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Well-Being Therapy in systemic sclerosis outpatients: a randomized controlled trial

Sara Romanazzo[1], Sara Ceccatelli[2], Giovanni Mansueto[1], Francesco Sera[3], Serena Guiducci[2], Marco Matucci Cerinic[4], Fiammetta Cosci [2,5,*]

[1]Department of Health Sciences, University of Florence, Florence, Italy
[2]Department of Experimental and Clinical Medicine, University of Florence, Florence, Italy
[3]Department of Statistics, Computer Science and Applications G. Parenti, University of Florence, Florence, Italy
[4]Immunology, Rheumatology, Allergology and Rare Diseases Unit, San Raffaele Hospital, Milan, Italy
[5]Department of Psychiatry and Neuropsychology, Maastricht University, Maastricht, The Netherlands

*Correspondence to: Fiammetta Cosci, Department of Health Sciences, University of Florence, via di San Salvi, 12, 50135 Florence, Italy.
E-mail: fiammetta.cosci@unifi.it

Abstract

Objectives: Systemic sclerosis (SSc) patients have psychological distress and poor well-being and need a tailored treatment. Psychological interventions, rarely tested for efficacy, showed poor benefits. The present randomized controlled trial tested the efficacy of Well-Being Therapy (WBT) in SSc patients.

Methods: Thirty-two outpatients were randomized (1:1) to WBT ($n = 16$) or Treatment As Usual (i.e. routine medical check-ups) (TAU) ($n = 16$). Primary outcome was well-being. Secondary outcomes included functional ability related to SSc, psychological distress, mental pain, suffering. All participants were assessed at baseline (T0). The WBT group was assessed after two months (end of WBT session 4) (T1), after four months (end of WBT session 8) (T2), after seven months (3-month follow-up) (T3) and after 10 months (6-month follow-up) (T4). The TAU group was assessed two (T1), four (T2), seven (T3) and ten (T4) months after baseline.

Results: WBT produced a significant improvement in subjective well-being ($P \leq 0.001$), personal growth ($P = 0.006$), self-acceptance ($P = 0.003$) compared with TAU, maintained at T3 as what concerns subjective well-being ($P = 0.012$). WBT produced a greater decrease in psychological distress ($P = 0.010$), mental pain ($P = 0.010$), suffering ($P \leq 0.001$) compared with TAU, maintained at T4 as what concerns suffering ($P \leq 0.001$). Participants reported high satisfaction with WBT.

Conclusion: The study provides preliminary evidence on the benefits of WBT as short-term approach for in- and out-patient SSc healthcare paths. Studies with larger samples are needed to have the evidence for recommending WBT to SSc patients.

Keywords: systemic sclerosis, Well-Being Therapy, well-being, psychological distress, mental pain, suffering.

Rheumatology key messages

- Preliminary results suggest the benefit of WBT in SSc patients when compared to treatment as usual
- Improvement in subjective well-being was observed in the WBT group when compared to treatment as usual
- Decrease in psychological distress, mental pain and suffering was observed in the WBT group when compared to treatment as usual

Introduction

Systemic sclerosis (SSc) is a rare, chronic, multi-system autoimmune disease with an incidence ranging from 8 to 56 new cases/million/year and a prevalence ranging from 38 to 341 cases/million [1]. SSc typically affects females (4:1 risk ratio) with a median onset at 30–50 years of age [2]. The disease is characterized by fibrosis of the skin and internal organs with consequent morphological and functional changes of hands and face (mainly) [3] and organs dysfunction [4–6] – implying physical functioning [7], work ability and employment status impairments [8].

The burden related to SSc can be increased by the occurrence of psychological issues, mainly distress. Depressive symptoms have been observed in 33.4–77.4% of cases [9–11] and anxiety in 36–80% of patients [12]. In addition, SSc patients present low capacity to cope with stress [13], particularly stress due to changes of physical aspect [14], and impaired health-related quality of life [15].

Few psychological interventions have been implemented in SSc patients, they were focused on psychoeducation [16,17] or distress reduction [18] and produced moderate effects. Psychoeducation provided patients with appropriate medical

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information about SSc, gave them the opportunity to discuss the symptoms and compare own experiences with other patients [16,17]. Mindfulness-based stress reduction therapy ameliorated anger control and expression, sleep quality and stress perception [18].

No interventions focusing on well-being empowerment/promotion have been implemented up to now, besides such interventions showed clinical utility or promising results in patients with chronic organic diseases [19–21]. Among interventions focusing on promoting and empowering psychological well-being rather than on distress, Well-Being Therapy (WBT) [22] is a short-term psychotherapeutic strategy which allows to work on and empower dimensions such as autonomy, environmental mastery, satisfactory interactions with other people and the milieu, individual growth, self-actualization, self-perception and acceptance, balance and integration of psychic forces. WBT showed to improve well-being and psychological distress in type 2 diabetes [19]; reduced self-rated pain intensity and favoured remission of residual depressive symptoms in a patient with recurrent depression and chronic pain [20]; decreased the number of migraine attacks and migraine disability, increased well-being and improved depressive symptoms in a patient with chronic migraine and depression [21].

In this framework, the present research aimed at testing the efficacy of Well-Being Therapy [22] in SSc patients to verify whether it can increase psychological well-being (primary outcome) and decrease psychological distress (secondary outcome).

Methods

Participants

Outpatients were enrolled at the Scleroderma Unit of the University Hospital Careggi (Florence, Italy) from June 2020 to October 2022. Inclusion criteria were: age $\geq$ 18 years; Italian mother-tongue; diagnosis of systemic sclerosis according to the 2013 American College of Rheumatology/European League Against Rheumatism classification criteria [6]; willingness to participate. Exclusion criteria were: co-occurrence of mental disorder(s) according to the Diagnostic and Statistical Manual of mental disorders, fifth edition (DSM-5) as diagnosed via the MINI International Neuropsychiatric Interview (MINI) [23]; currently undergoing a psychotherapy; change in drug therapy (including psychotropic medications) within the past three months; any other condition that might alter patient's ability to follow the study procedures. Participants were Caucasic. Among them, 15 (47%) lived in a rural area and 17 (53%) lived in an urban one. The trial was conducted according to CONSORT guidelines and registered in ClinicalTrials.gov (NCT04212247).

Design

This is a mono-centre, two-arm parallel, randomized controlled trial with Treatment as Usual (TAU) (i.e. routine medical check-ups, routine laboratory and instrumental examinations, when necessary analgesic/antidepressant/pregabalin prescription) as comparator. Participants gave a written informed consent at enrolment. Thereafter, demographic and clinical information were collected by clinical psychologists who administered the MINI [23]. Patients were assessed via the Health Assessment Questionnaire-Disability Index (HAQ-DI) [24], the 5-item World Health Organization Well-Being Index (WHO-5) [25], the Psychological Well-Being scales (PWB) [26], the Mental Pain Questionnaire (MPQ) [22], the Symptom Checklist-90-Revised (SCL-90-R) [27] and the Pictorial Representation of Illness and Self Measure (PRISM) [28].

Randomization to the experimental (WBT group) or the TAU condition was performed by an independent researcher using the random number generator of http://www.random.org and a block randomization of size two. The two groups were matched *a posteriori* for sex, age and severity of systemic sclerosis.

All patients included in the trial were assessed at enrolment (i.e. baseline—T0). Thereafter, the WBT group was assessed after two months (T1) (end of WBT session 4), after four months (T2) (end of WBT session 8), after seven months (T3) (3-month follow-up) and after 10 months (T4) (6-month follow-up). At T1, HAQ-DI, WHO-5, PWB, MPQ, SCL-90-R, PRISM and Kellner Scale [29] were proposed. At T2, MINI, HAQ-DI, WHO-5, PWB, MPQ, SCL-90-R, PRISM and Kellner Scale were administered. At T3 and T4, HAQ-DI, WHO-5, PWB, MPQ, SCL-90-R, PRISM and Kellner Scale were administered.

The TAU group was assessed two (T1), four (T2), seven (T3) and ten (T4) months after baseline evaluation. MINI, HAQ-DI, WHO-5, PWB, MPQ, SCL-90-R and PRISM were administered.

The protocol here used was unanimously approved by the Ethical Committee of the Tuscan Region members (protocol number 16425_spe of the 25/02/2020). All 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. The trial was conducted according to the CONSORT guidelines and registered in ClinicalTrials.gov (NCT04212247) (outcomes here presented as secondary were mentioned as primary in the registered protocol. This change is related to the fact that WBT, being aimed at empowering or promoting well-being, is expected to have primarily an effect on well-being and only secondary an effect on variables related to psychological distress). The full trial protocol can be accessed upon request to the corresponding author.

Intervention

WBT was delivered in 8 individual, in person sessions every other week, with a duration of 45–60 min each [22] by a psychological psychotherapist trained in WBT and having a clinical experience in conducting WBT of 3 years. Patients were encouraged to: identify episodes of well-being and set them in a situational context with the use of a structured diary; identify thoughts, beliefs and behaviours leading to premature interruption of well-being; restructuring thoughts interrupting well-being.

The TAU group implied the maintenance of the standard of care which included pharmacological treatment for SSc clinical manifestations, routine medical check-ups, routine laboratory and instrumental examinations.

Measures

Screening measure

The MINI International Neuropsychiatric Interview [23] is a structured interview allowing to formulate the diagnosis of mental disorders (i.e. major depressive episode/disorder, suicidal behavioural disorder, bipolar disorder, panic disorder, agoraphobia, social anxiety disorder, generalized anxiety

disorder, obsessive-compulsive disorder, post-traumatic stress disorder, alcohol/substance use disorder, psychotic disorder, anorexia/bulimia nervosa, being eating disorder, antisocial personality disorder) according to the DSM-5 [30].

Functional ability related to SSc

Functional ability related to SSc was assessed using the Health Assessment Questionnaire-Disability Index [24], a 20-item self-reported questionnaire assessing 8 domains: dressing, arising, eating, walking, hygiene, reach, grip, activities. Each domain is rated on a 0–3 scale, where 0 indicates 'without difficulty' and 3 indicates 'unable to do', and additional points can be added if aids or devices are needed for specific activities. The total score ranges between 0 and 3; increasing score indicates worse functionality.

Primary outcomes measures

Psychological well-being was assessed via the 5-item World Health Organization Well-Being Index [25]. Higher scores indicate higher well-being. The WHO-5 raw score ranges from 0 to 25 and is conventionally multiplied by 4 to give a percentage score from 0, representing the worst imaginable well-being, to 100, corresponding to the best imaginable state of subjective well-being [25].

Psychological well-being was assessed using the Psychological Well-Being scales [26], an 84-item self-report inventory measuring autonomy; environmental mastery; personal growth; positive relations with others; purposes in life; self-acceptance. Higher scores on each scale indicate greater well-being [26].

Secondary outcomes measures

Mental pain was assessed via the Mental Pain Questionnaire, a 10-item self-report questionnaire; the total score ranges from 0 to 10; higher scores indicate greater mental pain [22].

The Symptom Checklist-90-Revised [27] was used to assess psychological symptoms and distress. It is a 90-item self-report questionnaire with 9 subscales: somatization, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, psychoticism. The Global Severity Index is part of the SCL-90-R and assesses overall psychological distress, the score ranges from 0 to 4. High scores reflect high psychopathological distress and severity of self-reported symptoms.

The Pictorial Representation of Illness and Self Measure was used as a two-dimensional pictorial method to measure the burden of suffering [28]. Subjects are shown a white A4 board with a yellow disc at the bottom right-hand corner. They are asked to imagine that the board represents current life and the yellow disc represents the own 'Self'. Then, they receive a red disc which represents their current illness and are asked to place it on the board. The main outcome is the Self-Illness Separation (SIS), i.e. the distance between the yellow and the red disc centres (SIS range: 0–27 cm). SIS is inversely related to the burden of the illness and represents the current impact of the illness in patient's life [28]. For the present study, the PRISM+ version was used, and thus additional discs were proposed to represent physical pain, feeling of peace, work and leisure time.

Treatment satisfaction was assessed at post-treatment using the Client Satisfaction Questionnaire (CSQ-8) [31], an 8-item self-report questionnaire. The total score ranges from 8 to 32, with higher scores reflecting greater satisfaction.

Credibility and expectancy regarding the treatment received were verified using the Credibility-Expectancy Questionnaire (CEQ) [32], a 6-item self-report questionnaire. CEQ presents two rating Likert scales: one from 1 (not at all) to 9 (very much) and the other one from 0% (not at all) to 100% (very much), and therefore, the percentage ratings need to be linearly transformed (values from 40–60% are collapsed into one value, i.e. 5, and the sum score for each factor ranges from 3 to 27).

The Kellner Scale [29] was proposed to self-assess the overall change after the beginning of the treatment. It is a 9-point scale where 9 corresponds to 'a lot worse' and 1 corresponds to 'a lot better'.

Participants' engagement in treatment was calculated as the number of sessions completed during the intervention period. Participants completed all outcome measures at each assessment time point.

Statistical analysis

Sample size was calculated based on previous trials on psychotherapy for SSc [18] and WBT in patients with chronic disease [33]. Based on El Aoufy *et al.* [18], a large between-group difference on the secondary outcome (Cohen's $d = 2.76$) was expected between treatment and TAU; to achieve power = 0.9 at a 0.05 (two-tailed) level of statistical significance, a minimum sample size of 5 per group was required. Based on Fava *et al.* [33], a large between-group difference on the primary outcome (Cohen's $d = 1.38$) was expected between treatment and TAU; to achieve power = 0.9 at a 0.05 (two-tailed) level of statistical significance, a minimum sample size of 15 per group was required. However, substantially more participants were screened and enrolled to reach the final sample size.

Statistical analysis was performed in SPSS, version 26 (SPSS Inc). Descriptive statistics were calculated regarding baseline demographic and clinical characteristics. No missing data were present at T0, T1, T2 and T3, while one subject per arm did not complete the 6-month follow-up assessment (T4) and the corresponding data were missed.

Generalized Estimating Equation (GEE) models were used to examine possible interaction between treatment and time, i.e. whether treatment effect changed over time, considering possible non-independence due to repeated measurement for each subject. Treatment and time were considered categorical variables and were included in the GEE model with their interaction. A global test of interaction was performed using GEE models. Marginal estimates were obtained for each time point and treatment group, and differences between treatment groups were tested using Wald test on the linear combination of coefficients obtained in the GEE models. GEE analyses were conducted for primary and secondary measures (i.e. WHO-5, PWB, MPQ, SCL-90-R and PRISM).

A $P \leq 0.05$ was considered statistically significant.

Results

Baseline demographics and clinical characteristics

Each group included 16 subjects. Demographics are reported in Table 1.

Adherence, attrition, adverse events

All participants ($n = 32$, 100% of the whole sample) completed all sessions and 3-month follow-up assessment, while

Table 1. Demographic characteristics at baseline

	WBT (n = 16)	TAU (n = 16)	Overall (N = 32)
Sex, n (%)			
Female	16 (100)	16 (100)	32 (100)
Age, mean (SD)	58.37 (12.16)	57.88 (12.08)	58.13 (11.93)
Education, n (%)			
Primary school	1 (6.3)	1 (6.3)	2 (6.3)
Secondary school	2 (12.5)	3 (18.8)	5 (15.6)
High school	11 (68.8)	8 (50)	19 (59.4)
Degree	2 (12.5)	4 (25)	6 (18.8)
Marital status, n (%)			
Never married	2 (12.5)	1 (6.3)	3 (9.4)
Married	8 (50)	11 (68.8)	19 (59.4)
Divorced	3 (18.8)	1 (6.3)	4 (12.5)
Separated	1 (6.3)	1 (6.3)	2 (6.3)
Widowed	2 (12.5)	2 (12.5)	4 (12.5)
Employment status, n (%)			
Unemployed	1 (6.3)	2 (12.5)	3 (9.4)
Housewife	3 (18.8)	2 (12.5)	5 (15.6)
Student	1 (6.3)	0 (0.0)	1 (3.1)
Blue-collar worker	1 (6.3)	0 (0.0)	1 (3.1)
White-collar worker	5 (31.3)	3 (18.8)	8 (25)
Freelance	1 (6.3)	3 (18.8)	4 (12.5)
Self-employed	1 (6.3)	1 (6.3)	2 (6.3)
Retired	3 (18.8)	5 (31.3)	8 (25)

WBT: Well-being Therapy; TAU: treatment as Usual.

1 participant per arm ($n = 2$, 0.6%) dropped at 6-month follow-up (T4) (see Supplementary Fig. S1, available at *Rheumatology* online). No data were excluded from the analysis. No events with serious consequence or side effects occurred.

Over time changes of primary outcomes

Table 2 shows mean and standard deviation of primary outcome variables at T1, T2, T3 and T4 in WBT and TAU groups.

Table 3 reports GEE models for primary outcomes. WHO-5 subjective well-being increased under WBT at times T1, T2 and T3, and thereafter tended to return to T1 levels (Test for treatment-time interaction, $P \leq 0.001$). PWB personal growth increased between T2 and T3 under WBT (Test for treatment-time interaction, $P = 0.006$). PWB self-acceptance increased under WBT at T2 (Test for treatment-time interaction, $P = 0.003$).

Over time changes of secondary outcomes

Table 2 shows mean and standard deviation of secondary outcomes at T1, T2, T3 and T4 in the WBT and TAU groups.

Table 4 shows GEE models for secondary outcomes. Mental pain significantly decreased at T1 and T2 under WBT (Test for treatment-time interaction, $P = 0.010$). GEE showed significant decrease in SCL-90-R somatization, SCL-90-R obsessive-compulsive and SCL-90-R hostility at T2 in the WBT group (Test for treatment-time interaction, $P = 0.010$, $P = 0.022$ and $P = 0.036$, respectively). GEE showed significant decrease in SCL-90-R depression (Test for treatment-time interaction, $P = 0.030$) and SCL-90-R anxiety (Test for treatment-time interaction, $P = 0.048$) at T2 and T3 in the WBT group. SCL-90-R GSI significantly decreased at T2 in the WBT group (Test for treatment-time interaction, $P = 0.010$). The positive impact of WBT on PRISM illness-related suffering (Test for treatment-time interaction, $P \leq 0.001$), PRISM lack of feeling at peace (Test for treatment-time interaction, $P \leq 0.001$), PRISM physical pain-related suffering (Test for treatment-time interaction, $P \leq 0.001$) and PRISM lack of leisure-time functioning (Test for treatment-time interaction, $P \leq 0.001$) was observed during WBT. The positive impact of WBT on PRISM lack of job occupational functioning was observed at T4 (Test for treatment-time interaction, $P \leq 0.001$).

Treatment satisfaction, credibility, expectancy and change

Participants reported high satisfaction with WBT (CSQ-8 mean ± SD total score: 29.38 ± 2.33) and credibility (CEQ mean ± SD total score: 23.19 ± 3.67), medium-high level of expectancy regarding the treatment received (CEQ mean ± SD total score: 19.87 ± 6.08). Participants reported high level of overall change after WBT (Kellner's Scale mean ± SD total score: 2.25 ± 0.68).

Discussion

The present study provides preliminary support to the benefits and acceptability of WBT in SSc patients. The WBT group had a significant improvement in primary outcomes (i.e. well-being), which was maintained at 3-month follow-up as what concerns subjective well-being. The WBT group also reported a decrease in psychological distress, mental pain and suffering, and the latter was maintained at 3- and 6-month follow-ups.

Such benefits can be attributed to WBT, rather than to a natural improvement of the clinical manifestations, thanks to the use of TAU as comparator.

The results on subjective well-being and psychological distress extend preliminary examinations of WBT in patients with chronic diseases such as type 2 diabetes [19], chronic pain [20] and chronic migraine [21], and overall suggest that also SSc patients can take advantage from psychotherapeutic strategies promoting well-being because they learn how to cope with the disease and maintain a psychological balance.

Findings on mental pain and suffering, which showed to be reduced under WBT, are new in the literature because these specific variables were not measured in previous randomized controlled studies testing the efficacy of WBT. Since mental pain and suffering are strongly related to SSc symptom severity and to functional limitations, their decrease seems of benefit for SSc patients because it is associated with a decrease in disease burden.

The results can be interpreted considering that WBT allows SSc patients to become aware that the challenge is not eliminating SSc symptoms but learning and implementing strategies for empowering subjective well-being and living life consistently with own values and despite the organic disease [22].

In addition, an increase in well-being typically has an indirect decreasing effect on distress [19–21]. Indeed, also in SSc patients, WBT decreased mental pain and suffering which additionally strengthened the re-appraising of meaning and balance in life [22].

Unfortunately, WBT benefits were not maintained at 6-month follow-up in the sample under study which, being recruited at a third-level centre of care is characterized by relatively high SSc symptom severity and is composed by patients with a relatively long history of disease. Based on

Table 2. Mean and standard deviation of primary (well-being) and secondary (functional ability related to systemic sclerosis, psychological distress, mental pain, suffering) outcome variables

	T0 WBT (n = 16) M ± SD	T1 WBT (n = 16) M ± SD	T2 WBT (n = 16) M ± SD	T3 WBT (n = 16) M ± SD	T4 WBT (n = 15) M ± SD	T0 TAU (n = 16) M ± SD	T1 TAU (n = 16) M ± SD	T2 TAU (n = 16) M ± SD	T3 TAU (n = 16) M ± SD	T4 TAU (n = 15) M ± SD
WHO-5 total score	52.50 ± 19.37	59.25 ± 18.25	64.00 ± 18.30	57.00 ± 14.19	52.27 ± 20.97	51.75 ± 26.31	44.00 ± 22.62	41.00 ± 22.97	42.00 ± 20.24	49.87 ± 25.65
PWB autonomy	61.87 ± 9.37	62.63 ± 10.74	66.38 ± 8.99	65.19 ± 11.29	64.53 ± 10.25	64.69 ± 11.11	61.06 ± 14.42	61.94 ± 11.41	63.56 ± 10.92	67.80 ± 12.91
PWB environmental mastery	59.44 ± 10.76	59.50 ± 10.02	63.31 ± 9.68	60.75 ± 9.83	59.87 ± 10.03	60.56 ± 12.29	55.56 ± 13.63	56.75 ± 13.44	57.63 ± 14.89	61.60 ± 11.16
PWB personal growth	63.63 ± 11.30	62.31 ± 10.75	65.69 ± 7.83	66.56 ± 8.21	65.13 ± 8.68	61.00 ± 8.81	58.44 ± 11.26	57.19 ± 7.45	56.88 ± 11.05	61.27 ± 11.48
PWB positive relations with others	60.00 ± 11.28	61.00 ± 12.29	61.13 ± 10.34	63.94 ± 11.92	60.07 ± 10.35	64.81 ± 7.04	59.94 ± 9.71	58.63 ± 7.19	59.06 ± 9.29	63.27 ± 10.94
PWB purpose in life	58.19 ± 9.56	59.13 ± 9.52	61.63 ± 8.16	62.31 ± 9.20	60.20 ± 9.19	59.44 ± 13.18	53.06 ± 11.64	54.56 ± 12.45	55.25 ± 13.85	61.20 ± 11.37
PWB self-acceptance	59.06 ± 11.12	59.13 ± 10.18	64.31 ± 10.58	63.25 ± 11.45	60.93 ± 12.23	57.94 ± 14.54	53.50 ± 16.13	53.19 ± 14.48	55.56 ± 15.06	61.27 ± 11.92
MPQ total score	1.50 ± 1.79	0.69 ± 1.07	0.50 ± 0.89	0.88 ± 1.89	1.33 ± 2.13	2.13 ± 2.15	2.31 ± 2.49	2.56 ± 2.45	2.50 ± 2.94	2.20 ± 2.57
SCL-90-R somatization	0.84 ± 0.47	0.72 ± 0.42	0.55 ± 0.41	0.72 ± 0.53	0.79 ± 0.59	0.82 ± 0.54	0.90 ± 0.68	1.14 ± 0.71	1.12 ± 0.88	0.86 ± 0.76
SCL-90-R obsessive-compulsive	0.58 ± 0.44	0.59 ± 0.24	0.45 ± 0.32	0.61 ± 0.43	0.62 ± 0.54	0.58 ± 0.44	0.79 ± 0.78	0.89 ± 0.76	0.88 ± 0.66	0.62 ± 0.52
SCL-90-R interpersonal sensibility	0.38 ± 0.48	0.39 ± 0.38	0.34 ± 0.28	0.37 ± 0.43	0.55 ± 0.58	0.40 ± 0.36	0.49 ± 0.37	0.43 ± 0.47	0.56 ± 0.71	0.33 ± 0.49
SCL-90-R depression	0.64 ± 0.54	0.60 ± 0.36	0.45 ± 0.27	0.50 ± 0.36	0.70 ± 0.49	0.73 ± 0.51	0.85 ± 0.63	0.96 ± 0.85	1.00 ± 0.91	0.65 ± 0.66
SCL-90-R anxiety	0.43 ± 0.39	0.43 ± 0.28	0.32 ± 0.30	0.35 ± 0.31	0.50 ± 0.32	0.46 ± 0.35	0.53 ± 0.39	0.68 ± 0.63	0.70 ± 0.66	0.42 ± 0.33
SCL-90-R hostility	0.35 ± 0.75	0.31 ± 0.44	0.17 ± 0.19	0.27 ± 0.18	0.31 ± 0.40	0.22 ± 0.21	0.26 ± 0.18	0.41 ± 0.33	0.38 ± 0.36	0.33 ± 0.42
SCL-90-R phobic anxiety	0.18 ± 0.28	0.10 ± 0.15	0.08 ± 0.18	0.13 ± 0.23	0.18 ± 0.26	0.11 ± 0.23	0.24 ± 0.38	0.36 ± 0.59	0.42 ± 0.78	0.30 ± 0.59
SCL-90-R paranoid ideation	0.39 ± 0.47	0.46 ± 0.49	0.30 ± 0.26	0.40 ± 0.49	0.53 ± 0.48	0.37 ± 0.28	0.49 ± 0.35	0.47 ± 0.46	0.57 ± 0.62	0.41 ± 0.49
SCL-90-R psychoticism	0.28 ± 0.31	0.32 ± 0.24	0.23 ± 0.20	0.35 ± 0.33	0.40 ± 0.41	0.20 ± 0.23	0.37 ± 0.38	0.36 ± 0.43	0.45 ± 0.45	0.20 ± 0.21
SCL-90-R GSI	0.50 ± 0.38	0.48 ± 0.25	0.36 ± 0.21	0.46 ± 0.31	0.55 ± 0.37	0.49 ± 0.31	0.60 ± 0.43	0.70 ± 0.55	0.74 ± 0.62	0.51 ± 0.46
PRISM illness-related suffering	9.02 ± 6.03	12.09 ± 4.87	14.81 ± 4.36	16.65 ± 4.05	15.85 ± 5.06	8.20 ± 6.69	5.41 ± 3.65	4.30 ± 3.41	4.43 ± 4.92	6.43 ± 5.23
PRISM lack of feeling at peace	9.63 ± 6.93	6.89 ± 3.69	5.45 ± 2.71	4.81 ± 1.43	5.40 ± 2.31	8.23 ± 5.05	12.18 ± 4.91	14.86 ± 5.24	16.03 ± 6.51	13.95 ± 5.14
PRISM physical pain-related suffering	11.06 ± 5.99	13.07 ± 5.58	16.18 ± 4.78	17.95 ± 3.66	15.51 ± 5.26	9.90 ± 7.30	6.74 ± 4.08	5.10 ± 2.91	4.35 ± 3.55	8.06 ± 6.37
PRISM lack of job occupational functioning	7.27 ± 4.33	6.80 ± 3.45	5.81 ± 3.27	4.60 ± 2.66	3.42 ± 1.61	4.01 ± 2.00	4.52 ± 1.26	5.25 ± 1.94	5.23 ± 1.83	5.81 ± 1.99
PRISM lack of leisure-time functioning	8.32 ± 6.39	6.41 ± 4.00	4.73 ± 2.81	3.70 ± 1.90	4.61 ± 2.38	9.28 ± 5.91	12.81 ± 5.56	14.60 ± 4.64	15.67 ± 5.65	11.87 ± 5.59

WHO-5: 5-item World Health Organization Well-Being Index; PWB: Psychological Well-Being scales; MPQ: Mental Pain Questionnaire; SCL-90-R: Symptom Checklist-90-Revised; GSI: Global Severity Index; PRISM: Pictorial Representation of Illness and Self Measure; WBT: Well-being Therapy; TAU: treatment as Usual.

Table 3. Estimated marginal means (SE) of primary outcomes (well-being as measured via WHO-5 and PWB scales)

	T0 Means (SE)	*P*-value	T1 Means (SE)	*P*-value	T2 Means (SE)	*P*-value	T3 Means (SE)	*P*-value	T4 Means (SE)	*P*-value
WHO-5 total score										
WBT	52.50 (4.69)	*0.924*	59.25 (4.42)	*0.030*	64.00 (4.43)	*0.001*	57.00 (3.43)	*0.012*	53.80 (5.27)	*0.580*
TAU	51.75 (6.37)		44.00 (5.47)		41.00 (5.56)		42.00 (4.89)		49.30 (6.17)	
PWB autonomy										
WBT	61.88 (2.27)	*0.424*	62.63 (2.60)	*0.720*	66.38 (2.17)	*0.207*	65.19 (2.73)	*0.669*	65.01 (2.48)	*0.604*
TAU	64.69 (2.69)		61.06 (3.49)		61.94 (2.76)		63.56 (2.64)		67.09 (3.17)	
PWB environmental mastery										
WBT	59.44 (2.60)	*0.776*	59.50 (2.42)	*0.337*	63.31 (2.34)	*0.102*	60.75 (2.38)	*0.470*	60.78 (2.51)	*0.711*
TAU	60.56 (2.97)		55.56 (3.30)		56.75 (3.25)		57.63 (3.60)		62.14 (2.65)	
PWB personal growth										
WBT	63.63 (2.73)	*0.449*	62.31 (2.60)	*0.304*	65.69 (1.89)	*≤ 0.001*	66.56 (1.98)	*0.004*	65.95 (2.19)	*0.143*
TAU	61.00 (2.13)		58.44 (2.73)		57.19 (1.80)		56.88 (2.67)		60.76 (2.78)	
PWB positive relations with others										
WBT	60.00 (2.73)	*0.135*	61.00 (2.97)	*0.779*	61.12 (2.50)	*0.412*	63.94 (2.88)	*0.183*	60.76 (2.50)	*0.439*
TAU	64.81 (1.70)		59.94 (2.35)		58.62 (1.74)		59.06 (2.25)		63.61 (2.69)	
PWB purpose in life										
WBT	58.19 (2.30)	*0.751*	59.13 (2.30)	*0.096*	61.63 (1.97)	*0.050*	62.31 (2.30)	*0.079*	60.72 (2.21)	*0.916*
TAU	59.44 (3.19)		53.06 (2.81)		54.56 (3.01)		55.25 (3.35)		61.09 (2.69)	
PWB self-acceptance										
WBT	59.06 (2.69)	*0.800*	59.13 (2.46)	*0.233*	64.31 (2.56)	*0.010*	63.25 (2.77)	*0.093*	61.19 (2.93)	*0.972*
TAU	57.94 (3.52)		53.50 (3.90)		53.19 (3.50)		55.56 (3.64)		61.33 (2.79)	

WHO-5: 5-item World Health Organization Well-Being Index; PWB: Psychological Well-Being scales; WBT: Well-being Therapy; TAU: treatment as Usual. The sample size at T4 was $n = 15$ per arm.

Table 4. Estimated marginal means (SE) of secondary outcomes (functional ability related to systemic sclerosis, psychological distress, mental pain, suffering)

	T0 Means (SE)	*P*-value	T1 Means (SE)	*P*-value	T2 Means (SE)	*P*-value	T3 Means (SE)	*P*-value	T4 Means (SE)	*P*-value
MPQ total score										
WBT	1.50 (0.43)	*0.357*	0.69 (0.26)	*0.014*	0.50 (0.21)	*≤ 0.001*	0.88 (0.45)	*0.055*	1.25 (0.49)	*0.151*
TAU	2.13 (0.52)		2.31 (0.60)		2.56 (0.59)		2.50 (0.71)		2.42 (0.64)	
SCL-90-R somatization										
WBT	0.84 (0.11)	*0.904*	0.72 (0.10)	*0.350*	0.59 (0.09)	*0.006*	0.72 (0.13)	*0.115*	0.77 (0.14)	*0.745*
TAU	0.82 (0.13)		0.90 (0.16)		1.14 (0.17)		1.12 (0.21)		0.85 (0.18)	
SCL-90-R obsessive-compulsive										
WBT	0.58 (0.10)	*1.000*	0.59 (0.05)	*0.315*	0.45 (0.08)	*0.027*	0.61 (0.10)	*0.161*	0.59 (0.13)	*0.677*
TAU	0.58 (0.10)		0.79 (0.19)		0.89 (0.18)		0.88 (0.16)		0.67 (0.13)	
SCL-90-R interpersonal sensibility										
WBT	0.38 (0.11)	*0.925*	0.38 (0.09)	*0.417*	0.34 (0.06)	*0.464*	0.30 (0.09)	*0.192*	0.53 (0.14)	*0.288*
TAU	0.40 (0.08)		0.49 (0.09)		0.43 (0.11)		0.56 (0.17)		0.34 (0.11)	
SCL-90-R depression										
WBT	0.64 (0.13)	*0.596*	0.60 (0.08)	*0.166*	0.45 (0.06)	*0.019*	0.50 (0.08)	*0.037*	0.68 (0.12)	*0.918*
TAU	0.73 (0.12)		0.85 (0.15)		0.96 (0.20)		1.00 (0.22)		0.70 (0.15)	
SCL-90-R anxiety										
WBT	0.43 (0.09)	*0.807*	0.43 (0.07)	*0.392*	0.32 (0.07)	*0.037*	0.35 (0.07)	*0.049*	0.49 (0.07)	*0.665*
TAU	0.46 (0.08)		0.53 (0.09)		0.68 (0.15)		0.70 (0.16)		0.57 (0.17)	
SCL-90-R hostility										
WBT	0.35 (0.18)	*0.472*	0.31 (0.10)	*0.656*	0.17 (0.04)	*0.011*	0.27 (0.04)	*0.249*	0.28 (0.09)	*0.676*
TAU	0.21 (0.05)		0.26 (0.04)		0.41 (0.08)		0.38 (0.09)		0.34 (0.10)	
SCL-90-R phobic anxiety										
WBT	0.18 (0.06)	*0.483*	0.10 (0.03)	*0.182*	0.09 (0.04)	*0.066*	0.13 (0.05)	*0.147*	0.18 (0.06)	*0.327*
TAU	0.11 (0.05)		0.24 (0.09)		0.36 (0.14)		0.42 (0.18)		0.34 (0.15)	
SCL-90-R paranoid ideation										
WBT	0.39 (0.11)	*0.876*	0.47 (0.12)	*0.888*	0.67 (0.15)	*0.275*	0.40 (0.11)	*0.384*	0.49 (0.12)	*0.645*
TAU	0.37 (0.07)		0.49 (0.08)		0.47 (0.11)		0.57 (0.15)		0.42 (0.12)	
SCL-90-R psychoticism										
WBT	0.28 (0.07)	*0.422*	0.32 (0.06)	*0.609*	0.23 (0.05)	*0.257*	0.35 (0.08)	*0.438*	0.38 (0.09)	*0.446*
TAU	0.20 (0.05)		0.37 (0.09)		0.36 (0.10)		0.45 (0.11)		0.27 (0.09)	
SCL-90-R GSI										
WBT	0.50 (0.09)	*0.907*	0.48 (0.06)	*0.326*	0.36 (0.05)	*0.017*	0.46 (0.07)	*0.092*	0.53 (0.09)	*0.740*
TAU	0.49 (0.07)		0.60 (0.10)		0.70 (0.13)		0.74 (0.15)		0.58 (0.13)	
PRISM illness-related suffering										
WBT	9.02 (1.46)	*0.706*	12.09 (1.18)	*≤ 0.001*	14.81 (1.05)	*≤ 0.001*	16.65 (0.98)	*≤ 0.001*	15.80 (1.24)	*≤ 0.001*
TAU	8.20 (1.62)		5.41 (0.88)		4.30 (0.82)		4.43 (1.19)		6.61 (1.25)	

(continued)

Table 4. (continued)

	T0 Means (SE)	*P*-value	T1 Means (SE)	*P*-value	T2 Means (SE)	*P*-value	T3 Means (SE)	*P*-value	T4 Means (SE)	*P*-value
PRISM lack of feeling at peace										
WBT	9.63 (1.68)	*0.500*	6.89 (0.89)	*≤ 0.001*	5.45 (0.65)	*≤ 0.001*	4.81 (0.34)	*≤ 0.001*	5.29 (0.58)	*≤ 0.001*
TAU	8.23 (1.22)		12.18 (1.19)		14.86 (1.27)		16.03 (1.57)		13.66 (1.31)	
PRISM physical pain-related suffering										
WBT	11.06 (1.45)	*0.612*	13.07 (1.35)	*≤ 0.001*	16.80 (1.57)	*≤ 0.001*	17.95 (0.88)	*≤ 0.001*	15.68 (1.29)	*≤ 0.001*
TAU	9.90 (1.76)		6.74 (0.98)		5.10 (0.70)		4.35 (0.86)		7.49 (1.70)	
PRISM lack of job occupational functioning										
WBT	7.27 (1.36)	*0.033*	6.80 (1.08)	*0.053*	5.81 (1.02)	*0.651*	4.60 (0.83)	*0.552*	2.70 (0.82)	*0.004*
TAU	4.01 (0.70)		4.52 (0.44)		5.25 (0.68)		5.22 (0.64)		5.81 (0.69)	
PRISM lack of leisure-time functioning										
WBT	8.32 (1.54)	*0.648*	6.41 (0.96)	*≤ 0.001*	4.73 (0.68)	*≤ 0.001*	3.70 (0.46)	*≤ 0.001*	4.48 (0.55)	*≤ 0.001*
TAU	9.28 (1.43)		12.81 (1.34)		14.60 (1.12)		15.66 (1.36)		11.49 (1.46)	

MPQ: Mental Pain Questionnaire; SCL-90-R: Symptom Checklist-90-Revised; GSI: Global Severity Index; PRISM: Pictorial Representation of Illness and Self Measure; WBT: Well-being Therapy; TAU: treatment as Usual. The sample size at T4 was $n = 15$ per arm.

this, booster sessions have to be applied to support such patients overtime and the long-term outcomes need further evaluations. SSc is a chronic and deteriorating disease, and thus interventions aimed at supporting patients in coping with it are in need to last as long as the disease persists or worsens. Of course, it can be hypothesized that administering WBT as soon as the diagnosis is formulated might be of greater help because SSc patients can empower their skills at first stages of the disease when symptoms are less severe and invalidating, thus limiting the risk of secondary psychological distress occurrence and acquiring skills to be implemented at later and more severe stages of disease. Future research is warranted to support such hypothesis.

Given the preliminary nature of the present results, they should be interpreted under the light of some limitations. The study was monocentric and conducted at a third-level centre of care, thus the sample had on average high SSc severity and a long history of disease; for these reasons, it may not be entirely representative of the clinical population under study. The sample size is limited even though adequate to run the proposed analyses. It was difficult to engage SSc patients in a non-pharmacological trial and this may imply that only highly motivated SSc patients were enrolled. Since an active form of psychotherapeutic control group was not proposed, it is difficult to establish whether the results were determined by WBT or by non-specific psychological ingredients commonly applied to psychotherapy, including WBT.

Conclusion

This study demonstrated that WBT improved well-being, psychological distress, mental pain and suffering in patients with SSc. The results indicate that the intervention has a positive effect on mental health of patients with SSc and offer preliminary evidence in support of using WBT as a short-term approach for in- and out-patient SSc healthcare paths. Multicentre trials conducted in larger samples and including a psychological intervention other than WBT (e.g. psychoeducation, mindfulness-based stress reduction therapy) as comparator are needed to confirm the present preliminary and promising results.

Supplementary material

Supplementary material is available at *Rheumatology* online.

Data availability

The data underlying this article will be shared on reasonable request to the corresponding author.

The trial was conducted according to CONSORT guidelines and registered in ClinicalTrials.gov (NCT04212247).

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