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The assessment of psychological well-being in systemic sclerosis: a clinimetric validation

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Abstract

Background The assessment of psychological well-being has been largely neglected in clinical settings, particularly in patients with systemic sclerosis (SSc), where the focus of clinical attention was mainly on symptoms. This is the first study in which the validity, reliability, and sensibility of two patient-reported outcome measures (PROMs) of psychological well-being, the five-item World Health Organization Well-Being Index (WHO-5) and the six-item version of the Ryff's Psychological Well-Being Scales (PWB-6), have been tested according to clinimetric criteria to determine their current and potential clinical applications in SSc patients.

Methods A cross-sectional study was conducted involving 219 patients with a diagnosis of SSc. Rasch and Mokken analyses were performed to assess the clinimetric properties of the two PROMs and determine their clinical utility.

Results All items of WHO-5 and PWB-6 fitted the Rasch model, had an optimal scalability, and the dimensionality analyses yielded less than 5% of significant *t*-tests, thus indicating that the two PROMs were unidimensional measures. Person separation reliability indices revealed acceptable internal consistency and inspection of the person-item distribution map showed that WHO-5 and PWB-6 were reasonably well-targeted for use with SSc patients.

Conclusions Findings indicate that WHO-5 and PWB-6 are valid indices of psychological well-being that may provide unique prognostic information and help researchers and clinicians tailor personalized treatment strategies. The two PROMs can be used jointly but for different clinical purposes. WHO-5 is particularly suitable to assess the degree of subjective vitality, a positive feeling of aliveness and energy that may help SSc patients cope with their illness. The PWB-6 can be used to identify unique experiences of psychological well-being that may help SSc patients not only cope with their feelings of loneliness and uncertainty but also experience a meaningful life despite the progression of disease. In clinical research and daily practice, the baseline and follow-up use of WHO-5 and PWB-6 may thus lead to a substantial improvement in the quality of care of patients with SSc. Given the cross-sectional design of the present investigation, future prospective studies are, however, recommended to further assess the predictive validity and prognostic utility of the two PROMs.

Keywords Assessment, Clinimetrics, Patient-reported outcome measures, Psychological well-being, Systemic sclerosis, PWB-6, Rasch analysis, WHO-5

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Introduction

Systemic sclerosis (SSc) is a rare and chronic autoimmune disease characterized by an initial inflammatory phase followed by fibrosis and vasculopathy affecting the skin and multiple internal organ systems [1]. This clinical condition poses significant challenges to both clinicians and patients, who must cope with a progressive disease and an increasing subjective sense of uncertainty about clinical outcomes and complications that may occur [1, 2]. Being the patient's perception of SSc of paramount importance, clinical practice guidelines [2] and literature [3, 4] recommended the use of patient-reported outcome measures (PROMs) to ensure accurate and comprehensive assessment of patients' subjective experiences that provide unique insight about severity and impact of the disease. It has also been found that, for SSc patients, alterations in functional capacity and appearance are often more distressing and debilitating than changes in objective measures of disease, implying that PROMs may detect their unique needs and subjective experiences and reveal treatment effects that are not always fully captured by clinician-rated tools [4]. However, the interest in PROMs and their use in clinical research and practice, particularly in patients with SSc, are still limited and largely driven by a disease bias, being mainly focused on self-report measures of psychological distress (e.g., depression and anxiety) rather than on indices of subjective well-being [5]. In addition, a considerable amount of clinical attention was devoted to health-related quality of life [6], a concept that has been largely criticized for being too broad and unspecific, and therefore of limited clinical utility. Health-related quality of life is often defined based on the absence of symptoms, whereas psychological well-being usually refers to the presence of positive emotions and subjective life satisfaction [3, 6, 7]. Yet, there is strong evidence indicating that psychological well-being may play a significant role in improving treatment outcomes in SSc patients [3, 8]. In Santiago et al. [3], adaptive personality traits associated with overall subjective well-being were found to mitigate the perceived impact of SSc. Similarly, in a randomized controlled trial involving a small sample of SSc patients [8], a well-being-based intervention was found to produce not only a significant improvement in subjective experiences of well-being, personal growth, and self-acceptance but also a substantial decrease in psychological distress, mental pain and suffering. Previous research has also found a significant relationship between psychological well-being and lower levels of C-reactive protein and interleukin-6, two inflammatory biomarkers that are particularly relevant for patients with SSc given their known involvement in disease activity and impaired long function [9, 10].

The pursuit of psychological well-being in patients with SSc requires a careful assessment procedure, which should be performed using suitable evaluation methods. Over the years, a number of PROMs for the evaluation of psychological well-being have been proposed [11–13] but very few have been used in SSc [3, 8]. None were validated according to clinimetric principles of reliability, construct validity, and sensibility [14]. The clinimetric concept of sensibility refers to the assessment of the suitability of a measurement tool for clinical use and encompasses criteria such as feasibility, interpretability, brevity, and clarity of instructions that represent essential features to determine the ease with which a rating scale can be used and analyzed in a specific clinical setting and for a specific clinical purpose [14].

Clinimetrics [15], the science of clinical measurements [16], suggests that a patient-reported outcome measure must be reliable, valid and, more importantly, sensible (i.e., relatively brief, easy to administer and score, acceptable to both clinicians and patients). The five-item version of the World Health Organization Well-Being Index (WHO-5) is one of the most easy-to-use, short, and valid PROMs of subjective well-being [17]. The WHO-5 was originally developed for assessing well-being in primary care patients and it was subsequently extensively used in general population studies and in a wide range of clinical conditions, mainly as a screening measure for depression [17]. Two recent studies [18, 19] adopted the WHO-5 to evaluate psychological well-being in patients with SSc but measurement properties were not tested. Thus, clinimetric reliability, validity, and sensibility of WHO-5 in patients with SSc are not known.

The six-item version of the Ryff's Psychological Well-Being Scales, the PWB-6 [20], is another brief and easy-to-use index of subjective well-being, derived from the original 84-item version of the scale [21] using a single-item per dimension approach [22]. Stavraki et al. [20] tested the psychometric properties of the PWB-6 in a sample of 312 children and adolescents, showing that it was a unidimensional measure of psychological well-being. PWB-6 has never been used in patients with SSc, thus data regarding its clinimetric properties and clinical utility are not available for this specific clinical population.

This is the first validation study in which measurement properties and clinical utility of WHO-5 and PWB-6 have been assessed based on Clinimetric Patient-Reported Outcome Measures (CLIPROM) criteria [14]. The major aim was to determine the construct validity [14, 23, 24] testing whether WHO-5 and PWB-6 were valid indices of the latent dimensions of psychological well-being that the two PROMs are expected to measure. Reliability and sensibility or the ease of use of WHO-5 and PWB-6 were also tested. The following research questions have guided the present clinimetric analysis: to what extent can the WHO-5 and PWB-6 be used to detect and quantify the

subjective well-being of patients with SSc? What are the other potential clinical applications of the two PROMs in clinical practice and research involving SSc patients?

Methods

Study design

This is a single-center cross-sectional study, which is part of a larger, prospective randomized controlled trial aimed at testing the efficacy of a psychological well-being-based intervention in SSc patients [8].

Sample and recruitment procedures

The sample consisted of a total of 219 patients with a diagnosis of SSc. Participants were consecutive outpatients who were enrolled at the Scleroderma Unit of the Careggi University Hospital (Florence, Italy) from June 2020 to October 2022. The Scleroderma Unit of the Careggi University Hospital is a reference center of the Tuscany Region for patients with SSc. To be included in the present study, patients had to meet the following eligibility criteria: age ≥ 18 years; Italian mother-tongue; diagnosis of SSc according to 2013 classification criteria proposed by the American College of Rheumatology in collaboration with the European League Against Rheumatism [25]. Exclusion criteria were: having a psychiatric disorder (i.e., anxiety and mood disorders, obsessive-compulsive and related disorders, post-traumatic stress disorder, psychotic disorders, substance/alcohol use disorders, eating disorders, antisocial personality disorder) as detected with the Mini-International Neuropsychiatric Interview (MINI) [26]; having any other condition (e.g., intellectual disabilities or neurodevelopmental disorders or specific learning disorders with impairments in reading and/or in written expression based on information gathered during clinical interviewing) that might alter patient's ability to follow the study procedures. Data were collected by experienced clinical investigators, who were previously trained in the use of the MINI [26] and PROMs. All patients were asked to self-rate their experiences of subjective well-being and provided a written informed consent to participate. The research protocol (ID: WBTinSSC) was approved by the Ethics Committee of the Tuscany Region, Central Vast Area (Florence, Italy). All study procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2013.

Patient-Reported outcome measures

Two PROMs, the WHO-5 [11, 17] and the PWB-6 [20], which require an average completion time of less than 5 min, were selected for their brevity and simplicity, a data collection procedure that may significantly reduce the cognitive burden of SSc patients when responding to the survey.

WHO-5 is a widely used patient-reported outcome measure of psychological well-being [11, 17]. The items (i.e., (1) "I have felt cheerful and in good spirits"; (2) "I have felt calm and relaxed"; (3) "I have felt active and vigorous"; (4) "I woke up feeling fresh and rested"; (5) "My daily life has been filled with things that interest me") are positively worded [17]. Each item is rated on a 6-point Likert scale with responses ranging from 0 (i.e., at no time) to 5 (i.e., all of the time) based on how the respondent has been feeling over the past 2 weeks [17]. The Italian version of WHO-5 [27], freely available at https://www.psykiatri-regionh.dk/who-5/Documents/WHO5_Italian.pdf, was used in the present study. This version was found to be a valid cross-cultural measure of psychological well-being [27]. The Italian version of the WHO-5 was also found to have an acceptable scalability with a Loevinger's coefficient of homogeneity of 0.61 and dimensionality analysis showed that less than 5% of *t*-tests were significant, thus indicating that this is a unidimensional measure of subjective well-being [27].

PWB-6 [20, 28] is the brief version of the original 84-item Ryff's Psychological Well-Being Scales [21]. The PWB-6 includes the following items: (1) "In general, I feel confident and positive about myself"; (2) "I know that I can trust my friends, and they know they can trust me"; (3) "I have confidence in my opinions, even if they are contrary to the general consensus"; (4) "I have been able to build a living environment and a lifestyle for myself that is much to my liking"; (5) "I feel good when I think of what I have done in the past and what I hope to do in the future"; (6) "I have the sense that I have developed a lot as a person over time" [20]. Each item, rated on a 6-point Likert scale ranging from 1 (i.e., strongly disagree) to 6 (i.e., strongly agree), is intended to measure the following six dimensions of psychological well-being: self-acceptance (i.e., acknowledging and accepting multiple aspects of self, including good and bad qualities, and feeling positive about past life), positive relations with others (i.e., having satisfying relationships with others, being concerned about the welfare of others, and being capable of strong empathy, affection, and intimacy), autonomy (i.e., being a self-determining and independent person who regulates behavior from within and evaluates self by personal standards), environmental mastery (i.e., making effective use of surrounding opportunities, and being able to choose or create contexts suitable to personal needs and values), purpose in life (i.e., having aims and objectives for living, feeling there is meaning to present and past life, and holding beliefs that give life purpose), and personal growth (i.e., being open to new experiences, having sense of realizing his or her potential, and seeing self as growing and expanding over time)

[20, 28]. PWB-6 has shown adequate internal reliability and good factorial validity [20, 28]. The items constituting the PWB-6, which was used in the present research, have been obtained from the Italian version of the Ryff's Psychological Well-Being Scales [29]. There are no studies on the reliability and validity of the Italian version of PWB-6 and the present is the first study aimed at testing the clinimetric properties of this brief measure in patients with SSc. Given the lack of validated measures of psychological well-being in this clinical population, the findings of the present study may not only contribute to the clinimetric validation of PWB-6 but also favor the creation of new indices for a comprehensive assessment of subjective well-being in SSc patients.

Statistical analyses

A comprehensive assessment of construct validity was performed using both Rasch [23] and Mokken [24] analyses to test whether WHO-5 and PWB-6 can be used as dimensional measures for identifying and assessing levels of psychological well-being in patients with SSc.

Given the polytomous structure of WHO-5 [11, 17] and PWB-6 [20], including items with more than two response categories, the partial credit model, which assumes the distance between item thresholds to be different across all items [23], was used. Rasch analysis [23] was conducted using Rasch Unidimensional Measurement Models (RUMM2030) software to assess the following clinimetric properties:

- The overall fit to the model, which was tested using the chi-square item-trait interaction statistic [23]. The overall fit provides a summary measure of the extent to which the WHO-5 and PWB-6 conform to the Rasch model measurement expectations [23]. A non-significant chi-square probability value indicates overall fit to the Rasch model [23].
- Individual fit statistics were evaluated to detect specific item-level or person-level misfit to the Rasch model [23]. Individual fit statistics are presented for each item as a fit residual and as a chi-square probability statistic [23]. Fit residuals between ± 2.5 and non-significant chi-square probability values for each item of WHO-5 and PWB-6 indicate adequate fit to the Rasch model [23].
- The dimensionality of WHO-5 and PWB-6, which was evaluated using Principal Component Analysis (PCA) of standardized residuals to identify the two most different subsets of items (i.e., the most positively and negatively factor-loading items on the first component) [23]. Following this, paired *t*-tests were conducted to compare the scores across these two subsets [23]. If more than 5% of *t*-tests were significant, WHO-5 and PWB-6 were not considered unidimensional measures of psychological well-being.
- The local dependency was tested to assess whether the response to one item will determine the response on another item after controlling for the underlying construct under examination [23]. Based on the Rasch model assumption of local independence, there should be no significant correlation among item residuals [23]. A residual correlation value > 0.20 above the average of all item residual correlations indicates the presence of local dependency between items [23].
- Differential Item Functioning (DIF), which is a form of item bias, was evaluated to assess whether different groups of participants within the sample (e.g., males and females) respond differently to an item despite equal levels of the underlying trait under evaluation [23]. DIF was examined for sex (male and female), and age groups (below and above median age).
- Person Separation Reliability Index (PSI) was calculated to evaluate the internal consistency of WHO-5 and PWB-6 and estimate their clinimetric sensitivity (i.e., the ability of the two PROMs to discriminate among SSc patients with different levels of psychological well-being) [23]. A PSI ≥ 0.7 indicates that the two PROMs could reliably distinguish between different groups, while a PSI ≥ 0.85 suggests that the WHO-5 and PWB-6 could reliably discriminate between subjects with different levels of subjective well-being [23]. PSI values < 0.7 indicate that the reliability and clinimetric sensitivity of WHO-5 and PWB-6 are not acceptable [23].
- Sensibility was verified to test how well-targeted WHO-5 and PWB-6 items were for SSc patients [23]. Comparison of the mean location score for persons with the mean value of zero set for items provided an indication of how well targeted the items of WHO-5 and PWB-6 were for patients with SSc [23]. For a well-targeted measure, the mean location score for persons, as revealed by inspection of the person-item threshold distribution map, would also be around the value of zero [23]. A high positive mean score indicates that items of WHO-5 and PWB-6 are easy to be replied, while a negative mean score indicates that items are too hard to be replied [23].

Mokken analysis [24] was run using Stata statistical software, version 14.2 (Stata Corporation, College Station, TX). It was conducted to assess the scalability of WHO-5 and PWB-6, that is testing whether each item of the two PROMs belonged to the same underlying dimension and provided distinctive clinical information [24]. Mokken

Table 1 Demographics ($n = 219$)

Variable	Description	*n* (%)
Sex	Female	195 (89%)
	Male	24 (11%)
Age, Mean (SD)		58.55 (13.85)
Education	No graduation	19 (8.68%)
	Primary school	63 (28.77%)
	Highschool	89 (40.64%)
	Graduation	45 (20.55%)
	Postgraduation	3 (1.37%)
Work	Blue collar	15 (6.85%)
	White collar	46 (21.00%)
	Artisan	10 (4.57%)
	Freelance	18 (8.22%)
	Manager	2 (0.91%)
	Retired	74 (33.79%
	Student	7 (3.20%)
	Looking for job	4 (1.83%)
	Other	43 (19.63%)
Marital status	Single	31 (14.16%)
	Married or in a relationship	145 (66.21%)
	Divorced	13 (5.94%)
	Separated	8 (3.65%)
	Widow	22 (10.05%)
Residential area	Urban	121 (55.25%)
	Rural	98 (44.75%)
Summary score, Mean (SD)	WHO-5	56.50 (21.85)
	PWB-6	28.14 (5.83)

Note: PWB-6, six-item version of the Ryff's Psychological Well-Being Scales; SD, standard deviation; WHO-5, five-item version of the World Health Organization Well-Being Index

analysis was also performed to examine the extent to which WHO-5 and PWB-6 total scores were statistically sufficient measures of psychological well-being. According to Bech [24], Loevinger's coefficients of homogeneity [30], ranging from 0.30 to 0.39, suggest a just acceptable scalability, while a Mokken coefficient ≥ 0.40 indicates optimal scalability.

Results

Descriptive statistics

Demographic characteristics (e.g., age, sex, education) of the sample are shown in Table 1. A total of 221 patients with SSc were invited to participate to the study. The majority of them (i.e., 219 patients) agreed to participate in the study and fulfilled the inclusion criteria. Most of the patients were female (89%) and married or in a relationship (66.21%). The mean age was 58.55 years. Mean scores and standard deviations for WHO-5 and PWB-6 total scores are shown in Table 1.

Overall fit to the Rasch model

Summary fit statistics for the WHO-5 are shown in Table 2. A statistically significant item-trait interaction statistic ($\chi^2 = 20.10$; degrees of freedom [df] = 10; $p = 0.028$), which indicated an overall misfit to the Rasch model, was found. Summary fit residuals for items (0.36) and persons (-0.42) were both within acceptable limits (Table 2).

The summary fit statistics for PWB-6 are presented in Table 2. A non-significant item-trait interaction chi-square statistic ($\chi^2 = 12.93$; df = 12; $p = 0.374$) was detected, thus indicating overall fit of PWB-6 to the Rasch model (Analysis 1). PWB-6 fitted the Rasch model expectations ($\chi^2 = 13.70$; df = 10; $p = 0.187$) even after combining PWB-6 locally dependent items (Table 2, Analysis 2). The summary fit residuals for items and persons were both within the acceptable limits of ± 2.5 (Table 2, Analysis 1–2).

Individual fit to the Rasch model

Individual item fit statistics for the WHO-5 are reported in Table 3. Fit residuals and chi-square probability values showed adequate fit to the Rasch model for individual items of the WHO-5. Individual item fit statistics for PWB-6 are reported in Table 3. Fit residuals and chi-square probability values indicated adequate fit to the Rasch model for individual items of the PWB-6.

Table 2 Model fit statistics for WHO-5 and PWB-6

Action	Analysis	Model fit (overall)	Item fit residual, mean (SD)	Person fit residual, mean (SD)	PSI	Unidimensionality, significant t-tests (CI 95%)	Local dependency (residual correlations > 0.20 above average)
WHO-5							
Original sample ($n = 216$)	1	$\chi^2(10) = 20.10$, $p = 0.028$	0.36 (1.02)	-0.42 (1.11)	0.85	3.70 (1.68–14.32)	None
PWB-6							
Original sample ($n = 201$)	1	$\chi^2(12) = 12.93$, $p = 0.374$	0.43 (0.72)	-0.36 (1.16)	0.74	2.49 (-1.06-11.06)	PWB items 3 & 6
Subtest analysis, collapse of PWB items 3 & 6	2	$\chi^2(10) = 13.70$, $p = 0.187$	0.33 (0.48)	-0.36 (1.11)	0.73	1.49 (-3.06-9.06)	None

Note: χ^2, chi-square; *p*, probability; PWB-6, six-item version of the Ryff's Psychological Well-Being Scales; SD, standard deviation; PSI, person separation reliability index; WHO-5, five-item version of the World Health Organization Well-Being Index

Table 3 Individual item fit statistics for WHO-5 and PWB-6 ($n = 216$)

Item	Location	Fit residual	χ^2	Probability*
WHO-5				
WHO item 1	-0.019	0.042	4.748	0.093
WHO item 2	-0.074	-0.168	3.076	0.215
WHO item 3	0.142	-0.294	1.972	0.373
WHO item 4	0.450	0.053	3.095	0.213
WHO item 5	-0.499	2.155	7.205	0.027
PWB-6				
PWB item 1	0.382	0.024	1.602	0.449
PWB item 2	-0.126	1.016	1.002	0.606
PWB item 3	-0.025	1.327	1.790	0.409
PWB item 4	0.029	-0.426	6.567	0.038
PWB item 5	0.060	0.836	0.595	0.743
PWB item 6	-0.320	-0.197	1.378	0.502

*Bonferroni adjusted at 1% level. Note: PWB-6, six-item version of the Ryff's Psychological Well-Being Scales; WHO-5, 5-item version of the World Health Organization Well-Being Index

Table 4 Mokken analysis of WHO-5 and PWB-6 individual items

Patient-Reported Outcome Measures			
WHO-5		**PWB-6**	
Item	Coefficients of scalability	Item	Coefficients of scalability
WHO item 1	0.62	PWB item 1	0.46
WHO item 2	0.63	PWB item 2	0.43
WHO item 3	0.63	PWB item 3	0.41
WHO item 4	0.61	PWB item 4	0.50
WHO item 5	0.54	PWB item 5	0.34
		PWB item 6	0.49

Note: PWB-6, six-item version of the Ryff's Psychological Well-Being Scales; WHO-5, five-item version of the World Health Organization Well-Being Index

Dimensionality analysis

Dimensionality analysis of WHO-5 showed no significant *t*-tests outside the critical value of 5%, thus supporting the unidimensionality of this measure (Table 2). Dimensionality analyses of PWB-6 yielded less than 5% of significant *t*-tests, thus indicating that this is a unidimensional measure of psychological well-being (Table 2, Analysis 1–2).

Mokken coefficients of scalability

Table 4 shows the Mokken coefficients of scalability for individual items of WHO-5 and PWB-6. Loevinger's coefficients of homogeneity for WHO-5 items ranged from 0.54 to 0.63, thus indicating an optimal scalability. WHO-5 total score had a Loevinger's coefficient of homogeneity of 0.61, which indicated optimal scalability. Loevinger's coefficients of homogeneity for PWB-6 items ranged from 0.34 to 0.50, thus suggesting acceptable scalability (Table 4). PWB-6 total score had a Loevinger's coefficient of homogeneity of 0.44, which indicated optimal scalability.

Local dependency

No indication of local dependency between items of the WHO-5 was found (Table 2). Indication of local dependency was observed between items 3 and 6 of the PWB-6 (Table 2).

Differential item functioning (DIF)

There was no indication of statistically significant DIF for items of the WHO-5. PWB-6 item 5 showed a non-uniform DIF for sex (see also Supplementary Figure S1).

Person separation reliability index (PSI)

WHO-5 had a PSI of 0.85 indicating that this measure displayed high reliability and could, thus, reliably be used to differentiate between SSc patients with different levels of psychological well-being (Table 2). PWB-6 had a PSI of 0.74, which indicated that the scale displayed acceptable reliability and could, therefore, reliably be used to distinguish between groups of SSc patients with different levels of psychological well-being (Table 2, Analysis 1).

Sensibility or targeting of WHO-5 and PWB-6

Inspection of the person-item distribution map showed that the WHO-5 was well-targeted for use with SSc patients (see Supplementary Figure S2). Supplementary Figure S3 revealed that also the PWB-6 was reasonably well-targeted for use with SSc patients.

Correlation

A low correlation ($r = 0.35$) between WHO-5 and PWB-6 was found.

Discussion

Merkel [31] and then Thombs et al. [32] advocated the use of self-assessment instruments to gain a broader understanding of the impact of SSc and of therapeutic interventions on subjective well-being of patients with SSc. However, relatively little attention has been devoted to the development, validation, and use of PROMs of psychological well-being in clinical settings, particularly in patients with SSc, where studies testing the validity of measures of psychological well-being are remarkably lacking. This is the first study in which reliability, validity, sensibility, and clinical utility of PROMs of psychological well-being were tested in patients with SSc. The WHO-5 was found to have high reliability, making this index suitable for assessment at the individual level, particularly when used for distinguishing between SSc patients with different degrees of subjective well-being. No evidence of local dependency between items was found. There was no indication of DIF and dimensionality analysis indicated that the WHO-5 is a unidimensional measure of subjective well-being. The total score and items of the WHO-5 also displayed an optimal scalability with Loevinger's

coefficients of homogeneity ranging from 0.54 to 0.63. All the individual items fitted the Rasch model expectations but the initial inspection of WHO-5 showed poor overall fit. This issue requires further exploration in future validation studies to identify the potential reasons for misfit.

Findings regarding PWB-6 indicated acceptable reliability, making this index particularly suitable for use in groups of SSc patients with different degrees of psychological well-being. PWB-6 showed overall and individual item fit to the Rasch model expectations. Dimensionality analysis indicated that the PWB-6 is a unidimensional measure of psychological well-being and Mokken coefficients of homogeneity further confirmed the unidimensionality or scalability of the PWB-6 total score and individual items. It should be, however, noted that item 5 of the PWB-6 showed the lowest Loevinger's coefficient of homogeneity (H = 0.34), thus raising some concerns about its unique contribution to the underlying construct of psychological well-being. Future modifications of the PWB-6 may, therefore, consider revising the wording of item 5 to improve its scalability, thus ensuring better alignment with the underlying construct of psychological well-being. Attention should also be paid to local dependency, which was found between items 3 and 6, thus suggesting that the two items may provide clinically redundant information. In future iterations of the scale, clinical investigators should, therefore, consider removing one of the dependent items or grouping them together into a single item aiming to encapsulate the original two items. A non-uniform DIF for sex was also found in the item 5 of PWB-6. Revising the wording of this item may, thus, be necessary to eliminate this form of item bias.

Compared to PWB-6, WHO-5 demonstrated better reliability and scalability, with stronger coefficients of homogeneity, thus suggesting that it forms a more cohesive measure of psychological well-being. Nevertheless, the two PROMs were found to entail the clinimetric properties of construct validity, reliability, and sensibility [14], thus allowing their use in clinical research and practice with SSc patients.

Clinical applications

In response to the first research question of this clinimetric validation, WHO-5 and PWB-6 were found to be highly valid dimensional indices that can be used to assess the degree of psychological well-being in SSc patients. In clinical (e.g., psychotherapy or drug) trials and in daily practice, the use of WHO-5 and PWB-6 can significantly improve the detection of clinically relevant changes (including worsening or improvement) in psychological well-being over time (i.e., during treatment or over the course of the disease).

WHO-5 and PWB-6 can also be used as outcome measures to test the efficacy of medical or psychological interventions and to evaluate the impact of treatments on subjective well-being of patients with SSc. In his monograph on clinimetrics, Feinstein [33] recommended the use of outcome measures to describe patient's own assessment of well-being during treatment and Bech [34] consistently remarked that, if systematically used in clinical research and daily practice, effective tools to detect and monitor the course of psychological well-being can increase patients' compliance to prescribed medications and significantly improve the subjective experience of care. The need to understand the patients' perspectives and perceptions of living with SSc appeared to be particularly crucial to optimize the patient-clinician communication and interaction and to facilitate a shared-decision making process [3, 35–37]. WHO-5 and PWB-6 have the potential to be used to enhance patients' engagement and identify their priorities for care that may guide personalized treatment strategies targeting symptom decrease and well-being promotion [3].

In response to the second research question of the present study, other possible clinical applications of WHO-5 and PWB-6 include their use as prognostic measures to predict future outcomes or to identify high-risk SSc patients with low levels of psychological well-being. Findings on PSI indicated that WHO-5 and PWB-6 can be used to stratify patients into distinctively different prognostic groups based on their levels of subjective well-being.

Regarding sensibility, WHO-5 and PWB-6 items were found to be well-targeted (i.e., not too easy, not too hard) for use with patients with SSc. The brevity and simplicity make the WHO-5 and PWB-6 two easy-to-use indices that may have considerable screening utility. Bech et al. [38] argued that instruments measuring well-being are easier to use as screening instruments for mental disorders than those having distress or disability in their wording. The two PROMs, particularly the WHO-5 that was already used as a screening tool for depression [11, 17], may, thus, help clinicians early identify SSc patients with low levels of psychological well-being who are at risk of developing depression. Future studies are, however, warranted not only to further explore the screening utility of WHO-5 and PWB-6 but also to provide methodological guidelines to help clinical investigators and practitioners optimize the baseline and follow-up use of the two PROMs in clinical research and practice with SSc patients.

A weak positive association was found between WHO-5 and PWB-6, which were expected to be highly correlated being both measures of psychological well-being. This expectation was based on previous studies showing a strong correlation between the WHO-5 and

PWB measures of psychological well-being [20, 39, 40]. In the present study, the weak positive association suggests that the two PROMs capture distinct dimensions of psychological well-being with WHO-5 items probably reflecting a global experience of well-being characterized by feelings of subjective vitality (e.g., feeling active and vigorous, and waking up feeling fresh and rested) combined with a sense of personal satisfaction that makes life worth living (e.g., having a daily life filled with interesting things). Subjective vitality [41] and life satisfaction [42] may demarcate major prognostic and therapeutic differences among patients with SSc in terms of capacity to cope with various stages of illness and treatment and better tolerate invalidating and frequent symptoms such as pain and fatigue [43, 44]. The WHO-5 may, thus, help clinicians early recognize SSc patients with low subjective vitality, who may be at increased risk of experiencing fatigue and low energy levels. The WHO-5 may also be suitable to assess the protective role and adaptive value of subjective vitality, evaluating whether SSc patients who perceive greater subjective vitality have more favorable clinical outcomes than those with low subjective vitality.

As to the items of PWB-6, they cover the experiences of psychological well-being that Carol Ryff [21] derived from Marie Jahoda's [45] conceptualization of positive mental health, which included the dimensions of self-acceptance (i.e., having a positive attitude toward the self), positive relations with others (i.e., having warm, and trusting relationships with others), autonomy (i.e., being able to resist social pressures to think and act in certain ways), environmental mastery (i.e., having a sense of competence in managing the environment), purpose in life (i.e., experiencing a sense of directedness in life), and personal growth (i.e., having a feeling of continued development). The response to PWB-6, particularly to items 1 (to increase patients' self-acceptance in response to their altered physical appearances), 2 (to facilitate positive relations with others in order to counteract feelings of loneliness that are very common among SSc patients), and 5 (based on the evidence [46, 47] that having a purpose in life may mitigate physical suffering), may help investigators and clinicians tailor individualized treatment plans to the unique needs of patients with SSc. The PWB-6 might, therefore, be better suited to evaluate the beneficial effects that well-being experiences of self-acceptance, positive relations with others, autonomy, environmental mastery, purpose in life, and personal growth may have on the course of illness, particularly in SSc patients dealing with social isolation and existential concerns. This implies that the PWB-6 may also be used to early recognize SSc patients with low levels of psychological well-being, who may be more vulnerable to feelings of loneliness and also at increased risk of experiencing existential frustration.

Limitations

Given the cross-sectional design, this clinimetric validation study does not allow firm conclusions regarding WHO-5/PWB-6 predictive or incremental validity. This is the main limitation of the present study, implying that future research adopting a longitudinal design is needed to determine the prognostic utility of WHO-5 and PWB-6, particularly to determine the ability of the two PROMs not only to predict short- and long-term outcomes but also to evaluate quality of life trajectories and the protective role that experiences of psychological well-being may have on disease progression and treatment adherence. Another limitation of this clinimetric validation has to do with the adequacy of the sample size, which was not verified based on research that suggests the inclusion of 250–500 respondents as a reasonable guideline [23, 48, 49]. Given the number of items and thresholds of WHO-5 and PWB-6, a target sample size of at least 250 SSc patients is recommended in future studies for conducting a reliable Rasch analysis [23, 48, 49].

Conclusion

SSc is a healthcare challenge, which requires the expertise of different researchers and clinicians (including clinical psychologists) for a comprehensive evaluation and treatment of this complex clinical condition.

Psychological well-being of SSc patients is in need of being adequately assessed and promoted since it may provide protection against painful and distressing experiences and facilitate adaptive coping strategies that have the potential to improve short- and long-term outcomes.

The findings of this clinimetric validation indicate that WHO-5 and PWB-6 can be incorporated into the standard clinical assessment of patients with SSc since they may capture unique prognostic information and help tailoring individualized treatment strategies. The routine assessment of psychological well-being at each treatment and follow-up visit through the repeated use of WHO-5 and PWB-6 has also the potential to improve the quality of patient care.

Regarding the specific clinical applications of the two PROMs, WHO-5 and PWB-6 can be used jointly but for different clinical purposes. WHO-5 is indicated to assess the degree of subjective vitality, a positive feeling of aliveness and energy [41] that may help SSc patients cope with their illness. Regarding the PWB-6, it is particularly suitable to identify distinctive experiences of psychological well-being that may help SSc patients not only cope with their feelings of loneliness and uncertainty but also experience a meaningful life despite the progression of disease.

Abbreviations

SSc	Systemic Sclerosis
PROMs	Patient-Reported Outcome Measures

WHO-5	5-item version of the World Health Organization Well-Being Index
PWB-6	6-item version of the Ryff's Psychological Well-Being Scales
CLIPROM	Clinimetric Patient-Reported Outcome Measures
MINI	Mini-International Neuropsychiatric Interview
RUMM2030	Rasch Unidimensional Measurement Models
PCA	Principal Component Analysis
DIF	Differential Item Functioning
PSI	Person Separation Reliability Index
df	degrees of freedom

Supplementary Information

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Supplementary Material 1

Supplementary Material 2

Supplementary Material 3

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Author contributions
DC made substantial contributions to the conception of the study and drafted the first version of the manuscript. KSC made substantial contributions to the analysis and interpretation of data. SG made substantial contributions to the acquisition of data. AM made substantial contributions to the acquisition of data. SR made substantial contributions to the acquisition of data. FC made substantial contributions to the conception and design of the study and revised the manuscript critically for important intellectual content. All authors read and approved the final version of the manuscript.

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Data availability
The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The research protocol (ID: WBTinSSC) was approved by the Ethics Committee of the Tuscany Region, Central Vast Area (Florence, Italy). All study procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2013. All patients provided a written informed consent to participate.

Consent for publication
Not applicable.

Competing interests
The authors declare no competing interests.

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