Physical or Occupational Therapy Utilization in Systemic Sclerosis: A Scleroderma Patient-centered Intervention Network Cohort Study

Karima Becetti[1], Linda Kwakkenbos[2], Marie-Eve Carrier[3], Jessica K. Gordon[1], Joseph Nguyen[1], Carol A. Mancuso[1], Luc Mouthon[4,5], Christelle Nguyen[4], François Rannou[4], Joep Welling[6,7], Brett D. Thombs[3,8-11], Robert F. Spiera[1] and the SPIN Investigators[12]

[1]Hospital for Special Surgery, New York City, New York, USA; [2]Behavioural Science Institute, Clinical Psychology, Radboud University, Nijmegen, the Netherlands; [3]Lady Davis Institute for Medical Research, Jewish General Hospital, Montreal, Quebec, Canada; [4]Université Paris Descartes, Assistance Publique - Hôpitaux de Paris, Paris, France; [5]Service de Médecine Interne, Hôpital Cochin, Paris, France; [6]NVLE Dutch patient organization for systemic autoimmune diseases, Utrecht, The Netherlands; [7]Federation of European Scleroderma Associations, Brussel, Belgium; Departments of [8]Psychiatry, [9]Epidemiology, Biostatistics, and Occupational Health, [10]Medicine, and [11]Psychology, McGill University, Montreal, Quebec, Canada; [12]SPIN Investigators: Murray Baron, McGill University, Montreal, Quebec, Canada; Susan J. Bartlett, McGill University, Montreal, Quebec, Canada; Dan E. Furst, Division of Rheumatology, Geffen School of Medicine, University of California, Los Angeles, California, USA; Karen Gottesman, Scleroderma Foundation, Los Angeles, California, USA; Vanessa Malcarne, San Diego State

University, San Diego, California, USA; Maureen D. Mayes, University of Texas McGovern School of Medicine, Houston, Texas, USA; Warren R. Nielson, St. Joseph's Health Care, London, Ontario, Canada; Robert Riggs, Scleroderma Foundation, Danvers, Massachusetts, USA; Maureen Sauve, Scleroderma Society of Ontario, Hamilton, Ontario; Fredrick Wigley, Johns Hopkins University School of Medicine, Baltimore, Maryland, USA; Shervin Assassi, University of Texas McGovern School of Medicine, Houston, Texas, USA; Isabelle Boutron, Université Paris Descartes, and Assistance Publique - Hôpitaux de Paris, Paris, France; Angela Costa Maia, University of Minho, Braga, Portugal; Ghassan El-Baalbaki, Université du Québec à Montréal, Montreal, Quebec, Canada; Carolyn Ells, McGill University, Montreal, Quebec, Canada; Cornelia van den Ende, Sint Maartenskliniek, Nijmegen, The Netherlands; Kim Fligelstone, Scleroderma & Raynaud's UK, London, UK; Catherine Fortune, Scleroderma Society of Ontario, Hamilton, Ontario, Canada; Tracy Frech, University of Utah, Salt Lake City, Utah, USA; Dominique Godard, Association des Sclérodermiques de France, Sorel-Moussel, France; Geneviève Guillot, Sclérodermie Québec, Montreal, Quebec, Canada; Daphna Harel, New York University, New York, New York, USA; Marie Hudson, McGill University, Montreal, Quebec, Canada; Ann Impens, Midwestern University, Downers Grove, Illinois, USA; Yeona Jang, McGill University, Montreal, Quebec, Canada; Sindhu R. Johnson, Toronto Scleroderma Program, Mount Sinai Hospital, Toronto Western Hospital, and University of Toronto, Toronto, Ontario, Canada; Ann Tyrell Kennedy, Federation of European Scleroderma Associations, Dublin, Ireland; Annett Körner, McGill University, Montreal, Quebec, Canada; Maggie Larche, McMaster University, Hamilton, Ontario, Canada; Catarina Leite, University of Minho, Braga, Portugal; Carlo Marra, Memorial University, St. John's, Newfoundland, Canada; Karen Nielsen, Scleroderma Society of Ontario, Hamilton, Ontario, Canada; Janet Pope, University of Western Ontario, London, Ontario, Canada; Alexandra Portales, Asociación Española de Esclerodermia, Madrid, Spain; Tatiana Sofia Rodriguez Reyna, Instituto Nacional

de Ciencias Médicas y Nutrición Salvador Zubirán, Mexico City, Mexico; Anne A. Schouffoer, Leiden University Medical Center, Leiden, The Netherlands; Russell J. Steele, Jewish General Hospital and McGill University, Montreal, Quebec, Canada; Maria E. Suarez-Almazor, University of Texas MD Anderson Cancer Center, Houston, Texas, USA; Durhane Wong-Rieger, Canadian Organization for Rare Disorders, Toronto, Ontario, Canada; Christian Agard, Centre Hospitalier Universitaire - Hôtel-Dieu de Nantes, Nantes, France; Alexandra Albert, Université Laval, Quebec, Quebec, Canada; Marc André, Centre Hospitalier Universitaire Gabriel-Montpied, Clermont-Ferrand, France; Guylaine Arsenault, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Nouria Benmostefa, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Ilham Benzidia, Assistance Publique - Hôpitaux de Paris, Hôpital St-Louis, Paris, France; Sabine Berthier, Centre Hospitalier Universitaire Dijon Bourgogne, Dijon, France; Lyne Bissonnette, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Gilles Boire, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Alessandra Bruns, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Patricia Carreira, Servicio de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Marion Casadevall, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Benjamin Chaigne, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Lorinda Chung, Stanford University, Stanford, California, USA; Pascal Cohen, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Pierre Dagenais, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Christopher Denton, Royal Free London Hospital, London, UK; Robyn Domsic, University of Pittsburgh, Pittsburgh, Pennsylvania, USA; Sandrine Dubois, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; James V. Dunne, St. Paul's Hospital and University of British Columbia, Vancouver, British Columbia, Canada; Bertrand Dunogue, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Alexia Esquinca, Instituto Nacional de Ciencias Médicas y Nutrición Salvador Zubirán, Mexico City, Mexico; Regina Fare, Servicio

de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Dominique Farge-Bancel, Assistance Publique - Hôpitaux de Paris, Hôpital St-Louis, Paris, France; Paul Fortin, CHU de Québec - Université Laval, Quebec, Quebec, Canada; Anna Gill, Royal Free London Hospital, London, UK; Brigitte Granel-Rey, Aix Marseille Université, and Assistance Publique - Hôpitaux de Marseille, Hôpital Nord, Marseille, France; Claire Grange, Centre Hospitalier Lyon Sud, Lyon, France; Genevieve Gyger, Jewish General Hospital and McGill University, Montreal, Quebec, Canada; Eric Hachulla, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; Pierre-Yves Hatron, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; Ariane L Herrick, University of Manchester, Salford Royal NHS Foundation Trust, Manchester, UK; Adrian Hij, Assistance Publique - Hôpitaux de Paris, Hôpital St-Louis, Paris, France; Monique Hinchcliff, Northwestern University, Chicago, Illinois, USA; Alena Ikic, Université Laval, Quebec, Quebec, Canada; Niall Jones, University of Alberta, Edmonton, Alberta, Canada; Artur Jose de B. Fernandes, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Suzanne Kafaja, University of California, Los Angeles, California, USA; Nader Khalidi, McMaster University, Hamilton, Ontario, Canada; Benjamin Korman, Northwestern University, Chicago, Illinois, USA; Marc Lambert, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; David Launay, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; Patrick Liang, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Jonathan London, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Hélène Maillard, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; Nancy Maltez, University of Ottawa, Ottawa, Ontario, Canada; Joanne Manning, Salford Royal NHS Foundation Trust, Salford, UK; Isabelle Marie, CHU Rouen, Hôpital de Bois-Guillaume, Rouen, France; Maria Martin, Servicio de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Thierry Martin, Les Hôpitaux Universitaires de Strasbourg, Nouvel Hôpital Civil, Strasbourg, France; Ariel

Masetto, Université de Sherbrooke, Sherbrooke, Quebec, Canada; François Maurier, Hôpitaux Privés de Metz, Hôpital Belle-Isle, Metz, France; Arsene Mekinian, Assistance Publique - Hôpitaux de Paris, Hôpital St-Antoine, Paris, France; Sheila Melchor, Servicio de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Mandana Nikpour, St Vincent's Hospital and University of Melbourne, Melbourne, Victoria, Australia; Romain Paule, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Susanna Proudman, Royal Adelaide Hospital and University of Adelaide, Adelaide, South Australia, Australia; Alexis Régent, Assistance Publique - Hôpitaux de Paris, Hôpital Cochin, Paris, France; Sébastien Rivière, Assistance Publique - Hôpitaux de Paris, Hôpital St-Antoine, Paris, France; David Robinson, University of Manitoba, Winnipeg, Manitoba, Canada; Esther Rodriguez, Servicio de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Sophie Roux, Université de Sherbrooke, Sherbrooke, Quebec, Canada; Perrine Smets, Centre Hospitalier Universitaire Gabriel-Montpied, Clermont-Ferrand, France; Doug Smith, University of Ottawa, Ottawa, Ontario, Canada; Vincent Sobanski, Centre Hospitalier Régional Universitaire de Lille, Hôpital Claude Huriez, Lille, France; Virginia Steen, Georgetown University, Washington, DC, USA; Wendy Stevens, St Vincent's Hospital and University of Melbourne, Melbourne, Victoria, Australia; Evelyn Sutton, Dalhousie University, Halifax, Nova Scotia, Canada; Benjamin Terrier, Assistance Publique -Hôpitaux de Paris, Hôpital Cochin, Paris, France; Carter Thorne, Southlake Regional Health Centre, Newmarket, Ontario, Canada; John Varga, Northwestern University, Chicago, Illinois, USA; Pearce Wilcox, St. Paul's Hospital and University of British Columbia, Vancouver, British Columbia, Canada; Michelle Wilson, St Vincent's Hospital and University of Melbourne, Melbourne, Victoria, Australia; Julie Cumin, Jewish General Hospital, Montreal, Quebec, Canada; Claire Fedoruk, Jewish General Hospital, Montreal, Quebec, Canada; Rina S. Fox, Northwestern University Feinberg School of Medicine, Chicago, Illinois, USA; Shadi Gholizadeh, San Diego State University and University of California, San Diego, California,

USA; Lisa R. Jewett, Jewish General Hospital and McGill University, Montreal, Quebec, Canada; Brooke Levis, Jewish General Hospital and McGill University, Montreal, Quebec, Canada; Sarah D. Mills, University of North Carolina, Chapel Hill, North Carolina, USA; Mia R. Pepin, Jewish General Hospital, Montreal, Quebec, Canada; Kimberly Turner, Jewish General Hospital, Montreal, Quebec, Canada.

Keywords: scleroderma, systemic; physical therapy modalities; physical therapy specialty; occupational therapy; health services research; SPIN

Departments/Institutions:

Rheumatology Division, Department of Medicine, Hospital for Special Surgery, NY, NY, USA

Lady Davis Institute for Medical Research, Jewish General Hospital, Montreal, Quebec, Canada

Departments of Psychiatry, Epidemiology, Biostatistics, and Occupational Health, Medicine, and Psychology, McGill University, Montreal, Quebec, Canada

Source of Support: SPIN has been funded by grants from the Canadian Institutes of Health Research (TR3-119192, PJT-148504, PJT-149073) and the Arthritis Society. In addition, SPIN has received institutional contributions from the Lady Davis Institute for Medical Research of the Jewish General Hospital, Montreal, Canada and from McGill University, Montreal, Canada. SPIN has also received support from the Scleroderma Society of Ontario, the Scleroderma Society of Canada, and Sclérodermie Québec. Dr. Kwakkenbos was supported by a CIHR Banting Postdoctoral Fellowship. Dr. Thombs was supported by a Fonds de recherche du Québec Santé investigator salary award.

Conflict of Interests: Authors declare no conflicts of interest

Corresponding Author:

Karima Becetti, MD, MS

Tel: +974 5018 5757

Email: becettikarima@gmail.com

Address: P.O.Box 50216, Doha, Qatar

Short Running Head: PT/OT UTILIZATION IN SCLERODERMA

Abstract

Background: Systemic sclerosis (SSc) is characterized by significant disability due to musculoskeletal involvement. Physical and occupational therapy (PT/OT) have been suggested to improve function. However, the rate of PT/OT utilization has been shown to be low in SSc. We aimed to identify demographic, medical, and psychological variables associated with PT/OT use in SSc.

Methods: Participants were patients with SSc enrolled in the Scleroderma Patient-centered Intervention Network Cohort. We determined the rate and indication of PT/OT use in the 3 months prior to enrollment. Multivariable logistic regression was used to identify variables independently associated with PT/OT utilization.

Results: Of the 1,627 SSc patients included in the analysis, 23% used PT/OT in the preceding 3 months. PT/OT use was independently associated with higher education (odds ratio [OR] 1.08, 95% confidence interval [CI] 1.04-1.12), having moderately severe small joint contractures (OR 2.09, 95% CI 1.45 – 3.03), severe large joint contractures (OR 2.33, 95% CI 1.14 – 4.74), fewer digital ulcerations (OR 0.70, 95% CI 0.51 – 0.95), and higher disability (OR 1.54, 95% CI 1.18 – 2.02) and pain (OR 1.04, 95% CI 1.02 – 1.06) scores. The highest rate of PT/OT utilization was reported in France (43%) and lowest in the USA (17%).

Conclusions: Despite the potential of PT/OT interventions to improve function, less than 1 in 4 SSc patients enrolled in a large international cohort used PT/OT services in the last 3 months. Patients who utilized PT/OT had more severe musculoskeletal manifestations and higher pain and disability.

Introduction

Systemic sclerosis (SSc, or scleroderma) is a chronic systemic disease characterized by dysregulated fibrosis, autoimmunity, inflammation and vasculopathy (1). Musculoskeletal involvement is nearly universal in SSc. Skin fibrosis, joint and muscle pain, arthritis, hand deformities, joint contractures and reduced range of motion are common manifestations and result in significant disability (2). Hand pain and joint stiffness are amongst the five highest rated symptoms and are described by more than 80% of patients with SSc (3). Skin fibrosis of the face and oral tissues causes difficulties with eating, speaking, dental care and oral hygiene (4).

Musculoskeletal rehabilitation and physical and occupational therapy (PT/OT) are recommended for the management of musculoskeletal impairment in SSc (5). Multiple rehabilitation techniques including range of motion exercises, connective tissue massages, joint manipulation, splinting, heat/paraffin wax baths, and generalized PT have been suggested to improve pain and joint motion in small randomized control trials, case reports and case series (5). Similarly, hand and orofacial exercises have been suggested to improve gingival health (6,7). A trial of 220 patients with SSc found that a 4-week PT program significantly reduced disability one-month post-randomization, although there was not an effect on disability at 12-month follow up (8).

Variable rates of PT/OT utilization have been reported in the literature with most studies showing less than 50% of patients with SSc using PT/OT despite it being one of the primary available interventions to address musculoskeletal manifestations (9–11).

In this study, we aimed to determine the rates and indications of PT/OT utilization among patients with SSc enrolled in one of the largest SSc cohorts worldwide, the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. We also aimed to identify demographic, medical and psychological variables associated with the use of these services.

Material and Methods

Patients and Procedures:

The present study included a convenience sample of patients enrolled in the SPIN Cohort. As previously described (12), this large international cohort is composed of patients recruited from more than 40 centers in Canada, the USA, and Europe. To be eligible for enrollment, patients must have been classified as having SSc according to the 2013 American College of Rheumatology/European League Against Rheumatism (ACR/EULAR) classification criteria (13), be18 years of age or older and fluent in English, Spanish, or French, and have the ability to provide informed consent and respond to SPIN questionnaires online. Medical variables are completed by the SPIN physician or coordinator, initiating enrolment in the SPIN Cohort. Patients are then invited via email to register and complete the SPIN Cohort questionnaires online. SPIN patients who completed baseline questionnaires from January 2014 through September 2017 were included in this study. The SPIN Cohort study was approved by the institutional review boards of the Jewish General Hospital, Montreal, Canada (ethics protocol #: CODIM-FLP-12-123), the Hospital for Special Surgery, NY, USA (IRB study #: 2014-326), and all other participating centers. All patients provided informed consent.

Measures:

PT/OT Utilization:

Patients were asked to answer three questions on PT/OT utilization: “In the last 3 months, have you seen a physical therapist, physiotherapist, occupational therapist, ergotherapist, kinesiotherapist or other health care professional for rehabilitation services?”; “If yes, was this for?” (patients could pick one or more options from hands, feet, mouth/face, wound care, activities of daily living, other); and “If yes, how many times in the last 3 months did you receive rehabilitation services?”.

Demographic Variables:

Demographic variables were completed by patients and included race/ethnicity, years of education, current occupation, and housing location. Race/ethnicity were entered differently in different countries and were categorized according to the corresponding country's definitions. A consolidated race/ethnicity variable that included White, Black, and other was created for statistical analysis. Enrolling physicians captured the site of enrollment, gender, and date of birth at baseline.

Medical Variables:

Disease-specific variables included disease subtype (diffuse or limited), duration since first non-Raynaud's disease manifestation, the presence of Raynaud's Phenomenon, Modified Rodnan Skin Score (MRSS), digital ulcerations (DUs), tendon friction rubs, joint contractures, cardiopulmonary disease, and overlap syndromes. Diffuse SSc was defined as skin sclerosis involving the limbs proximal to the elbows and knees and/or the chest and/or trunk at any time, whereas limited SSc was defined as skin sclerosis confined to the limbs distal to the elbows. MRSS is a clinical measure of skin thickness from 0 to 51, with higher scores indicating more severe thickness. The presence of overlap syndromes with rheumatoid arthritis (RA), systemic lupus erythematosus (SLE), and/or idiopathic inflammatory myositis was also determined.

Disability Measures:

Patients completed the Scleroderma Health Assessment Questionnaire (SHAQ) and Cochin Hand Function Scale (CHFS-II). The SHAQ assesses physical disability with a composite score from 0 to 3, with higher scores indicating greater disability (14,15). The CHFS-II assesses hand disability with a score from 0 to 90, with higher scores indicating higher disability (15,16). The SHAQ and CHFS-II have been validated in patients with SSc (14–16).

Psychological Measures:

To assess symptoms of depression, patients completed the Patient Health Questionnaire (PHQ-8). Scores range from 0 to 24, with higher scores indicating more depressive symptoms (17). Patients also completed the Patient Reported Outcomes Measurement Information System (PROMIS-29) measure version 2, which assessed 7 patient-reported outcome domains including the anxiety, fatigue, and pain intensity domains used in this study. Scores are standardized with a mean of 50 and a standard deviation of 10 with the mean score representing the average of a general US population and higher scores reflecting more of the measured domain (18). The Satisfaction with Appearance (SWAP) scale was used to measure body image distress. Scores can range from 0 to 84 with higher scores indicating greater dissatisfaction with appearance (19). The Social Interaction Anxiety Scale-6 (SIAS-6) was used to assess distress due to social interactions by rating patients' experience in social situations from 0 to 24 with higher scores indicating higher social anxiety symptoms (20). Patients also completed the Self-Efficacy for Managing Chronic Disease (SEMCD) scale to assess their confidence in self-managing disease-specific symptoms. Scores ranged from 1 to 10, with higher scores indicating higher self-efficacy. These scores have been validated in SSc (18,19,21–25).

Statistical Analysis:

Descriptive analyses included means, standard deviations (SD), minimum and maximum for continuous variables and frequencies and percentages for discrete variables. Prior to initiation of inferential analyses, data completeness and normality for continuous measures was evaluated. Demographic, psychological, disability and medical variables were compared between patients who participated in PT/OT in the prior 3 months and those who didn't using a chi square test and independent samples t-test. Multivariable binary logistic regression was subsequently used to identify variables independently associated with PT/OT utilization using an *a priori* defined model that included age, gender, education level, employment status, disease subset, disease

duration, joint contractures, DUs, SHAQ, the PROMIS domain on pain intensity, PHQ-8, and SEMCD. Country of enrollment was analyzed after pre-defined variables were identified. To improve the precision of parameter estimates and fit of the data in the model, backward stepwise procedure was used to build best-fitting parsimonious models to best predict predictors of PT/OT usage. Missing variables were not imputed or replaced. *P*-values of 0.05 or below were considered statistically significant. All statistical analyses were performed using SAS version 9.3 (Cary, NC, USA).

Results

Sample Characteristics:

At the time of data extraction, 1,641 patients had completed baseline assessment of which 1,627 patients answered the PT/OT utilization questions and were included in the analysis. 43% were from the USA, 27% from Canada, 17% from France, 10% from the UK, and 2% from Spain. Patients had a mean (± SD) age of 54.9 ± 12.5 years. Female patients constituted 87% of the patients and diffuse SSc was reported in 41% of patients. Mean disease duration was 11.4 ± 8.8 years. The mean MRSS was 7.8 ± 8.4. Interstitial lung disease and pulmonary arterial hypertension were reported in 36% and 10% of patients, respectively.

PT/OT Utilization:

Of the 1,627 patients, 381 (23%) used PT/OT in the 3 months prior to enrollment in SPIN. The mean number of times these services were used among patients who used it in the last 3 months was 9.8 ± 10.7. The rate of PT/OT use varied between the different countries and was highest in France (43%) followed by Spain (28%), Canada (23%), UK (19%), and USA (17%). In the entire cohort, hand PT/OT was the most common indication and was reported by 59% of patients. Feet and activities of daily living were other common indications and were reported by 27% and 30% of patients respectively (**Figure 1**).

***Factors associated with PT/OT utilization*:**

In bivariate analyses, no differences were observed in age, sex, race, education, or housing location among patients who used PT/OT in the 3 months prior to enrollment (**Table 1**). Compared to employed patients, we observed a higher rate of unemployed (28% vs 20%, p = 0.01) and disabled (36% vs 20%, $p < 0.01$) patients in the PT/OT utilization group. While all other sites reported a higher percentage of PT/OT utilization compared to the USA, only Canada (23% vs 17%, $p = 0.02$) and France (43% vs 17%, $p < 0.01$) had statistically significant higher utilization of PT/OT services.

Patients who used PT/OT were more likely to have the diffuse compared to the limited form of the disease (29% vs 19%, $p < 0.01$) and early (≤ 3 years) compared to late (> 3 years) disease (29% vs 23%, $p = 0.04$). Compared to patients who did not use these services, they were also more likely to have shorter disease duration (10.1 ± 8.4 years vs 12.0 ± 8.9 years, $p = 0.01$) and higher MRSS (10.0 ± 9.9 vs 7.1 ± 7.8, $p < 0.01$). The PT/OT group had more tendon friction rubs in the past (27% vs 21%, $p < 0.01$) and currently (31% vs 21%, $p = 0.049$) compared to never, moderately severe small joint contractures (37% vs 19%, $p < 0.01$) and severe small joint contractures (42% vs 19%, $p < 0.01$) compared to no/mild small joint contractures, and moderately severe large joint contractures (40% vs 21%, $p < 0.01$) and severe large joint contractures (37% vs 21%, $p = 0.01$) compared to no/mild large joint contractures. Patients who used PT/OT were slightly more likely to have interstitial lung disease than not (27% vs 22%, p = 0.02). No differences in Raynaud's Phenomenon, DUs, pulmonary arterial hypertension, and overlap syndromes were seen.

With respect to functional impairment (**Table 2)**, patients who used PT/OT had higher hand dysfunction (CHFS-II 20.3 ± 18.9 vs 11.6 ± 14.6, $p < 0.01$) and functional disability (SHAQ 1.0 ± 0.7 vs 0.7 ± 0.7, $p < 0.01$) scores. These patients were also found to have statistically significantly more pain (59.3 ± 8.6 vs 54.5 ± 9.7, $p < 0.01$), fatigue (57.5 ± 10.1 vs

54.4 ± 11.2, $p < 0.01$), and anxiety (54.1 ± 10.2 vs 51.3 ± 10.0, $p < 0.01$) as measured by PROMIS-29, higher depression symptoms (PHQ-8 7.7 ± 5.8 vs 5.9 ± 5.3, $p < 0.01$), and more body image distress (SWAP 35.6 ± 18.6 vs 30.1 ± 18.7, $p < 0.01$). Lastly, patients who used PT/OT had lower self-efficacy compared to those who did not (SEMCD 5.8 ± 2.2 vs 6.7 ± 2.3, $p < 0.01$).

The *a priori* defined and final multivariable regression models are described in **Table 3**. Using a backward stepwise technique, higher education, more small joint contractures, fewer DUs, higher disability, and more pain were significantly associated with PT/OT utilization after adjusting for age and sex. There was an 8% increase in the likelihood of PT/OT utilization for every additional year of education (OR 1.08, 95% CI 1.04 – 1.12). The likelihood of PT/OT utilization was significantly higher in the presence of moderately severe small joint contractures (OR 2.09, 95% CI 1.45 – 3.03) and severe large joint contractures (OR 2.33, 95% CI 1.14 – 4.74). Higher disability (OR 1.54, 95% CI 1.18 – 2.02) and pain (OR 1.04, 95% CI 1.02 – 1.06) scores were associated with more PT/OT use. On the other hand, the presence of DUs was found to decrease the odds of PT/OT utilization (OR 0.70, 95% CI 0.51 – 0.95). The association between PT/OT use and country of enrollment was analyzed after *a priori* variables were defined and included in the final model for analysis. Compared to patients from the USA, patients from Canada were 62% more likely to use PT/OT services (OR 1.62, 95% CI 1.12 – 2.32) while those from France were 4 times more likely to use PT/OT (OR 4.38, 95% CI 2.99 – 6.42).

Discussion

In the present cross-sectional study, we found that 23% of SSc patients in a large international cohort used PT/OT services in the 3 months prior to enrollment and that hand PT/OT was the most common rehabilitation service used by these patients. Our findings also demonstrated that PT/OT utilization was significantly and independently associated with higher

education, more severe musculoskeletal involvement, and higher disability and pain scores. We showed that geographical differences existed in the rate of PT/OT use.

Variable rates of PT/OT utilization in SSc have been described in the literature. Bassel et al reported a rate of 28% PT/OT referral and 12% current utilization among 317 Canadian SSc patients with hand involvement surveyed between September 2008 and August 2009 (10). Consistent with our findings, the study by Bassel et al also showed that the presence of more hand problems was associated with PT/OT referrals (10). Through another survey study of 813 Canadian patients with SSc, Johnson et al showed that 36% of patients have seen a PT and 22% an OT since SSc diagnosis (9). A higher rate of PT/OT utilization was shown in Western Europe. Among 198 Dutch patients with SSc surveyed from June and August of 2011, 75% reported contact with a PT and 36% with an OT since SSc onset. Of these patients, 53% and 13% reported contact with a PT and an OT, respectively, in the 12 months prior to study survey (11). Between-study differences in sampling and methodology can explain some of the variability in the rates of PT/OT utilization. For example, in our study, the evaluation was restricted to the 3 months prior to enrollment while other studies looked at use over longer periods of time. However, this variability is in accordance with our study findings of different rates of PT/OT use in the different countries evaluated, which reflects regional variations in SSc management, access to rehabilitation services, and health care costs.

Rehabilitation services use has been more extensively studied in RA, a more common rheumatic disease with a predominance of musculoskeletal manifestations. In RA, the rates of PT and OT use among 8,001 German RA patients were estimated at 44% and 15%, respectively, from 1993 to 1998 (26). Similar utilization rates were observed among 1,200 patients with RA from Amsterdam in 1997 (40% PT and 17% OT use in the preceding year) (27). In the US, a 2011 study of 772 patients with RA showed that 15% of patients used PT in the preceding 6 months (28). The variability can be explained by differences in the management and prognosis of

RA between the pre- and post-biologics eras but might also reflect country differences in PT/OT utilization as seen in our study. Similar to our findings, disease activity, disability, and higher education were also predictive of PT use in RA (28). In both RA and SSc, patients with these characteristics are in more need for (and thus more likely to be referred to) PT/OT and are possibly more likely to ask for/participate in these services due to their higher education and/or better access.

In the SPIN Cohort, hand PT/OT was the most commonly used rehabilitation service, reflecting the importance of hand symptoms in SSc. Hand pain and stiffness were previously shown to be frequently reported by SSc patients, with more than 50% associating them with moderate to severe impact on quality of life (3). In our study, patients who received PT/OT had more severe small joint contractures, tendon friction rubs, and higher hand dysfunction. Of interest, the presence of DUs was independently associated with less PT/OT use. Wound care is recommended for the management of DUs (29) and is offered by different specialists including specially trained physical therapists. However, wound care was an indication for PT/OT for about 6% of the SPIN cohort only, limiting conclusions on its association with DUs. Based on limited studies, exercise had no direct effect on healing ulcers (30). In contrary, since avoiding trauma is one of the recommended non-pharmacological interventions in the management of DUs (29), it is conceivable that concerns regarding exacerbation of ulcers due to the trauma of PT/OT activities result in hesitancy to refer to or participate in therapy when DUs are active.

No association was seen between PT/OT use and age, sex, race/ethnicity and overlap syndromes with other rheumatic diseases with significant musculoskeletal involvement. Although patients who used PT/OT were slightly more likely to have interstitial lung disease than not, no independent association was found between PT/OT use and SSc-associated cardiopulmonary disease.

Patients who used PT/OT were found to have higher psychological distress and lower

self-efficacy than those who did not. Since all measures were obtained at the same time point, conclusions on a cause-effect relationship between distress, self-efficacy, and PT/OT use are not possible. The higher distress and lower self-efficacy may reflect a more severe disease and higher disease impact in those who were more likely to be referred to, and thus more likely to use, these services. In SSc, lower self-efficacy has been found to correlate with greater physical limitation, increased pain and fatigue, and more depression (31). Moreover, better self-efficacy has been associated with better health outcomes, improved adherence to home exercise programs, and reduced health services utilization and cost in patients with chronic diseases including arthritis (32–34). Most of these studies focused on hospitalizations and emergency room and physician visits.

The large sample size, number of variables, and *a priori* selected variables are important strengths. We included demographic variables, disease-specific variables including a detailed assessment of musculoskeletal manifestations, disability measures, and a number of psychological variables that were thought and/or previously shown to affect health services utilization. SSc diagnosis and medical variables were ascertained by physicians and all variables were provided at one time-point allowing for a real-time assessment of their association with PT/OT use.

There are important limitations to consider when interpreting the findings of this study. The SPIN Cohort is a convenience sample of patients recruited from specialized SSc centers, which might limit the generalizability of our findings. These patients have access to tertiary healthcare systems and are likely different from those seen in other settings. The rate of PT/OT use might, therefore, be lower in patients receiving care in generalized rheumatology practices. As previously stated, we were only able to look at PT/OT utilization in the 3 months prior to enrollment and the evaluation of PT/OT referrals was limited. A previous study showed differences between the rates of PT/OT referrals and utilization indicating possible differences in

the perceived need for PT/OT between physicians and patients among other factors worth exploring. In SPIN, questions on PT/OT utilization were restricted to supervised rehabilitation services only and might not have captured home-based physical therapy and exercising which have been reported by more than half of SSc patients in the SPIN cohort in a study by Azar et al (35). In addition, it is important to recognize that the lack of data on income, medical insurance, and access to rehabilitation services are important limitations of this study given the critical impact these variables have been shown to have on PT/OT and other health services use. In adult patients with low back pain in the USA, out-of-pocket expenditure and type of insurance were shown to be predictive of PT visits, with privately and Medicare insured people being more likely to have more visits compared to those publically insured and uninsured (36). Similarly, SSc patients with private insurance were almost twice as likely to be referred to PT/OT in the study by Bassel et al (10). Because of the complexity and variability of insurance coverage within each healthcare system, looking at the rate of PT/OT use across the different countries does not fully overcome this limitation.

In conclusion, we showed that fewer than 25% of patients with SSc in a large international cohort used PT/OT services in the 3 months prior to enrollment and that geographical differences exist in the rate of utilization. Future research evaluating the impact of rehabilitation services on functional outcomes in SSc is needed to fully understand the importance of these interventions in SSc and strengthen the evidence based on which management guidelines are developed. Furthermore, in addition to identifying barriers to PT/OT in different healthcare settings, interventions are needed to improve the utilization of these services among patients with SSc. Such interventions include developing effective SSc-specific online and home-based physical therapy and exercise programs, which is one of the primary aims of SPIN. These programs could help overcome some of the barriers to PT/OT use, including cost and access.

References

1. Abraham D, Varga J. Systemic sclerosis: a prototypic multisystem fibrotic disorder. J Clin Invest 2007;117:557-567.

2. Morrisroe KB, Nikpour M, Proudman SM. Musculoskeletal Manifestations of Systemic Sclerosis. Rheum Dis Clin North Am 2015;41:507-518.

3. Bassel M, Hudson M, Taillefer SS, Schieir O, Baron M, Thombs BD. Frequency and impact of symptoms experienced by patients with systemic sclerosis: results from a Canadian National Survey. Rheumatology (Oxford) 2011;50:762-767.

4. 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.

5. Poole JL. Musculoskeletal rehabilitation in the person with scleroderma. Curr Opin Rheumatol 2010;22:205-212.

6. Poole J, Conte C, Brewer C, Good CC, Perella D, Rossie KM, et al. Oral hygiene in scleroderma: The effectiveness of a multi-disciplinary intervention program. Disabil Rehabil 2010;32:379-384.

7. Yuen HK, Weng Y, Bandyopadhyay D, Reed SG, Leite RS, Silver RM. Effect of a multi-faceted intervention on gingival health among adults with systemic sclerosis. Clin Exp Rheumatol 2011;29:S26-32.

8. 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;69:1050-1059.

9. Johnson SR, Carette S, Dunne JV. Scleroderma: health services utilization from patients' perspective. J Rheumatol 2006;33:1123-1127.

10. Bassel M, Hudson M, Baron M, Taillefer SS, Mouthon L, Poiraudeau S, et al. Physical and occupational therapy referral and use among systemic sclerosis patients with impaired hand function: results from a Canadian national survey. Clin Exp Rheumatol 2012;30:574-577.

11. Willems LM, Kwakkenbos L, Bode C, van den Hoogen FH, van den Ende CH. Health care use and patients ' perceptions on quality of care in systemic sclerosis. Clin Exp Rheumatol 2013;31:S64-70.

12. Kwakkenbos L, Jewett LR, Baron M, Bartlett SJ, Furst D, Gottesman K, et al. The Scleroderma Patient-centered Intervention Network (SPIN) Cohort: protocol for a cohort multiple randomised controlled trial (cmRCT) design to support trials of psychosocial and rehabilitation interventions in a rare disease context. BMJ Open 2013;3:1-11.

13. van den Hoogen F, Khanna D, Fransen J, Johnson SR, Baron M, Tyndall A, et al. 2013 classification criteria for systemic sclerosis: An american college of rheumatology/European league against rheumatism collaborative initiative. Arthritis Rheum 2013;65:2737-2747.

14. Pope J. Measures of systemic sclerosis (scleroderma): Health Assessment Questionnaire (HAQ) and Scleroderma HAQ (SHAQ), Physician- and Patient-Rated Global Assessments, Symptom Burden Index (SBI), University of California, Los Angeles, Scleroderma Clinical Trials Consortium Gastrointestinal Scale (UCLA SCTC GIT) 2.0, Baseline Dyspnea Index (BDI) and Transition Dyspnea Index (TDI) (Mahler's Index), Cambridge Pulmonary Hypertension Outcome Review (CAMPHOR), and Raynaud's Condition Score (RCS). Arthritis Care Res 2011;63:S98-111.

15. Rannou F, Poiraudeau S, Berezné A, Baubet T, Le-Guern V, Cabane J, et al. Assessing disability and quality of life in systemic sclerosis: Construct validities of the Cochin Hand Function Scale, Health Assessment Questionnaire (HAQ), systemic sclerosis HAQ, and medical outcomes study 36-item short form health survey. Arthritis Rheum 2007;57:94-102.

16. Poole JL. Measures of hand function: Arthritis Hand Function Test (AHFT), Australian Canadian Osteoarthritis Hand Index (AUSCAN), Cochin Hand Function Scale, Functional Index for Hand Osteoarthritis (FIHOA), Grip Ability Test (GAT), Jebsen Hand Function Test (JHFT), and Michigan Hand Outcomes Questionnaire (MHQ). Arthritis Care Res (Hoboken) 2011;63:S189-199.

17. Kroenke K, Spitzer RL, Williams JB, Lowe B. The Patient Health Questionnaire Somatic, Anxiety, and Depressive Symptom Scales: a systematic review. Gen Hosp Psychiatry 2010;32:345-359.

18. Khanna D, Maranian P, Rothrock N, Cella D, Gershon R, Khanna PP, et al. Feasibility and construct validity of PROMIS and “legacy” instruments in an academic scleroderma clinic. Value Health 2012;15:128-134.

19. Heinberg LJ, Kudel I, White B, Kwan A, Medley K,Wigley F, et al. Assessing body image in patients with systemic sclerosis (scleroderma): validation of the Adapted Satisfaction with Appearance Scale. Body Image 2007;4:79-86.

20. Fergus TA, Valentiner DP, McGrath PB, Gier-Lonsway SL, Kim HS. Short forms of the social interaction anxiety scale and the social phobia scale. J Pers Assess 2012;94:310-

320.

21. Kwakkenbos L, Thombs BD, Khanna D, Carrier ME, Baron M, Furst D, et al. Validation of the Patient-Reported Outcomes Measurement Information System-29 (PROMIS-29) in Scleroderma and Associations with Clinical Characteristics: A Scleroderma Patient-centered Intervention Network (SPIN) Cohort Study. Rheumatology [in press].

22. Morrisroe K, Stevens W, Hug M, Sahhar J, Ngian GS, Zochling J, et al. Validity of the PROMIS-29 in a large Australian cohort of patients with systemic sclerosis. J Scleroderma Relat Disord 2017;2:188-195.

23. Milette K, Hudson M, Baron M, Thombs BD, Canadian Scleroderma Research Group. Comparison of the PHQ-9 and CES-D depression scales in systemic sclerosis : internal consistency reliability , convergent validity and clinical correlates. Rheumatology (Oxford) 2010;49:789-796.

24. Gholizadeh S, Mills SD, Fox RS, Jewett L, Kwakkenbos L, Carrier ME, et al. A Psychometric Analysis of the Social Interaction Anxiety Scale (SIAS-6) in Systemic Sclerosis : Results from the Scleroderma Patient-Centered Intervention Network (SPIN) Cohort [abstract]. Arthritis Rheumatol 2015;67:1303-1304.

25. Riehm KE, Kwakkenbos L, Carrier ME, Bartlett SJ, Malcarne VL, Mouthon L, et al. Validation of the Self-Efficacy for Managing Chronic Disease Scale : A Scleroderma Patient-Centered Intervention Network Cohort Study. Arthritis Care Res (Hoboken) 2016;68:1195-1200.

26. Zink A, Listing J, Niewerth M, Zeidler H. The national database of the German Collaborative Arthritis Centres: II. Treatment of patients with rheumatoid arthritis. Ann Rheum Dis 2001;60:207-213.

27. Jacobi CE, Triemstra M, Rupp I, Dinant HJ, Van Den Bos GA. Health care utilization among rheumatoid arthritis patients referred to a rheumatology center: Unequal needs, unequal care? Arthritis Rheum 2001;45:324-330.

28. Iversen MD, Chhabriya RK, Shadick N. Predictors of the use of physical therapy services among patients with rheumatoid arthritis. Phys Ther 2011;91:65-76.

29. Hughes M, Herrick AL. Digital ulcers in systemic sclerosis. Rheumatology (Oxford) 2017;56:14-25.

30. Moran ME. Scleroderma and evidence based non-pharmaceutical treatment modalities for digital ulcers: a systematic review. J Wound Care 2014;23:510-541.

31. Buck U, Poole JL, Mendelson C. Factors Related to Self-Efficacy in Persons With Scleroderma. Musculoskeletal Care 2010;8:197-203.

32. Brady TJ, Murphy L, O'Colmain BJ, Beauchesne D, Daniels B, Greenberg M, et al. A meta-analysis of health status, health behaviors, and healthcare utilization outcomes of the Chronic Disease Self-Management Program. Prev Chronic Dis 2013;10:120112.

33. Marks R. Self-efficacy and arthritis disability: An updated synthesis of the evidence base and its relevance to optimal patient care. Health Psychol Open 2014;1:1-18.

34. Medina-Mirapeix F, Escolar-Reina P, Gascón-Cánovas JJ, Montilla-Herrador J, Jimeno-Serrano FJ, Collins SM. Predictive factors of adherence to frequency and duration components in home exercise programs for neck and low back pain: an observational study. BMC Musculoskelet Disord 2009;10:155.

35. Azar M, Rice DB, Kwakkenbos L, Carrier ME, Shrier I, Bartlett SJ, et al. Exercise habits and factors associated with exercise in systemic sclerosis : a Scleroderma Patient-centered Intervention Network (SPIN) cohort study investigators. Disabil Rehabil 2018;40:1997-2003.

36. Dolot J, Viola D, Shi Q, Hyland M. Impact of Out-of-Pocket Expenditure on Physical Therapy Utilization for Nonspecific Low Back Pain: Secondary Analysis of the Medical Expenditure Panel Survey Data. Phys Ther 2016;96:212-221.

Table 1. Differences in demographic and medical variables between patients who used PT/OT and those who did not*

Variable	PT/OT (n = 381)	No PT/OT (n = 1246)	*p*-value
Age (years)	54.5 (12.7)	55.1 (12.4)	0.48
Sex:			
Female	337 (24%)	1086 (76%)	Reference
Male	44 (22%)	160 (78%)	0.50
Country of enrollment:			
USA	120 (17%)	585 (83%)	Reference
Canada	100 (23%)	339 (77%)	0.02
UK	32 (19%)	137 (81%)	0.56
France	119 (43%)	159 (57%)	<0.01
Spain	10 (28%)	26 (72%)	0.10
Race/Ethnicity:**			
White	308 (23%)	1034 (77%)	Reference
Black	28 (24%)	88 (76%)	0.77
Other	45 (27%)	123 (73%)	0.27
Education (years)	15.1 (3.8)	15.0 (3.4)	0.78
Current occupation:			
Employed	141 (20%)	562 (80%)	Reference
Unemployed	61 (28%)	157 (72%)	0.01
Retired	94 (24%)	298 (76%)	0.13
On disability	59 (36%)	103 (64%)	< 0.01
Other	26 (18%)	121 (82%)	0.51
Housing location:			
Non-urban	225 (22%)	784 (78%)	Reference
Urban	155 (26%)	444 (74%)	0.10
Disease subtype:			
Limited	179 (19%)	750 (81%)	Reference
Diffuse disease	188 (29%)	453 (71%)	< 0.01

Disease duration* (years)**	10.1 (8.4)	12.0 (8.9)	0.01
Late disease (> 3 years)	296 (23%)	990 (77%)	Reference
Early disease (≤ 3 years)	66 (29%)	158 (71%)	0.04
MRSS	10.0 (9.9)	7.1 (7.8)	< 0.01
Raynauds Phenomenon:			
Negative	6 (25%)	18 (75%)	Reference
Positive	373 (23%)	1218 (77%)	0.86
Any digital ulcerations:			
Negative	250 (24%)	790 (76%)	Reference
Positive	127 (23%)	437 (77%)	0.49
Tendon friction rubs:			
Never	230 (21%)	874 (79%)	Reference
Currently	53 (27%)	118 (73%)	< 0.01
In the past	50 (31%)	133 (69%)	0.049
Small joints contractures:			
No-mild (0-25%)	214 (19%)	938 (81%)	Reference
Moderate (25-50%)	103 (37%)	176 (63%)	< 0.01
Severe (>50%)	48 (42%)	66 (58%)	< 0.01
Large joints contractures:			
No-mild (0-25%)	281 (21%)	1043 (79%)	Reference
Moderate (25-50%)	54 (40%)	80 (60%)	< 0.01
Severe (>50%)	21 (37%)	36 (63%)	0.01
Interstitial lung disease:			
No	221 (22%)	801 (78%)	Reference
Yes	154 (27%)	419 (73%)	0.02
Pulmonary hypertension:			
No	329 (24%)	1026 (76%)	Reference
Yes	39 (25%)	119 (75%)	0.91
Overlap Syndromes			
Rheumatoid arthritis:			
No	348 (23%)	1149 (77%)	Reference

Yes	30 (29%)	72 (71%)	0.16
Lupus:			
No	368 (24%)	1185 (76%)	Reference
Yes	12 (24%)	39 (76%)	0.98
Inflammatory myositis:			
No	350 (23%)	1157 (77%)	Reference
Yes	25 (28%)	64 (72%)	0.29

*Values presented as number (row %) for categorical variables and mean (SD) for continuous variables. Response rate varied from 80% to 100% for the different variables.
**Consolidated race and ethnicity variable.
***Disease duration since first non-Raynaud's manifestation. MRSS = Modified Rodnan Skin Score

Table 2. Differences in disability and psychological variables between patients who used PT/OT and those who did not*

Variable	PT/OT (n = 381)	No PT/OT (n = 1246)	*p*-value
Hand dysfunction (CHFS-II)	20.3 (18.9)	11.6 (14.6)	< 0.01
Disability index (SHAQ)	1.0 (0.7)	0.7 (0.7)	< 0.01
Depression (PHQ-8)	7.7 (5.8)	5.9 (5.3)	< 0.01
PROMIS-29			
Anxiety	54.1 (10.2)	51.3 (10.0)	< 0.01
Fatigue	57.5 (10.1)	54.4 (11.2)	< 0.01
Pain	59.3 (8.6)	54.5 (9.7)	< 0.01
Body Image Distress (SWAP)	35.6 (18.6)	30.1 (18.7)	< 0.01
Social Anxiety (SIAS-6)	3.0 (4.6)	2.5 (3.8)	0.09
Self-Efficacy (SEMCD)	5.8 (2.2)	6.7 (2.3)	< 0.01

* Values presented as number (%) for categorical variables and mean (SD) for continuous variables. Response rate varied from 80% to 100% for the different variables.
CHFS-II = Cochin Hand Function Scale; SHAQ = Scleroderma Health Assessment Questionnaire; PHQ-8 = Patient Health Questionnaire; PROMIS-29 = Patient Reported Outcomes Measurement Information System; SWAP = Satisfaction with Appearance; SIAS-6 = Social Interaction Anxiety Scale; SEMCD = Self-Efficacy for Managing Chronic Disease

Table 3. Factors independently associated with PT/OT use in the SPIN cohort

	Model 1[a]		Model 2[b]	
Variable	**OR (95% CI)**	***p*-value**	**OR (95% CI)**	***p*-value**
Age	1.00 (0.99 – 1.02)	0.54	-	-
Sex (male)	0.95 (0.62 – 1.46)	0.81	-	-
Country of enrollment:				
USA			Reference	
Canada			1.62 (1.12 – 2.32)	0.01
UK			0.87 (0.52 – 1.45)	0.60
France			4.38 (2.99 – 6.42)	< 0.01
Spain	-	-	2.35 (0.99 – 5.58)	0.052
Education	1.04 (1.00 – 1.09)	0.04	1.08 (1.04 – 1.12)	< 0.01
Employment status:				
Full time/Part time employed	Reference			
Unemployed	1.11 (0.72 – 1.72)	0.63		
Retired	1.03 (0.68 – 1.57)	0.88		
On disability	1.09 (0.68 – 1.76)	0.71		
Other	0.68 (0.39 – 1.18)	0.17	-	-
Early SSc (disease duration ≤ 3 years)	1.24 (0.85 – 1.82)	0.26	1.43 (0.98 – 2.09)	0.06
Diffuse disease (vs. Limited)	1.12 (0.82 – 1.52)	0.48	-	-
Small joints contractures:				
No-mild (0-25%)	Reference		Reference	
Moderate (25-50%)	2.11 (1.46 – 3.05)	< 0.01	2.09 (1.45 – 3.01)	< 0.01
Severe (>50%)	2.16 (1.28 – 3.64)	< 0.01	1.61 (0.95 – 2.72)	0.08
Large joint contractures:				
No-mild (0-25%)	Reference		Reference	
Moderate (25-50%)	1.28 (0.80 – 2.05)	0.31	1.22 (0.76 – 1.96)	0.42
Severe (>50%)	1.77 (0.87 – 3.58)	0.11	2.33 (1.14 – 4.74)	0.02
Any digital ulcerations	0.68 (0.51 – 0.92)	0.01	0.70 (0.51 – 0.95)	0.02
Disability index (SHAQ)	1.23 (0.93 – 1.63)	0.15	1.54 (1.18 – 2.02)	< 0.01
Depression (PHQ-8)	1.02 (0.98 – 1.05)	0.34	-	-

Pain (PROMIS-29)	1.04 (1.02 – 1.06)	< 0.01	1.04 (1.02 – 1.06)	< 0.01
Self-Efficacy (SEMCD)	1.00 (0.92 – 1.08)	0.93	-	-

[a] *a priori* defined model
[b] Reduced model following a stepwise regression procedure, adjusted for age and sex
OR = Odds Ration; CI = Confidence Interval; SHAQ = Scleroderma Health Assessment Questionnaire; PHQ-8 = Patient Health Questionnaire; PROMIS-29 = Patient Reported Outcomes Measurement Information System; SEMCD = Self-Efficacy for Managing Chronic Disease

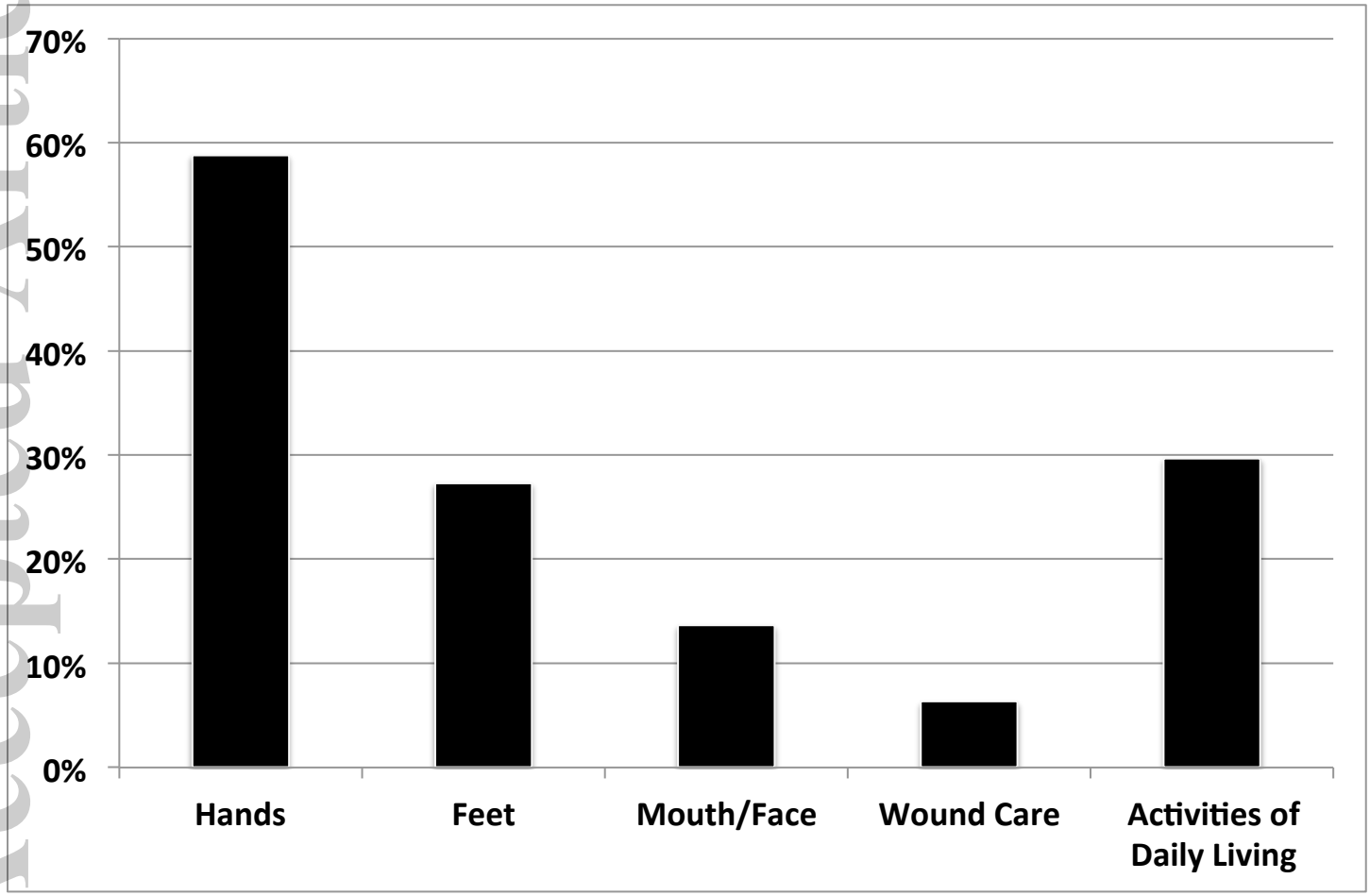

Figure 1. PT/OT indications in the 3 months prior to enrollment in the SPIN cohort