



# Validation of Turkish version of the Scleroderma Health Assessment Questionnaire

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Received: 14 November 2018 / Revised: 10 February 2019 / Accepted: 24 February 2019 / Published online: 6 March 2019  
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## Abstract

**Objectives** The Scleroderma Health Assessment Questionnaire (SHAQ) is a functional scale which consists of five scleroderma-specific items (overall disease severity, Raynaud's phenomenon, digital ulcers, respiratory and intestinal involvement) in addition to Health Assessment Questionnaire Disability Index (HAQ-DI). The objective of this study was to perform an adaptation and validation of a Turkish version of the SHAQ.

**Method** We validated psychometric properties of the scale with 70 consecutive systemic sclerosis (SSc) patients, who fulfilled the 2013 ACR/EULAR classification criteria for SSc. We evaluated test–retest reliability with the intraclass correlation coefficient (ICC), discriminant validity by stratifying patients according to organ involvements and disease subtypes, and convergent validity by testing the correlation between SHAQ and related components of Short Form 36 version 2 (SF-36v2). Internal consistency of the questionnaire was evaluated by Cronbach's alpha coefficient.

**Results** The SHAQ-global, visual analogue scales (VAS) of pulmonary, digital ulcer, and Raynaud's phenomenon were significantly correlated with the physical component score of the SF-36v2 ( $r = -0.274$ ,  $r = -0.295$ ,  $r = -0.326$ ,  $r = -0.308$ ,  $p < 0.05$ , respectively) for the convergent validity. The instruments could not discriminate between disease subtypes, except the digital ulcer VAS which was significantly higher in patients with dcSSc ( $1.00 \pm 0.93$  vs  $0.55 \pm 0.88$ ,  $p = 0.026$ ) for the discriminant validity. The HAQ-DI, SHAQ-global, digital ulcer VAS, and pulmonary VAS showed moderate correlation with an increase in the number of the organs involved ( $r = 0.319$ ,  $r = 0.329$ ,  $r = 0.341$ ,  $r = 0.278$ ,  $p < 0.05$ , respectively). We demonstrated high reproducibility for HAQ-DI (ICC = 0.962, 95% confidence interval = 0.934–0.978) and the other items of SHAQ. The overall internal consistency of the SHAQ was satisfactory (Cronbach's alpha = 0.953).

**Conclusions** The Turkish version of the SHAQ met the requirements of validity and reproducibility.

**Keywords** Scleroderma Health Assessment Questionnaire Disability Index · Systemic sclerosis · Turkish · Validation

## Introduction

Systemic sclerosis (SSc) is a chronic connective tissue disorder characterized by three cardinal features: vasculopathy, autoimmunity, and fibrosis [1] with higher mortality rates than expected from the general population and also compared with some of the other rheumatic diseases [2, 3]. The multisystem

involvement associated with SSc has severe physical and psychosocial impact on affected patients' quality of life. One of the challenges in management of SSc is the scarcity of tools to evaluate disease activity, predict the outcome, and measure changes during the course of the disease despite functional disability being a significant consequence of SSc. Previously, there have been initiatives to improve the scales used for measuring functional disability in SSc, because of its relatively greater impact in this patient group [4]. The Health Assessment Questionnaire (HAQ) is one of the scales validated to assess disability and function in SSc [5]. The Health Assessment Questionnaire Disability Index (HAQ-DI) was originally developed for rheumatoid arthritis to evaluate patients' day-to-day capabilities in their normal environment [6]. After Poole and Steen used HAQ-DI in SSc patients for the first time, Steen and Medsger further developed the scale by

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adding five scleroderma-specific, patient-generated items as a measure of overall disease severity [7]. These SSc-specific items included assessment of Raynaud's phenomenon (RP), digital ulcers, and respiratory and intestinal problems. The Scleroderma-HAQ (SHAQ) has been validated for SSc in English and also translated into several languages and adapted cross-culturally in different countries [8–10]. SHAQ has additional utility and content validity compared with HAQ since it includes questions for SSc-specific manifestations [11]. There have been suggestions for more structured measures, which would be created by pooling together the HAQ-DI and patient-generated visual analogue scale (VAS) tools [12].

Currently, there is no specific questionnaire to investigate functional disability and disease-related quality of life measures in Turkish SSc patients. The aim of this study was to adapt and validate the Turkish version of SHAQ and evaluate its internal consistency, reliability, and validity.

## Material and methods

### Patients

Seventy consecutive patients who fulfilled the 2013 American College of Rheumatology/European League against Rheumatism (ACR/EULAR) SSc classification criteria were enrolled in the study beginning in March 2017 until December 2017 [13]. Patients with a good command of spoken Turkish were included in the study because they would need to understand the abstract concept of a VAS. The study was approved by the local ethics committee (Kocaeli University School of Medicine Ethics Committee, Kocaeli, TURKEY) as stated in the protocol of research number (GOKAEK 2017/347).

During the clinical examinations, on the day of routine follow-up, the patients were asked whether they wanted to participate in the study. The content and the aims of the study were explained to each of the patients. After the participants gave consent, they were asked to fill in the self-administered questionnaires, the Turkish versions of the SHAQ and the SF-36v2. Mean duration required to fill in both of the questionnaires was approximately 30 min. Fifty-one of the patients re-filled the SHAQ questionnaire 2 weeks after the first visit, with the assumption that their disease-related condition remained stable.

### Clinical assessment

Clinical and laboratory data were obtained by clinical examination and from the medical records of the patients. The following data were recorded: gender, age, symptoms within the last week, organ involvement, the type of SSc (diffuse or limited), and duration of the disease. The disease duration was defined from the first onset of non-RP symptom related

to SSc. The patients were classified as diffuse cutaneous SSc (dcSSc) or limited cutaneous SSc (lcSSc) according to the distribution of skin involvement by LeRoy's criteria [14]. The extent of the skin involvement was evaluated by using the modified Rodnan skin score (mRSS) by the same rheumatologist [15].

The following definitions were used to determine the specific visceral involvements: gastrointestinal involvement indicated by distal esophageal hypomotility or aperistalsis documented by either radiographic or manometric study, gastrointestinal symptoms defined by heartburn, dysphagia, episodes of pseudo-occlusion, anal incontinence, diarrhea, and/or fecal incontinence; pulmonary involvement evidenced by ground-glass, honeycombing, or traction bronchiectasis on thoracic high-resolution computed tomography and pulmonary function test showing restrictive pulmonary disease pattern characterized by forced vital capacity (FVC) of < 70% of predicted normal and/or carbon monoxide diffusing capacity (DLCO) of < 80% of predicted normal; musculoskeletal involvement defined as restriction of the skeletal motion due to myalgia, muscular weakness, peripheral arthralgia, arthritis, calcinosis, or contracture of the joints; pulmonary hypertension indicated by systolic arterial pulmonary pressure of greater than 45 mmHg measured by the tricuspid regurgitant jet flow on echocardiography because of the strong correlation between the right heart catheterization and this estimated cut-off level [16] and/or mean pulmonary artery pressure at rest of  $\geq 25$  mmHg and pulmonary wedge pressure of  $\leq 15$  mmHg on right heart catheterization; and heart involvement defined by the presence of arrhythmia and/or pericarditis and/or myocardial dysfunction observed on Doppler echocardiogram. Raynaud's phenomenon crisis and digital lesions defined by pitting scars, digital tip ulceration, and/or digital gangrene on the examination during the past week were recorded.

### Definitions of the scales

#### HAQ-DI

The standard disability index of HAQ-DI is a self-administered questionnaire, which consists of 20 items divided into eight domains of dressing and grooming, arising, eating, walking, hygiene, reach, grip, and activities. The answers for each question ranges between "without any difficulty (0)," "with some difficulty (1)," "with much difficulty (2)," and "unable to do (3)." The highest score of the questions in each domain are added and then divided by 8 to determine the HAQ-DI. When the patient needs any devices or the help of somebody for activities in any of the domains, it is automatically scored 2. The Turkish version of the HAQ-DI has already been validated and adapted [17].

## Scleroderma-HAQ

The SHAQ was constructed by addition of the five following questions related to symptoms: “In the past week, how much have your—Raynaud’s phenomenon, digital ulcers, gastrointestinal symptoms, lung symptoms, and overall scleroderma symptoms—interfered with your activity?” The answer is marked on a VAS with a length of 15 cm. The ends of the line are “does not interfere” and “very severe limitations.” The final VAS score is calculated by multiplying the value by 0.2. The score ranges from 0 to 3 representing a minimum to maximum limitation, respectively. The value of each VAS score is reported separately [7]. The SHAQ-global score which was calculated by adding five SSc-related VAS to eight HAQ-DI domains and dividing the sum by 13 [8 HAQ-DI domains + 5 SSc VAS]/13] was  $0.8 \pm 0.6$  [12].

## Short Form 36 version 2

Short Form 36 version 2 (SF-36v2) is a self-administered questionnaire consisting of 36 items that evaluate the quality of life in eight areas: physical functions, physical role, bodily pain, general health, vitality, social functions, emotional role, and mental health. Each area was assessed by its specific score ranging from 0 to 100, from poorer to better health status. The four physical health scales are summarized to calculate the physical component score (PCS) and the four mental health scales for the mental component score (MCS). The Turkish version of SF-36v2 was validated and updated previously for use in musculoskeletal research [18].

## Translation-adaptation

We employed the Turkish version of HAQ-DI, which is currently used in rheumatic diseases. We translated the five additional VAS according to the guidelines for the process of cross-cultural adaptation of self-report measures [19, 20]. The initial English version was first translated into Turkish by two translators one of whom was informed and the other was uninformed about the objective of the questionnaire. Both of the translators and a physician who participated in the study worked on a synthesis of these two translations. They concluded on a common translation. The back translation was carried out by two uninformed translators with the source language (English) as their mother tongue. After this process, the translations were reviewed by a group of experts consisting of a methodologist and an experienced rheumatologist to reach a consensus on any discrepancy. Subsequently, the original and back translation versions were compared, and a final version was rewritten after having reached a consensus.

According to the consensus, the Turkish version of the SHAQ was clear and understandable enough and required

no additional correction. However, the patients were mostly unaware of the term “Raynaud’s phenomenon,” because it was derived from a foreign proper name. We decided to give a brief explanation of the terms which the patients had difficulty in understanding, being careful to adhere to the rules of self-administered questionnaires.

## Validation of the psychometric properties of the Turkish SHAQ

Each patient who participated was asked to fill in the SF-36v2 and HAQ-DI questionnaires and in addition to answer the five SSc-specific SHAQ VAS questions. The patients were supervised by a trainee (F.K.) who provided a limited but adequate explanation about the questionnaires.

## Convergent validity

Convergent validity was assessed by the analysis of the association between the HAQ-DI and S-VAS scores with SF-36v2 health-related quality of life domains. We assumed that HAQ-DI scores were expected to be more closely correlated with the physical items of SF-36v2 than the mental items. The five VASs of the SHAQ would correlate more strongly with the physical items of SF-36v2 compared to the mental items of SF-36v2 and the SHAQ VAS scores were expected to show high correlation with HAQ-DI.

## Discriminant validity

The values of SF-36v2 physical items, HAQ-DI, and SHAQ VAS scores were compared between patients with and without SSc-related organ involvements as defined above. Close associations were expected between these scales and the number of specific organs involved. The SF-36v2 PCS and HAQ-DI were expected to be worse in diffuse SSc subtype. However, the SHAQ VAS was not expected to be better than HAQ-DI in discriminating between limited cutaneous SSc (lcSSc) and diffuse cutaneous SSc (dcSSc) [11].

## Test-retest reliability

We evaluated the test-retest reliability of both the HAQ-DI and SHAQ by administering it at baseline and reapplying after 2 weeks of the first visit. A trained interviewer initially administered the HAQ-DI, SF-36v2, and the five questions of the SHAQ to all patients. The re-administration was carried out during face-to-face-interview in 51 patients in the outpatient clinic by the same interviewer. We measured the strength of agreement between repeated measures using the intraclass correlation coefficient (ICC), with ICC of 0.7 or greater representing a high level of agreement [21].

## Internal consistency

Internal consistency of the questionnaire was evaluated by Cronbach's alpha coefficient.

## Statistical analysis

Descriptive statistics for clinical and demographic characteristics of the patients were presented as frequency and percentage (%) for categorical variables and mean with standard deviation (mean  $\pm$  SD) or median with interquartile range (median [IQR = Q3–Q1]) according to the distribution of the continuous variables. Demographic and clinical features were compared between SSc subgroups using the Mann–Whitney *U* test (Wilcoxon rank sum test), independent sample *T* test, or chi-square test as appropriate. The Spearman's rank correlation coefficients were used to examine the degree of associations between the SHAQ scores with the HAQ-DI, SF-36v2, VAS scores, and the number of organs involved. The test–retest reliability was assessed by the intraclass correlation coefficient (ICC). Correlations less than or equal to 0.29 were considered to be low; between 0.30 and 0.49, as moderate, and greater than or equal to 0.50, as high [22]. Statistical analyses were performed using “SPSS version 20.0 software package” (IBM Inc., Chicago, IL, USA). Two-sided *p* values less than 0.05 were considered statistically significant ( $p < 0.05$ ).

## Results

A total of 70 consecutive patients filled in the HAQ-DI, SF-36v2, and SHAQ questionnaires. Demographic data and disease characteristics are shown in Table 1. Most of the patients were unemployed (81.4%). However, ten of them were manual workers, one tradesman, one accountant, and one academician (data not shown).

## Convergent validity

The HAQ-DI, SHAQ-global, and SSc-related five VASs exhibited correlation with SF-36v2 physical and with mental-related scores (Table 2). There was mild to a moderate negative correlation between the SF-36v2 physical component score and SHAQ-global, RP-VAS, digital ulcer VAS, and pulmonary VAS scores ( $r = -0.274$ ,  $r = -0.295$ ,  $r = 0.326$ , and  $r = 0.308$ , respectively). The SF-36v2 mental component score correlated negatively with HAQ-DI, SHAQ-global, and overall disease severity VAS score ( $r = -0.374$ ,  $r = -0.362$ , and  $r = -0.285$ , respectively). All correlations were statistically significant at the 0.01 and 0.05 levels (see Table 2).

**Table 1** Demographic data and clinical characteristics of SSc patients

Variables	N, %
Sex (women)	59 (84.3%)
Age, years	57.2 $\pm$ 13.7
Diffuse cutaneous SSc (dcSSc)	29 (41.4%)
Disease duration, years	8.7 $\pm$ 4.4
Raynaud's phenomenon	69 (98.6%)
Raynaud's attack <sup>a</sup>	20 (28.6%)
Sclerodactyly	37 (52.9%)
Digital ulcers	17 (24.3%)
New onset digital ulcers <sup>a</sup>	9 (12.9%)
Pitting scars	35 (52.3%)
Telangiectasia	42 (60%)
Musculoskeletal symptoms	22 (31.4%)
Calcinosis	3 (4.3%)
Hand contracture	7 (10%)
Inflammatory arthritis	7 (10%)
Digestive symptoms <sup>a</sup>	33 (47.1%)
Pulmonary involvement	34 (48.3%)
Pulmonary hypertension	4 (5.7%)
FVC, (%)	89.4 $\pm$ 17.4
DLCO, (%)	63.8 $\pm$ 16.7
Antinuclear antibodies (positive)	63 (96.9%)
Anticentromere antibodies (positive)	28 (42.2%)
Antitopoisomerase I antibodies (positive)	26 (39.1%)
mRSS	14.6 $\pm$ 8.8
HAQ-DI (0–3)	0.88 [1.16–0.25]
SHAQ-Raynaud's phenomenon VAS (0–3)	0.70 [1.20–0.00]
SHAQ-digital ulcer VAS (0–3)	0.20 [1.55–0.00]
SHAQ-digestive VAS (0–3)	0.20 [0.60–0.00]
SHAQ-pulmonary VAS (0–3)	0.20 [1.00–0.00]
SHAQ-overall disease severity VAS (0–3)	1.00 [2.00–0.05]
SHAQ-global (0–3)	0.72 [1.15–0.36]
SF-36v2 PCS	38.5 $\pm$ 11.8
SF-36v2 MCS	29.3 $\pm$ 16.8

FVC forced vital capacity, DLCO carbon monoxide diffusing capacity, mRSS modified Rodnan skin score, HAQ-DI Health Assessment Questionnaire Disability Index, SHAQ Scleroderma Health Assessment Questionnaire, VAS visual analogue scale, SF-36v2 PCS Short Form 36 version 2 physical component score, SF-36v2 MCS Short Form 36 version 2 mental component score

<sup>a</sup> Symptoms are evaluated as positive if they occurred in the last week

## Discriminant validity

The mean digital ulcer VAS score in patients with dcSSc was higher than in those with limited disease (1.00  $\pm$  0.93 vs 0.55  $\pm$  0.88,  $p = 0.026$ ). The HAQ-DI, SHAQ-global, and the other SSc-related VAS scores did not differ between the SSc subtypes (Table 3). The HAQ-DI, SHAQ-global, digital ulcer VAS, and pulmonary VAS scores correlated positively with

**Table 2** Correlation of HAQ-DI and SSc-specific VASs with SF-36v2

Correlation coefficient							
	HAQ-DI	SHAQ-global	RP-VAS	DU-VAS	Digestive VAS	Pulmonary VAS	Overall disease severity VAS
Physical functions	−0.452**	−0.431**	−0.012	−0.127	0.001	−0.188	−0.323**
Role limitation-physical	−0.397**	−0.376**	0.015	−0.165	−0.004	−0.228	−0.380**
Body pain	−0.226	−0.312*	−0.185	−0.324**	−0.264*	−0.275*	−0.378**
General health	−0.150	−0.198	−0.149	−0.169	−0.084	−0.137	−0.235
Vitality	−0.113	−0.157	−0.024	−0.181	−0.112	−0.218	−0.265*
Social functions	−0.246*	−0.365**	−0.328**	−0.477**	−0.247*	−0.323**	−0.303*
Role limitation-emotional	−0.217	−0.270*	−0.196	−0.187	−0.134	−0.282*	−0.180
Mental health	−0.176	−0.280*	−0.326**	−0.333**	−0.296*	−0.298*	−0.403**
SF-36v2 MCS	−0.374**	−0.362**	0.052	−0.185	0.001	−0.140	−0.285*
SF-36v2 PCS	−0.183	−0.274*	−0.295*	−0.326**	−0.216	−0.308*	−0.197

\*\*Correlation is significant at the 0.01 level (two-tailed); \*correlation is significant at the 0.05 level (two-tailed)

HAQ-DI Health Assessment Questionnaire Disability Index, SHAQ Scleroderma Health Assessment Questionnaire, RP Raynaud’s phenomenon, VAS visual analogue scale, DU digital ulcer, SF-36v2 PCS Short Form 36 version 2 physical component score, SF-36v2 MCS Short Form 36 version 2 mental component score

the number of visceral involvements ( $r = 0.319$ ,  $r = 0.341$ , and  $r = 0.278$ , respectively) (Table 4). Among the scleroderma-related VAS scores, digital ulcer VAS scores correlated moderately with mRSS ( $r = 0.364$ ,  $p = 0.018$ ).

**Reproducibility**

The test–retest procedure, performed in 51 patients (47 for SHAQ-global), demonstrated small variations in the HAQ-DI score, global SHAQ score, and in the SSc-related VAS scores. Reproducibility was high for each of the five VAS, with few differences between the

**Table 3** The comparison of health-related quality of life and functional status (disability) measures between disease subsets

	dcSSc (n = 29)	lcSSc (n = 41)	p
HAQ-DI	0.92 ± 0.71	0.78 ± 0.68	0.376
SHAQ-global	0.72 ± 0.53	0.90 ± 0.64	0.300
Raynaud’s phenomenon VAS	0.95 ± 0.84	0.67 ± 0.64	0.202
Digital ulcer VAS	1.00 ± 0.93	0.55 ± 0.88	0.026
Digestive VAS	0.52 ± 0.65	0.40 ± 0.63	0.275
Pulmonary VAS	0.79 ± 0.77	0.48 ± 0.71	0.063
Overall disease severity VAS	1.16 ± 0.92	1.04 ± 0.94	0.572
SF-36v2 PCS	30 ± 15.5	28.8 ± 17.8	0.267
SF-36v2 MCS	40.2 ± 10.4	37.3 ± 12.7	0.830

HAQ-DI Health Assessment Questionnaire Disability Index, SHAQ Scleroderma Health Assessment Questionnaire, VAS visual analogue scale, SF-36v2 PCS Short Form 36 version 2 physical component score, SF-36v2 MCS Short Form 36 version 2 mental component score, dcSSc diffuse cutaneous systemic sclerosis, lcSSc limited cutaneous systemic sclerosis

various scales (Table 5). The overall internal consistency of the HAQ-S was satisfactory with a Cronbach’s alpha value of 0.953.

**Discussion**

Self-administered questionnaires to evaluate consequences on daily life activities provide a simple and useful standardized tool to evaluate each patient’s perspective of disease severity. This facilitates patient to patient comparison and allows for assessment of a single patient’s experience of disease progression, if applied over time. The present study confirms the value of the SHAQ and demonstrates its psychometric properties in Turkish SSc patients. This study is the first validation of the SHAQ in Turkish. It provides a comprehensive outcome measure which evaluates a large group of SSc patients globally with HAQ-DI and specifically with an additional five SSc-specific VAS scores.

The validation of the Turkish SHAQ version required no change for cross-cultural adaptation, consistent with the previous reports [9]. In order to adhere to the rules of the self-administered questionnaire, a trained interviewer accompanied the patients and gave a brief explanation to avoid any bias. Although the questionnaire was clear and understandable, patients required the explanation in order to be able to use the VAS and understand what the words “Raynaud’s phenomenon” meant to them as the patients were mostly unaware of the phrase “Raynaud’s phenomenon.” These difficulties might be related to the demographics of our study population, the majority of whom were unemployed women with low socio-cultural status. Most of them were administered this kind of a questionnaire or had heard the medical

**Table 4** Correlation of the measures with the number of visceral involvements

Instruments/scales	Correlation with number of visceral involvements	<i>p</i>
HAQ-DI	0.319**	0.007
SHAQ-global	0.329**	0.006
Raynaud's phenomenon VAS	0.220	0.072
Digital ulcer VAS	0.341**	0.004
Digestive VAS	0.119	0.333
Pulmonary VAS	0.278*	0.021
Overall disease severity VAS	0.166	0.176

\*\*Correlation is significant at the 0.01 level (2-tailed); \*correlation is significant at the 0.05 level (2-tailed)

HAQ-DI Health Assessment Questionnaire Disability Index, SHAQ Scleroderma Health Assessment Questionnaire, VAS visual analogue scale

terms for the first time in their lives when taking part in this study.

In our evaluation of the psychometric properties of the SHAQ, we detected moderate convergent validity when compared to the SF-36v2. SHAQ correlated with both SF-36v2 PCS and MCS. There was a moderate correlation between the SF-36v2 physical component score and SHAQ-global, RP-VAS, digital ulcer VAS, and pulmonary VAS scores. However, we observed a correlation between the SF-36v2 MCS with HAQ-DI, SHAQ-global, and overall disease severity VAS score. This result highlighted the effect of the patient's emotional state at the time of questionnaire completion which mainly affects the evaluation of his/her own disability. Similar results were reported by Georges et al. and they warned about the potential effect of this error over the test–retest [12]. Although we did not experience any adverse result due to this error, we are aware of any uncontrolled estimation resulting mainly from this problem. We hypothesize that the consistent results of retest reliability is due to the shortness of period between the two applications. There is also evidence that

some of the SSc-related involvements (e.g., gastrointestinal) may lead to more impact on mental status than physical [23].

We evaluated discriminant validity by testing the change in HAQ-DI, SHAQ-global, and disease-specific VAS scores according to the disease subtypes and the number of organs involved. Diffuse cutaneous SSc is known to have severe organ involvement and poor prognosis compared to the limited subtype. Therefore, the outcome measures are expected to be differentiative between the disease subtypes [24]. However, except for the digital ulcer VAS which was significantly higher in patients with the diffuse subtype compared to the patients with limited subtype, we could not demonstrate a strong difference between dcSSc and lcSSC similar to the results of previous studies [9, 12]. Although the patients with dcSSc had more severe hand involvement, including hand contractures, RP attacks in the last week, and greater mRSS, the musculoskeletal involvement exhibited no difference between the disease subtypes. This may be owing to the fact that, in our study, the musculoskeletal involvement restricting motion and leading to disability was due to a wide range of factors. Similarly, interstitial lung disease and pulmonary symptoms were more common in dcSSc patients. However, the pulmonary VAS failed to show discrimination of this severe involvement between the disease subtypes. This result may be due to our tendency to determine pulmonary involvement mainly based on imaging techniques rather than functional tests, as many clinicians do. In accordance with this result, the forced vital capacity and carbon monoxide diffusing capacity were within the normal range and showed no difference between the disease subtypes. In order to assist the discriminant validity of the questionnaires, we showed satisfactory correlations of SHAQ, SHAQ-global, digital ulcer VAS, and pulmonary VAS with increasing number of organs involved. Moreover, we examined the test–retest reliability of the HAQ-DI and all of the SHAQ scores and demonstrated good reproducibility with intraclass correlation coefficients (ICC) of > 0.70.

The promising results of our study highlight the potential value of validated, SSc-specific, and patient-reported instruments for use in local clinical settings. As they are fairly quick and easy to administer, clinicians can utilize them as a part of

**Table 5** Reproducibility. Test–retest score differences were measured among patients who filled in a second questionnaire 2 weeks after the first one

	Intraclass correlation coefficients (95% CI, lower–upper bound)	Number of the patients analyzed
HAQ-DI	0.962 (0.934–0.978)	51
SHAQ-global	0.900 (0.820–0.944)	47
Raynaud's phenomenon VAS	0.974 (0.955–0.985)	51
Digital ulcer VAS	0.873 (0.778–0.928)	51
Digestive VAS	0.874 (0.779–0.928)	51
Pulmonary VAS	0.813 (0.674–0.893)	51
Overall disease severity VAS	0.816 (0.678–0.895)	51

HAQ-DI Health Assessment Questionnaire Disability Index, SHAQ Scleroderma Health Assessment Questionnaire, VAS visual analogue scale

routine care in SSc patients' evaluation in an outpatient setting. In conclusion, this study has demonstrated that the Turkish version of the SHAQ is a valid and reliable tool to assess SSc-related quality of life. These findings support the use of this version of the SHAQ in clinical practice and research settings in Turkey.

**Acknowledgments** This study had no financial support. The authors thank all investigators for their contribution to the study. The corresponding author certifies that all authors approved the entirety of the submitted material and contributed actively to the study. We would like to thank Mr. Jeremy Jones, of the Kocaeli University Academic Writing Department, for the revision of the English in this paper.

## Compliance with ethical standards

**Disclosures** None.

**Ethical approval** This article does not contain any studies with animals performed by any of the authors. Informed consent was obtained from all individual participants included in the study.

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## References

- Gabrielli A, Avvedimento EV, Krieg T (2009) Scleroderma. *N Engl J Med* 360:1989–2003
- Hudson M, Thombs BD, Steele R, Panopalis P, Newton E, Baron M (2009) Quality of life in patients with systemic sclerosis compared to the general population and patients with other chronic conditions. *J Rheumatol* 36:768–772
- Denton CP, Khanna D (2017) Systemic sclerosis. *Lancet* 390:1685–1699
- Silman A, Akesson A, Newman J, Henriksson H, Sandquist G, Nihill M, Palfrey S, Lomas R, Wollheim F, Black C (1998) Assessment of functional ability in patients with scleroderma: a proposed new disability assessment instrument. *J Rheumatol* 25:79–83
- Poole JL, Williams CA, Bloch DA, Hollak B, Spitz P (1995) Concurrent validity of the Health Assessment Questionnaire Disability Index in scleroderma. *Arthritis Care Res* 8:189–193
- Bruce B, Fries JF (2003) The Stanford Health Assessment Questionnaire: dimensions and practical applications. *Health Qual Life Outcomes* 1:20
- Steen VD, Medsger TA Jr (1997) The value of the Health Assessment Questionnaire and special patient-generated scales to demonstrate a change in systemic sclerosis patients over time. *Arthritis Rheum* 40:1984–1991
- Kuwana M, Sato S, Kikuchi K, Kawaguchi Y, Fujisaku A, Misaki Y, Hatamochi A, Kondo H, Takehara K (2003) Evaluation of functional disability using the health assessment questionnaire in Japanese patients with systemic sclerosis. *J Rheumatol* 30:1253–1258
- Rocha LF, Marangoni RG, Sampaio-Barros PD, Levy-Neto M, Yoshinari NH, Bonfa E, Steen V, Kowalski SC (2014) Cross-cultural adaptation and validation of the Brazilian version of the Scleroderma Health Assessment Questionnaire (SHAQ). *Clin Rheumatol* 33:699–706
- Ng X, Thumboo J, Low AH (2012) Validation of the scleroderma health assessment questionnaire and quality of life in English and Chinese-speaking patients with systemic sclerosis. *Int J Rheum Dis* 15:268–276
- Johnson SR, Hawker GA, Davis AM (2005) The health assessment questionnaire disability index and scleroderma health assessment questionnaire in scleroderma trials: an evaluation of their measurement properties. *Arthritis Rheum* 53:256–262
- Georges C, Chassany O, Mouthon L, Tiev K, Toledano C, Meyer O, Marjanovic Z, Heneggar C, Papo T, Crickx B, Sereni D, Cabane J, Farge D (2005) Validation of French version of the Scleroderma Health Assessment Questionnaire (SSc HAQ). *Clin Rheumatol* 24:3–10
- van den Hoogen F, Khanna D, Fransen J, Johnson SR, Baron M, Tyndall A et al (2003) Classification criteria for systemic sclerosis: an American College of Rheumatology/European League Against Rheumatism collaborative initiative. *Ann Rheum Dis* 72:1747–1755
- LeRoy EC, Black C, Fleischmajer R, Jablonska S, Krieg T, Medsger TA Jr et al (1998) Scleroderma (systemic sclerosis): classification, subsets and pathogenesis. *J Rheumatol* 15:202–205
- Clements P, Lachenbruch P, Siebold J, White B, Weiner S, Martin R, Weinstein A, Weisman M, Mayes M, Collier D (1995) Inter and intraobserver variability of total skin thickness score (modified Rodnan TSS) in systemic sclerosis. *J Rheumatol* 22:1281–1285
- Hsu VM, Moreyra AE, Wilson AC, Shinnar M, Shindler DM, Wilson JE, Desai A, Seibold JR (2008) Assessment of pulmonary arterial hypertension in patients with systemic sclerosis: comparison of noninvasive tests with results of right-heart catheterization. *J Rheumatol* 35:458–465
- Küçükdeveci AA, Sahin H, Ataman S, Griffiths B, Tennant A (2004) Issues in cross-cultural validity: an example from the adaptation, reliability, and validity testing of a Turkish version of the Stanford Health Assessment Questionnaire. *Arthritis Rheum* 51:14–19
- Celik D, Coban Ö (2016) Short Form Health Survey version-2.0 Turkish (SF-36v2) is an efficient outcome parameter in musculoskeletal research. *Acta Orthop Traumatol Turc* 50:558–561
- Beaton DE, Bombardier C, Guillemin F, Ferraz MB (2000) Guidelines for the process of cross-cultural adaptation of self-report measures. *Spine (Phila Pa 1976)* 25:3186–3191
- Wild D, Grove A, Martin M, Eremenco S, McElroy S, Verjee-Lorenz A, Erikson P (2005) ISPOR Task Force for Translation and Cultural Adaptation. Principles of good practice for the translation and cultural adaptation process for patient-reported outcomes (PRO) measures: report of the ISPOR Task Force for Translation and Cultural Adaptation. *Value Health* 8:94–104
- Fayers PM, Machin D (2007) Scores and measurements: validity, reliability, sensitivity. In: *Quality of life: the assessment, analysis, and interpretation of patient-reported outcomes*, 2nd edn. John Wiley & Sons, Chichester, pp 77–107
- Cohen J (2003) *Applied multiple regression/correlation analysis for the behavioral sciences*, 3rd edn. Erlbaum L Associates, Mahwah, p 703
- Khanna D, Furst DE, Clements PJ, Park GS, Hays RD, Yoon J, Relaxin Study Group, Scleroderma Clinical Trials Consortium et al (2005) Responsiveness of the SF-36v2 and the Health Assessment Questionnaire Disability Index in a systemic sclerosis clinical trial. *J Rheumatol* 32:832–840
- Pope J (2011) Measures of systemic sclerosis (scleroderma). *Arthritis Care Res (Hoboken)* 63:S98–S111