

Evaluation of a mail-delivered, print-format, self-management program for persons with systemic sclerosis

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Abstract Patient self-management programs usually require participants to attend group sessions, which can be difficult for individuals with mobility issues. In addition, many programs are not disease specific. The purpose of this study was to evaluate the effects of a mail-delivered self-management program for individuals with scleroderma (SSc). The program consisted of a workbook and exercise DVD that provided information on medical aspects of the disease, dysphagia, fatigue management, advocacy, activities of daily living, oral hygiene, skin and wound care, psychosocial changes, exercises, and other features of the condition. Participants provided feedback on the effects of the self-management program by responding to questions on demographic and six self-report questionnaires, keeping a health log, and participating in a program evaluation interview. A total of 49 participants completed the program and returned the postintervention questionnaires. Participants consistently reported that the program was easy to use. Depression, fatigue, and pain decreased, and hand function, self-efficacy for controlling pain, and self-efficacy “other” improved; however, the only statistically significant change was in self-efficacy for pain. This is the first study to develop and assess the effects of a mail-delivered format for self-management for people with SSc. A self-management program should help individuals with SSc develop self-management strategies to manage this complex disease and advocate for themselves to promote better health.

Keywords Scleroderma · Self-efficacy · Self-management

Introduction

Chronic disease self-management programs have demonstrated that programs based on common problems in coping across diseases can be effective [1–3]; however, the opportunity to provide disease-specific management information is lost. Indeed, several studies showed that individuals frequently desire disease-specific information and often feel marginalized when the specific impact of the disease on their lives is not part of the program [4, 5]. Systemic sclerosis (SSc) is one chronic rheumatic disease with unique manifestations such as tight skin, facial changes, dysphagia, lung disease, and severe Raynaud’s phenomenon, which are not addressed by the existing chronic disease self-management programs. These manifestations make a program that incorporates principles of self-management, along with content specific to the unique aspects of SSc, optimal.

Several group-format self-management programs developed for people with SSc reported increases in self-efficacy and reduced helplessness [6–8]. However, group formats are dependent on people going to central locations, and because the prevalence of SSc is low, people with SSc may not have access to SSc-specific support groups or education programs.

This paper reports on a study designed to determine the effectiveness of a mail-delivered self-management program for persons with SSc. We developed the self-management program based on information gathered from focus groups [9] and content areas included in published studies on self-management in SSc [6–8]. The program consisted of a workbook and exercise DVD (Table 1). Each chapter contained learning activities, with action plans modeled after the arthritis self-management programs. Because of the joint contractures and microstomia that occur with SSc,

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Table 1 Self-management workbook chapters

What is scleroderma
Emergencies
Advantages and disadvantages of clinical trials
Coping and body image/appearance
Fatigue and energy conservation
Activities of daily living
Mouth and teeth care
Exercise
Handouts for hand and leg exercise
Management of Raynaud's phenomenon and finger ulcers
Dysphagia and dietary management
Self-advocacy

the DVD demonstrated face, mouth, hand, arm, and leg exercises.

Materials and methods

Sample and setting

Individuals with SSc were recruited from the Scleroderma Foundation website and through announcements sent to local chapters of the Scleroderma Foundation. Inclusion criteria included age 21 or older, US resident, diagnosis of SSc, ability to communicate in English, and willingness to complete the study protocol. Sixty-nine participants who met the inclusion criteria were sent the consent form, questionnaires, and a self-addressed return mailer. Sixty-two participants provided written consent, returned completed questionnaires, and were sent the workbook and DVD. Participants were expected to complete the program in 3 to 4 months. On completion of the program, participants were sent posttest questionnaire packets and, if possible, scheduled for a program evaluation telephone interview. This study was approved by the institution's human research and protections committee.

Instruments

Demographics and baseline disease information was collected on age, gender, type of SSc, length of time since disease onset, self-rated health, health distress, communication with physicians, education level, marital status, and ethnicity.

Health log. While enrolled in the study, participants kept a log of health-related events, such as visits to a physician, days sick, days confined to the home, and visits to the emergency department.

The *Arthritis Self-Efficacy Scale* measured perceived self-efficacy to perform specific tasks or behaviors to cope with the consequences of SSc [10]. Participants rate their

perceived ability to perform each item on a scale ranging from 10 (*very uncertain*) to 100 (*very certain*) in increments of 10. Higher scores indicate greater self-efficacy or the confidence that one can perform the specific behavior.

The *Health Assessment Questionnaire* (HAQ) measured functional ability [11]. It consists of eight categories of daily living: dressing and grooming, arising, eating, walking, hygiene, reach, grip, and outside activity. Subjects rate how much difficulty they have with each item from 0 (*no difficulty*) to 3 (*cannot do*).

The *Scleroderma Functional Assessment Questionnaire*, or UK Scleroderma Functional Score, measured functional ability with upper extremity activities [12]. Items are scored on a 4-point scale, from 0 (*able to perform in a normal manner*) to 3 (*impossible to perform*).

Pain was assessed using the 10-cm horizontal analog pain scale on the HAQ. Pain severity is rated from 0 (*no pain*) to 10 (*very severe pain*).

The *Multidimensional Assessment of Fatigue Scale* measured fatigue degree, severity, distress, and interference with daily activities. Scores range from 1 (*not a great deal, not severe*) to 10 (*a great extent, severe*) [13].

The *Center for Epidemiologic Studies Depression Scale* (CES-D) measured depression [14]. A higher score indicates greater symptoms of depression.

Program evaluation interviews were conducted to determine the participants' experiences with and opinions about the program. Twenty-nine brief telephone interviews were conducted approximately 4 to 6 weeks after program completion using a structured interview guide. Where appropriate, participants were encouraged to develop their answers and probing questions used as necessary. Each interview lasted approximately 15 min. Interviews were tape recorded and transcribed. Content analysis was used to identify the key ideas presented during the interviews.

Results

Forty-nine participants completed the program and returned postintervention questionnaires. There were no significant differences between those who completed the program and those who did not, except for marital status (Table 2). Significantly more married persons completed the program, suggesting a supportive partner may be important in completing such a program.

Table 3 shows the results of *t* tests comparing baseline and postintervention scores. After the intervention, depression, fatigue, and pain decreased, whereas hand function, self-efficacy for controlling pain, and self-efficacy "other" improved; however, the only statistically significant change was in self-efficacy for pain. Participants felt they were better able to manage pain after the intervention. There were also no

Table 2 Comparison of selected baseline characteristics of participants categorized by completers and noncompleters of the self-management program

Characteristic	Completers (<i>n</i> =49)	Noncompleters (<i>n</i> =13)	<i>p</i> value
Mean age (SD), years	53.9 (12.5)	49.3 (7.3)	0.24
Mean years of education (SD)	15.2 (2.7)	18.1 (3.3)	0.16
Mean disease duration (SD), years	6.9 (7.1)	7.4 (4.5)	0.79
Gender, <i>n</i> (%)			0.13
Female	45 (92)	9 (72)	
Male	4 (8)	3 (25)	
Ethnicity, <i>n</i> (%)			0.44
White	40 (82)	8 (67)	
Hispanic	3 (6)	1 (8)	
African American	4 (8)	2 (17)	
Asian/Pacific	1 (2)	1 (8)	
Native American	1 (2)	0 (0)	
Education, <i>n</i> (%), years			0.16
±12	11 (22)	1 (8)	
13–17	25 (51)	4 (33)	
≥18	13 (27)	7 (58)	
Marital status, <i>n</i> (%)			0.03
Never married	4 (8)	1 (8)	
Married	36 (73)	5 (52)	
Widowed	3 (6)	1 (8)	
Separated	0 (0)	2 (17)	
Divorced	6 (12)	3 (25)	
Employment, <i>n</i> (%)			0.36
Full time	19 (39)	7 (58)	
Part time	5 (10)	1 (8)	
Disability	6 (12)	2 (17)	
Retired	12 (24)	0 (0)	
Looking for work	2 (4)	0 (0)	
Other	5 (10)	2 (17)	
Mean scores (SD)	5.2 (2.7)	5.4 (2.3)	0.83
Self-efficacy pain			
Self-efficacy function	7.6 (3.0)	7.0 (2.5)	0.51
Self-efficacy other	6.4 (2.6)	6.6 (2.4)	0.84
Self-efficacy total	6.6 (2.1)	6.5 (1.7)	0.92

SD standard deviation

significant changes in the number of doctor visits, emergency department visits, overnight stays in the hospital, or total night stays in the hospital over the previous 6 months.

Analysis of the program evaluation interviews

Participants consistently reported that the program was easy to use. The spiral-bound format of the workbook was especially appealing to participants with hand deformities because it was easier for them to handle than a bound book.

Most participants thought that the chapters were properly ordered, the content flowed well, and the reading level appropriate. The exercise DVD was very popular. Many participants were unaware of the mouth exercises and found

these very useful. One participant said, “They [the workbook and the DVD] were both quite easy to use, perhaps the DVD was the better part of the program just because seeing something is easier to comprehend than especially the material covered by the DVD.”

While creating the program, we experienced a dilemma regarding what information to include and the depth of information to provide. We believed that the most appropriate audience for the program would be participants who were newly diagnosed, in the process of learning about their illness, and learning how to manage the various physical changes and psychosocial implications. However, our sample did not match this profile as most of the participants had long-standing illness.

Table 3 Pre–post changes for outcome measures

Outcome measures	Preintervention, mean (SD)	Postintervention, mean (SD)	Change, mean (SD)	<i>p</i> value
Self-efficacy pain	5.2 (2.7)	6.4 (2.7)	1.2 (2.9)	0.006
Self-efficacy function	7.6 (3.0)	7.4 (2.2)	−0.2 (2.8)	0.59
Self-efficacy other	6.4 (2.6)	7.0 (2.2)	0.6 (3.0)	0.18
Self-efficacy total	6.6 (2.1)	6.5 (2.5)	−0.1 (2.8)	0.86
Multidimensional fatigue scale (MAF)	26.1 (11.5)	24.6 (11.6)	−1.5 (8.1)	0.20
Depression (CES-D)	14.4(10.9)	14.3 (10.8)	−0.1 (7.9)	0.93
Activity disability (HAQ)	0.9 (0.7)	0.9 (0.7)	0.0 (0.4)	0.56
Pain	2.9 (2.6)	2.8 (2.6)	−0.05 (1.9)	0.87
Hand disability (SFAQ)	6.8 (5.8)	6.4 (5.5)	−0.3 (2.8)	0.39
Number doctor visits	6.6 (6.5)	5.9 (6.2)	−0.6 (5.5)	0.19
Number ED visits	0.2 (0.6)	0.2 (0.6)	0	1.0
Number of overnight stays in hospital	0.0 (0.2)	0.1 (0.5)	0.1 (0.5)	0.38
Total number of nights spent in hospital	0.1 (0.4)	0.4 (1.8)	0.3 (1.8)	0.38

MAF Multidimensional Assessment of Fatigue Scale, *CES-D* Center for Epidemiologic Studies Depression Scale, *HAQ* Health Assessment Questionnaire, *SFAQ* Scleroderma Functional Assessment Questionnaire, *ED* emergency department

Regardless, many were able to garner useful information from the program and reported the content reaffirmed information that they had gathered or learned on their own. For others, it provided additional depth of information to what they already knew. Participants also noted that the program could and did serve as a good reference for basic information and as a resource as new symptoms arise and participants are confronted with new challenges as a consequence of their illness. Other participants said it provided additional information, such as the mouth exercises, as previously mentioned, or stretching exercises in general. A few participants with severe contractures indicated that they had never been referred to occupational or physical therapy and had never been taught the benefits of stretching exercises. Additional content suggested by participants included navigating airports and managing symptoms while traveling, aerobic exercises, dealing with facial changes, and pulmonary hypertension.

Discussion

This study of the effects of a workbook-and-DVD self-management program for SSc showed that depression, fatigue, and pain decreased, whereas hand function, self-efficacy for controlling pain, and self-efficacy other improved after the intervention. However, the only statistically significant improvement was in self-efficacy for pain. The prevalence of pain has been reported to be as high as 60 to 75 % in persons with SSc, and pain has been reported to be one of the five most frequent symptoms that interfere with daily activities [15]. An improvement in self-efficacy for

pain is noteworthy because higher self-efficacy for pain has been reported to predict better coping, functioning, and less depression, and reduced avoidance behaviors [16].

Participants' experience of using the self-management program was positive. Participants stated that they learned new information, appreciated the exercise DVD, and felt the content and reading level were appropriate. Suggestions for additional content were made.

Our findings are similar to a recent study assessing a group-format education program for SSc [8]. That study, which also used a pretest–posttest design, reported less helplessness and higher acceptance of limitations after participating in the program [8]. Similar to our study, there were no significant changes in depressive symptoms, pain, and physical functioning (HAQ). However, our findings are contrary to studies by Lorig et al. for mail-based and written programs, which did find improvements in self-efficacy [1, 17, 18]. There may be several explanations. First, although the self-management program was based on self-efficacy theory, the workbook format did not allow participants to share their stories, goals, and successes or to encourage each other. Adding a teleconference once a week or developing an Internet format with a chat room or discussion board would allow participants to interact. Second, our sample size of 49, although large enough to detect an effect size of 0.45, was small compared with the other studies on the arthritis self-management programs. However, our sample was larger than the samples used in the other studies on self-management programs for SSc. Third, although self-efficacy scores were slightly higher in our sample compared with other studies in persons with rheumatoid arthritis, recruitment methods might have yielded a sample that was already using strategies to manage the disease. Furthermore,

our sample was well educated and self-selected and perhaps motivated to acquire additional information. Regardless, the characteristics of our participants were similar to participants in several larger studies of persons with SSc except we had a higher percent of females [19, 20]. However, higher participation in research studies by females has been reported [21]. Future studies might want to examine how to make education programs attractive and available to more diverse groups. Our participants also had minimal disability as measured by the HAQ, low pain levels, and scores less than 16 on the CES-D. Additionally, the disease duration of our participants was almost 7 years. Perhaps, the intervention would be more effective for persons newly diagnosed. Indeed, several participants commented during the postintervention interview that they wished they had had the booklet and DVD when they were first diagnosed. Fourth, and most important, although our outcome measures were similar to the outcome measures used in other studies on self-management programs [2, 17], several investigators have suggested that outcomes should focus on knowledge and strategies gained from the programs and psychosocial measures [22–24].

The limitations of this study are the sample size, lack of a control group, and the use of telephone interviews for feedback. However, our study and previous research have shown a need for this type of program [6–8]. Future researchers could use an interactive Internet-based self-management program format to reach a larger number of people with SSc or could conduct a randomized control trial. While telephone interviews allowed us to gather in-depth feedback about the program, feedback was not obtained from all participants and could have been biased. However, studies show similar content is achieved for telephone and face-to-face interviews [25]. Future studies could use evaluation forms to reach all participants and ensure anonymity.

In conclusion, this is the first study to develop and assess the effects of a mail-delivered format for self-management for people with SSc. The findings from this study are promising to warrant future studies with larger sample sizes, since the nonsignificant changes are in the expected directions, the interviews were positive, and the methodology can be used for people who do not have access to other modalities such as support groups. A self-management program should help persons with SSc develop self-management strategies to manage this complex disease and advocate for themselves to promote better health and well-being. In addition, the content of the program could also be educational to health professionals who work with people with SSc.

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