Evaluation of the Coping Strategies Questionnaire-Revised for fatigue in systemic sclerosis: a Scleroderma Patient-centred Intervention Network (SPIN) cohort study

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Abstract

Objective
To adapt and evaluate the Coping Strategies Questionnaire-Revised (CSQ-R), designed to assess pain coping, for assessing coping with fatigue in systemic sclerosis (SSc).

Methods
We adapted CSQ-R items for fatigue, and a panel of people with SSc verified content validity. Scleroderma Patient-centred Intervention Network Cohort participants completed the CSQ-R-Fatigue. We evaluated factor structure with confirmatory factor analysis (CFA), assessed differential item functioning (DIF) by English and French language and disease subtype, and evaluated internal consistency and test-retest reliability.

Results
863 participants were included. Most were female (n=756; 88%), and 36% (n=308) had diffuse SSc. We replicated the 6-factor CSQ-R structure (Tucker-Lewis Index =0.95, Comparative Fit Index =0.97, Root Mean Square Error of Approximation =0.05). We found substantive DIF across multiple factors, however, for language and disease subtype (11 items on 6 factors for language, 10 items on 5 factors for subtype). Factor-score differences due to DIF by language and subtype were ≥0.20 standardised mean differences for 4 factors each. Test-retest reliability for factors based on intraclass correlation was between 0.68 [95% CI 0.58, 0.76] and 0.91 [95% CI 0.88, 0.93]; n=183.

Conclusion
The CSQ-R-Fatigue may not be appropriate to assess coping with fatigue in SSc due to possible biases related to language and disease severity. An additional concern is that the CSQ-R-Fatigue focuses on psychological coping and does not assess active coping strategies. Research is needed to identify or develop tools to evaluate coping strategies for managing fatigue in SSc.

Key words
assessment, coping, fatigue, patient-reported outcomes, systemic sclerosis

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Introduction

Systemic sclerosis (SSc; scleroderma) is a rare chronic autoimmune disease characterised by fibrosis and thickening of the skin and internal organs including the lungs, gastrointestinal tract, kidneys, and heart (1). Fatigue, defined as persistent exhaustion that exceeds exertion level and is not relieved by sleep (2), is a common and debilitating symptom, but little is known about how people with SSc cope with fatigue (3, 4). How people select coping strategies and their effectiveness depend on the stressor and situation (5), but there are no validated measures of coping with fatigue from chronic diseases, including SSc. The 27-item Coping Strategies Questionnaire-Revised (CSQ-R) (6) is commonly used to assess coping with chronic pain (7) and includes 27 items that reflect 6 domains: distraction, catastrophising, ignoring, distancing, coping self-statements, and praying.

The objectives of the present study were to evaluate whether the CSQ-R can be adapted for coping with fatigue from SSc (CSQ-R-Fatigue) and evaluate measurement properties by (1) performing a confirmatory factor analysis (CFA) to evaluate factor structure; (2) assessing differential item functioning (DIF) for English and French versions and by SSc disease subtype (diffuse, limited or sine) to determine if scores are comparable across languages and subtype; and (3) evaluating internal consistency and test-retest reliability.

Materials and methods

We used cross-sectional data from the Scleroderma Patient-centred Intervention Network (SPIN) Cohort (8-10) to evaluate English- and French-language versions of the CSQ-R-Fatigue for factor structure, language- and subtype-based DIF, and internal consistency. Due to substantive DIF by language and subtype, we compared unadjusted and DIF-adjusted scores to Patient-Reported Outcomes Measurement Information System-29 v. 2.0 (PROMIS-29v2) domains. We administered the CSQ-R-Fatigue a second time, 1-2 weeks after the initial assessment, to a subset of participants to evaluate test-retest reliability.

We reported study results based on guidance from the COnsensus-based Standards for the selection of health status Measurement INstruments (COSMIN) Study Design checklist (11). Methods in SPIN Cohort studies are similar; thus, we adhered to reporting guidance from the Text Recycling Research Project (12).

Participants and procedure
The SPIN Cohort includes individuals with SSc from 50 sites in 7 countries (Australia, Canada, France, Mexico, Spain, UK, USA). Participants must be fluent in English, French, or Spanish; be ≥18 years of age; meet 2013 American College of Rheumatology/European League Against Rheumatism criteria for SSc (13), verified by a SPIN site physician; and have internet access. Participants are invited to enrol by recruiting site physicians or nurse coordinators. After providing written informed consent, site staff submit an online medical data form to enrol participants, and participants receive email instructions to activate their online SPIN account. They complete baseline assessments and follow-up assessments every 3 months. We analysed data from participants from 48 sites who completed measures in English or French from January 11, 2024, to April 11, 2024, when the CSQ-R-Fatigue was included in the SPIN Cohort. We excluded 2 Spanish-language sites because the CSQ-R is not available in Spanish.

To assess test-retest reliability, we aimed to administer the CSQ-R-Fatigue to approximately 200 participants 1-2 weeks after initially completing the scale. Participants received an email invitation via Qualtrics 7 days later to complete the scale a second time. We sent email invitations until approximately 200 participants completed the survey a second time. The questionnaire remained accessible to invited participants for 7 days to ensure responses within a 7- to 14-day window following initial completion. We sent a reminder email to non-responders 4 days after the initial invitation. As an incentive, 5 respondents were randomly selected to receive a $100 Amazon gift card, or their country's currency equivalent.

The SPIN Cohort was approved by the Research Ethics Committee of the Centre Intégré Universitaire de Santé et de Services Sociaux du Centre-Ouest-de-l'Île-de-Montréal (no. MP-05-2013-150) and by the ethics committees of all recruiting sites. The present study was approved as an amendment.

Measures
At baseline, SPIN Cohort participants provide sociodemographic information, including race or ethnicity, education, and marital status. Physician-reported data collected at baseline include age; sex; height; weight; date of initial onset of non-Raynaud phenomenon symptoms; SSc subtype (diffuse, limited, sine); digital tip ulcers; telangiectasias; abnormal skin pigmentation; current tendon friction rubs; presence of joint contractures, gastrointestinal involvement, interstitial lung disease, pulmonary arterial hypertension, and overlap syndromes; and history of scleroderma renal crisis. Participants complete patient-reported outcome measures at baseline and all follow-ups.

- CSQ-R-Fatigue
The CSQ-R is composed of 27 items on 6 domains related to cognitive strategies for coping with pain, including coping self-statements (4 items), catastrophising (6 items), distancing (4 items), distraction (5 items), ignoring pain (5 items), and praying (3 items; 14). CSQ-R item responses reflect behaviour frequency with no specified time period on a 7-point frequency scale ranging from 0 (never) to 6 (always). Higher scores on each domain indicate greater coping strategy use. There is a French version (15), which had been forward and backward translated, but members of our translation group identified several items for which, when we back translated to English ourselves, led to inconsistencies with the original English scale. We edited 1 item to use more inclusive gender language, 4 for item comparability, and 1 for grammar (Supplementary Table S1). To adapt items to assess coping with fatigue rather than pain, we replaced the word 'pain' with 'fatigue' (Suppl. Tables S2 and S3 for full English and French CSQ-R-Fatigue scales).

To assess content validity of the CSQ-R-Fatigue, we conducted expert reviews by inviting people with SSc who had previously participated as SPIN research partners to review the English versions of the CSQ-R and the CSQ-R-Fatigue. For each CSQ-R-Fatigue item, we asked if the item a) was clear and b) would also apply to fatigue. Seven of twenty-one invited responded and confirmed both criteria for 22 of 27 items. One respondent indicated that the other 5 items (items 11-14, 20), which all addressed the concept of 'thinking outside the body' were unclear; however, the other six respondents rated these items as clear. Therefore, we retained all 27 items.

- PROMIS-29v2
The PROMIS-29v2 (16) measures patient-reported health status over the past 7 days on 7 domains (physical function, anxiety, depression, fatigue, sleep disturbance, social role and activity participation, pain interference), each with 4 items, plus a single pain intensity item. Items are scored from 1 to 5 with different response options by domain. The pain intensity item is measured on an 11-point rating scale (0=no pain, 10=worst imaginable pain). Higher scores represent more of a domain; that is, better physical function and greater anxiety, for example. Raw domain sum scores are converted into T-scores standardised for the general US population (mean=50, standard deviation [SD]=10). English and French PROMIS-29v2.0 versions have been validated in SSc (17, 18).

Statistical analyses
We calculated descriptive statistics, including means and SDs for continuous variables and frequencies with percentages for categorical variables, for the total sample and separately for English- and French-language participants.

- CFA
We used CFA to verify the hypothesised 6-factor structure of the CSQ-R-Fatigue item loading onto designated domains (14). CSQ-R-Fatigue responses consist of ordinal Likert data. Thus, items were modelled with

the weighted least squares estimator, a diagonal weight matrix, and robust standard errors. Model fit was assessed using the Tucker-Lewis Index (TLI), Comparative Fit Index (CFI) and Root Mean Square Error of Approximation (RMSEA) with thresholds for well-fitting models of TLI and CFI ≥0.95 and RMSEA ≤0.06 (19), although CFI ≥0.90 and RMSEA ≤0.08 are often regarded as indicators of acceptable fit (20). Modification indices were used to identify if allowing error covariances between similar items would improve model fit.

- DIF analysis
DIF analyses compare patterns of item responses between subgroups and examine whether individuals with similar levels of a latent construct respond similarly to each item, irrespective of group affiliation. We conducted DIF analyses to evaluate whether any item response patterns were associated with language or disease subtype, controlling for latent domain scores. We used the Multiple-Indicator Multiple-Cause (MIMIC) model, which is based on structural equation modelling (21). The group variable (*e.g.* English *vs.* French, diffuse versus limited or sine) is incorporated into the CFA model as an observed variable, and the base MIMIC model comprises the CFA factor model along with a direct effect of group on each latent factor, thus adjusting for group differences on latent factor levels. To evaluate item DIF, we additionally regressed each item on group (language or disease subtype), one at a time, and determined the presence of DIF based on a statistically significant ($p<0.05$) link from the group variable to the item. When DIF was detected for one or more items in a domain, the item exhibiting DIF with the lowest p-value was considered to have DIF, the link between the group variable and that item was incorporated into the model, and other items were re-assessed for DIF. This procedure was repeated until no additional items exhibited DIF. We evaluated items for language-based then disease subtype-based DIF. All models also included age and years since onset of first non-Raynaud's symptoms

Table I. Sociodemographic and disease characteristics for the full sample and by assessment language.

Characteristics		Full sample (n=863)		English (n=493)		French (n=370)
	n	Mean (SD) or n (%)	n	Mean (SD) or n (%)	n	Mean (SD) or n (%)
Sociodemographic variables						
Age (years)	863	61.9 (12.1)	493	63.5 (11.1)	370	59.7 (13.0)
Sex	863		493		370	
Female		756 (87.6%)		438 (88.8%)		318 (85.9%)
Male		107 (12.4%)		55 (11.2%)		52 (14.1%)
Education (years)	853	15.2 (3.7)	489	15.8 (3.2)	364	14.4 (4.2)
Marital status	856		490		366	
Married or living as married		602 (70.3%)		354 (72.2%)		248 (67.7%)
Single, divorced/separated, widowed		254 (29.7%)		136 (27.7%)		118 (32.2%)
Race or ethnicity[a]	842		480		362	
White		727 (86.3%)		418 (87.1%)		309 (85.4%)
Non-white		115 (13.7%)		62 (12.9%)		53 (14.6%)
Country	863		493		370	
USA		230 (26.7%)		230 (46.7%)		0 (0.0%)
France		321 (37.2%)		3 (0.6%)		318 (85.9%)
Canada		216 (25.0%)		164 (33.3%)		52 (14.1%)
UK		80 (9.3%)		80 (16.2%)		0 (0.0%)
Australia		16 (1.9%)		16 (3.2%)		0 (0.0%)
Body mass index (kg/m²)	863	25.0 (5.2)	493	25.4 (5.4)	370	24.5 (4.9)
Years since first non-Raynaud's symptoms	802	16.3 (9.5)	456	18.5 (9.5)	346	13.5 (8.7)
Disease subtype	858		489		369	
Diffuse		308 (35.9%)		207 (42.3%)		101 (27.4%)
Limited or sine		550 (64.1%)		282 (57.7%)		268 (72.6%)
Gastrointestinal involvement	863		493		370	
Yes		735 (85.2%)		434 (88.0%)		301 (81.4%)
No		128 (14.8%)		59 (12.0%)		69 (18.6%)
Digital ulcers	818		458		360	
Yes		110 (13.4%)		65 (14.2%)		45 (12.5%)
No		708 (86.6%)		393 (85.8%)		315 (87.5%)
Tendon friction rubs	761		417		344	
Current		74 (9.7%)		39 (9.4%)		35 (10.2%)
Past		72 (9.5%)		52 (12.5%)		20 (5.8%)
Never		615 (80.8%)		326 (78.2%)		289 (84.0%)
Small joint contractures	813		453		360	
None or mild		610 (75.0%)		346 (76.4%)		264 (73.3%)
Moderate		150 (18.5%)		81 (17.9%)		69 (19.2%)
Severe		53 (6.5%)		26 (5.7%)		27 (7.5%)
Large joint contractures	798		448		350	
None or mild		704 (88.2%)		401 (89.5%)		303 (86.6%)
Moderate		76 (9.5%)		36 (8.0%)		40 (11.4%)
Severe		18 (2.3%)		11 (2.5%)		7 (2.0%)
History of SSc renal crisis	850		486		364	
Yes		30 (3.5%)		18 (3.7%)		12 (3.3%)
No		820 (96.5%)		468 (96.3%)		352 (96.7%)
Interstitial lung disease	834		476		358	
Yes		254 (30.5%)		142 (29.8%)		112 (31.3%)
No		580 (69.5%)		334 (70.2%)		246 (68.7%)
Pulmonary arterial hypertension	833		471		362	
Yes		65 (7.8%)		37 (7.9%)		28 (7.7%)
No		768 (92.2%)		434 (92.1%)		334 (92.3%)
Overlap[b]	863		493		370	
Yes		171 (19.8%)		108 (21.9%)		63 (17.0%)
No		692 (80.2%)		385 (78.1%)		307 (83.0%)

[a]Race or ethnicity data were self-reported in each country using standard categories used in that country. Therefore, categories differed between countries.
[b]Participant had one or more of rheumatoid arthritis, Sjögren's syndrome, systemic lupus erythematosus, or idiopathic inflammatory myositis.

due to differences by language on those variables. We used mean substitution for missing time since non-Raynaud's symptom onset (n=61 missing) to preserve sample size.

Once all items with DIF were identified, cumulative influence of DIF on total CSQ-R-Fatigue factor scores was assessed by comparing latent factor scores between language and disease subtype groups in the baseline CFA model not adjusted for DIF and the DIF-adjusted model. We evaluated strength of association between latent factor scores from each model using Pearson's product-moment correlation coefficient and agreement using intraclass correlation coefficient (ICC). We also assessed the standardised mean difference (SMD) on latent factor scores between groups in baseline and DIF-adjusted models.

Table II. Coping Strategies Questionnaire-Revised for Fatigue confirmatory factor analysis base model and DIF-corrected model factor loadings and DIF evaluation for items by domains.

	Base model[a]		DIF-corrected model[b]	
	Factor loading	95% CI	Factor loading	95% CI
Catastrophising				
3. It's terrible and I feel it's never going to get any better	0.71	0.66, 0.75	0.74	0.68, 0.79
6. It's awful and I feel that it overwhelms me	0.79	0.75, 0.82	0.88	0.83, 0.92
7. I feel my life isn't worth living	0.63	0.57, 0.69	0.65	0.59, 0.70
14. I worry all the time about whether it will end	0.79	0.75, 0.83	0.77	0.73, 0.81
21. I feel I can't stand it anymore	0.81	0.78, 0.85	0.82	0.78, 0.85
24. I feel like I can't go on	0.76	0.72, 0.81	0.77	0.73, 0.81
Coping self-statements				
4. I tell myself to be brave and carry on despite the fatigue	0.71	0.67, 0.76	0.72	0.66, 0.77
5. I tell myself that I can overcome the fatigue	0.78	0.73, 0.82	0.78	0.74, 0.82
11. I tell myself I can't let fatigue stand in the way of what I have to do	0.71	0.66, 0.77	0.72	0.67, 0.78
20. Although I am fatigued, I just keep on going	0.69	0.64, 0.74	0.71	0.66, 0.76
Distancing				
1. I try to feel distant from the fatigue, almost as if the fatigue was in somebody else's body	0.81	0.77, 0.84	0.95	0.88, 1.02
9. I try not to think of it as my body, but rather as something separate from me	0.75	0.71, 0.80	0.81	0.77, 0.86
18. I imagine that the fatigue is outside of my body	0.77	0.73, 0.81	0.84	0.79, 0.88
27. I pretend it's not a part of me	0.82	0.78, 0.85	0.94	0.87, 1.01
Distraction				
2. I try to think of something pleasant	0.80	0.76, 084	0.83	0.79, 0.87
15. I replay in my mind pleasant experiences in the past	0.74	0.69, 0.79	0.66	0.60, 0.72
16. I think of people I enjoy doing things with	0.81	0.77, 0.84	0.76	0.72, 0.81
25. I think of things I enjoy doing	0.84	0.80, 0.87	0.83	0.79, 0.86
26. I do something I enjoy, such as watching TV or listening to music	0.62	0.57, 0.68	0.65	0.60, 0.71
Ignoring				
10. I don't think about the fatigue	0.51	0.43, 0.58	0.52	0.44, 0.59
12. I don't pay any attention to it	0.73	0.68, 0.77	0.74	0.69, 0.79
13. I pretend it's not there	0.84	0.81, 0.88	0.85	0.81, 0.90
19. I just go on as if nothing happened	0.80	0.76, 0.83	0.81	0.77, 0.85
22. I ignore it	0.79	0.75, 0.83	0.80	0.76, 0.84
Praying				
8. I pray to God it won't last long	0.80	0.75, 0.85	0.77	0.73, 0.82
17. I pray for the fatigue to stop	0.95	0.91, 0.99	0.91	0.86, 0.95
23. I rely on my faith in God	0.58	0.52, 0.65	0.76	0.68, 0.85
Direct effects on items associated with French language[c]				
Catastrophising				
3. It's terrible and I feel it's never going to get any better	-	-	-0.08	-0.15, -0.01
6. It's awful and I feel that it overwhelms me	-	-	-0.21	-0.27, -0.14
Coping self-statements				
4. I tell myself to be brave and carry on despite the fatigue	-	-	0.32	0.22, 0.41
20. Although I am fatigued, I just keep on going	-	-	-0.20	-0.30, -0.11
Distancing				
1. I try to feel distant from the fatigue, almost as if the fatigue was in somebody else's body	-	-	-0.34	-0.45, -0.23
27. I pretend it's not a part of me			-0.30	-0.40, -0.20
Distraction				
15. I replay in my mind pleasant experiences in the past	-	-	0.51	0.42, 0.61
16. I think of people I enjoy doing things with	-	-	0.33	0.24, 0.43
25. I think of things I enjoy doing	-	-	0.17	0.09, 0.25
Ignoring				
13. I pretend it's not there	-	-	0.31	0.23, 0.40
Praying				
23. I rely on my faith in God	-	-	-0.38	-0.49, -0.27
Direct effects on items associated with diffuse disease subtype[d]				
Coping self-statements				
4. I tell myself to be brave and carry on despite the fatigue	-	-	0.26	0.17, 0.35
20. Although I am fatigued, I just keep on going	-	-	-0.20	-0.29, -0.11
Distancing				
9. I try not to think of it as my body, but rather as something separate from me	-	-	0.21	0.14, 0.28
18. I imagine that the fatigue is outside of my body	-	-	0.26	0.19, 0.33
Distraction				
2. I try to think of something pleasant	-	-	-0.15	-0.25, -0.05
15. I replay in my mind pleasant experiences in the past	-	-	0.31	0.22, 0.40
16. I think of people I enjoy doing things with	-	-	0.18	0.10, 0.26
26. I do something I enjoy, such as watching TV or listening to music	-	-	-0.12	-0.20, -0.05
Ignoring				
13. I pretend it's not there	-	-	0.18	0.10, 0.26
Praying				
23. I rely on my faith in God	-	-	-0.34	-0.46, -0.21

CFA: confirmatory factor analysis; CI: confidence interval.
[a]Unstandardised model with fixed variance and regression of the latent coping domains (coping self-statements, distancing, catastrophizing, distraction, ignoring, praying) on language and disease subtype, not corrected for differential item functioning (DIF).
[b]Unstandardised model with fixed variance and regression of the latent coping domains (coping self-statements, distancing, catastrophizing, distraction, ignoring, praying) on language (corrected for DIF on items 4, 6, 8, 11, 13, 15, 17, 20, 25, 26, and 27) and on disease subtype (corrected for DIF on items 2, 4, 9, 13, 15, 16, 18, 20, 23, 26).
[c]Positive parameters reflect higher item scores among French-language participants.
[d]Positive parameters reflect higher item scores among participants with diffuse disease subtype.

To generate SMD estimates, we set latent factor variances equal to 1. Additionally, we compared associations between unadjusted and DIF-adjusted model domain scores with PROMIS-29v2 domain scores using Pearson's correlations.

Because we found consequential DIF, we conducted item analyses and assessed reliability for the overall sample and separately by language and subtype subgroups.

- Item analyses
Means, SDs, item intercorrelations, and corrected item-total correlations were calculated for each CSQ-R-Fatigue item. We also assessed floor and ceiling effects, defined as ≥15% of participants having the lowest or highest possible score on each CSQ-R-Fatigue domain (22).

- Reliability
To assess internal consistency, we computed Cronbach's alpha (23). For evaluating test-retest reliability, we utilised ICC (23, 24). We chose ICC as the measure of test-retest reliability based on its ability to capture both the degree of correlation and agreement between measurements (25). All statistical analyses were carried out utilising R-studio v. 2023.12.1.402 (26).

Sample size
- CFA
We planned to perform a multiple-factor CFA with 3 to 6 indicators per factor using a sample that we expected would include 800 to 1000 participants. This number easily exceeded the minimum number suggested in recommendations and standards for excellent agreement between true model characteristics and estimates (27-29).

- Test-retest reliability
ICC ≥0.70 reflects acceptable test-retest reliability, but ≥0.80 is preferable (30). We sought to do test-retest with 200 people to generate a 95% CI of width ± 0.05 for ICC of 0.80 (31, 32).

Participant feedback
We received unsolicited emails from 3 participants about CSQ-R-Fatigue con-tent, and we evaluated those along with input from members of our team who live with SSc.

Results
Sample characteristics
Of 1425 active SPIN Cohort participants, 863 (61%) completed the CSQ-R-Fatigue and were included in the study. There were 756 female participants (88%), mean (SD) age was 61.9 (12.1) years, mean (SD) time since first non-Raynaud's phenomenon symptom onset was 16.3 (9.5) years, and 308 individuals had diffuse SSc (36%). Participants were from France (37%), USA (27%), Canada (25%), UK (9%), and Australia (2%). Sociodemographic and disease characteristics were similar across English- (n=493, 57%) and French-language participants (n=370, 43%), except for age, years since first non-Raynaud's symptoms, and disease subtype (Table I).

There were 183 participants for test-retest. Characteristics were similar to those of the total sample (Suppl. Table S4).

CSQ-R-Fatigue measurement properties
- CFA and DIF analysis
CFA results (n=863) are shown in Table II. In the initial 6-factor CFA, the hypothesised multi-factor model fit well (TLI 0.95, CFI 0.96, RMSEA 0.06) without freeing any items to covary. The model with latent coping domains regressed on language, age, years since first non-Raynaud's symptoms and disease subtype also demonstrated good fit (n=858, 5 participants missing disease subtype not included; TLI 0.97, CFI 0.97, RMSEA 0.05). Table II shows baseline CFA factor loadings and parameters in the DIF-corrected model. We identified 11 items on 6 domains with statistically significant language-based DIF and 10 items on 5 domains with statistically significant disease subtype-based DIF.

As shown in Table III, estimated differences between French- and English-language respondents between unadjusted and DIF-adjusted models on CSQ-R-Fatigue subscales were 0.20 SMDs or larger for catastrophising, distancing, ignoring, and praying, with positive SMDs indicating that French-language respondents had greater scores than would be expected without DIF. Estimated differences between limited-sine and diffuse SSc respondents between unadjusted and DIF-adjusted models on CSQ-R-Fatigue subscales were 0.20 SMDs or larger for catastrophising, coping self-statements, ignoring, and praying, with positive SMDs indicating that diffuse disease subtype respondents had greater scores. Associations between unadjusted and DIF-adjusted scores were between 0.96 and >0.99 for Pearson's correlations and 0.96 and >0.99 for ICC for both language and disease subtype. Raw differences in Pearson's correlations between unadjusted and DIF-adjusted scores and PROMIS-29v2 domain scores were ≤ 0.05 for all correlations (Fig. 1 and Suppl. Table S5).

- Item analysis
Mean item and total CSQ-R-Fatigue domain scores and item-rest correlations in the full sample are shown in Table IV and by language and disease subtype in Supplementary Table S6. Correlations between individual items ranged from r=0.36 (items 10 and 19) to r=0.72 (items 15 and 16 and items 16 and 25; not shown). Item-rest correlations within subscales ranged from r=0.49 (item 10) to r=0.79 (item 25), and this was similar by subgroups (not shown). The percentage of individuals with the highest possible score for each domain ranged from 0.3% (catastrophising) to 7% (coping self-statements). The percentage of individuals with the lowest possible score for each domain ranged from 1% (coping self-statements) to 33% (praying). Item response frequencies are shown in Supplementary Table S7.

- Reliability
Cronbach's alpha was 0.81 (95% CI 0.78, 0.85) for self-statements, 0.87 (95% CI 0.84, 0.90) for distancing, 0.88 (95% CI 0.85, 0.91) for catastrophising, 0.87 (95% CI 0.84, 0.90) for distraction, 0.85 (95% CI 0.82, 0.89) for ignoring, and 0.82 (95% CI 0.78, 0.85) for praying. For test-retest re-

Table III. Standardised mean difference, intra-class correlations, and Pearson's correlations for the Coping Strategies Questionnaire-Revised for Fatigue with and without DIF adjustment.

Domain	Language[a]		Subtype[b]		ICC	Pearson's correlation coefficient
	Without DIF adjustment	With DIF adjustment	Without DIF adjustment	With DIF adjustment		
	SMD (95% CI)	SMD (95% CI)	SMD (95% CI)	SMD (95% CI)		
Catastrophising	0.22 (0.15, 0.29)	0.58 (0.51, 0.65)	0.01 (-0.07, 0.08)	0.34 (0.26, 0.42)	0.99	>0.99
Coping self-statements	0.04 (-0.04, 0.12)	-0.13 (-0.24, -0.02)	-0.04 (-0.12, 0.04)	-0.16 (-0.27, -0.05)	>0.99	>0.99
Distancing	0.34 (0.27, 0.41)	0.54 (0.45, 0.63)	-0.02 (-0.09, 0.05)	-0.03 (-0.12, 0.05)	0.96	0.97
Distraction	0.24 (0.17, 0.31)	0.03 (-0.08, 0.14)	0.00 (-0.07, 0.08)	0.06 (-0.06, 0.17)	0.96	0.96
Ignoring	0.08 (0.01, 0.16)	-0.20 (-0.30, -0.10)	-0.07 (-0.15, 0.00)	-0.28 (-0.38, -0.18)	0.99	0.99
Praying	-0.04 (-0.12, 0.04)	0.32 (0.22, 0.42)	0.01 (-0.07, 0.08)	0.33 (0.23, 0.44)	0.98	0.98

CI: confidence interval; DIF: differential item functioning; ICC: intraclass correlation coefficient; SMD: standard mean difference.
[a]French-English latent factor scores. [b]Diffuse-limited/sine latent factor scores.
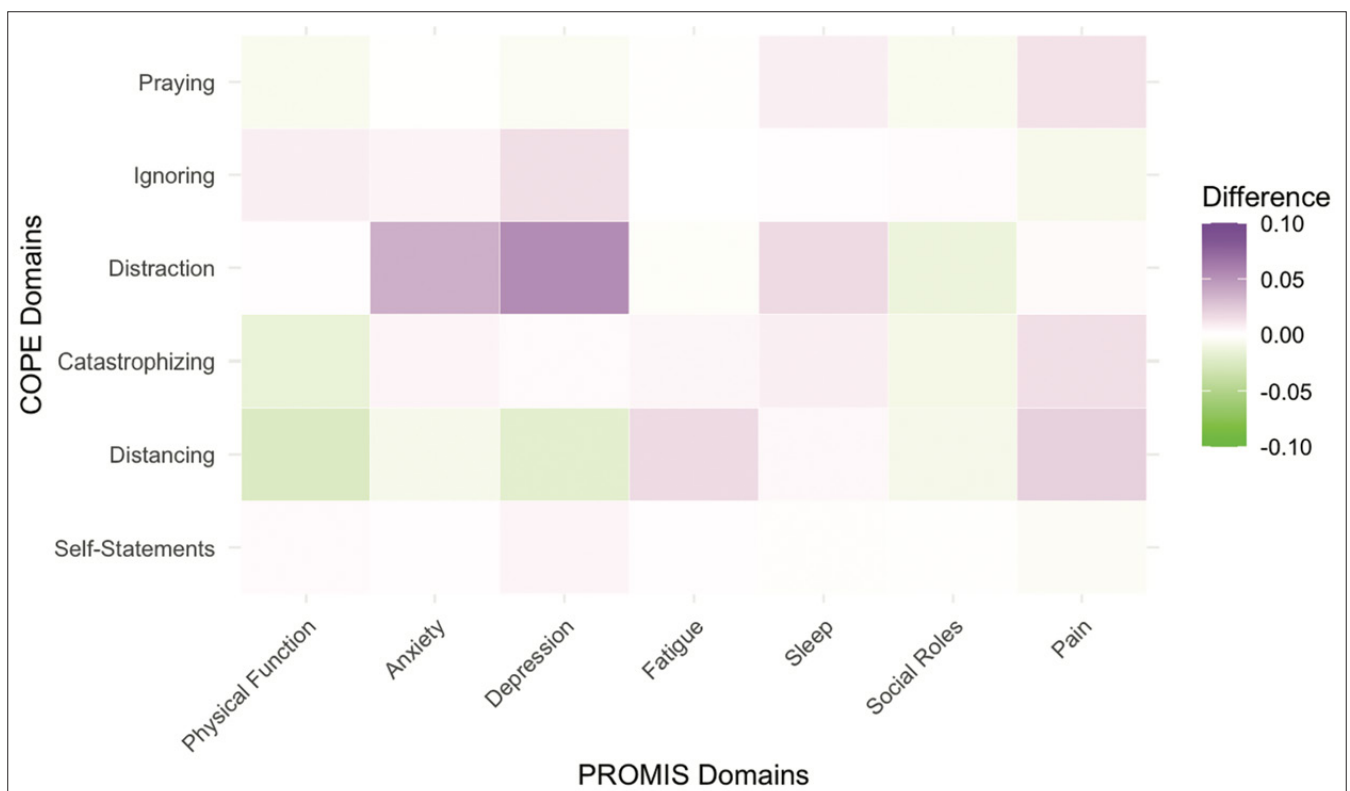
Fig. 1. Differences in correlations between unadjusted and DIF-adjusted models of Coping Strategies Questionnaire-Revised for Fatigue scores and Patient-Reported Outcomes Measurement Information System-29 domains. *DIF: differential item functioning.

liability (n=183), the ICC was 0.74 (95%CI 0.65, 0.81) for self-statements, 0.82 (95%CI 0.75, 0.86) for distancing, 0.91 (95%CI 0.88, 0.93) for catastrophising, 0.85 (95%CI 0.79, 0.88) for distraction, 0.68 (95%CI 0.58, 0.76) for ignoring and 0.90 (95%CI 0.90, 0.93) for praying.

- Participant feedback
All 3 participants who contacted us expressed concern that the CSQ-R-Fatigue includes only psychological or cognitive aspects and does not include important active coping strategies, such as taking breaks, that they use to manage fatigue in SSc. Patient research team partners who initially reviewed the items as applicable to fatigue agreed that the items do not cover the spectrum of fatigue coping strategies used by people with SSc.

Discussion
The hypothesised 6-factor structure of the CSQ-R fit well, but we found substantial DIF across multiple domains based on language (11 items) and disease subtype (10 items). We compared adjusted and non-adjusted domains and found that the differences for language were between 0.20 and 0.36 SMD for catastrophising, distancing, distraction, ignoring, and praying and for disease subtype between 0.21 and 0.33 SMD for catastrophising, ignoring and praying. Correlations between unadjusted and DIF-adjusted CSQ-R-Fatigue scores were ≥0.96 for all domains for

Table IV. Item characteristics of the Coping Strategies Questionnaire-Revised for Fatigue.

Items	Mean ± SD score [a]	Item-rest correlation [b]
Catastrophising	1.5 ± 2.6	
3. It's terrible and I feel it's never going to get any better	2.0 ± 1.8	0.67
6. It's awful and I feel that it overwhelms me	1.9 ± 1.7	0.75
7. I feel my life isn't worth living	0.9 ± 1.5	0.61
14. I worry all the time about whether it will end	1.5 ± 1.6	0.69
21. I feel I can't stand it anymore	1.5 ± 1.6	0.76
24. I feel like I can't go on	1.1 ± 1.5	0.72
Coping self-statements	3.9 ± 2.6	
4. I tell myself to be brave and carry on despite the fatigue	3.9 ± 1.7	0.63
5. I tell myself that I can overcome the fatigue	3.7 ± 1.6	0.70
11. I tell myself I can't let fatigue stand in the way of what I have to do	3.8 ± 1.6	0.62
20. Although I am fatigued, I just keep on going	4.1 ± 1.5	0.59
Distancing	1.7 ± 3.7	
1. I try to feel distant from the fatigue, almost as if the fatigue was in somebody else's body	2.2 ± 2.0	0.68
9. I try not to think of it as my body, but rather as something separate from me	1.5 ± 1.9	0.77
18. I imagine that the fatigue is outside of my body	1.4 ± 1.8	0.72
27. I pretend it's not a part of me	1.8 ± 2.0	0.73
Distraction	3.4 ± 3.4	
2. I try to think of something pleasant	3.4 ± 1.9	0.68
15. I replay in my mind pleasant experiences in the past	2.7 ± 2.0	0.67
16. I think of people I enjoy doing things with	3.2 ± 1.9	0.78
25. I think of things I enjoy doing	3.5 ± 1.9	0.79
26. I do something I enjoy, such as watching TV or listening to music	4.2 ± 1.5	0.59
Ignoring	3.0 ± 3.3	
10. I don't think about the fatigue	2.9 ± 1.8	0.49
12. I don't pay any attention to it	2.9 ± 1.8	0.74
13. I pretend it's not there	2.7 ± 1.9	0.71
19. I just go on as if nothing happened	3.4 ± 1.8	0.68
22. I ignore it	2.9 ± 1.9	0.72
Praying	1.8 ± 4.7	
8. I pray to God it won't last long	1.6 ± 2.0	0.73
17. I pray for the fatigue to stop	2.0 ± 2.1	0.69
23. I rely on my faith in God	1.9 ± 2.3	0.60

[a]On a 7-point scale, where 0=never, 3=sometimes, and 6=always. [b]Item-rest correlations were done within subscales.

language and disease subtype, and correlations with PROMIS-29 domains were within 0.05 of each other.

The factor structure and reliability of the CSQ-R-Fatigue in SSc were consistent with studies on the CSQ-R for pain (5, 14). We did not identify any previous studies, however, that have evaluated the CSQ-R for DIF by language or disease severity in pain or any other condition. The CSQ-R-Fatigue had large language and disease subtype DIF in SSc. The CSQ-R in French was previously forward and back translated (15), and this was reviewed by our team with only minor adjustments. Thus, it is not likely that translation issues led to such widespread DIF. It is more likely that DIF reflected cultural differences between how French-speakers (Quebec, France) and English-speakers (Canada, USA, UK, Australia) may think about coping from a psychologi-

cal standpoint. This is consistent with evidence of differences across cultural and racial or ethnic groups in responses to pain stimuli and descriptions of pain experiences (33, 34). We also found DIF by disease subtype, which reflects SSc disease severity. This may reflect that coping strategies are not independent of symptom severity and people with greater fatigue may be more likely to resort to some strategies than people with less fatigue. Thus, scores on some CSQ-R-Fatigue items may reflect, to some degree, severity of fatigue rather than preferences for coping strategies across individuals with similar fatigue. Our findings have important implications for research. Given the magnitude of DIF we identified, the CSQ-R-Fatigue should not be used for comparisons of domain scores across groups since these comparisons may reflect language or disease subtype differences

on some items rather than interpretable differences in coping domains. If used in correlational studies, DIF-adjusted scores could be used, albeit cautiously, to reduce DIF-related bias, and investigators might also include a covariate for language or subtype in models. It is possible that the DIF we identified is specific to SSc and fatigue, but it is also possible that this is a characteristic of the CSQ-R measure itself. Since the CSQ-R is commonly used for pain, research should examine whether this scale similarly has language-based or pain severity-based DIF that may require reconsideration of how it is used. In addition to issues related to DIF, 3 study participants contacted us and raised the question of whether the CSQ-R-Fatigue is appropriate for assessing fatigue coping in SSc. All 3 noted that the CSQ-R-Fatigue focuses on psychological coping strategies but

not active coping strategies they commonly use to deal with fatigue. Our team's patient partners agreed with this criticism. Psychological coping strategies involve cognitive and emotional responses to manage stress, whereas active coping strategies include practical actions taken to mitigate symptoms. Participants mentioned that they commonly use strategies such as sleeping, napping, or resting; prioritising tasks; taking breaks; and planning their day to accommodate fatigue. These coping strategies align with what is taught in fatigue management programs (35, 36) but are not addressed in the CSQ-R or other commonly used coping scales (6). The Illness Management Questionnaire (IMQ), which is designed to assess coping with chronic fatigue syndrome, focuses on specific strategies on how to actively cope, but has been used on a limited basis (37). One option might be to adapt the IMQ and assess its validity for coping with fatigue in SSc.

Our study has several notable strengths. It includes a multinational cohort with participants from 48 clinical sites, a large sample size, an assessment of test-retest reliability, and a comparison of measurement properties in both English- and French-speaking participants and by disease subtype. However, there are limitations to consider. First, the SPIN Cohort is a convenience sample of individuals with SSc receiving treatment at SPIN recruiting centres who can complete online measures, potentially introducing bias and limiting generalisability. Second, the DIF analysis was limited to English- and French-language versions of the CSQ-R-Fatigue and adults with SSc, which may restrict the applicability of the results to other languages. Third, we only examined uniform DIF in this study with the assumption of a constant relationship between measures and grouping variables; we did not examine nonuniform DIF. Non-uniform DIF, which involves the presence of an interaction, however, is unusual and not easily integrated into a sum-score scale format compared to uniform DIF (21).

Overall, the results of the study indicate that the CSQ-R can be adjusted for fatigue and that evidence of factor structure and reliability are similar for the CSQ-R-Fatigue and original CSQ-R in pain. We identified substantial DIF, however, by language and disease subtype, due to which comparisons on coping domains across these variables should be avoided. Furthermore, the CSQ-R-Fatigue domain scores should be interpreted cautiously if used in correlational analyses. We do not believe that DIF has been examined in the original CSQ-R in pain, and it is possible that it would exhibit similar issues. An additional concern about the CSQ-R-Fatigue questionnaire is that it assesses psychological coping strategies and does not include active coping strategies used by many individuals with SSc to manage fatigue. More research is needed on tools that capture the active coping strategies that individuals with SSc use to manage fatigue to determine the use of all coping strategies and their effectiveness.

References

1. ALLANORE Y, SIMMS R, DISTLER O *et al.*: Systemic sclerosis. *Nat Rev Dis Primer* 2015; 1: 15002. https://doi.org/10.1038/nrdp.2015.2
2. SWAIN MG: Fatigue in chronic disease. *Clin Sci* 2000; 99(1): 1-8.
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* 2011; 50(4): 762-67. https://doi.org/10.1093/rheumatology/keq310
4. FAIRLEY JL, HANSEN D, PROUDMAN S *et al.*: The burden and determinants of fatigue in incident and prevalent systemic sclerosis. *Clin Exp Rheumatol* 2024; 42(8): 1669-74. https://doi.org/10.55563/clinexprheumatol/6528od
5. FOLKMAN S, LAZARDUS RS, DUNKEL-SCHETTER C, DELONGIS A, GRUEN RJ: Dynamics of a stressful encounter: cognitive appraisal, coping, and encounter outcomes. *J Pers Soc Psychol* 1986; 50(5): 992-1003. https://doi.org/10.1037/0022-3514.50.5.992
6. MONTICONE M, FERRANTE S, GIORGI I, GALANDRA C, ROCCA B, FOTI C: The 27-item Coping Strategies Questionnaire – Revised: confirmatory factor analysis, reliability and validity in Italian-speaking subjects with chronic pain. *Pain Res Manag J Can Pain Soc* 2014; 19(3): 153-58. https://doi.org/10.1155/2014/956828
7. KATO T: Frequently used coping scales: a meta-analysis. *Stress Health J Int Soc Investig Stress* 2015; 31(4): 315-23. https://doi.org/10.1002/smi.2557
8. KWAKKENBOS L, JEWETT LR, BARON M *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(8): e003563. https://doi.org/10.1136/bmjopen-2013-003563
9. DOUGHERTY DH, KWAKKENBOS L, CARRIER ME *et al.*: The Scleroderma Patient-Centered Intervention Network Cohort: baseline clinical features and comparison with other large scleroderma cohorts. *Rheumatology* 2018; 57(9): 1623-31. https://doi.org/10.1093/rheumatology/key139
10. The Scleroderma Patient-centered Intervention Network. Accessed October 9, 2024. https://www.spinsclero.com/
11. GAGNIER JJ, LAI J, MOKKINK LB, TERWEE CB: COSMIN reporting guideline for studies on measurement properties of patient-reported outcome measures. *Qual Life Res* 2021; 30: 2197-18. https://doi.org/10.1007/s11136-021-02822-4
12. Text Recycling Research Project. Best practices for researchers. Accessed October 9, 2024. https://textrecycling.org/resources/best-practices-for-researchers/
13. VAN DEN HOOGEN F, KHANNA D, FRANSEN J *et al.*: 2013 Classification criteria for systemic sclerosis: an American College of Rheumatology/European League Against Rheumatism collaborative initiative. *Arthritis Rheum* 2013; 65(11): 2737-47. https://doi.org/10.1002/art.38098
14. RILEY JLI, ROBINSON ME: CSQ: five factors or fiction? *Clin J Pain* 1997; 13(2): 156-62. https://doi.org/10.1097/00002508-199706000-00010
15. IRACHABAL S, KOLECK M, RASCLE N, BRUCHON-SCHWEITZER M: Stratégies de coping des patients douloureux: adaptation française du coping strategies questionnaire (CSQ-F) [Pain coping strategies: French adaptation of the coping strategies questionnaire (CSQ-F)]. *Encephale* 2008; 34(1): 47-53. https://doi.org/10.1016/j.encep.2006.11.002
16. HealthMeasures. Intro to PROMIS. Accessed October 9, 2024. https://www.healthmeasures.net/explore-measurement-systems/promis/intro-to-promis
17. HINCHCLIFF M, BEAUMONT JL, THAVARAJAH K *et al.*: Validity of two new patient reported outcome measures in systemic sclerosis: the PROMIS-29 profile and the FACIT-Dyspnea. *Arthritis Care Res* 2011; 63(11): 1620-28. https://doi.org/10.1002/acr.20591
18. KWAKKENBOS L, THOMBS BD, KHANNA D *et al.*: Performance of the patient-reported outcomes measurement information system-29 in scleroderma: a scleroderma patient-centered intervention network cohort study. *Rheumatology* 2017; 56(8): 1302-11. https://doi.org/10.1093/rheumatology/kex055
19. HU LT, BENTLER PM: Cut-off criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives: *Struct Equ Modeling* 1999; 6(1): 1-55. https://doi.org/10.1080/10705519909540118
20. BROWNE MW, CUDECK R: Alternative ways of assessing model fit. *Sociol Method Res* 1992; 21(2): 230-58. https://doi.org/10.1177/0049124192021002005
21. MONTOYA AK, JEON M: MIMIC Models for

Uniform and Nonuniform DIF as Moderated Mediation Models. *Appl Psychol Meas* 2020; 44(2): 118-36.
https://doi.org/10.1177/0146621619835496

22. TERWEE CB, BOT SD, BOER MR DE *et al.*: Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007; 60(1): 34-42.
https://doi.org/10.1016/j.jclinepi.2006.03.012

23. CRONBACH LJ: Coefficient alpha and the internal structure of tests. *Psychometrika* 1951; 16(3): 297-334.
https://doi.org/10.1007/BF02310555

24. FISHER RA: Statistical methods for research workers. *In*: KOTZ S, JOHNSON NL (Eds.) Breakthroughs in Statistics: Methodology and Distribution. Springer Series in Statistics. Springer, 1992: 66-70. https://
doi.org/10.1007/978-1-4612-4380-9_6

25. KOO TK, LI MY: A guideline of selecting and reporting intraclass correlation coefficients for reliability research. *J Chiropr Med* 2016; 15(2): 155-63.
https://doi.org/10.1016/j.jcm.2016.02.012

26. Posit team. RStudio: Integrated development environment for R. Accessed October 9, 2024. http://www.posit.co/

27. KLINE RB: Beyond significance testing: statistics reform in the behavioral sciences. 2nd ed. *APA PsycNet* 2013.

https://doi.org/10.1037/14136-000

28. MUNDFROM DJ, SHAW DG, KE TL: Minimum sample size recommendations for conducting Factor Analyses. *Int J Test* 2005; 5(2): 159-68.
https://doi.org/10.1207/s15327574ijt0502_4

29. WOLF EJ, HARRINGTON KM, CLARK SL, MILLER MW: Sample size requirements for structural equation models: an evaluation of power, bias, and solution propriety. *Educ Psychol Meas* 2013; 76(6): 913-34.
https://doi.org/10.1177/0013164413495237

30. DE VET HCW, TERWEE CB, MOKKINK LB, KNOL DL: Measurement in medicine: a practical guide. 1st Ed., Cambridge University Press, 2011.
https://doi.org/10.1017/CBO9780511996214

31. POLIT DF: Getting serious about test-retest reliability: a critique of retest research and some recommendations. *Qual Life Res* 2014; 23(6): 1713-20.
https://doi.org/10.1007/s11136-014-0632-9

32. GIRAUDEAU B, MARY JY: Planning a reproducibility study: how many subjects and how many replicates per subject for an expected width of the 95 per cent confidence interval of the intraclass correlation coefficient. *Stat Med* 2001; 20(21): 3205-14.
https://doi.org/10.1002/sim.935

33. CAMPBELL CM, EDWARDS RR: Ethnic dif-

ferences in pain and pain management. *Pain Manag* 2012; 2(3): 219-30.
https://doi.org/10.2217/pmt.12.7

34. FILLINGIM RB: Individual differences in pain: understanding the mosaic that makes pain personal. *Pain* 2017; 158: S11-18.
https://doi.org/10.1097/j.pain.0000000000000775

35. HEWLETT S, ALMEIDA C, AMBLER N *et al.*: Reducing arthritis fatigue impact: two-year randomised controlled trial of cognitive behavioural approaches by rheumatology teams (RAFT). *Ann Rheum Dis* 2019; 78(4): 465-72. https://
doi.org/10.1136/annrheumdis-2018-214469

36. BACHMAIR EM, MARTIN K, AUCOTT L *et al.*: Remotely delivered cognitive behavioural and personalised exercise interventions for fatigue severity and impact in inflammatory rheumatic diseases (LIFT): a multicentre, randomised, controlled, open-label, parallel-group trial. *Lancet Rheumatol* 2022; 4(8): 534-45. https://doi.org/10.1016/S2665-9913(22)00156-4

37. RAY C, WEIR W, STEWART D, MILLER P, HYDE G: Ways of coping with chronic fatigue syndrome: development of an illness management questionnaire. *Soc Sci Med* 1993; 37(3): 385-91. https://
doi.org/10.1016/0277-9536(93)90268-9