Pain and Self-Efficacy Among Patients With Systemic Sclerosis

A Scleroderma Patient-Centered Intervention Network Cohort Study

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Background: Pain is one of the most common symptoms affecting patients with systemic sclerosis; however, little is known about the relationship between self-efficacy and pain and changes in pain over time.

Objectives: The purpose of this study was to describe the relationships between self-efficacy and pain in patients with systemic sclerosis, as well as determine whether changes in self-efficacy mediate changes in pain.

Methods: A prospective longitudinal study was conducted using data from the Scleroderma Patient-Centered Intervention Network Cohort. The baseline sample included 1,903 adults, with a trajectory subsample of 427 who completed 3-month assessments across 3 years. Hierarchical (sequential) forward multivariable regression, covarying for participant characteristics, was conducted to determine the association between self-efficacy and patient characteristics on pain outcomes. Trajectory models, covarying for participant characteristics, were used to examine changes in self-efficacy and pain outcomes across time and whether self-efficacy mediated the pain trajectories.

Results: Mean time since diagnosis was 9.5 years, with 39.2% diagnosed with diffuse cutaneous systemic sclerosis. Greater self-efficacy was associated with less pain interference and intensity. Increasing age, female gender, finger ulcers, and small joint contractures were related to greater pain interference and intensity. Esophageal gastrointestinal symptoms were associated with more pain interference. Self-efficacy and pain trajectories remained stable across time, and self-efficacy did not mediate the pain trajectories.

Discussion: This study identified self-efficacy, age, gender, finger ulcers, small joint contractures, and esophageal gastrointestinal symptoms as important correlates associated with pain in patients with systemic sclerosis. In addition, this study found that self-efficacy and pain outcomes remained stable over time, providing important insights into the longitudinal pain experiences of patients with systemic sclerosis.

Key Words: pain • scleroderma • self-efficacy • systemic sclerosis

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Systemic sclerosis (SSc; scleroderma) is a rare, progressive, multisystem autoimmune disease characterized by immune dysfunction, vasculopathy, and fibrosis of the skin and internal organs (Van Den Hoogen et al., 2013). Although prevalence rates vary worldwide (7–489 cases per million; Chifflot et al., 2008), SSc is more common in women with a peak disease onset often between 20 and 50 years old (Alba et al., 2014). SSc is associated with significant morbidity and mortality, as well as high healthcare costs and reduced productivity resulting in a total cost of $1.9 billion per year in North America (Fischer et al., 2017). Patients with SSc exhibit a variety of clinical manifestations but are often divided into two main subsets, limited or diffuse cutaneous SSc (lcSSc or dcSSc, respectively), based on the extent of skin involvement (Denton

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& Khanna, 2017). There is no cure for SSc, and major goals of treatment are aimed at ameliorating symptoms, reducing functional disability, and improving health-related quality of life (Almeida et al., 2015).

Pain is one of the most common and debilitating symptoms experienced by patients with SSc, leading to functional disability and reduced health-related quality of life (Haythornthwaite et al., 2003). Pain is experienced by up to 83% of patients with SSc with more than a third of patients rating their pain as moderate to severe (Schieir et al., 2010). The most common source of pain is joint pain (Ostojic et al., 2019); other sources of pain include finger ulcers, joint contractures, gastrointestinal symptoms, synovitis, joint tenderness, and Raynaud's phenomenon (Malcarne et al., 2007; Schieir et al., 2010). Patients with SSc have described their pain as excruciating, debilitating, and draining (Suarez-Almazor et al., 2007; Sumpton et al., 2017). In addition, nearly half of patients with SSc experience pain on a daily basis (Ostojic et al., 2019), underscoring the importance of effective pain management in this population. Pain management in SSc includes a variety of treatments, such as pharmacological therapies, exercise and rehabilitation programs, and psychosocial interventions; however, additional emphasis on psychosocial correlates, such as self-efficacy, on pain in patients with SSc is needed to better understand and treat pain in this population (Merz et al., 2018).

Although SSc is a chronic and progressive disease in which patients are challenged with managing their symptoms on a daily basis, patients with SSc report less confidence in their ability to perform self-care tasks related to managing their pain (Thombs et al., 2017). More severe symptoms, such as higher levels of pain, have also been associated with lower levels of self-efficacy or one's perceived confidence in performing a specific behavior or task (Bandura, 1997) in patients with SSc (Buck et al., 2010; Thombs et al., 2017). Self-efficacy is an essential component of chronic disease self-management (Moore et al., 2016), and gaining a better understanding of the relationship between pain and self-efficacy is essential for future development of self-management interventions in this population.

In addition, little is known about changes in self-efficacy and pain over time. To our knowledge, only two studies have explored changes in pain over time (Merz et al., 2017; Sekhon et al., 2010). Sekhon et al. (2010) found little change in pain among 109 patients with SSc who were evaluated at two consecutive visits (ranging from 4.2 to 10.9 months between visits), and Merz et al. (2017) found a slight improvement in pain among patients with early disease who were followed for three annual visits. However, these studies were limited by small sample sizes and limited data collection points (i.e., two consecutive visits or three annual visits). In addition, these studies did not explore self-efficacy and its potential mediating effect on pain, which has been supported in other rheumatic diseases (Somers et al., 2010). As such, the aims of this study were to (a) describe the relationship between self-efficacy for managing pain and pain outcomes (i.e., pain interference and pain intensity), (b) examine self-efficacy for managing pain and pain trajectories across 3 years, and (c) determine whether changes in self-efficacy for managing pain mediate changes in pain outcomes across 3 years in patients with SSc.

METHODS

Design

This study utilized prospective longitudinal cohort data collected at enrollment and each subsequent 3-month follow-up assessment across 3 years (baseline to 36 months) from patients with SSc enrolled in the Scleroderma Patient-Centered Intervention Network (SPIN) Cohort. Self-efficacy for managing pain, pain interference, and pain intensity scores were evaluated at each assessment time point. The baseline assessment included patient data collected upon enrollment in the SPIN Cohort. This study was approved by the institutional review board of Duke University Health System; the SPIN Cohort study was approved by the research ethics committee of the Jewish General Hospital, Montreal, Canada, and by the institutional review boards of each participating SPIN Center.

Cross-sectional analyses were conducted to determine whether self-efficacy was associated with pain outcomes at baseline. Trajectory analyses were performed to examine change in self-efficacy, pain interference, and pain intensity across 36 months. Finally, we explored whether change in self-efficacy was a mechanism that influenced pain trajectories across 36 months. Patient characteristics associated with pain scores based on published findings were included as covariates in the analyses. A 36-month period was selected based on two considerations: (a) examining a time period that was long enough to capture change or flares in pain outcomes and (b) 78% of those enrolled had not completed a 36-month assessment at the time of the analysis.

Data Source

The SPIN Cohort was established in 2013 to collect patient-reported data to better understand the problems faced by patients with SSc and to develop and test interventions to improve quality of life for patients with SSc (Kwakkenbos et al., 2019). The SPIN Cohort includes adult patients with SSc from 45 SPIN Centers in the United States, Canada, the United Kingdom, France, Spain, Mexico, and Australia. The SPIN sample is a convenience sample in which eligible participants are invited to participate by a local SPIN physician or supervised nurse coordinator. Written informed consent is obtained, and the SPIN physician or supervised nurse coordinator complete and submit an online medical data form, which initiates patient registration in the SPIN Cohort. After completion of online registration, participants receive an

automated welcoming e-mail with instructions on how to activate their online SPIN account and how to complete the SPIN Cohort online patient-reported measures. Participants complete the online patient-reported measures at enrollment and every 3 months.

Eligibility criteria for inclusion in the cohort were (a) a diagnosis of SSc according to the 2013 American College of Rheumatology/European League Against Rheumatism classification criteria (Van Den Hoogen et al., 2013) confirmed by a SPIN-affiliated physician, (b) 18 years of age or older, (c) ability to give informed consent, and (d) fluent in English, French, or Spanish. Exclusion criteria included not having access to the Internet or otherwise not being able to respond to the patient-reported measures via the Internet.

Selected Sample

This study included a baseline sample of 1,903 patients with SSc who completed an enrollment assessment from January 2013 through December 2018, and a trajectory subsample composed of 427 who completed the enrollment and Month 36 assessments. From the baseline sample, participants with self-efficacy and pain (interference and intensity) scores at enrollment were included in the trajectory subsample. Of those, participants without self-efficacy and pain scores at Month 36 were omitted from the trajectory subsample. Trajectory subsample participants, however, were not required to have self-efficacy and pain scores at each interim assessment. A sensitivity analysis was conducted to compare baseline participants included and excluded from the trajectory subsample (see data analysis plan and results). Supplemental Digital Content Figure S1, http://links.lww.com/NRES/A399, provides a flow diagram of the final analysis samples for this study.

Data Collection

Participants completed patient-reported measures at enrollment (baseline) and every 3 months thereafter via the Internet. Each online assessment included items to measure self-reported outcomes.

Baseline Patient Characteristics Participants provided demographic data upon enrollment. The SPIN-affiliated physician reported age and gender as well as clinical data, such as the SSc subset (lcSSc or dcSSc), time since SSc diagnosis, and the presence or absence of disease manifestations.

Self-Efficacy for Managing Pain One item from the Self-Efficacy for Managing Chronic Disease (SEMCD) Scale (Ritter & Lorig, 2014) was used to measure self-efficacy for managing pain. The SEMCD Scale consisted of six items measuring one's confidence in performing certain activities regularly at the present time. The single item related to self-efficacy for managing physical discomfort or pain was used to measure self-efficacy for managing pain. More specifically, this item examined: "how confident do you feel that you can keep the physical discomfort or pain of your disease from interfering with things you want to do" (Ritter & Lorig, 2014). This item was rated on a scale from 1 (*not confident at all*) to 10 (*totally confident*), with higher scores indicating greater self-efficacy. The SEMCD Scale is a valid and reliable measure in patients with SSc with established convergent validity and high internal consistency (Cronbach's $\alpha = .93$; Riehm et al., 2016); however, the reliability and validity of the single item related to pain has not been tested. For the participants in the trajectory subsample, self-efficacy was assessed at baseline and every 3 months, except for Months 3, 9, and 15. The trajectory subsample was composed of participants who had been enrolled in the cohort for at least 36 months, which was before the inclusion of self-efficacy in the assessment packet at Months 3, 9, and 15.

Pain Interference and Intensity The pain interference domain and pain intensity item of the Patient-Reported Outcomes Measurement Information System-29 Version 2.0 (PROMIS-29v2; Hays et al., 2018) were used to evaluate pain in the past 7 days. The pain interference domain score was composed of four items rated on a scale of 1 (*not at all*) to 5 (*very much*), with higher scores indicating greater pain interference. Raw domain scores, calculated by adding the ratings for the four pain interference items, had a possible range of 4–20. These scores were converted to *T* scores standardized for the general U.S. population (mean = 50, *SD* = 10). The single pain intensity item was measured on an 11-point scale, ranging from 0 (*no pain*) to 10 (*worst imaginable pain*). Psychometric studies demonstrated that the latter provides valid and reliable measures of these pain constructs in patients with SSc, with demonstrated convergent validity and Cronbach's α values ranging from .86 to .96 for domain scores (Kwakkenbos et al., 2017). Pain interference and pain intensity were assessed at enrollment and every 3 months.

Data Analysis

Nondirectional tests were performed with significance set at .05 for all tests. The significance level was not adjusted for multiple testing because of the exploratory nature of this study. Effect sizes were calculated to address clinical significance. Analyses were conducted using SAS Version 9.4 software.

Baseline Patient Characteristics Descriptive statistics were used to summarize participant characteristics of the baseline sample and trajectory subsample. A sensitivity analysis using general linear models for scalar measures and chi-square tests for categorical data were performed to compare the baseline characteristics of those included in the trajectory subsample to those excluded from the subsample.

Baseline Covariates Eight patient characteristics were selected a priori as covariates in the baseline and trajectory analysis. The three demographic covariates were age (at completion of baseline assessment), female gender, and married/living with

a partner. The five clinical covariates were the presence of (a) finger ulcers (i.e., digital pulp ulcers distal to the distal interphalangeal joints or anywhere else on the finger), (b) moderate or severe small joint contractures (i.e., to the distal interphalangeal joints, proximal interphalangeal joints, metacarpophalangeal joints, and/or wrists), (c) esophageal gastrointestinal symptoms (i.e., dysphagia, heartburn, and/or reflux, because of SSc at any time, now or in the past), (d) time since diagnosis (i.e., time since diagnosis to completion of baseline assessment), and (e) diffuse SSc subset (i.e., defined as skin sclerosis involving the limbs proximal to the elbows and knees and/or the chest and/or trunk, at any time). These covariates were examined in previous studies exploring pain and have been associated with pain outcomes (Malcarne et al., 2007; Merz et al., 2017; Ostojic et al., 2019; Schieir et al., 2010). Although considered, the modified Rodnan skin score was not included because of the 18% missing rate and its high overlap with disease subset.

Baseline Sample: Self-Efficacy and Pain Relationships Bivariate regression was used to examine the relationship of (a) self-efficacy for managing pain with pain interference and intensity scores and (b) eight participant characteristics with these pain scores at enrollment in the baseline sample. Next, hierarchical (sequential) forward multivariable regression models were conducted to determine the influence of self-efficacy on pain scores, covarying for the eight participant characteristics. Variance inflation factors (VIF) scores were used to check for multicollinearity (VIF $\geq$ 5.0 indicating concern) among the predictors in each model. For this hierarchical approach, Model 1 included self-efficacy; Model 2 included self-efficacy and demographic covariates; and Model 3 (final model) included self-efficacy, demographic covariates, and clinical covariates. Adjusted R^2 (aR^2, adjusting for the number of predictors in the model) was used as an indicator of effect size and clinical significance. Small, medium, and large effects were indicated by an aR^2 of .02, .13, and .26, respectively. These aR^2 values are equivalent to Cohen's f^2 values of 0.02 (small), 0.15 (medium), and 0.35 (large) effects (Lenhard & Lenhard, 2015).

Trajectory Analyses Random coefficients regression models for longitudinal data, a type of multilevel, mixed-effects model for repeated measures, were employed to determine the trajectories of change in self-efficacy and pain outcomes across 36 months in the trajectory subsample, covarying for patient characteristics. A hierarchical forward (sequential) modeling approach was used to build toward a final trajectory model for each outcome that included time and the eight patient characteristics. Model 1 included time; Model 2 included time and demographic covariates; and Model 3 (final model) included time, demographic covariates, and clinical covariates. Fixed effects were time (months) and covariates (baseline patient characteristics), whereas random effects were participant and Participant × Time. Baseline was defined as Month 0. As needed, trajectories were fitted for a nonlinear pattern of change. The assumption of data missing at random was evaluated.

Trajectory analyses, covarying for patient characteristics, were applied to test for the mediating effect of change in self-efficacy on pain trajectories across 36 months, in accordance with recommended guidelines and path criteria for establishing mediation (Baron & Kenny, 1986; Bennett, 2000).

Statistical Power The baseline sample of 1,903 provided greater than 80% statistical power to examine the relationship between self-efficacy and patient characteristics with the pain outcomes using a hierarchical forward regression, assuming nine explanatory variables (self-efficacy and eight covariates), small effect sizes (aR^2 = .02), and two-tailed tests with significance set at .05 per test for the final and most complex models (Model 3). Based on these assumptions, it was estimated that a sample size of 788 would be required to achieve 80% power (G*Power 3 software; Faul et al., 2007). The trajectory subsample of 427 with time and eight covariates in the model did not provide 80% power, assuming two-tailed tests, significance set at .05 per test, and the smallest clinical meaningful effect size for the time would be small effect sizes (aR^2 = .01), as determined using SuperMix software for longitudinal analyses (Hedeker & Gibbons, 2021).

RESULTS

Baseline Patient Characteristics

The baseline sample was composed of 1,903 adults with SSc. Of those, the 427 who completed a Month 36 outcome assessment were included in the trajectory subsample. Table 1 presents the characteristics of the baseline sample and trajectory subsample at enrollment. The mean age of the baseline sample was 54.8 years (range: 18.3–88.6), with a high proportion of the sample being female (87.5%) and White (83.6%), which is consistent with other large SSc cohorts (Dougherty et al., 2018). The mean time from diagnosis was 9.5 years (range: 0.0–55.8), and 39.2% had dcSSc. The patients who performed serial measures over 3 years were comparable to the overall sample.

Table 2 details self-efficacy for managing pain and pain scores at enrollment for the baseline sample and trajectory subsample. Pain interference scores for the baseline sample were converted to *T* scores and revealed that the mean pain interference *T* score of 55.5 (*SD* = 9.7) was significantly higher than the mean of 50 (*SD* = 10, $z = 24.0$, $p < .001$) estimated for the general U.S. population. A sensitivity analysis did not reveal significant differences in the baseline patient characteristics, self-efficacy scores, and pain scores for those included in the trajectory subsample (N = 427) compared to those excluded from the subsample (N = 1,476).

TABLE 1. Patient Characteristics

Characteristics	Baseline sample (n = 1,903)	Trajectory subsample (n = 427)
Age, in years	54.8 (12.6)	56.7 (11.7)
Female	1666 (87.5%)	377 (88.3%)
Race		
White	1589 (83.6%)	379 (89.0%)
Black	115 (6.1%)	20 (4.7%)
Other	197 (10.4%)	27 (6.3%)
Married/living with partner	1358 (71.4%)	326 (76.4%)
Time since diagnosis, in years	9.5 (8.0)	10.0 (8.3)
Diffuse subset	739 (39.2%)	159 (37.8%)
Raynaud's phenomenon	1853 (98.0%)	416 (98.4%)
Modified Rodnan skin score	7.2 (8.2)	7.3 (7.7)
Distal digital tip ulcers	655 (35.0%)	144 (33.7%)
Digital tip ulcers anywhere	306 (16.7%)	62 (15.0%)
Tendon friction rubs	395 (23.7%)	83 (21.8%)
Moderate–severe small joint contractures	472 (26.2%)	97 (24.1%)
Moderate–severe large joint contractures	227 (12.9%)	48 (11.9%)
Esophageal GI symptoms	1602 (85.0%)	366 (85.9%)
Stomach GI symptoms	572 (31.1%)	119 (28.8%)
Intestinal GI symptoms	727 (39.0%)	157 (37.6%)

Note. n (%) reported for categorical measures; mean (standard deviation) provided for scalar measures. GI = gastrointestinal.

Baseline Analysis: Bivariate Relationships

Bivariate regression indicated that self-efficacy for managing pain was significantly related to pain interference ($\beta = -.60$, $p < .001$, $aR^2 = .36$) and intensity ($\beta = -.55$, $p < .001$, $aR^2 = .30$), with an $aR^2 \geq .26$ indicating large effects (Table 3, Model 1). As self-efficacy increased (greater confidence), pain interference and pain intensity scores significantly decreased (less pain). As expected, pain interference and pain intensity scores were positively correlated ($\beta = +.85$, $p < .001$, $aR^2 = .72$).

Patient characteristics were significantly associated with both pain interference and intensity ($p \leq .05$). Exceptions were age and time since diagnosis. Younger age was significantly related to greater pain interference ($\beta = -.05$, $p = .034$, $aR^2 = .01$); however, age was not associated with intensity ($\beta = -.04$, $p = .093$, $aR^2 = .00$). Time since diagnosis was not significantly related to pain interference ($\beta = +.03$, $p = .188$, $aR^2 = .00$) or intensity ($\beta = +.04$, $p = .124$, $aR^2 = .00$). Finger ulcers explained 3% of variability of pain interference and intensity (both $\beta = +.17$, $p < .001$, $aR^2 = .03$, small effects). Moderate or severe small joint contractures also explained 3% of variability of both pain interference and intensity (both $\beta = +.18$, $p < .001$, $aR^2 = .03$, small effects). Although statistically significant, the remaining characteristics individually explained 1% or less of the variability of the pain scores ($aR^2 \leq .01$, weak effects).

TABLE 2. Baseline Self-Efficacy for Managing Pain and Pain Scores

Baseline measure	Baseline sample Mean (*SD*) (n = 1,903)	Trajectory subsample Mean (*SD*) (n = 427)
Self-efficacy scores	6.1 (2.7)	6.3 (2.7)
Pain interference scores	9.4 (4.7)	9.2 (4.8)
Pain interference *T* scores	55.5 (9.7)	55.1 (9.8)
Pain intensity scores	3.6 (2.6)	3.5 (2.7)

Note. Self-efficacy range was 1–10, with higher scores indicating greater self-efficacy. Pain interference range was 4–20, with higher scores indicating greater pain interference. Pain intensity range was 0–10, with higher scores representing greater pain intensity. *SD* = standard deviation.

Baseline Analysis: Multivariable Relationships

Table 3 provides the hierarchical forward multivariable regression results. The final full covariate-adjusted model (Model 3) indicated that self-efficacy was significantly related to both pain interference ($\beta = -.58$, $p < .001$) and intensity ($\beta = -.53$, $p < .001$), after adjusting for all eight patient characteristics. Specifically, increasing self-efficacy was associated with less pain interference and intensity. This final model explained 38% of the variability of pain interference ($aR^2 = .38$) and 32% of the variability of intensity ($aR^2 = .32$), which was a 2% improvement in aR^2 for both pain outcomes compared to the initial model that included only self-efficacy (Model 1). VIF scores were <1.2 for the final model, indicating that multicollinearity was not a concern.

The final model indicated the following statistically significant relationships between each covariate and the pain outcomes, after adjusting for self-efficacy and other patient characteristics:

TABLE 3. Baseline Analysis: Hierarchical Forward Multivariable Regression Models

			Model 1 (n = 1,903)			Model 2 (n = 1,899)			Model 3 (n = 1,689)		
Pain outcome	Block	Explanatory variable	aR^2 = .36			aR^2 = .36			aR^2 = .38		
			b	*SE*	β	*b*	*SE*	β	*b*	*SE*	β
Interference	1	Self-efficacy	−1.05 ***	0.03	−.60	−1.05***	0.03	−.60	−1.01***	0.03	−.58
	2	Age, in years				+0.01	0.01	+.03	+0.02*	0.01	+.04
		Female				+0.88 ***	0.26	+.06	+0.89***	0.28	+.06
		Married/living with partner				−0.50**	0.19	−.05	−0.36	0.20	−.03
	3	Finger ulcers							+0.85***	0.27	+.07
		Small joint contractures							+0.50*	0.23	+.05
		Esophageal GI symptoms							+0.82***	0.25	+.06
		Time since diagnosis							+0.01	0.01	+.12
		Diffuse subset							+0.37	0.20	+.04
Pain outcome	Block	Explanatory variable	aR^2 = .30			aR^2 = .31			aR^2 = .32		
			b	*SE*	β	*b*	*SE*	β	*b*	*SE*	β
Intensity	1	Self-efficacy	−0.53 ***	0.02	−.55	−0.54***	0.02	−.55	−0.51 ***	0.02	−.53
	2	Age				+0.01*	0.00	+.04	+0.01*	0.00	+.04
		Female				+0.58***	0.15	+.07	+0.58***	0.16	+.07
		Married/living with partner				−0.33**	0.11	−.06	−0.23*	0.12	−.04
	3	Finger ulcers							+0.42**	0.15	+.06
		Small joint contractures							+0.39**	0.13	+.07
		Esophageal GI symptoms							+0.28	0.15	+.04
		Time since diagnosis							+0.01	0.01	+.02
		Diffuse subset							+0.10	0.12	+.02

Note. Block 1 = self-efficacy; Block 2 = baseline demographic characteristics; Block 3 = baseline clinical characteristics; Self-efficacy: higher scores = greater self-efficacy for managing pain; Pain interference: higher scores = greater pain interference; Pain intensity: higher scores = greater pain intensity. Self-efficacy and age were continuous variables. Remaining characteristics coded 0 = *absent* and 1 = *present*; aR^2: .02 = small, .13 = medium, .26 = large effect sizes. aR^2 = adjusted R^2; *b* = unstandardized regression coefficient; *SE* = standard error; β = standard regression coefficient; GI = gastrointestinal.

*$p \leq .05$. **$p \leq .01$. ***$p \leq .001$.

(a) increasing age was associated with greater pain interference and intensity; (b) females reported greater pain interference and intensity; (c) adults married/living with a partner reported less pain intensity, but this covariate was not related to pain interference; (d) finger ulcers were associated with greater pain interference and intensity; (e) moderate or severe small joint contractures were related to greater pain interference and intensity; and (f) esophageal gastrointestinal symptoms were associated with greater pain interference, but not intensity. Neither time since diagnosis nor diffuse subset were significantly related to pain outcomes. In contrast to the bivariate results, the results from the full regression model indicated (a) older age was significantly associated with both pain outcomes, (b) married/living with a partner was only related to pain intensity, (c) esophageal gastrointestinal symptoms were associated with pain interference only, and (d) dcSSc was not related to pain interference or intensity.

Trajectories Analysis: Change in Self-Efficacy and Pain Outcomes

Supplemental Digital Content Table S1, http://links.lww.com/NRES/A400, presents descriptive statistics and the completion rate for self-efficacy, pain interference, and pain intensity at each assessment across the 36 months. All 427 patients in the trajectory subsample completed the baseline (Month 0) and final (Month 36) assessments. The completion rate was 85% or higher for most of the other assessments included in each analysis. For each outcome, a linear trajectory model best fitted the longitudinal data.

Table 4 presents the trajectory model results for self-efficacy and pain outcomes, applying a forward model building approach. The time effect in the initial model (Model 1) and two covariate-adjusted models (Models 2 and 3) for each outcome was not statistically significant, indicating no significant change across the 36 months in self-efficacy, pain interference, or pain intensity. With regard to magnitude of change across the 36 months, minimal to very small effect sizes were observed for each outcome (intraclass correlation coefficients < .15). Figure 1 presents the adjusted mean trajectory for each outcome, covarying for the patient characteristics (Model 3).

Three baseline covariates were significantly related to all three outcomes in the final model (Model 3). Finger ulcers were associated with greater pain interference (unstandardized regression coefficient b = +1.81, SE = 0.63, p = .005), greater pain intensity (b = +1.21, SE = 0.37, p = .001), and less self-efficacy (b = −0.75, SE = 0.37, p = .042). Moderate-to-severe small joint contractures were associated with greater pain interference (b = +1.43, SE = 0.52, p = .007), greater pain intensity (b = +1.00, SE = 0.31, p = .001), and less self-efficacy (b = −0.75, SE = 0.30, p = .012). Finally, esophageal gastrointestinal

TABLE 4. Trajectory Analysis Hierarchical Forward Mixed-Effects Models

			Model 1 ($n = 427$)	Model 2 ($n = 424$)	Model 3 ($n = 377$)
Outcome	**Block**	**Explanatory variable**	***p***	***p***	***p***
Interference	1	Time, in months	.382	.548	.719
	2	Age, in years		.215	.591
		Female		.832	.956
		Married/living with partner		.044	.215
	3	Finger ulcers			.005
		Small joint contractures			.007
		Esophageal GI symptoms			.001
		Time since diagnosis			.195
		Diffuse subset			.844
Intensity	1	Time, in months	.298	.323	.158
	2	Age, in years		.400	.824
		Female		.942	.761
		Married/living with partner		.033	.181
	3	Finger ulcers			.001
		Small joint contractures			.001
		Esophageal GI symptoms			.006
		Time since diagnosis			.147
		Diffuse subset			.126
Self-efficacy	1	Time, in months	.101	.217	.984
	2	Age, in years		.033	.135
		Female		.110	.158
		Married/living with partner		.039	.107
	3	Finger ulcers			.042
		Small joint contractures			.012
		Esophageal GI symptoms			.001
		Time since diagnosis			.125
		Diffuse subset			.444

Note. Mixed-effects trajectory models with time and baseline patient characteristics (covariate) as fixed effects; patient and Patient × Time interaction as random effects. Significant results are bolded. Block 1 = time, in months; Block 2 = baseline demographic characteristics; Block 3 = baseline clinical characteristics; GI = gastrointestinal.

symptoms were associated with greater pain interference (b = +1.91, SE = 0.57, p = .001), greater pain intensity (b = +0.93, SE = 0.33, p = .006), and less self-efficacy (b = −1.13, SE = 0.33, p = .001). Thus, finger ulcers, small joint contractures, and esophageal gastrointestinal symptoms were related to higher levels of pain and less self-efficacy for managing pain.

Change in Self-Efficacy as a Mediator of Pain

Change in self-efficacy over 36 months did not mediate the trajectories of pain interference or intensity, covarying for patient characteristics. Not all four criteria (paths) for establishing mediation were met. Path A (impact of time on self-efficacy) was not met because of the minimal, nonsignificant change in self-efficacy over time (noted above). Path B (association between self-efficacy and pain outcomes) was met for improvements in self-efficacy and were significantly associated with reductions in pain interference (b = −0.43, SE = 0.07, p < .001, aR^2 = .09, small effect) and intensity (b = −0.24, SE = 0.04, p < .001, aR^2 = .08, small effect). Path C (impact of time on pain outcomes) and Path C′ (impact of time on pain outcomes, covarying for self-efficacy over time) were not met because of the minimal, nonsignificant change in pain interference and intensity over time (noted above).

DISCUSSION

SSc is a chronic and progressive disease in which patients frequently experience pain and are challenged with self-managing their symptoms throughout the disease course. In this study, self-efficacy for managing pain and pain interference and intensity did not significantly change over the 3-year period. One potential explanation for the stable pain trajectories found in our study is that our sample consisted of patients who had lived with their disease for approximately 9 years. Previous research found that patients experience higher levels of disease activity, more rapid progression, and worsening skin thickening within the first 5 years of symptom onset, after which the overall disease course remains relatively stable (Medsger, 2003). Given patients had stable disease, the occurrence of chronic pain is unsurprising. Chronic pain or pain that occurs for more than 3 months (Treede et al., 2015) has been reported in up to 75% of

patients with SSc and is often refractory to treatment (Thombs et al., 2008). Although our trajectories supported the chronicity of pain, patients have also described their disease as progressive in nature with superimposed flares lasting for 3 days to 3 months (Suarez-Almazor et al., 2007). Although qualitative studies have provided descriptions of symptom flares in patients with SSc, quantitative studies have yet to quantify or characterize such flares. One potential explanation for this is that the longitudinal data used to establish trajectories in SSc do not include time points frequent enough to capture periods of intense symptoms, subsequently resulting in relatively stable trajectories as seen in this study. Future studies that measure these distinct periods of symptom flares are needed to guide the timing and delivery of pain management interventions in this population.

In our study, the presence of finger ulcers, small joint contractures, and esophageal gastrointestinal symptoms were associated with baseline pain outcomes. Although our study underscored the association between finger ulcers and pain, we did not explore the frequency or chronicity of finger ulcers. This is an important consideration given that previous research has identified that patients who experience more frequent digital ulcers, such as those with recurrent or chronic ulcers, often have a higher disease burden and a greater need for intervention (Matucci-Cerinic et al., 2016).

In addition, we found that small joint contractures and esophageal gastrointestinal symptoms (i.e., dysphagia, heartburn, and/or reflux) were significantly associated with pain interference and intensity. Previous studies have also described an association between joint contractures and esophageal gastrointestinal symptoms (Ashida et al., 2007; Johnson et al., 2006); however, these studies did not explore differences in pain outcomes separately among small and large joint contractures or explore the extent and severity of gastrointestinal symptoms. This is an important consideration given that small joint contractures of the hand often do not improve with pharmacological therapy (Young et al., 2016), and many patients do not respond to existing treatments for esophageal gastrointestinal symptoms (Denaxas et al., 2018). Future studies might explore differences in pain outcomes in patients who experience more frequent and/or chronic finger ulcers and differences in the extent and severity of gastrointestinal symptoms extending beyond esophageal symptoms to inform future pain management interventions in patients with SSc.

Greater self-efficacy for managing pain was associated with less pain interference and lower pain intensity. Patients with SSc report worse self-efficacy than patients with other chronic diseases (i.e., multiple sclerosis, cardiovascular disease, and breast cancer), particularly related to performing self-care tasks for pain (Thombs et al., 2017). Despite the correlation between self-efficacy and pain in this population (Buck et al., 2010), self-management interventions aimed at improving self-efficacy have shown mixed results. For example, there was no significant difference in self-efficacy in a randomized controlled trial comparing an Internet-based self-management program with a patient-focused educational book (Khanna et al., 2019). However, online self-management programs for patients with chronic diseases (i.e., arthritis, diabetes, hypertension, lung and heart disease), such as the Chronic Disease Self-Management Program, have been associated with improvements in self-efficacy and health outcomes (Lorig et al., 2006), with sustained improvements in outcomes over time (Barlow et al., 2005). Although self-efficacy for managing pain did not change over time and was not a mediator of the pain trajectories in our study, future studies might explore the timing and effect of self-management interventions on pain outcomes, such as the delivery of a self-management intervention during a period of high disease activity (i.e., within the first 5 years of disease onset).

Limitations

Our study had many strengths, including a large sample, validated SSc diagnosis by a healthcare provider, inclusion of patient-reported measures, and the use of longitudinal, multicenter data. The SPIN Cohort is a convenience sample of patients receiving care at a SPIN Center with access to the Internet and the ability to complete online measures, which may limit the generalizability of findings. In addition, participants in this study had a mean disease duration of 9.5 years, and results may differ for patients with a shorter or longer disease duration. Although patient-reported measures were used, our study only used a single item of the SEMCD Scale; future studies should consider use of the full scale. Finally, although the baseline sample size provided power to detect statistically significant relationships with small, nonclinically meaningful effects, such findings should be interpreted in terms of their clinical relevance. The sample size for the trajectory analysis did not provide adequate power for the very small time effects observed. Thus, this analysis was exploratory in nature.

Conclusion

This study examined the relationship between self-efficacy for managing pain and pain outcomes upon enrollment and over 3 years in adults with SSc. Our findings indicate that self-efficacy for managing pain was strongly and inversely related to pain outcomes and that self-efficacy for managing pain and pain outcomes remained stable over a 3-year period. We identified important correlates (i.e., self-efficacy for managing pain, age, gender, finger ulcers, small joint contractures, and esophageal gastrointestinal symptoms) associated with pain outcomes in adults with SSc that may serve as important factors to consider in the development of pain management interventions. In addition, our findings provide important insights into the longitudinal pain experiences of patients with SSc and can inform the assessment and management of pain in this population.

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