease (1,2). In particular, skin thickening and joint contractures in the upper extremities limit the ability to perform daily activities and are associated with disability (3,4) and reduced quality of life (5–7).

Evidence-based rehabilitation interventions for the upper extremity in patients with SSc are limited. Treatments for which some evidence supports their effects include thermal modalities such as paraffin wax baths

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Significance & Innovations

- In a pilot test of occupational therapy treatment consisting of thermal modalities, tissue mobilization, range of motion exercises, functional activities, and home exercises, improvements in reported upper extremity and physical function and some objective performance measures were observed in a small cohort of patients with early systemic sclerosis.
- A regimen of 8 weekly occupational therapy sessions plus home exercises was highly feasible for participants, despite the burden of a long travel distance to the clinic.
- The standardized therapy manual created in this study has the potential to be disseminated to the occupational therapy community after further testing in larger studies, which could increase clinical application of an evidence-based intervention in patients with early systemic sclerosis.

(8-10), range of motion (ROM) exercises (11,12), and manual therapies including tissue mobilization and lymphatic drainage (13-15). Moreover, there have been few high-quality clinical trials evaluating such interventions in patients with SSc (16,17). Most such studies were not randomized, had small sample sizes, used many different outcome measures, and involved various forms of treatments, treatment delivery, and dose, all of which limit the ability to make comparisons. To date, only one large multisite randomized controlled trial tested a rehabilitation intervention in SSc, in which 220 participants were randomized either to an individualized 4-week physical and occupational therapy intervention or to usual care (18). That study demonstrated that intensive rehabilitative treatment for SSc involving exercise not exclusive to the upper extremity had, at minimum, short-term benefits in terms of reported disability and some objective mobility measures. However, the intervention did not include evidence-based treatments such as thermal modalities or tissue mobilization, and a large portion of the intervention was devoted to splinting, although little evidence supports the use of splinting in SSc (17,19,20).

To address the shortcomings in the knowledge base regarding upper extremity interventions in SSc, our study team was interested in testing an intervention that comprised evidence-based components and could eventually be easily disseminated to clinical practice. The majority of occupational therapists who come into contact with a patient with SSc may have little to no experience treating this disease, due to its rarity. Thus, part of our study was devoted to developing and testing a standardized treatment manual that included instruction for therapists regarding adaptations for patients with different upper extremity problems that would facilitate translation into practice once the intervention is fully evaluated and support for effectiveness can be established.

The purpose of this pilot study was to test the feasibility and preliminary effects of standard provision of an in-

person 8-week occupational therapy treatment with prescribed home exercises to improve upper extremity function in patients with early SSC who had contractures. The intervention thought by our team to be most effective for SSc patients involved a minimum of 8 in-person visits with the occupational therapist. Because of the rarity of SSc and the fact that many patients with SSc travel long distances to the health system clinic, it was necessary to examine the feasibility of this intervention. In addition to feasibility, we examined the preliminary effects of treatment over time using the Shortened Disability Arm Shoulder Hand measure (QuickDASH) of upper extremity function, Patient-Reported Outcomes Measurement Information System (PROMIS) physical function measure; objective measures of upper extremity mobility, strength, and coordination; and skin thickening. We hypothesized that the treatment would be feasible to deliver, and that it would demonstrate preliminary effects on patient-reported functional measures.

PATIENTS AND METHODS

Design. This pilot study used a single-group pretest/ posttest design with a target sample of 20 patients with SSc. Data for the outcome measures were collected at baseline, 4 weeks (mid-treatment), and 8 weeks (immediately following treatment).

Sample. Participants were recruited from the Scleroderma Center at the University of Michigan Health System from September 2016 to May 2017. Potential participants were contacted either by phone (if they were included in an established research registry at the Scleroderma Center) or in person at their clinic visit if they appeared to meet the inclusion criteria based on review of their electronic medical records. To be eligible for this study, participants had to be 18 years of age or older, have SSc, have a contracture of the hand and another joint in at least one arm (e.g., wrist or elbow) with the ability to demonstrate active ROM in that arm, speak English, have no active hand ulcers and no concurrent medical issues, and be willing to travel to the Scleroderma Center for treatment. We focused on patients with early SSc with a diffuse cutaneous distribution, because our hypothesis was that early upper extremity contractures are related to active and progressive skin and joint disease and are amenable to treatment, whereas late disease reflects greater damage and does not improve with therapy. Early SSc was considered to be disease with a duration of <5 years after onset, which is similar to the designation used in a previous study (21).

Procedure. The research coordinator met with potential participants who were initially eligible based on telephone screening or a review of their electronic medical records, prior to a clinic visit. After eligibility was confirmed and informed consent was obtained, participants were scheduled for a baseline visit with the occupational therapist. The therapist administered questionnaires to evaluate upper extremity function (QuickDASH) and overall physical function (PROMIS), conducted active and passive ROM assessments, assessed skin thickness and grip/pinch strength, and

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Table 1. Treatment protocol*				
Technique				
Thermal modalities				
Hot packs, focused on areas with limitations				
Paraffin, focused on digital limitations				
PhysioTouch, applied proximal to distal in areas with pathological skin in sections				
Passive ROM exercises				
Hold end position of joint for 3–10 seconds (dependent on skin and joint				
integrity); repeat for each affected joint/digit				
Active ROM exercises				
Functional activities (manual dexterity activities such as working with small				
foam cubes or jar openers, or rolling putty) (limited due to time)				
Tailored active and passive ROM exercises based on limitations in upper extremity mobility				

administered tests of hand coordination. These outcome assessments were performed at baseline, 4 weeks, and 8 weeks. Treatment was conducted each week over 8 weeks at the University of Michigan Scleroderma Center, an outpatient rehabilitation clinic. Treatment involved preparatory thermal modalities, tissue mobilization, and upper extremity mobility exercises beginning with passive ROM and ending with active ROM (see Table 1). Tissue mobilization was performed using a PhysioTouch device, (also called LymphaTouch; Healthy Life Devices Ltd). PhysioTouch is a negative pressure device that has been used primarily to decrease swelling in tissue (22) but is currently being used at our healthcare system as a treatment for patients with SSc, because it delivers mild tissue mobilization (23), which may provide better mobilization than that provided using manual techniques. The therapist also instructed participants how to perform a home ROM exercise program that was tailored as needed for each participant based on the severity of contractures and level of arm mobility. Participants were instructed to complete daily exercise sessions at home.

Development of a standardized treatment manual. The treating therapist, therapist consultant (a certified hand therapist with >30 years of experience treating patients with scleroderma), and the principal investigator developed an initial guide for treatment, as shown in Table 1. The treatment components were chosen based on support for their effects in the literature (thermal modalities, tissue mobilization, and ROM) and reflected current practices at our clinic. During each treatment session, the therapist logged the duration of each treatment component and noted any adaptations made to treatment based on each individual's disease severity or specific impairments. After all participants completed treatment, the treatment manual was reviewed, and details were included to provide instruction on how to deliver the intervention. An excerpt of the treatment manual is provided in the Supplementary Appendix (available on the Arthritis Care & Research web site at http://onlinelibrary.wiley.com/doi/10.1002/acr.23522/ abstract).

Feasibility measures. We tested feasibility against a priori criteria: 1) at least 50% of participants who were eligible for the study would enroll, 2) at least 80% of

participants would attend all treatment sessions, and 3) the sessions that included both treatment and outcome assessments would not last, on average, >2 hours. We determined how many individuals who were initially approached, either through a telephone call or an in-person clinic visit, were interested in participating. We also assessed what percentage of participants completed all 8 sessions. We examined the feasibility of providing treatment that can potentially be provided via outpatient visits. Thus, we assessed the length of time needed to complete all procedures performed during the in-person sessions.

Primary outcome. The primary outcome was upper extremity function as measured by the QuickDASH questionnaire, a reliable and validated self-reported measure used in the population of patients with SSc (24,25). QuickDASH is an 11-item questionnaire in which difficulties in several tasks involving the upper extremity are rated, along with interference with daily life activities and the severity of symptoms. Items are averaged and converted to a 0–100-point scale; a higher score indicates worse function. This measure is responsive to change, and a 16-point increase is the minimal clinically important difference in patients with shoulder and arm limitations (26).

Secondary outcomes. The secondary outcomes included general reported physical function as assessed by the PROMIS physical function version 2.0 8-item short form; in the US population, the mean \pm SD score is 50 \pm 10, with a higher score denoting better function (27). A 2-point improvement in the T score is considered to be clinically meaningful (28). The main measure of ROM was total active hand motion in the right and left hands. This was calculated by summing the total active ROM for each finger and thumb, using a goniometer (260 degrees in each finger and 135 degrees in the thumb) (29); a total score of 1,125 for each hand was possible. The therapist also took photographs of participants that showed each ROM in the upper extremity at each outcome assessment, as another way of evaluating change over time. Coordination was measured using the 9-hole peg test, a commonly used test of dexterity in which an individual needs to put 9 pegs in holes on a peg board while being timed (30). Handgrip

strength was measured in pounds of pressure, using a Jamar hand dynamometer (Lafayette Instruments) according to a standardized protocol in which the participant squeezes the dynamometer while seated with his or her elbow at a 90degree angle (31). The value used was the maximum of 6 trials (3 trials for both the left and right upper extremities).

Exploratory outcomes. Outcomes that were considered to be exploratory were measures that were thought by the team of therapists to be important in upper extremity function but may not have been as directly impacted by the treatment. These outcomes included: 1) active ROM of wrist flexion and elbow flexion for each upper extremity, as measured using a goniometer, 2) lateral pinch strength, as measured using a pinch gauge in an average of 3 trials (31), and 3) skin thickness, as assessed using the modified Rodnan skin thickness score (MRSS) (32). The MRSS was assessed at baseline and at 8 weeks by a clinic rheumatologist who was not part of the study team.

Sample size determination. The target sample size was 20 participants; this sample size was thought to be sufficient to establish feasibility over the 1-year study period. With 20 participants, we determined that at 80% power we could detect an effect of 0.67 SD units, which is an ~16-point change in the QuickDASH measure, a cutoff value reported for the minimally clinically important difference in patients with shoulder limitations (25).

Statistical analysis. Descriptive statistics were used to examine the feasibility of study processes and treatment protocol, and compared feasibility with our a priori criteria for success. To examine the change over time from the baseline to the 4-week and 8-week assessments in our primary and secondary outcome measures, we used linear mixed models using all available data, which served as an intent-to-treat analysis. For exploratory outcomes, we performed a per-protocol analysis in which completer data

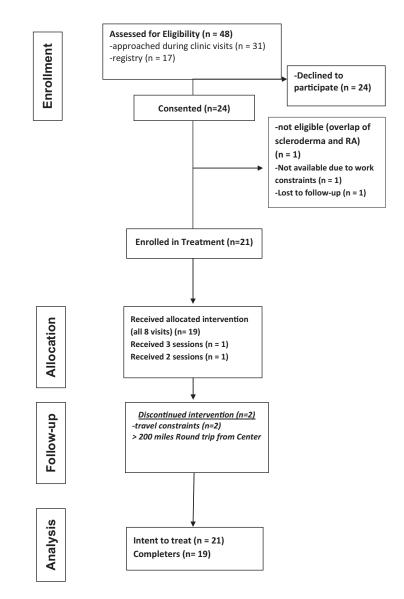


Figure 1. Flow diagram showing participant enrollment, allocation, and follow-up.

were examined for change over time using one-way repeated-measures analyses of variance or paired *t*-tests.

RESULTS

Participant flow and characteristics. Figure 1 shows participant flow through the study. Forty-eight potentially eligible participants identified by chart review or from the early scleroderma research registry were either approached at a clinic visit or by telephone. Of these, 24 subjects (50%) were interested and consented to participate. A main reason for not participating was travel burden. Individuals who were interested were screened in person and completed the informed consent process. One person did not meet the study eligibility criteria, and 2 individuals chose not to participate due to not being able to get time off from work or being unable to schedule visits. Twentyone of the eligible participants were enrolled. Despite their interest in participating, 2 of the subjects were lost to follow-up due to travel constraints (the roundtrip distance to the treatment center ranged from 200 miles to 550 miles per session).

The baseline characteristics of the sample (n = 21) are shown in Table 2. Participants were predominantly female, and 38% identified as a racial minority, with almost one-fourth of the sample identifying as African American. Participants ranged in age from 20 years to 75 years (mean 47.9 years). All participants had diffuse cutaneous SSc. The mean \pm SD duration of SSc was 3.1 ± 2.3 years. The mean \pm SD MRSS was 17.6 ± 9.7 , indicating moderate skin disease. The majority of patients were being treated with immunosuppressive therapy or were participants in ongoing clinical trials for their aggressive skin disease.

Feasibility outcomes. Nineteen participants (91% of the enrolled sample) completed the protocol as intended, attending all 8 in-person sessions. These participants traveled a mean \pm SD of 103.4 \pm 82.5 miles roundtrip for each session, with 37% traveling between 100 miles and ~340 miles each session. There were a few protocol deviations due to timing of the sessions. One participant stopped and then restarted treatment 2 months later due to travel issues but then was able to attend all 8 sessions. Fifteen of the 19 participants who attended all sessions attended them weekly, whereas the remaining 4 participants had at least one cancellation and rescheduled for the next available slot (usually the following week). There was a protocol deviation due to treatment of a participant with an active hand ulcer, which is a relatively common phenomenon in SSc. This patient received modified treatment modalities (e.g., no paraffin treatment to the affected hand). Both of these participants received modified treatment modalities (such as no paraffin treatment to affected hands). We also evaluated the time required to administer sessions in which evaluation plus treatment were combined (at baseline, 4 weeks, and 8 weeks) for feasibility of administration. Ten percent of the evaluation-plus-treatment sessions (6 of 59 total sessions) lasted longer than 2 hours, which exceeded our feasibility target; however, 5 of those sessions occurred at baseline, and

Table 2. Baseline characteristics of the 21 patients*				
Age, mean \pm SD (range) years	$47.9 \pm 16.1 \; \text{(20-75)}$			
Female sex	18 (86)			
Race				
Black/African American	5 (23.8)			
White	13 (61.9)			
Other	3 (14.3)			
Ethnicity				
Non-Hispanic/non-Latino	3 (13.4)			
Hispanic/Latino	1 (4.8)			
Other	7 (33.3)			
Married	10 (47.6)			
High school education or less	8 (38.1)			
MRSS, mean \pm SD (range 1–37)†	17.6 ± 9.7			
Disease duration, mean \pm SD years	3.1 ± 2.3			
Diffuse cutaneous SSc	21 (100)			
Interstitial lung disease, %	42.9			
Gastrointestinal involvement, %	81.0			
Raynaud's phenomenon	19 (90.5)			
Use of immunosuppressive agents				
MMF	10 (48)			
MTX	5 (23.8)			
MMF + MTX	2 (9.5)			
Investigational agents	4 (19.1)			
Abatacept ⁺	1 (4.8)			
None	4 (19.1)			
 * Except where indicated otherwise, val- MRSS = modified Rodnan skin thickness nolate mofetil; MTX = methotrexate. † Twenty patients were assessed. 				

the therapist was able to improve process efficiency at almost all of the subsequent sessions.

Adverse events and unanticipated problems. There were no adverse events or unanticipated problems related to treatment. The fingernail of one participant fell off prior to presentation at the last session of treatment, but this event was considered to be unrelated to the treatment provided. Thus, after this issue was detected, the final treatment session and the last outcome assessment did not include any activity that involved the affected digit.

Effects of treatment. Table 3 shows the results of each outcome using linear mixed models. Participants had a mean 6.6-point improvement on the QuickDASH at 4 weeks, which was not significant; however, participants continued to improve from 4 weeks to 8 weeks, with a mean 14-point improvement from baseline (t[2,36] = 3.53, P = 0.0012). Using a previously cited clinically meaningful cut point of a 16-point improvement on the QuickDASH (25), 47% of participants who completed the intervention (9 of 19) met this threshold.

For the PROMIS physical function measure, participants had a significant improvement from baseline over the 8-week treatment period. Similar to the observed Quick-DASH trends, change from baseline to 4 weeks was not significant; however, improvements continued from 4 weeks to 8 weeks, with a significant effect (t[2,36] = -3.08, P = 0.004). The mean improvement over time was 3.1 points on the PROMIS, demonstrating a change of one-third

	Baseline	Mid-treatment (4 weeks)	Post-treatment (8 weeks)	Р
Primary outcome				
QuickDASH score†	49.3 (4.6)	42.7 (4.8)	35.2 (4.8)	0.0012
Secondary outcomes				
PROMIS physical function score‡	38.0 (1.3)	38.5 (1.4)	41.1 (1.4)	0.004
Total active motion§				
Left hand	736.5 (41.0)	797.3 (41.3)	778.0 (41.3)	0.013
Right hand	745.2 (43.1)	775.5 (43.4)	758.0 (43.4)	0.49
9-hole peg test, seconds				
Left hand	25.4 (1.6)	21.5 (1.6)	22.9 (1.6)	0.03
Right hand	23.6 (1.6)	21.9 (1.6)	21.8 (1.6)	0.15
Handgrip strength¶	45.8 (4.1)	45.4 (4.1)	43.3 (4.1)	0.06

* Linear mixed models were used. PROMIS = Patient-Reported Outcomes Measurement Information System.

+ A higher score indicates worse function.

‡ A higher score indicates better function.

§ Calculated by summing the total range of motion for each finger and thumb (260 degrees for each finger and

135 degrees for the thumb, with a total possible score of 1,175 degrees). ¶ Maximum value for either hand.

Maximum value for either hand

of an SD, which is larger than the minimally important difference of 2 points on the PROMIS 20-item physical function scale observed in a sample of patients with rheumatoid arthritis sample (28). Fifty-three percent of the participants in our sample achieved a 2-point increase on the PROMIS physical function scale after 8 weeks.

Among the objective upper extremity measures, left total active hand function and left 9-hole peg test scores were significantly improved after the intervention. On average, participants gained 41.5 degrees of active hand motion at 8 weeks, and their performance on the 9-hole peg test with the left hand was 2.5 seconds faster. There are no established clinically important differences for either of these measures. Figure 2 shows examples of improvement from baseline to 8 weeks in active ROM. No significant improvement in active hand motion or coordination in the right hand after 8 weeks was observed. Handgrip strength did not improve, and participants had a slightly weaker handgrip at 8 weeks, although the difference was not statistically significant (P = 0.06). For the exploratory outcomes, there were no statistically significant changes in wrist or elbow flexion or lateral pinch measures. Skin thickness, as evaluated using the MRSS, was assessed in 15 participants at the posttest visit, and a paired *t*-test on completer data showed no significant change and slight worsening from baseline to 8 weeks (mean \pm SD 17.9 \pm 7.9 at baseline and 20.1 \pm 9.5 8 weeks; t[1,14] = -1.4 [P = 0.18]).

DISCUSSION

In this study, we examined the feasibility and preliminary effects of an 8-session in-person occupational therapy treatment to improve upper extremity outcomes in patients with SSc. In general, the study and the treatment were feasible, supported by our ability to enroll and retain participants despite their burden of traveling to the center. Of 47 eligible participants, 51% were enrolled; this percentage was slightly above our target of 50%. Attendance at all 8 sessions by the enrolled patients exceeded our expectations, in that 91% of the sample met this metric despite the burden of traveling a mean distance of >100 miles roundtrip for each session. In an attempt to reduce participant travel burden, we consolidated treatment and outcome visits at baseline, 4 weeks, and 8 weeks. We examined the feasibility of conducting these combined sessions in a 2-hour period, and 90% of the sessions met the criterion that the sessions would not take more than 2 hours to complete. The sessions that exceeded the time limit occurred early in the study, and timing improved as the therapist was able to streamline the processes.

The treatment provided showed the strongest effects in reported improvement in upper extremity function and general physical function. In approximately one-half of the sample, improvements in the QuickDASH and PROMIS physical function measures (47% and 53%, respectively) were considered to be clinically important, based on established cutoffs in other populations (26,28). Because of the lack of studies in SSc that evaluated these clinically important differences, it is not clear whether this finding is an accurate reflection of which participants benefited as a result of the treatment. For instance, depending on the study, different values for minimal clinically important differences for QuickDASH have been reported; in one study, cutoff score of 8 was reported, and another study demonstrated a minimal clinically important difference of 14 (24,33). Thus, our chosen cutoff of 16 points is likely conservative, and more patients in our sample may have benefited.

Improvements were shown in some but not all objective measures, and most improvement occurred in hand mobility and coordination. Significant effects were observed only in the left hand, although trends for improvement were similar in both hands. Interestingly, more gains occurred in the first 4 weeks of treatment. However, gains in these measures continued from 4 weeks to 8 weeks, showing that the additional sessions were valuable. It remains unclear how many sessions are optimal for sustaining gains made during treatment. Most studies investigating upper extremity rehabilitation interventions in SSc were designed to measure

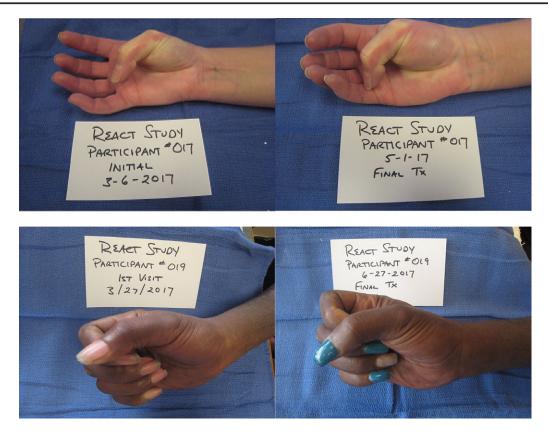


Figure 2. Photographs of the right hands of participants 17 and 19, showing improvement from baseline to 8 weeks in upper extremity mobility.

short-term efficacy, with end points ranging from 4 weeks to 3 months (9,10,12-15), and most clinic-based interventions lasted 3-8 weeks; however, the intensity of these interventions was variable. The highest-quality randomized controlled trial investigating upper extremity rehabilitation interventions in SSc showed that a 3-week intensive intervention, consisting of 36 treatment hours and a prescribed daily home exercise program, had short-term effects on disability (as rated using the Health Assessment Questionnaire) that diminished over time (at 6 months and 12 months of follow-up) (18). Although long-term adherence to the home exercise program was poor, participants who did adhere to the daily home exercises had better effects over time compared with those who did not adhere, which supports the inclusion of home exercise in future interventions. In future studies, home exercise programs likely need to be more engaging for participants in order to improve adherence.

A main strength of this study is that we tested a treatment informed by available evidence supporting specific treatment components in SSc, which capitalized on the extensive experience of our therapist team who commonly provide upper extremity treatment for patients with SSc. In addition, the creation of a standardized manual, as was done in this study, will be important for further testing of this intervention and has the potential to provide an evidence-based guide for therapists who treat patients with SSc on a broad scale.

The ability to draw conclusions based on this study is limited due to its single-group design. Thus, the assessment of outcomes does not provide definitive evidence of the efficacy of treatment. Further, due to the size and scope of this study, the therapist also served as the outcome assessor and therefore did not perform assessments in a blinded manner. Tracking of home exercise needs to be strengthened in future studies, because we did not formally assess adherence. Thus, it was not possible to disentangle the effects of home exercise from the effects of in-person sessions. Understanding the effects of in-person intervention versus home exercise will be important in future research studies, because participation in the intervention was precluded mainly because of the travel distance to the center. Given that our sample included all patients with diffuse SSc who were in the early stages of the disease (within 5 years of diagnosis), our findings can be generalized only to this population. In addition, we are not certain whether these improvements were maintained after the in-person sessions were completed.

In conclusion, this pilot single-group trial supported the feasibility of an 8-session occupational therapy intervention to address upper extremity function in patients with SSc. Preliminary effects were observed at 8 weeks, with reported improvements in upper extremity disability, physical function, and objective measures of hand mobility and coordination. Although definitive treatment effects cannot be determined from this study, therapists who are unfamiliar with treating patients with SSc may benefit from reviewing information on the treatment provided in order to gain knowledge of progression of treatment components and recommended adaptations based on individual differences. Further larger studies are needed that include a control or comparator group and that examine the durability of treatment effects.

AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be published. Dr. Murphy had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. **Study conception and design.** Murphy, Dodge, Cutter, Khanna. **Acquisition of data.** Murphy, Barber, Homer.

Analysis and interpretation of data. Murphy, Barber, Homer, Dodge, Khanna.

REFERENCES

- 1. Denton C, Khanna D. Systemic sclerosis. Lancet 2017;390: 1685–99.
- 2. Bálint Z, Farkas H, Farkas N, Minier T, Kumánovics G, Horváth K, et al. A three-year follow-up study of the development of joint contractures in 131 patients with systemic sclerosis. Clin Exp Rheumatol 2014;32:S68–74.
- 3. Sandqvist G, Eklund M, Åkesson A, Nordenskiöld U. Daily activities and hand function in women with scleroderma. Scand J Rheumatol 2004;33:102–7.
- 4. Sandqvist G, Scheja A, Hesselstrand R. Pain, fatigue and hand function closely correlated to work ability and employment status in systemic sclerosis. Rheumatology 2010;49:1739–46.
- Frantz C, Avouac J, Distler O, Amrouche F, Godard D, Kennedy AT, et al. Impaired quality of life in systemic sclerosis and patient perception of the disease: a large international survey. Semin Arthritis Rheum 2016;46:115–23.
- 6. Khanna D, Yan X, Tashkin D, Furst DE, Elashoff R, Roth MD, et al. Impact of oral cyclophosphamide on health-related quality of life in patients with active scleroderma lung disease: results from the Scleroderma Lung Study. Arthritis Rheumatol 2007;56:1676–84.
- Maddali-Bongi S, Del Rosso A, Mikhaylova S, Francini B, Branchi A, Baccini M, et al. Impact of hand and face disabilities on global disability and quality of life in systemic sclerosis patients. Clin Exp Rheumatol 2014;32:S15–20.
- 8. Horvath J, Balint Z, Szep E, Deiszinger A, Minier T, Farkas N, et al. Efficacy of intensive hand physical therapy in patients with systemic sclerosis. Clin Exp Rheumatol 2017; 35 Suppl 106:159–66.
- 9. Mancuso T, Poole JL. The effect of paraffin and exercise on hand function in persons with scleroderma: a series of single case studies. J Hand Ther 2009;22:71–8.
- Sandqvist G, Åkesson A, Eklund M. Evaluation of paraffin bath treatment in patients with systemic sclerosis. Disabil Rehabil 2004;26:981–7.
- 11. Mugii N, Hasegawa M, Matsushita T, Kondo M, Orito H, Yanaba K, et al. The efficacy of self-administered stretching for finger joint motion in Japanese patients with systemic sclerosis. J Rheumatol 2006;33:1586–92.
- 12. Stefanantoni K, Sciarra I, Iannace N, Vasile M, Caucci M, Sili Scavalli A, et al. Occupational therapy integrated with a self-administered stretching programme on systemic sclerosis patients with hand involvement. Clin Exp Rheumatol 2016;34:15761.
- Bongi SM, Del Rosso A, Galluccio F, Tai G, Sigismondi F, Passalacqua M, et al. Efficacy of a tailored rehabilitation program for systemic sclerosis. Clin Exp Rheumatol 2009;27: S44–50.
- 14. Bongi SM, Del Rosso A, Passalacqua M, Miccio S, Cerinic MM. Manual lymph drainage improving upper extremity edema and hand function in patients with systemic sclerosis in edematous phase. Arthritis Care Res (Hoboken) 2011;63:1134–41.

- 15. Bongi SM, Rosso A, Galluccio F, Sigismondi F, Miniati I, Conforti ML, et al. Efficacy of connective tissue massage and Mc Mennell joint manipulation in the rehabilitative treatment of the hands in systemic sclerosis. Clin Rheumatol 2009;28:1167–73.
- Willems LM, Vriezekolk JE, Schouffoer AA, Poole JL, Stamm TA, Boström C, et al. Effectiveness of nonpharmacologic interventions in systemic sclerosis: a systematic review. Arthritis Care Res (Hoboken) 2015;67:1426–39.
- 17. Poole JL. Musculoskeletal rehabilitation in the person with scleroderma. Curr Opin Rheumatol 2010;22:205–12.
- Rannou F, Boutron I, Mouthon L, Sanchez K, Tiffreau V, Hachulla E, et al. Personalized physical therapy versus usual care for patients with systemic sclerosis: a randomized controlled trial. Arthritis Care Res (Hoboken) 2017;69:1050–9.
- Harvey LA, Katalinic OM, Herbert RD, Moseley AM, Lannin NA, Schurr K. Stretch for the treatment and prevention of contractures. Cochrane Database Syst Rev 2017;CD007455.
- Seeger MW, Furst DE. Effects of splinting in the treatment of hand contractures in progressive systemic sclerosis. Am J Occup Ther 1987;41:118-21.
- Khanna D, Hays RD, Furst DE. Functional disability and other health-related quality-of-life domains: points to consider for clinical trials in systemic sclerosis. Rheumatology 2017;56:v17-22.
- Iivarinen JT, Korhonen RK, Jurvelin JS. Modeling of interstitial fluid movement in soft tissue under negative pressure: relevance to treatment of tissue swelling. Comput Methods Biomech Biomed Engin 2016;19:1089–98.
- Adams A. Technical note: PhysioTouch. Rehabil Oncol 2016; 34:49–50.
- 24. Kennedy CA, Beaton DE, Smith P, Van Eerd E, Tang K, Inrig T, et al. Measurement properties of the QuickDASH (Disabilities of the Arm, Shoulder and Hand) outcome measure and cross-cultural adaptations of the QuickDASH: a systematic review. Qual Life Res 2013;22:2509–47.
- 25. Varjú C, Bálint Z, Solyom AI, Farkas H, Kárpáti E, Berta B, et al. Cross-cultural adaptation of the disability of the arm, shoulder, and hand (DASH) questionnaire into Hungarian and investigation of its validity in patients with systemic sclerosis. Clin Exp Rheumatol 2008;26:776–84.
- 26. Franchignoni F, Vercelli S, Giordano A, Sartorio F, Bravini E, Ferriero G. Minimal clinically important difference of the disabilities of the arm, shoulder and hand outcome measure (DASH) and its shortened version (quickDASH). J Orthop Sports Phys Ther 2014;44:30–9.
- 27. Cella D, Riley W, Stone A, Rothrock N, Reeve B, Yount S, et al. The Patient-Reported Outcomes Measurement Information System (PROMIS) developed and tested its first wave of adult self-reported health outcome item banks: 2005-2008. J Clin Epid 2010;63:1179–94.
- Hays RD, Spritzer KL, Fries JF, Krishnan E. Responsiveness and minimally important difference for the Patient-Reported Outcomes Measurement Information System (PROMIS) 20item physical functioning short form in a prospective observational study of rheumatoid arthritis. Ann Rheum Dis 2014;74:104–7.
- Adams L, Greene L, Topoozian E. Clinical assessment recommendations. In: American Society of Hand Therapists. Chicago: ASHT Clinical Assessment Recommendation; 1992. p. 56–70.
- Grice KO, Vogel KA, Le V, Mitchell A, Muniz S, Vollmer MA. Adult norms for a commercially available nine hole peg test for finger dexterity. Am J Occup Ther 2003;57:570–3.
- Mathiowetz V, Weber K, Volland G, Kashman N. Reliability and validity of grip and pinch strength evaluations. J Hand Surg Am 1984;9:222–6.
- 32. Clements P, Lachenbruch P, Seibold J, Zee B, Steen VD, Brennan P, et al. Skin thickness score in systemic sclerosis: an assessment of interobserver variability in 3 independent studies. J Rheumatol 1993;20:1892–6.
- Sorensen AA, Howard D, Tan WH, Ketchersid J, Calfee RP. Minimal clinically important differences of 3 patient-rated outcomes instruments. J Hand Surg Am 2013;38:641–9.