ORIGINAL ARTICLE

Using Optimal Test Assembly Methods for Shortening Patient-Reported Outcome Measures: Development and Validation of the Cochin Hand Function Scale-6: A Scleroderma Patient-Centered Intervention Network Cohort Study

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Objective. To develop and validate a short form of the Cochin Hand Function Scale (CHFS), which measures hand disability, for use in systemic sclerosis, using objective criteria and reproducible techniques.

Methods. Responses on the 18-item CHFS were obtained from English-speaking patients enrolled in the Scleroderma Patient-Centered Intervention Network Cohort. CHFS unidimensionality was verified using confirmatory factor analysis, and an item response theory model was fit to CHFS items. Optimal test assembly (OTA) methods identified a maximally precise short form for each possible form length between 1 and 17 items. The final short form selected was the form with the least number of items that maintained statistically equivalent convergent validity, compared to the full-length CHFS, with the Health Assessment Questionnaire (HAQ) disability index (DI) and the physical function domain of the 29-item Patient-Reported Outcomes Measurement Information System (PROMIS-29).

Results. There were 601 patients included. A 6-item short form of the CHFS (CHFS-6) was selected. The CHFS-6 had a Cronbach's alpha of 0.93. Correlations of the CHFS-6 summed score with HAQ DI (r = 0.79) and PROMIS-29 physical function (r = -0.54) were statistically equivalent to the CHFS (r = 0.81 and r = -0.56). The correlation with the full CHFS was high (r = 0.98).

Conclusion. The OTA procedure generated a valid short form of the CHFS with minimal loss of information compared to the full-length form. The OTA method used was based on objective, prespecified criteria, but should be further studied for viability as a general procedure for shortening patient-reported outcome measures in health research.

INTRODUCTION

Patient-reported outcomes (PROs) assess patient health, wellbeing, and treatment response based on patient perspectives (1,2). In rheumatic diseases, PROs such as quality of life and functional ability are as important to many patients as survival

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Significance & Innovations

- Valid and reliable short forms of patient-reported outcome measures can reduce patient burden and increase research capacity.
- There are no standard methods for developing short forms to maximize information.
- This study showed that optimal test assembly (OTA) and equivalence testing methods may be used to create short forms objectively and reproducibly.
- The Cochin Hand Function Scale-6 was developed using OTA and equivalency methods and is a brief, valid short form for measuring hand disability in systemic sclerosis.

In rare diseases, including systemic sclerosis (SSc; scleroderma), cohorts designed to collect medical and PRO data from large numbers of patients require collaborations that span countries, languages, and clinical settings. SSc is a chronic, multisystem autoimmune disorder characterized by fibrotic changes to the skin, joints, and tendons, as well as vascular injury (5,6). Large SSc multinational cohorts include the European League Against Rheumatism (EULAR) Scleroderma Trials and Research Cohort (7) and the Scleroderma Patient-Centered Intervention Network (SPIN) Cohort (8) (see Appendix A for a list of the SPIN investigators) among others (9).

Digital ulcers, contractures, and deformities of the hand, which lead to decreased flexion, limited extension, and reduced thumb abduction, play a major role in functional disability among patients with SSc (10–12). Disability related to impaired hand function may be present in close to 90% of SSc patients (12,13). The Cochin Hand Function Scale (CHFS) (14) was developed to measure functional ability of

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Address correspondence to Daphna Harel, PhD, 246 Greene Street, 3rd Floor, New York NY, 10003. E-mail: daphna.harel@ nyu.edu. the hand among patients with rheumatic diseases and has been validated (15,16) and used extensively in SSc (8,17–19). The CHFS consists of 18 items that, when completed by the patient or clinician, assess the ability to perform daily handrelated activities. There are, however, notable redundancies among the 18 items. For example, item 13 (Can you write a short sentence with a pencil or an ordinary pen?) and item 14 (Can you write a letter with a pencil or an ordinary pen?) may not together provide significantly more information beyond what is captured by either item independently.

Shortening PRO measures would increase the number of outcomes that can be measured in studies; however, no standard methods currently exist (20–23). Traditionally, items have been deleted based on item-total correlations or the goal of maintaining or improving factorial structure by removing items with low factor loadings or high residuals (23,24) or through qualitative analysis of item content (23). Modern techniques, such as item response theory (25), allow for detailed item evaluation to identify problematic items, but still leave the final selection of items to the researcher's prerogative rather than objective and reproducible criteria.

Optimal test assembly (OTA) (26), used frequently for item selection in designing high-stakes educational tests (27), incorporates the results of item response theory models to select the optimal subset of an item pool that best satisfies objective, prespecified constraints, such as content- or precision-related requirements. To the best of our knowledge, OTA has not been used previously in health research. Nonetheless, these methods have the potential to empirically guide the shortening of previously validated PRO measures by optimizing performance based on objective, replicable procedures.

The objective of the present study was to develop a short form of the CHFS using OTA. To do this, we 1) verified the unidimensionality of the scale using confirmatory factor analysis, 2) applied OTA methods in order to obtain maximally precise candidate short forms of each possible length, and 3) selected the shortest possible short form that demonstrated statistical equivalency, based on tests of convergent validity, to the full-length scale.

PATIENTS AND METHODS

Sample and procedure. The sample consisted of patients enrolled in the SPIN Cohort (8) from 21 centers in Canada, the US, and the UK who completed study questionnaires from March 2014 through June 2015. To be included in the SPIN Cohort, patients must have a confirmed diagnosis of SSc, according to 2013 American College of Rheumatology (ACR)/EULAR classification criteria (28), be ≥ 18 years of age, have the ability to give informed consent, be fluent in English or French, and be able to respond to questionnaires via the internet. Eligible patients are invited by SPIN-center physicians or supervised nurse coordinators to participate, and written informed consent is obtained. To initiate patient registration, the local SPIN physician or nurse coordinator completes an online medical data record and an automated e-mail is then sent to the patient with instructions for activating their account. Participants complete SPIN Cohort measures online upon enrollment and subsequently once every 3

months. Only patients with complete CHFS data at baseline in English were included in the present study.

Measures. SPIN physicians provided medical information, including time since onset of the first non–Raynaud's phenomenon symptom, SSc subtype (limited or diffuse) (29), modified Rodnan skin score, and presence of puffy fingers, sclerodactyly, skin thickening of the fingers, fingertip pitting scars, digital ulcers, and small joint contractures (30). Patients provided demographic data and completed PROs.

The 18-item CHFS (14) measures the ability to perform daily hand-related activities. Items reflecting 5 content areas (kitchen, dressing oneself, hygiene, the office, and other) are scored on a Likert scale from 0 (yes, without difficulty) to 5 (impossible). Total CHFS scores range from 0 to 90, and higher scores indicate more hand disability. The CHFS, when clinician completed, has shown excellent intra- and interrater reliability (intraclass correlation coefficients of 0.97 and 0.96, respectively) (14), good convergent validity with functional disability measures, and sensitivity to changes in hand function (14,31). Validity and reliability of the self-report version have been confirmed in SSc (15,16).

The Health Assessment Questionnaire (HAQ) disability index (DI) and the physical function domain of the 29-item Patient Reported Outcomes Measurement Information System (PROMIS-29; profile version 1.0 or 2.0) were used to establish convergent validity. The HAQ DI assesses disability within 8 categories measured over the past 7 days: dressing/ grooming, arising, eating, walking, hygiene, reach, grip, and common daily activities. Each item is rated on a 4-point scale, ranging from 0 (without any difficulty) to 3 (unable to do). The highest score from each category, indicating greater disability, determines the score for that category, and the total score is the mean of the 8 category scores, ranging from 0 (no disability) to 3 (severe disability). The HAQ DI is widely used in patients with rheumatologic diseases, and is a valid measure of functional disability in SSc (32).

The physical function domain of the PROMIS-29 assesses functional ability. This domain consists of 4 items measuring capacity to complete day-to-day activities, scored on a Likert scale from 1 (unable to do) to 5 (without any difficulty). The summed score of the 4 items is standardized based on norms from the general US population (mean \pm SD 50 \pm 10). Higher scores indicate greater physical function. The PROMIS-29 and its subscales have been shown to be valid measures of health status in SSc (33).

Statistical analysis. To verify the assumption of unidimensionality for the CHFS, a single-factor confirmatory factor analysis model was fit to the CHFS data using a robust weighted least squares estimator (34). Modification indices were calculated to recognize item pairs for which measurement errors correlated highly. If there was theoretical justification for shared effects within these pairs of items, we allowed their errors to co-vary if this improved model fit (35). Model fit was evaluated via a mean- and variance-adjusted chi-square test statistic (34), the Comparative Fit Index (CFI), the Tucker-Lewis Index (TLI), and the root mean square error of approximation (RMSEA). The CFI, TLI, and RMSEA indices were prioritized, as the chi-square test may reject models despite good fit because it is highly dependent on sample size (36). Values of CFI and TLI \geq 0.95 and RMSEA \leq 0.10 were considered to indicate good fit (37,38).

Next, a generalized partial credit item response theory model (25,39) was fit to all 18 CHFS items. For each item, the model estimates 1) thresholds for the levels of disability in hand function at which patients are more likely to endorse a given response category instead of the category below, and 2) a discrimination parameter, which measures the strength of the relationship between that item and the underlying construct, that is, functional disability of the hand (θ). Item information functions were then estimated. The test information functions, measures the total amount of Fisher's information contained in the CHFS as a function of the latent trait, θ . Fisher's information, which is inversely related to the SE of measurement, summarizes the degree of precision of measurements of the latent construct (25).

OTA was then used to create candidate short forms of each possible length by selecting items to maximize the TIF (26). Thus, for each possible short-form length, a single optimal short form, which included a subset of the 18 total CHFS items, was generated. OTA methods use mixed integer programming to optimize an objective function subject to a series of user-defined constraints. In this case, we adopted a maximin procedure (26,40) to select items that maximized the height of each short form's TIF, maintaining the same relative shape of the full-form TIF. This approach yields short forms that measure the latent trait with the same relative precision across the continuum of the latent trait as the full form, while minimizing absolute loss of information. Based on previously established guidelines for best performance of this OTA procedure, the relative shape of the TIF was anchored at 5 points across the spectrum of disability in hand function ($\theta = -1, 1, 2, 3, \text{ and } 5$) (26). For each candidate short form, the height of the TIF and percentage of the fullform TIF maintained in the short form were calculated, as well as the average total information across the latent trait spectrum, as a percentage of full-form total information.

For each candidate short form and the full-length form, patients were scored in 2 ways. First, summed scores were computed since summed scores are typically relied upon for clinical use. Second, factor scores of disability in hand function, which are assumed to have a standard normal distribution in the population, were estimated for each patient via an application of Bayes' theorem. Because of the well-known limitations of the summed score under the generalized partial credit model, factor scores were considered to provide a more consistent estimate of the latent trait (41,42).

For each candidate short form and the full-length CHFS, Cronbach's α was used to assess the internal consistency reliability. For concurrent validity, Pearson's product-moment correlation coefficients between summed scores and factor scores on each candidate form with summed and factor scores for the full-length form were calculated. Convergent validity was assessed via correlations between summed and factor scores on each candidate form and summed scores on the HAQ DI and the PROMIS-29 physical function domain. Patients with missing data for either of the convergent validity measures were not included in the corresponding correlation calculations. We hypothesized that higher scores on the CHFS and its short forms would be associated with higher scores on the HAQ DI (more disability) but with lower scores

Table 1. Patient demographic and disease cha	aracteristics (n = 601)*
Age, mean ± SD (range) years Women, no. (%) Education >12 years, no. (%) Currently employed, no. (%) Married/cohabitating, no. (%) Time since onset of first non-Raynaud's phenomenon symptoms, years, mean ± SD (range)† Patients with diffuse SSc, no. (%)‡ MRSS, mean ± SD (range)§ Puffy fingers, no. (%)¶ Sclerodactyly, no. (%)¶ Sclerodactyly, no. (%)# Skin thickening of the fingers, no. (%)** Fingertip pitting scars, no. (%)†+ Digital ulcers, no. (%)‡‡ Moderate to severe small joint contractures, no. (%)§§ CHFS score, mean (median) ± SD (range)	$55.4 \pm 11.9 (18.6-84.7)$ $524 (87)$ $483 (80)$ $248 (41)$ $442 (74)$ $11.8 \pm 8.8 (0.1-46.2)$ $250 (42)$ $8.2 \pm 9.2 (0-48)$ $371 (65)$ $503 (84)$ $338 (56)$ $250 (42)$ $229 (40)$ $145 (25)$ $14.4 (8) \pm 16.7 (0-88)$
In diffuse SSc subset, mean (median) ± SD (range) In limited SSc subset, mean (median) ± SD (range) HAQ DI score, mean (median) ± SD (range)¶¶ PROMIS-29 physical function score,	$\begin{array}{c} 14.4 \ (8) \pm 16.7 \ (0-88) \\ 20.5 \ (15) \pm 19.2 \ (0-88) \\ 10.0 \ (4) \pm 12.9 \ (0-62) \\ 0.81 \ (0.75) \pm 0.69 \ (0-3) \\ 42.5 \ (41.8) \pm 8.7 \ (22.9-56.9) \end{array}$
mean (median) ± SD (range)## * SSc = systemic sclerosis; MRSS = modified Rodnan skin score; 0 HAQ = Health Assessment Questionnaire; DI = disability index; PRON comes Measurement Information System. † N = 555. ‡ N = 597. § N = 503. ¶ N = 571. # N = 598. ** N = 600. +± N = 501.	
<pre>++ N = 591. ++ N = 579; considered to have digital ulcer if had digital pulp (v joints, or elsewhere on the finger, and provided a response to both \$\$ N = 574. ¶¶ N = 596. ## N = 595.</pre>	

for the PROMIS-29 physical function domain (greater physical function).

OTA methods generate optimal candidate short forms of each possible length, but do not readily provide criteria by which to select the best short-form length. By nature of eliminating items, the short forms necessarily will have lower information as compared with the full CHFS. Beforehand, there is no obvious threshold at which one would conclude that a short form has adequate information, so the validity of the short forms must be assessed in order to find a balance between shortening the scale and retaining its measurement ability. Thus, 2 criteria were used for selecting which candidate short form should be chosen. First, we required that the selected short form maintain high concurrent validity (r > 0.90) and high internal consistency ($\alpha \ge 0.90$) and demonstrate statistical equivalence with the full CHFS for measures of convergent validity. Equivalence testing, which has origins in clinical trials, is used to test whether the difference between 2 effect measures (e.g., treatment effect for 2 drugs) is within a prespecified range (43). The equivalence testing paradigm, contrary to traditional hypothesis testing, tests a null hypothesis that there will be a difference between the 2 effect measures equal to or greater than a prespecified threshold against the alternative of equivalence or no difference. In our study, we specified a null hypothesis that the magnitude of the difference between each convergent validity correlation for the candidate short form and its corresponding

correlation for the full CHFS would be ≥ 0.05 (44). To assess statistical significance, we applied a Bonferonni correction factor for each of 66 possible comparisons (summed score and factor score \times 2 measures \times 16 short forms of length 2–17 items plus single-item score for 2 measures; $P < 7.58 \times 10^{-4}$) to maintain the family-wise Type I error rate of $\alpha = 0.05$.

The confirmatory factor analysis was done using Mplus 7 (34). All other analyses were done using R, version 3.2.1 (45). The generalized partial credit model was fit using the ltm package (46). The OTA analysis was conducted using the lpSolveAPI package (47).

RESULTS

There were 601 patients who completed the CHFS. Of these, 596 (99%) also completed the HAQ DI, and 595 (99%) completed the PROMIS-29 physical function domain. The mean age was 55.4 years, 87% were women, and 42% had diffuse SSc. The mean \pm SD score on the CHFS was 14.4 \pm 16.7. CHFS scores in patients with diffuse SSc were substantially higher than patients with limited SSc (see Table 1 for descriptive statistics).

Confirmatory factor analysis. A unidimensional confirmatory factor analysis model of the CHFS items, where

Item number	Description	Discrimination parameter	
1	Can you hold a bowl?	2.33	
2	Can you seize a full bottle and raise it?	1.84	
3	Can you hold a plate full of food?	2.25	
4	Can you pour liquid from a bottle into a glass?	2.10	
5	Can you unscrew the lid from a jar opened before?	1.27	
6	Can you cut meat with a knife?	1.98	
7	Can you prick things well with a fork?	2.65	
8	Can you peel fruit?	1.92	
9	Can you button your shirt?	1.76	
10	Can you open and close a zipper?	1.99	
11	Can you squeeze a new tube of toothpaste?	1.61	
12	Can you hold a toothbrush efficiently?	1.66	
13	Can you write a short sentence with a pencil or an ordinary pen?	1.52	
14	Can you write a letter with a pencil or an ordinary pen?	1.23	
15	Can you turn a round door knob?	2.11	
16	Can you cut a piece of paper with scissors?	2.29	
17	Can you pick up coins from a table top?	1.41	
18	Can you turn a key in a lock?	2.32	

covariance of item residuals was restricted to zero, resulted in less than ideal fit (χ^2 [135df] = 1,509.3, P < 0.0001, TLI = 0.966, CFI = 0.970, and RMSEA = 0.130). Modification indices suggested that allowing residuals of items 9 and 10 and items 13 and 14 to co-vary would improve model fit. Items 9 and 10 both assess the ability to perform actions involved in dressing one-self, and the content of items 13 and 14 involves writing with a pencil. The model was refitted, allowing the residuals for these 2 item pairs to co-vary, and fit improved (χ^2 [133df] = 866.0, P < 0.0001, TLI = 0.982, CFI = 0.984, and RMSEA = 0.096). Factor loadings for the items were all very high (>0.82) with the majority >0.90.

Item response theory model and OTA. The generalized partial credit model was fit to the 18 items of the CHFS. Item content along with the discrimination parameters of the model are shown in Table 2. The items with greatest discriminative ability and therefore the greatest influence on the TIF were items 1 (Can you hold a bowl?), 3 (Can you hold a plate full of food?), 7 (Can you prick things well with a fork?), 16 (Can you cut a piece of paper with scissors?), and 18 (Can you turn a key in a lock?). Figure 1 shows the individual item information functions generated by the generalized partial credit model and the aggregate TIF.

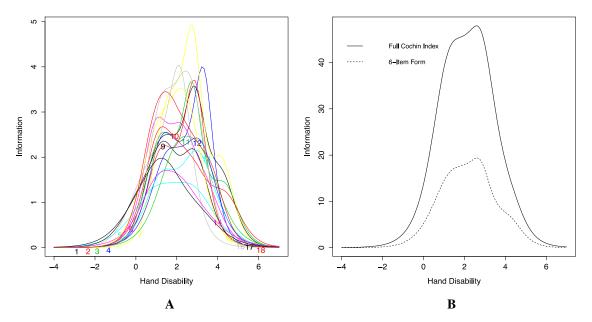


Figure 1. Item and test information curves of the Cochin Hand Function Scale (CHFS), showing the 18 individual item information curves, labeled by color (A) and the comparison of the test information functions (B) of the full CHFS (solid line) and CHFS-6 (broken line).

Table 3. Optimal short forms of each length																		
	Item number (X indicates inclusion)																	
	Hold			4 Pour liquid	5 Unscrew lid					10 Zipper		tooth-					17 Pick up coins	18 Turn key
1																		Х
2							Х	Х										
3							Х		Х									Х
4							Х		Х	Х					Х			
5	Х						Х	Х	Х									Х
6	Х		Х				Х	Х	Х									Х
7	Х		Х				Х		Х						Х	Х	Х	
8	Х		Х				Х		Х						Х	Х	Х	Х
9	Х		Х	Х			Х	Х	Х							Х	Х	Х
10	Х		Х	Х			Х	Х	Х	Х					Х	Х	Х	
11	Х		Х	Х			Х	Х	Х	Х					Х	Х	Х	Х
12	Х	Х	Х	Х		Х	Х	Х	Х	Х					Х		Х	Х
13	Х	Х	Х	Х	Х		Х	Х	Х	Х					Х	Х	Х	Х
14	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х					Х	Х	Х
15	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х				Х	Х	Х	Х
16	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х		Х		Х	Х	Х	Х
17	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х		Х	Х	Х	Х	Х
18	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х	Х

Considering each possible subset of the 18 items, the OTA procedure selected the short form of each size that maximally maintained the shape of the TIF for the full-length form. The items chosen for each of the short-form sizes are shown in Table 3. Several patterns emerged from the OTA selection procedure. First, items 12, 13, and 14 were only selected in the longest short forms and therefore quickly dropped from smaller short forms. These items all had low discrimination parameters (Table 2). Second, items 1, 3, 7, and 9 were

included in all forms of at least 6 items. All of these items had very high information at certain points on the continuum of disability or had fairly high information consistently across the continuum. Third, some items seemed to alternate in their selection into short forms. For example, items 15 and 18 were often mutually exclusive in the smaller short-form sizes. The content of both items relates to opening a lock or door, and there was little added measurement value in including both.

Short form length		Average				
	$\theta = -1$	$\theta = 1$	$\theta = 2$	$\theta = 3$	$\theta = 5$	information (% of full form)
1	0.14 (4.6)	3.16 (8.1)	3.13 (6.8)	2.34 (5.4)	0.25 (4.5)	6.8
2	0.37 (12.4)	4.51 (11.6)	7.66 (16.7)	5.71 (13.0)	0.77 (13.6)	13.3
3	0.57 (19.1)	7.57 (19.5)	8.87 (19.3)	8.72 (19.9)	1.11 (19.7)	19.7
4	0.72 (24.2)	9.66 (24.9)	11.61 (25.3)	10.63 (24.3)	1.73 (30.8)	24.9
5	1.00 (33.4)	12.10 (31.2)	15.39 (33.5)	13.61 (31.1)	1.88 (33.4)	32.1
6	1.08 (36.0)*	14.43 (37.2)*	17.92 (39.0)*	16.82 (38.4)*	2.69 (47.8)*	38.6*
7	1.30 (43.6)	16.62 (42.8)	19.59 (42.7)	19.69 (44.9)	2.73 (48.5)	43.2
8	1.44 (48.1)	19.79 (51.0)	22.72 (49.5)	22.03 (50.3)	2.98 (53.0)	50.0
9	1.69 (56.6)	21.50 (55.4)	25.60 (55.8)	24.64 (56.2)	3.07 (54.6)	55.6
10	1.85 (61.7)	23.58 (60.7)	28.34 (61.8)	26.55 (60.6)	3.70 (65.8)	60.8
11	1.99 (66.3)	26.75 (68.9)	31.47 (68.6)	28.90 (66.0)	3.95 (70.3)	67.6
12	2.14 (71.7)	28.65 (73.8)	32.83 (71.5)	31.29 (71.4)	4.15 (73.8)	72.0
13	2.40 (80.3)	30.13 (77.6)	35.19 (76.7)	33.77 (77.1)	4.25 (75.7)	76.7
14	2.43 (81.2)	31.39 (80.8)	36.80 (80.2)	35.72 (81.5)	4.77 (84.9)	81.0
15	2.60 (86.9)	34.13 (87.9)	40.29 (87.8)	38.15 (87.1)	4.85 (86.2)	87.1
16	2.72 (90.9)	35.66 (91.8)	42.03 (91.6)	40.30 (92.0)	5.14 (91.5)	91.6
17	2.87 (96.0)	37.30 (96.1)	44.15 (96.2)	41.67 (95.1)	5.32 (94.7)	95.6
18	2.99 (100.0)	38.83 (100.0)	45.89 (100.0)	43.82 (100.0)	5.62 (100.0)	100.0

	HAQ DI scor	e correlation	PROMIS-29 physical function correlation			
Short form length	Summed score	Factor score	Summed score	Factor score		
1	0.093 (P>0.99)	NA	-0.071 (P = 0.89)	NA		
2	$0.077 \ (P > 0.99)$	$0.080 \ (P > 0.99)$	$-0.060 \ (P = 0.78)$	-0.069 (P = 0.90)		
3	$0.040 \ (P = 0.09)$	$0.045 \ (P = 0.28)$	-0.038 (P = 0.13)	-0.045 (P = 0.34)		
4	$0.045 \ (P = 0.24)$	$0.051 \ (P = 0.53)$	$-0.043 \ (P = 0.25)$	$-0.047 \ (P = 0.40)$		
5	$0.025 \ (P < 0.0001)$	0.026 (<i>P</i> < 0.0001)	$-0.025 \ (P < 0.001)$	$-0.026 \ (P < 0.01)$		
6	0.016 (P<0.0001)†	0.015 (P<0.0001)†	-0.017 (P < 0.0001)†	-0.016 (P < 0.0001)		
7	0.023 (<i>P</i> < 0.0001)	$0.025 \ (P < 0.0001)$	-0.035 (P < 0.01)	-0.041 (P = 0.12)		
8	0.020 (<i>P</i> < 0.0001)	0.020 (<i>P</i> < 0.0001)	$-0.031 \ (P < 0.001)$	-0.036 (P = 0.02)		
9	0.015 (P < 0.0001)	$0.014 \ (P < 0.0001)$	$-0.019 \ (P < 0.0001)$	-0.019 (P < 0.0001)		
10	0.017 (<i>P</i> < 0.0001)	0.017 (<i>P</i> < 0.0001)	$-0.021 \ (P < 0.0001)$	-0.022 (P < 0.0001)		
11	0.014 (<i>P</i> < 0.0001)	0.015 (<i>P</i> < 0.0001)	$-0.019 \ (P < 0.0001)$	-0.020 (P < 0.0001)		
12	0.005 (P < 0.0001)	0.005 (P < 0.0001)	$-0.008 \ (P < 0.0001)$	-0.007 (P < 0.0001)		
13	0.005 (<i>P</i> < 0.0001)	0.005 (<i>P</i> < 0.0001)	$-0.008 \ (P < 0.0001)$	-0.009 (P < 0.0001)		
14	$0.001 \ (P < 0.0001)$	$0.000 \ (P < 0.0001)$	-0.005 (P < 0.0001)	-0.005 (P < 0.0001)		
15	$0.002 \ (P < 0.0001)$	0.001 (<i>P</i> < 0.0001)	$-0.008 \ (P < 0.0001)$	-0.009 (P < 0.0001)		
16	0.002 (P < 0.0001)	0.000 (<i>P</i> < 0.0001)	-0.006 (P < 0.0001)	-0.005 (P < 0.0001)		
17	$-0.001 \ (P < 0.0001)$	-0.001 (P < 0.0001)	-0.001 (P < 0.0001)	-0.002 (P < 0.0001)		

versions $[r_{full}-r_{short}]$, and *P* values for equivalency within ± 0.05 . HAQ = Health Assessment Questionnaire; DI = disability index; PROMIS-29 = 29-item Patient Reported Outcomes Measurement Information System; NA = not applicable. \pm Indicates values of the final selected short form.

The TIFs for each candidate short form are summarized in Table 4. As expected, the height of the TIF and percentage of information at each of 5 points across the latent spectrum and average information across the entire spectrum show a consistent decrease in information as the length of the form decreases. This drop in information translates into an increase in the SE of measurement for the latent trait as the length of the short form decreases. However, despite this loss of information, all internal and external validity correlations remained consistently high, even for short forms containing a small number of items (see Supplementary Tables 1 and 2, available on the *Arthritis Care & Research* web site at http://onlinelibrary.wiley.com/doi/10.1002/acr.22893/abstract).

Selection of final short form. The equivalency analysis presented in Table 5 assesses which candidate short forms maintained a reasonably equivalent level of concurrent and convergent validity. The 6-item short form and all short forms with at least 9 items demonstrated statistically significant equivalence, following Bonferroni correction, to all correlations between summed and factor scores of the full CHFS with the HAQ DI and PROMIS-29 physical function scores. Although the 5-item short form satisfied statistical equivalence for 3 of the correlations, the correlation between the factor scores and the PROMIS-29 physical function scores failed to demonstrate equivalence to the corresponding full CHFS correlation ($P = 0.003 > 7.58 \times 10^{-4}$). Thus, the 6-item optimal short form (CHFS-6; see Supplementary Table 3, available on the Arthritis Care & Research web site at http://onlinelibrary. wiley.com/doi/10.1002/acr.22893/abstract) was the shortest candidate form to fulfill our equivalence requirement.

The CHFS-6 had a Cronbach's α of 0.932 and a correlation with the full 18-item CHFS scores for summed scores of r = 0.980 (95% confidence interval [95% CI] 0.976, 0.983) and factor scores of r = 0.970 (95% CI 0.965, 0.975) (see Supplementary Tables 1 and 2, available on the *Arthritis Care & Research* web site at http://onlinelibrary.wiley.com/doi/10.1002/acr.22893/abstract). The summed scores on the CHFS-6 maintained strong positive correlations with the HAQ DI (r = 0.790, 95% CI 0.758, 0.819) and moderate negative correlations with the PROMIS-29 physical function domain (r = -0.544, 95% CI -0.599, -0.485). The TIF of the CHFS-6 as compared to the full-length form is shown in Figure 1. The CHFS-6 retained, on average, 38.6% of the Fisher information of the full form (Table 4), corresponding to 1.61 times the SE of measurement on average between the short and full forms.

DISCUSSION

This study demonstrated how OTA methods, which have been used extensively in high-stakes educational testing, may also be used to create valid PRO short forms based on objective, prespecified constraints in health research. The main finding of the study was that the 18-item CHFS could be shortened to a 6-item version (see Supplementary Table 3, available on the *Arthritis Care & Research* web site at http://onlinelibrary. wiley.com/doi/10.1002/acr.22893/abstract) with minimal loss of information and maintaining high internal validity and similar convergent validity with the HAQ DI and PROMIS-29 physical function domain. The summed scores of the CHFS-6 and the full CHFS correlated at r = 0.980. Cronbach's α was 0.932 compared to 0.974 for the full CHFS, and all correlations with convergent validity measures were similar and within

prespecified ranges for statistical equivalence to the correlations between these measures and the full CHFS. The items of the CHFS-6 included 3 related to eating (holding a bowl, holding a plate of food, and holding a fork), 1 related to food preparation (peeling fruit), 1 related to dressing (buttoning shirts), and 1 related to the ability to use a key to unlock doors.

Compared to patients with greater hand disability, the CHFS-6 had relatively higher standard error among patients with minimal disability ($\theta < -1$). This occurred for 2 reasons. First, the short-forming procedure prioritized maximizing information where the original form measures hand disability well (θ between -1 and 5). Second, the minimal estimated factor score for patients in our sample was $\theta = -1.26$, resulting in little data from patients in the lower end.

The exact specification of the OTA procedure that we used resulted in candidate short forms that were not required to be nested, meaning that the items of one short form were not required to be contained in all larger short forms. For example, item 8 is in the 2-item short form, but does not appear in the 3-item or 4-item forms. This reflects the shape-maintaining property of our OTA specification that allows for a push-andpull dynamic between items that have more information at different locations of the latent trait continuum. However, if the creation of nested short forms is desired, the methodology used in this study could be easily adapted to satisfy this constraint by selecting each subsequent optimal candidate short form only from the items appearing on the optimal short form 1 item longer. Similarly, OTA does not automatically consider content validity, but these constraints, such as setting a maximum number of items per content area, may be added as desired. However, if a short form indeed performs virtually equivalently to its parent form, despite the lack of content in some subareas due to elimination of items, this suggests that the content may not be necessary for optimal measurement, regardless of theoretical suggestions otherwise.

Although this study focused on the development and validation of a short form of the CHFS for patients with SSc, the OTA approach that we used could be applied with other patient populations and other measures for developing short forms of PRO measures. The present study, however, represents only a first step in using OTA methods to attempt to standardize processes for developing optimally functioning shortened PRO measures in health research. The SPIN Cohort is a convenience sample, and therefore may not be representative of the SSc population. Patients in the present study, for instance, had somewhat lower hand disability, on average, compared to other SSc cohorts where the CHFS has been used (15,16).

Further research is needed to determine the robustness of the OTA procedure with other measures and patient cohorts and to compare to other short-forming methods. The Bonferroni correction used to account for issues of multiple testing may be overly conservative, albeit easily applied, and alternative approaches may be preferred. Furthermore, additional research is needed on how best to use OTA to shorten scales in the context of multidimensionality. Finally, OTA is an exploratory, data-driven approach, and results of this study should be replicated.

In summary, this study demonstrated how OTA methods can be used to develop and validate short forms of PRO measures based on prespecified and objective criteria for determining both the number of items to include in the short form and the specific items to be included. Application of OTA methods to the 18-item CHFS in a large sample of SSc patients resulted in a 6-item version with minimal loss of information and minimal change to indices of reliability and convergent validity compared to the 18-item form.

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. Harel 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.

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