Arthritis Care & Research
Vol. 65, No. 4, April 2013, pp 585–593
DOI 10.1002/acr.21874
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ORIGINAL ARTICLE

Measuring Illness Behavior in Patients With Systemic Sclerosis

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Objective. **Illness behaviors (cognitive, affective, and behavioral reactions) among individuals with systemic sclerosis (SSc; scleroderma) are of clinical concern due to relationships between these behaviors and physical and mental quality of life, such as pain and symptoms of depression. Self-report measures with good psychometric properties can aid in the accurate assessment of illness behavior. The Illness Behavior Questionnaire (IBQ) was designed to measure abnormal illness behaviors; however, despite its longstanding use, there is disagreement regarding its subscales. The goal of the present study was to evaluate the validity of the IBQ in a cohort of patients with SSc.**

Methods. **Patients with SSc (n = 278) completed the IBQ at enrollment into the Genetics Versus Environment in Scleroderma Outcome Study. Structural validity of previously derived factor solutions was investigated using confirmatory factor analysis. Exploratory factor analysis was utilized to derive SSc-specific subscales.**

Results. **None of the previously derived structural models were supported for SSc patients. Exploratory factor analysis supported an SSc-specific factor structure with 5 subscales. Validity analyses suggested that the subscales were generally independent of disease severity, but were correlated with other health outcomes (i.e., fatigue, pain, disability, social support, and mental health).**

Conclusion. **The proposed subscales are recommended for use in SSc, and can be utilized to capture illness behavior that may be of clinical concern.**

INTRODUCTION

Systemic sclerosis (SSc; scleroderma) is a chronic, rheumatic condition characterized by the thickening of skin and fibrosis of internal organs. It is most common among women between ages 30 and 50 years, but is relatively rare with an overall prevalence of 150 to 300 cases per million (1,2). There are 2 subtypes: limited cutaneous SSc is milder and has less severe organ involvement, and diffuse cutaneous SSc is characterized by more extensive skin and organ involvement and worse prognosis (3). Individuals with SSc report problems across multiple domains, including fatigue (4), pain (5), disability (6), sleep (7), interpersonal functioning (8), anxiety and depression (9), and more generally, physical and mental health–related quality of life (10). There is also an increasing awareness that disease severity is inadequate for discriminating patients who are at risk of poor adjustment, suggesting a need to also emphasize psychosocial variables (6).

Illness behaviors, defined as cognitive, emotional, and

Dr. Assassi's work was supported by the NIH/National Institute for Arthritis and Musculoskeletal and Skin Diseases, Center of Research Translation in Scleroderma (grants KL2-RR-024149 and K23-AR-061436). Dr. Mayes' work was supported by the NIH/National Institute for Arthritis and Musculoskeletal and Skin Diseases, Center of Research Translation in Scleroderma (grant P50-AR-054144).

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Dr. Mayes has received consultant fees, speaking fees, and/or honoraria (more than $10,000 each) from Actelion; Rush University, Chicago, Illinois; and Medtelligence (grant reviews); and has received book royalties from Oxford University Press.

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Submitted for publication April 30, 2012; accepted in revised form October 3, 2012.

Significance & Innovations

- Illness behaviors may be associated with quality of life outcomes in systemic sclerosis (SSc; scleroderma); such behaviors have been measured in other disease populations using the Illness Behavior Questionnaire (IBQ).
- The psychometric properties of the IBQ have not been evaluated in SSc.
- Results support the use of the IBQ in SSc, and that behaviors that may be most relevant to SSc quality of life are symptom bother, health worry, interpersonal functioning, other life worries, and affective inhibition.

behavioral reactions (11), can occur in response to chronic diseases such as SSc. Although illness behavior is neutral by definition, some behaviors are more adaptive than others (12). For example, concerns about health may encourage a patient with SSc to seek necessary medical help, or could lead to excessive doctor visits and anxiety. It may be helpful to divulge one's feelings about his/her disease to others, but excessive disclosure may lead to social network problems. Such extreme responses, termed abnormal illness behavior, also include actions to maintain the sick role, or a level of disability that exceeds the given pathology (12). The Illness Behavior Questionnaire (IBQ) is a widely used tool that was developed to measure these reactions (13). The IBQ contains 62 yes/no items, including all 14 items of the Whiteley Index (14). The history and development of the IBQ have been discussed elsewhere (15). The IBQ was developed in a relatively small sample (n = 100) of pain clinic patients using principal components analysis with varimax rotation, which yielded 7 subscales (Pilowsky and Spence [13] initially used items 1–52 in their analysis and removed 22 items due to poor loadings. Items 53–62 were written afterwards based on face validity and added to the subscales to improve internal consistency reliability. Thus, only 40 of the 62 items were ultimately used in the original 7 subscales). The 7 subscales are general hypochondriasis (anxious health-related concern), disease conviction (belief that a "real" disease is present), psychological versus somatic functioning (tendency to somaticize), denial (tendency to attribute life stress to physical problems), affective inhibition (inability to express personal feelings to others), affective disturbance (anxiety, depression), and irritability (anger, friction).

The IBQ has been associated with physical and psychological quality of life across a variety of conditions, such as health care utilization and disability (16), postoperative outcomes (17), health-related quality of life (18), psychopathology (19), anxiety (20), depression (21), fatigue (4,22), pain (23), and social support (24). Unfortunately, the psychometric properties of the IBQ have not been well established. The original factor structure (13) has been shown to be unstable across studies. Although internal structure is only one consideration when evaluating a measure's overall performance (25), this does suggest that the interpretability of the IBQ for other disease groups may be uncertain. Several alternate structures have been proposed (26–28), although most researchers utilize the original subscales. The original subscales have been used in patients with cancer (29), gastroesophageal reflux disease (17), myocardial infarction (30), stroke (16), lupus (31), fibromyalgia (32), osteoarthritis and rheumatoid arthritis (33), chronic fatigue syndrome, multiple sclerosis (34), and back pain (23,35).

There are several possibilities as to why the IBQ has not been well-replicated in different populations and diseases. The IBQ may have been overfactored (26), which can lead to unreliable or split factors (36). Because IBQ items are binary, poor factor specification is especially problematic given the high influence of item-level error on a factor (26). It is also plausible that previous samples were not large enough to reproduce the structure of the IBQ. The original subscales were developed using data from 100 patients, although the structure did later replicate in 1,578 pain and psychiatric patients (37). Another study (26) also used a relatively small sample (n = 200), but others reported findings from large (n = 675–1,061) samples (27,28). Another consideration is that the factorial instability is due to a disconnect between methodological and practical considerations, and the challenges inherent to measuring complex psychological constructs (25). Alternately, it has been suggested that the inconsistent factor structure of the IBQ is due to disease-specific illness behaviors unique to the physical process, treatment, and functional and social implications (15). Accordingly, some items may be more or less relevant for a given disease. For example, the disease conviction subscale may not apply to individuals with an identified pathology; it is reasonable that a person with a diagnosed disease would indeed have a strong belief that they have a disease. Thus, a new research agenda has been proposed (15,27) that entails investigating the need for disease-specific subscales to best capture the experiential, cognitive, and behavioral aspects of a given illness. Because understanding illness behavior may aid in providing total clinical care, and so that patients with maladaptive illness behaviors may be identified and offered additional intervention or referral, it would be beneficial to determine whether the IBQ can be used in patients with SSc.

The study's first aim was to evaluate the various IBQ factor structures. If the internal structure is not upheld, which could suggest problems with previously derived solutions for patients with SSc, the second aim of the study was to uncover a plausible factor structure specifically for SSc. The third aim was to establish convergent and divergent validity of the subscales derived from the best fitting model, via correlations of derived subscales with disease severity, and other quality of life variables. We predicted that the dimensions of the IBQ would have little to no correlation with disease severity, as has been shown with other psychosocial variables (6). We also predicted that greater endorsement of illness behaviors would be related to worse fatigue, pain, disability, social support, and mental health, as has been previously demonstrated (4,16,20–24).

MATERIALS AND METHODS

Participants. This investigation utilized data provided by participants from the Genetics versus Environment in Scleroderma Outcome Study (GENISOS), a prospective early-disease (within 5 years of onset) cohort study that represents collaboration among the University of Texas Health Science Center at Houston, the University of Texas Medical Branch at Galveston, and the University of Texas Health Science Center at San Antonio. Enrollment is ongoing. Data are collected annually via a clinical exam and survey packet during regular outpatient visits, and intermittently as inpatient services (as needed) at the hospitals staffed by the clinician investigators. Patients with SSc who lived within the geographic catchment area of 1 of the 3 centers were recruited from the rheumatology faculty clinics, the county hospital, and chapters of the Scleroderma Foundation (38). Participants had to be age ≥18 years.

Procedure. Baseline data from the GENISOS study were used (38). International Review Board approval was obtained at all participating institutions, including San Diego State University and University of California, San Diego, for analysis of archival data. All participants gave written informed consent. Participants received clinical examinations by the physician investigators, including evaluations of clinical manifestations (e.g., sclerodactyly, skin thickening, Raynaud's phenomenon, and gastrointestinal involvement), comorbidities, an electrocardiogram, a chest radiograph, and blood samples, as well as completion of a packet of psychosocial measures.

Measures. *IBQ.* The IBQ is a 62-item self-report measure designed to assess illness behavior (for basic item data, see Supplementary Appendix A, available in the online version of this article at http://onlinelibrary.wiley.com/doi/10.1002/acr.21874/abstract). Using a yes/no format, respondents indicate whether an item describes their experience, with "abnormal" behaviors being scored 1 point.

Modified Rodnan skin thickness score (MRSS). The MRSS (39) total score is an established indicator of skin disease severity in SSc and is calculated by measuring the extent and severity of skin thickening on 17 body surfaces by palpation on a 4-point scale (0 = uninvolved to 3 = severe thickening). Higher scores indicate greater severity.

Forced vital capacity (FVC; % predicted). Percent-predicted FVC is a validated measure for severity of SSc-related interstitial lung disease (40). It indicates the ratio of the volume of air that the study subject can forcibly exhale after a maximum inspiration to the same volume in age-, sex-, weight-, height-, and ethnicity-matched unaffected controls. All pulmonary measurements met criteria outlined by the American Thoracic Society/European Respiratory Society and were reviewed by a pulmonologist. Lower scores indicate greater severity of SSc-related interstitial lung disease.

Fatigue Severity Scale (FSS). The FSS (41) is a widely used 29-item self-report questionnaire wherein respondents rate (on a Likert scale where 1 = completely disagree and 7 = completely agree) the extent of their agreement with statements regarding their level of fatigue. It has demonstrated adequate test–retest reliability, discriminant validity, and convergent validity (41). The FSS yields an overall score and 4 factor analytically–derived subscales. The total score, in which higher total scores represent more severe global fatigue, was used in the current study. Internal consistency (α = 0.90) was good.

Medical Outcomes Study Short Form 36 (SF-36). The SF-36 (42) is a 36-item self-report health-related quality of life measure that yields 8 factor analytically–derived subscales and 2 composite scores of physical and mental health. The questions follow a variety of response formats; scoring algorithms are required for generating the subscales. It is reliable and valid for SSc (43). The bodily pain (α = 0.87) and mental health (α = 0.79) subscales were utilized. Higher scores indicate better domain-related quality of life.

Modified Health Assessment Questionnaire (MHAQ). The MHAQ (44) is an 8-item self-report index of overall disability. Respondents rate their functional ability to perform tasks on a 4-point scale (0 = without any disability to 3 = unable to do). It has been validated for use in SSc (44,45) and has been shown to have a 1-factor structure (46). Internal consistency (α = 0.91) was good in the current sample. Higher scores reflect greater disability.

Interpersonal Support Evaluation List (ISEL). The ISEL (47) is a widely used 40-item self-report measure of perceived social support, wherein respondents rate whether a statement is "probably true" or "probably false," based on their experience. The ISEL yields 4 subscales and a total score of overall support, which has been supported using confirmatory factor analysis (48). The total score was used for the current study and demonstrated good internal consistency (α = 0.87). Higher scores indicate better social support.

Data analysis. Factor analysis was conducted to achieve aims one (evaluate the various IBQ factor structures) and two (uncover a plausible factor structure specifically for SSc). Theory-driven confirmatory factor analysis (CFA) was utilized to evaluate previously derived IBQ factor structures. If CFA models do not provide sufficient fit, it is reasonable to follow up with exploratory factor analysis (EFA) (49,50). Data-driven EFA was conducted to explore alternate structures by estimating the number of underlying latent variables within the data and thus identifying SSc-specific subscales.

Because the IBQ contains binary data, traditional factor analytic techniques are inappropriate, as the assumptions of linearity and normality are violated (51). A tetrachoric correlation matrix, in which it is assumed that a normally distributed continuous latent variable underlies the "truncated" binary items, should therefore be used (51). Moreover, ordinary least squares and maximum likelihood estimation approaches are not recommended due to dependencies and systematic residuals among observed variables (52). Consequently, we used a tetrachoric correlation matrix with a weighted least squares means and variance-adjusted estimation procedure in MPlus 6.1 (53) that is robust to non-normal and nonindependent data. Internal

Table 1. Sample characteristics*

Variable	Mean ± SD or no. (%)
Age, years	49.05 ± 12.92
Age at disease onset, years	46.42 ± 13.03
MRSS	15.49 ± 11.84
FVC, % predicted	83.06 ± 21.66
Women	233 (83.8)
Men	45 (16.2)
Ethnicity	
White	135 (48.6)
Hispanic	82 (29.5)
African American	53 (19.1)
Asian	7 (2.5)
American Indian	1 (0.3)
Marital status	
Married/partnered	159 (58.2)
Never married	30 (11.0)
Divorced/separated	72 (26.4)
Widowed	12 (4.4)
Education	
Less than high school	44 (16.1)
High school diploma	143 (52.4)
Associate's degree	26 (9.5)
Bachelor's degree	38 (13.9)
Postgraduate	22 (8.1)
Family income	
<$14,999	67 (24.1)
$15,000–$29,999	65 (23.4)
$30,000–$49,999	56 (20.1)
$50,000–$99,999	51 (18.3)
≥$100,000	29 (10.4)
Disease subtype	
Diffuse	160 (57.6)
Limited	118 (42.4)

* MRSS = modified Rodnan skin thickness score; FVC = forced vital capacity.

consistency for all factors in all models was evaluated using the Kuder-Richardson 20 formula.

Evaluation of model fit. For CFA and EFA, it is recommended that samples number at least 200 (54), although samples greater than 250 are preferred for binary data (55). The current sample is near the low end of this desired range, but does meet these recommendations. Because chi-square tests may not be suitable to determine model fit, descriptive fit indices were also calculated (56). The comparative fit index (CFI) (57) and root mean square error of approximation (RMSEA) (58) were used, as other descriptors (e.g., root mean square residual [59]) are unfit for binary data (55). A model fit well if CFI values were ≥0.95 and RMSEA values were ≤0.05, based on widely accepted guidelines (55).

Exploratory analysis. Previous researchers have used different combinations of items in their EFAs of the IBQ. In the original study, items 1–52 were included in the analysis, and items 53–62 were added afterwards to increase the number of items per subscale and to improve internal consistency (13). Prior and Bond (27) used a similar strategy by including items 1–52 in their analysis and later adding items 54 and 59, based on face validity and internal consistency. Zonderman et al (28) found that the solutions for 2 analyses (the first on items 1–52 and the second on items 1–63) were identical and reported the latter solution. Main and Waddell (26) removed 25 items due to poor reliability and/or incidence, leaving 37 items for the analyses. Given the heterogeneity of approaches, and Pilowsky's (37) suggestion that the IBQ may be particularly useful as an item pool, all 62 items were analyzed in the EFA so that results were not reliant on face validity. Models with 1–7 factors were tested to reflect the various numbers of dimensions found in previous studies. A factor needed at least 3 items (preferably 4) to reduce the likelihood of overfactoring (26). In EFA, items with loadings of the strict criterion of >0.40 were used to inhibit errors in factor estimation. Cross-loadings were determined as loadings greater than half of the primary loading. Although

Table 2. Weighted least squares means and variance-adjusted exploratory factor analysis (χ^2) on 62 items of the IBQ*

Model	RMSEA	CFI	χ^2	df	*P*
1 factor	0.041	0.815	2,695.99	1,829	< 0.001
2 factor	0.031	0.897	2,250.30	1,768	< 0.001
3 factor	0.026	0.930	2,033.10	1,708	< 0.001
4 factor	0.022	0.955	1,860.94	1,649	< 0.001
5 factor	0.018	0.970	1,732.58	1,591	0.007
6 factor	0.014	0.983	1,614.28	1,534	0.075
7 factor	0.012	0.987	1,536.84	1,478	0.140

* IBQ = Illness Behavior Questionnaire; RMSEA = root mean squared error of approximation; CFI = comparative fit index.

Table 3. Summary of loadings for 5 rotated factors*

	1	2	3	4	5
IBQ3	0.632†	0.159	−0.017	0.192	0.038
IBQ16	0.988†	−0.295	0.016	−0.003	0.077
IBQ26	0.970†	−0.270	−0.011	−0.020	−0.076
IBQ41	0.727†	0.036	0.068	0.114	−0.100
IBQ50	0.613†	0.014	0.068	0.053	−0.074
IBQ1	0.180	0.796†	−0.129	−0.083	−0.040
IBQ8	0.036	−0.647†	−0.110	0.042	−0.114
IBQ21	0.040	0.613†	0.012	−0.007	0.088
IBQ24	0.128	0.533†	0.130	−0.008	−0.071
IBQ34	0.267	0.745†	−0.179	−0.076	0.033
IBQ4	−0.152	0.077	−0.647†	0.038	0.145
IBQ48	0.154	0.051	0.534†	0.205	0.030
IBQ51	−0.196	0.200	0.729†	0.043	−0.082
IBQ56	0.001	−0.021	0.743†	0.044	−0.019
IBQ61	−0.128	0.096	0.730†	0.129	−0.035
IBQ27	0.083	−0.122	0.032	0.875†	−0.011
IBQ43	−0.017	−0.217	0.099	0.735†	0.045
IBQ55	0.408	0.146	0.086	−0.705†	0.059
IBQ60	0.072	0.018	−0.008	0.790†	−0.004
IBQ22	−0.024	0.110	−0.197	0.038	−0.785†
IBQ36	0.143	−0.016	−0.011	0.214	0.551†
IBQ53	0.137	0.046	−0.191	0.229	0.650†
IBQ62	−0.105	0.041	0.006	0.165	0.768†

* IBQ = Illness Behavior Questionnaire.
† Indicates membership on a subscale.

Table 4. SSc-specific subscales from GENISOS data and membership in previous scales*

New subscale/original IBQ item	Original scale membership			
	Pilowsky and Spence, 1975 (12)	Zonderman et al, 1985 (28)	Main and Waddell, 1987 (26)	Prior and Bond, 2010 (27)
Symptom bother				
IBQ3: Does your illness interfere with your life a great deal?	Disease conviction	Illness disruption	Life disruption	Affirmation of illness
IBQ16: Are you bothered by many pains and aches?	Psychological vs. somatic functioning†	Illness disruption	Life disruption	Affirmation of illness
IBQ26: Do you experience a lot of pain with your illness?		Illness disruption		Affirmation of illness
IBQ41: Do you find that you are bothered by many different symptoms?	Disease conviction		Affective and hypochondriacal disturbance	Affirmation of illness
IBQ50: Do you often have the symptoms of a very serious disease?				Affirmation of illness
Health worry				
IBQ1: Do you worry a lot about your health?		Health worry		Concern for health
IBQ8: Is it easy for you to forget about yourself and think about all sorts of other things?		Illness disruption†	Affective and hypochondriacal disturbance†	
IBQ21: Are you afraid of illness?	General hypochondriasis	Health worry		Concern for health
IBQ24: Do you think that you worry about health more than other people?	General hypochondriasis	Health worry		Concern for health
IBQ34: Do you often worry about the possibility that you have got a serious illness?		Health worry	Affective and hypochondriacal disturbance	Concern for health
Interpersonal functioning				
IBQ4: Are you easy to get along with when you are ill?	Irritability†	Irritability†	Affective and hypochondriacal disturbance†	
IBQ48: Do you worry or fuss over small details that seem unimportant to others?		Irritability	Affective and hypochondriacal disturbance	General affective state
IBQ51: Do you find that you get angry easily?	Irritability	Irritability	Affective and hypochondriacal disturbance	General affective state
IBQ56: Are you more irritable towards other people?	Irritability	Irritability	Affective and hypochondriacal disturbance	
IBQ61: Do you often find that you lose patience with other people?	Irritability	Irritability	Affective and hypochondriacal disturbance	
Other life worries				
IBQ27: Except for your illness, do you have any problems in your life?	Denial†	Absence of life problems	Life disruption†	General affective state
IBQ43: Do you have any family problems?	Denial†	Absence of life problems	Life disruption†	General affective state
IBQ55: Would all your worries be over if you were physically healthy?	Denial	Absence of life problems†	Life disruption	
IBQ60: Do you have personal worries that are not caused by physical illness?	Denial†	Absence of life problems	Life disruption†	
Affective inhibition				
IBQ22: Can you express your personal feelings easily to other people?	Affective inhibition†	Affective inhibition†	Social inhibition†	
IBQ36: When you are angry, do you tend to bottle up your feelings?	Affective inhibition	Affective inhibition	Social inhibition	
IBQ53: Do you prefer to keep your feelings to yourself?	Affective inhibition	Affective inhibition	Social inhibition	
IBQ62: Is it hard for you to show people your personal feelings?	Affective inhibition	Affective inhibition	Social Inhibition	

* SSc = systemic sclerosis (scleroderma); GENISOS = Genetics Versus Environment in Scleroderma Outcome Study; IBQ = Illness Behavior Questionnaire.
† Reverse-scored in the respective analyses for each study listed.

underfactoring (i.e., including too few factors in a model) has not typically been a criticism of the IBQ, it can lead to problems, such as the combination of multiple factors (36); therefore, the pattern matrix was also inspected for interpretability. Items derived from the factor analysis were further evaluated for their contribution to the internal consistency of their subscale. Based on recommendations for decreasing redundancy among subscale items, items were retained if their removal from a subscale resulted in decreased internal consistency, and eliminated if internal consistency was unchanged upon removal (49). Subscale intercorrelations were then evaluated; models with intercorrelations with high multicollinearity ($r = >0.7$) were considered unsuitable.

Table 5. Associations between IBQ scales for SSc and disease-related outcomes*

IBQ	Modified Rodnan skin score	Forced vital capacity	Fatigue	Pain	Disability	Social support	Mental health
Symptom bother	0.14†	−0.05	0.42‡	−0.57‡	0.42‡	−0.15†	−0.31‡
Health worry	0.11	−0.01	0.10	−0.17§	0.21§	−0.25‡	−0.42‡
Interpersonal functioning	−0.05	0.06	0.22‡	−0.14†	0.16§	−0.19§	−0.33‡
Other life worries	−0.09	0.00	0.08	−0.00	0.00	−0.13†	−0.21§
Affective inhibition	0.05	−0.14†	0.09	−0.15†	0.15†	−0.32‡	−0.30‡

* IBQ = Illness Behavior Questionnaire; SSc = systemic sclerosis (scleroderma).
† $P < 0.05$.
‡ $P < 0.001$.
§ $P < 0.01$.

RESULTS

Descriptive characteristics are available in Table 1. Skin thickening ($t[274] = -13.79$; mean ± SD diffuse 22.03 ± 11.10, mean ± SD limited 6.74 ± 5.39) and FVC ($t[262] = 2.65$; diffuse mean ± SD 80.09 ± 20.71, mean ± SD limited 87.16 ± 22.36) indicated greater disease severity in the diffuse subtype ($P < 0.001$ and $P < 0.01$, respectively).

CFAs of original and alternate models. First, CFA was used to examine the model fit of the 7 dimensions comprised of 40 items, as suggested by Pilowsky and Spence (13). Internal consistencies were poor (0.200 – 0.697); only affective disturbance (0.759) was reliable. Model fit was poor statistically ($\chi^2[719] = 1{,}048.04$, $P < 0.001$; n = 278) and descriptively (CFI = 0.893 and RMSEA = 0.041). Interfactor correlations ranged from |0.20–1.06|, suggesting high redundancy among factors (correlation coefficients among factors that are >1 indicate that the factors are indistinguishable; therefore, model fit is unacceptable). Because internal consistency and solution were both poor, most dimensions were inadmissible, thus alternate structures were considered.

The 6 dimensions comprised of 47 IBQ items as suggested by Zonderman et al (28) were tested first. Internal consistency was better (0.632 – 0.796). Model fit was poor statistically ($\chi^2[1{,}019] = 1{,}538.46$, $P < 0.001$; n = 278) and descriptively (CFI = 0.871 and RMSEA = 0.043). Interfactor correlations ranged from |0.21–0.81|.

The 6 dimensions comprised of 33 IBQ items as suggested by Main and Waddell (26) were tested next. Internal consistencies ranged from 0.566 to 0.814. Model fit was poor statistically ($\chi^2[492] = 1{,}093.12$, $P < 0.001$; n = 278) and descriptively (CFI = 0.782 and RMSEA = 0.066). Interfactor correlations ranged from |0.32–0.72|.

Finally, the 3 dimensions comprised of 31 IBQ items as suggested by Prior and Bond (27) were tested. Internal consistency was good (0.733–0.805); however, model fit was poor statistically ($\chi^2[431] = 804.70$, $P < 0.001$; n = 278) and descriptively (CFI = 0.893 and RMSEA = 0.056). Interfactor correlations ranged from |0.69–0.74|.

Exploratory analysis of IBQ items. In the exploratory analysis, raw data (not reverse scored) were analyzed. Because none of the models fit adequately, EFA was utilized to determine if a better model could be derived (Table 2). The 4-factor model was the first to meet the descriptive fit criteria, therefore models 4–7 were evaluated for interpretability. Inspection of the simple structure of these models showed an adequate number of items on the 4- and 5-factor models. For the 6- and 7-factor models, several dimensions yielded only 2 to 3 items. Given the issues of over-factoring (26), these models were not evaluated further.

Both the 4- and 5-factor models were reviewed on the basis of simple structure and interpretability. Both contained 3 identical factors. However, the largest factor from the 4-factor model was split into 2 meaningful factors in the 5-factor model, suggesting that the 4-factor model was underfactored. At this point, 33 items were removed due to insufficient loadings or cross-loadings. Each factor was then further refined based on internal consistency, as described above. The final solution used 23 items. The factor loadings are shown in Table 3.

SSc-specific subscales of the IBQ. Table 4 describes the subscales, and items shared with subscales from previous solutions. Intercorrelations among the SSc subscales (range for r = 0.00–0.38) were reasonable.

Symptom bother. Three items that loaded onto this subscale were removed as they did not improve internal consistency. Thus, the first subscale retained the 5 best items out of the 8 that met the interpretability criteria. Higher scores indicate greater intensity and life interference of disease-related symptoms. Internal consistency (0.778) was adequate.

Health worry. One item that loaded onto the second subscale was removed as it did not improve internal consistency. Thus, the second subscale retained the 5 best items out of the 6 that met interpretability criteria. Higher scores indicate that a respondent is more preoccupied with health in general. Internal consistency (0.725) was adequate.

Interpersonal functioning. Two items that loaded onto the third subscale were removed as they did not improve internal consistency. Thus, the third subscale retained the 5 best items out of the 7 that met interpretability criteria. Higher scores indicate more interpersonal problems. Internal consistency (0.720) was adequate.

Other life worries. Four items loaded onto the fourth subscale. Higher scores indicate a greater number of non-illness problems. Internal consistency (0.715) was adequate.

Affective inhibition. Four items loaded onto the fifth subscale. Higher scores reflect greater difficulty expressing emotion to others. Internal consistency (0.662) for this subscale was weaker.

Relationships of subscales to health outcomes. Correlations between the subscales and other measures were performed to establish convergent and divergent validity (Table 5). As predicted, the proposed subscales were not generally associated with disease severity. As predicted, the subscales were related to fatigue, pain, disability, social support, and mental health in the expected directions. Higher scores on the subscales were associated with worse outcomes, with stronger relationships among related domains (e.g., relationships between symptom bother and pain, or between affective inhibition and social support).

DISCUSSION

The current study expands on efforts to create a useful measure that characterizes illness behaviors by examining the psychometric properties of the IBQ (13) in patients with SSc. None of the previous solutions adequately fit data from patients with SSc. Failing to replicate the factor structure of a measure is one element that may call its performance into question; thus, the approach became exploratory. The physiologic and psychological aspects of specific diseases vary widely, thus it is reasonable for different diseases to have different factor structures and resultant subscales for the IBQ (15,55). Therefore, only those items that were meaningful for SSc patients were included to ensure sharper measurement of the relevant aspects of illness behavior for SSc. On the basis of a number of statistical and theoretical decisions, an SSc-specific structure was derived. The proposed subscales comprised illness-related (symptom bother, health worry), social (interpersonal functioning), and affective (other life worries, affective inhibition) domains.

Internal consistency of the subscales was acceptable, although affective inhibition was lower but satisfactory, given the small number of items and exploratory nature of the study (60). Although higher internal consistencies have been reported for longer subscales (27), this is unsurprising given that items were added after factor analysis based on face validity, with the specific intention of increasing internal reliability. Shorter forms that are sufficiently valid and reliable to achieve measurement objectives are generally preferable in clinical contexts.

The validity analyses suggested that SSc-specific subscale scores were generally unrelated to skin thickness and pulmonary function. This suggests that disease severity only partially explains illness behavior. Fatigue, pain, disability, social support, and mental health were generally associated with the subscales, such that greater endorsement of the illness behavior domains was predictive of poorer outcomes. Taken together, these findings suggest that these subscales provide an acceptable assessment of illness behavior in SSc. However, score interpretation should be considered in the larger context of a patient's current physical status and psychological comorbidities.

Given the rarity of SSc, a notable strength of the current study is the large, representative sample of patients. However, there were some limitations. Only cross-sectional data were utilized. The sample size was on the low end of recommendations for latent variable analyses. Despite these limitations, this study provides preliminary support for the utility of the IBQ for patients with SSc. Future work should focus on confirming this factor structure in a different sample of patients with SSc, and on comparing the measurement model between diffuse and limited subtypes. Additionally, researchers and clinicians should begin building more integrative models of illness behavior, with attention to the physical, psychological, and social aspects of SSc to enhance total patient care. Within such a framework, clinicians will be better equipped to identify at-risk patients to implement appropriate interventions to target problematic illness behaviors (61), underscoring the need for a reliable and valid screening tool.

In conclusion, this study evaluated the factorial validity of the IBQ in a sample of patients with SSc derived from the GENISOS cohort. The original factor structure of the IBQ was not supported, providing one piece of evidence that may call the factor structure into question. Therefore, an SSc-specific factor structure was uncovered, which demonstrated convergent and divergent validity. These subscales offer clinicians a relatively concise way to identify patients who may benefit from additional intervention.

AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be submitted for publication. Dr. Malcarne had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study conception and design. Merz, Malcarne, Roesch, Sharif, Draeger, McNearney, Assassi, Mayes.

Acquisition of data. Sharif, Harper, Draeger, Gonzalez, Nair, Assassi, Mayes.

Analysis and interpretation of data. Merz, Malcarne, Roesch, Draeger, McNearney, Assassi.

ADDITIONAL DISCLOSURE

Dr. McNearney is currently employed by Eli Lilly but was not at the time of the study. Eli Lilly had no financial interest in this project and had no input in the design, content, data collection, or analysis, and had no role in the writing or approval of this article, with all opinions and conclusions expressed herein those of the authors.

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