Reasons for Not Participating in Scleroderma Patient Support Groups: A Cross-sectional Study

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ABSTRACT

Objective: Peer-led support groups are an important resource for many people with scleroderma (systemic sclerosis; SSc). Little is known, however, about barriers to participation. The objective of this study was to identify reasons why some people with SSc do not participate in SSc support groups.

Methods: A 21-item survey was used to assess reasons for non-attendance among SSc patients in Canada and the United States. Exploratory factor analysis (EFA) was conducted using the software MPlus 7 to group reasons for non-attendance into themes.

Results: A total of 242 (202 women) people with SSc completed the survey. EFA results indicated that a three-factor model best described the data ($\chi^2(150)=302.7$, $p < 0.001$; CFI=0.91; TLI=0.88; RMSEA=0.07; factor intercorrelations 0.02 to 0.43). The three identified themes, reflecting reasons for not attending SSc support groups, included: (1) Personal Reasons (9 items; e.g., already having enough support), (2) Practical Reasons (7 items; e.g., no local support groups), and (3) Beliefs about Support Groups (5 items; e.g., support groups are too negative). On average, respondents rated 4.9 items as Important or Very Important reasons for non-attendance. The two items most commonly rated as Important or Very Important were (1) Already having enough support from family, friends, or others, and (2) Not knowing of any SSc support groups offered in my area.

Conclusion: SSc organizations may be able to address limitations in accessibility and concerns about SSc support groups by implementing online support groups, better informing patients about support group activities, and training support group facilitators.

SIGNIFICANCE AND INNOVATIONS

- Peer-led support groups are an important resource for many people living with scleroderma (systemic sclerosis, SSc); however, there is limited research on why people with SSc cannot attend or choose not to attend these groups.
- Based on exploratory factor analysis, people with SSc do not attend support groups due to: (1) Personal Reasons (e.g., being too busy with other responsibilities, already having enough alternative support), (2) Practical Reasons (e.g., not knowing of any local support groups, not having a reliable way to get to the meetings), and (3) Beliefs about Support Groups (e.g., thinking that support groups are not helpful or too negative).
- Limits to accessibility and concerns about support groups may be addressed by implementing online groups, better informing patients about the activities of support groups, and by providing training to the group facilitators.

People with rare diseases have many of the same concerns as people with any chronic medical disease. They also face unique challenges, including gaps in knowledge about their disease and how to treat it, insufficient access to effective treatment, and a lack of professionally organized supportive care [1-4]. Social implications include stigmatization, social exclusion, reduced professional opportunities, and potentially dramatic modifications to family, social, and professional roles [2, 5-7].

Many people with chronic diseases join support groups to help them cope with and manage their disease [8, 9]. Support groups may be particularly important for people with rare diseases who are unlikely to have other disease-specific support [3, 5]. Support groups typically include educational or information sharing activities and opportunities for members to give and receive emotional support. They are based on the principle that people enduring similar challenges can help one another via social contact and the sharing of experiences [10, 11]. Support groups for people with chronic diseases may be held face-to-face, online, or via teleconference, may be led by professionals or peers, and may follow structured or unstructured formats [5, 11-13]. No trials have assessed the effectiveness of support groups on knowledge and health outcomes in rare diseases. However, people with rare diseases report that perceived benefits of these groups include getting to know others with similar disease-related experiences, gaining information about the disease and treatment options, giving and receiving emotional support, and advocating for improved health care for people with rare diseases [14-24].

Systemic sclerosis (SSc; or scleroderma) is a rare chronic autoimmune disease characterized by fibrosis and internal organ dysfunction. A recent Canadian study estimated prevalence to be 74 cases per 100,000 for women and 13 per 100,000 for men

[25]. As in other rare diseases, many people with SSc initially experience disabling but unexplained symptoms and often struggle to obtain a diagnosis, which frequently takes several years [26, 27]. Once diagnosed, the future is filled with ongoing uncertainty and unpredictability as there is no clear course to the development and progression of the disease [28-30]. Marked disfigurement from the disease sets people with SSc apart as different, and some have described changes in physical appearance so drastic that they are no longer recognized by former acquaintances and colleagues [31]. Support groups play a central role for many SSc patients, and currently there are approximately 30 SSc support groups in Canada and 150 in the United States, almost all of which are peer-led groups that meet in local settings [32, 33].

To evaluate and improve support group services, knowledge about why people choose not to attend or are unable to attend these groups is required. In more common conditions, studies of cancer and HIV/AIDS support groups have identified a number of reasons for not participating, including: (1) believing that one already has sufficient support, (2) access and practical barriers, such as distance and not having time to attend the group meetings, (3) not being aware that support groups exist or knowledgeable about the activities of support groups, (4) not wanting to focus on one's disease, (5) perceiving support groups to be overly negative, (6) concerns about not having anything in common with other group members, (7) issues related to stigmatization, privacy, and confidentiality, and (8) personal factors, such as being too sick, tired, or depressed to attend a group [34-36].

Only one study to date [37] has explored reasons why some people with SSc do not participate in support groups. That study reported that the most common reasons for not participating in SSc support groups were not being interested or having no perceived need for support; not having access to a support group; and not being aware that SSc support groups exist. Other reasons included practical barriers, such as not having enough time or being too ill or disabled to attend; emotional factors such as being scared to see patients who are very ill; and having negative perceptions about support groups. An important limitation of the study, however, was that it was based on a single-item that asked respondents to categorize reasons for non-attending as “I’m not interested,” “None are easily available,” or “Other – (please specify).”

Due to the additional challenges faced by people with SSc, many turn to support groups as a way to help them cope with the disease, obtain emotional and practical support, and reduce feelings of isolation. The nature of living with a rare disease, however, may present unique barriers to participating in support groups. Identifying barriers to support group attendance among patients with SSc, including both barriers that are shared with more common diseases and unique barriers, is an important step to better understand how support groups should be structured to increase accessibility and improve the ability of these groups to effectively meet members’ needs. Thus, the objective of the present study was to comprehensively assess reasons for non-participation and barriers to attendance of SSc support groups among people living with SSc.

PATIENTS AND METHODS

Participant Sample

People with SSc were recruited to complete an anonymous survey, which was accessible via the online survey tool *Qualtrics* between April and August 2015. Respondents were recruited through 1) postings on the Scleroderma Society of Canada’s and the Scleroderma Foundation’s websites and other social media venues (e.g., Facebook, Twitter); 2) distribution of flyers at the Scleroderma Foundation’s annual conference; 3) announcements in SSc patient newsletters; 4) emails to support group facilitators and members across Canada and the US; 5) postings on Canadian provincial SSc society websites; and 6) postings in SSc-related chat rooms.

Respondents who accessed the survey website could complete the survey in English or French. After clicking on the survey link and selecting their preferred language, respondents were shown a brief consent form that described study objectives and provided instructions on how to complete the survey. Respondents were given the option to close their browser and not participate or to provide consent by clicking an arrow to continue with the survey. The survey was set up using cookies to prevent respondents from completing the survey more than once, in order to reduce the possibility of duplicate responses. To be included in the present analysis, survey respondents had to confirm that they had been diagnosed with SSc and that they resided in Canada or the United States.

The survey was approved by the Ethics Committee of the Jewish General Hospital in Montréal, Québec. Respondents were not required to provide written informed consent because the survey was done anonymously and did not involve collection of any data that could be used to identify respondents, such as names, dates of birth, or telephone numbers.

The Scleroderma Support Group Non-Attenders Survey

Initial items for the survey were obtained from items that were used in a similar survey related to cancer support groups [38], generated from published results of a qualitative study on reasons for not attending cancer support groups [39], and generated from responses to a single item on reasons for not attending SSc support groups, which was administered as part of the Canadian Scleroderma Patient Survey of Health Concerns [37]. In that survey, respondents who indicated that they had not participated in SSc support groups were asked to specify their reasons for not attending from the following response options: (1) I'm not interested; (2) None are easily available; and (3) Other (please specify). Respondents' open-ended responses to the "Other" option were analyzed using thematic analysis [37].

All initial survey items were reviewed by research team members, who edited individual items, made recommendations to remove items that were less relevant for SSc or were repetitive, and generated new items to reflect content important to SSc that was not included in the initial item set. Items were reviewed iteratively on several occasions by all research team members until consensus on a final item pool was reached. Team members who participated in this process included representatives from the Scleroderma Society of Canada, the Scleroderma Society of Ontario, and the Scleroderma Foundation; a Patient Advisory Board that consisted of six current SSc support group peer facilitators; and researchers with expertise in SSc.

The final survey (see Appendix 1) consisted of 21 core items that assessed possible reasons for not attending SSc support groups. In addition, there were three items that assessed reasons for not attending support groups among survey respondents who had previously attended a SSc support group. There were also four additional items that were only relevant to subgroups of survey respondents (respondents with children and respondents who differed from most other patients due to age, sex, or race/ethnicity).

Response options for all 28 closed-ended items included Not Important, Somewhat Important, Important, and Very Important (scored 0-3).

Data Analysis

Exploratory factor analysis (EFA) was conducted to group survey items into general themes that reflected reasons for not attending support groups. EFA was done with MPlus 7 [40] using weighted least squares mean with variance adjusted estimation, which accounts for the ordinal nature of the survey items, and geomin oblique rotation. The number of factors was chosen based on the scree plot (eigenvalues), model adequacy, and overall interpretability. Model adequacy was assessed using a chi-square goodness-of-fit test and three fit indices, including the Tucker-Lewis Index (TLI) [41], the Comparative Fit Index (CFI) [42], and the Root Mean Square Error of Approximation (RMSEA) [43]. Since the chi-square test is highly sensitive to sample size and can lead to the rejection of well-fitting models, practical fit indices were emphasized [44]. Models with a TLI and CFI close to 0.95 or higher, and RMSEA close to 0.06 or lower are representative of good fitting models [45]. A CFI of 0.90 or above [46] and a RMSEA of 0.08 or less [47] may also be considered to represent reasonably acceptable model fit.

Since the purpose of the present study was not to develop a measure with scoring properties, but rather to identify general themes, we did not remove items or calculate Cronbach’s alpha. Items were included in the theme for which they had the highest factor loading, or the degree that responses were explained by the factor. In a post-hoc analysis, we compared the proportion of patients who rated each item as not important or somewhat important versus important or very important by education level (college and below versus university or postgraduate degree).

RESULTS

Sample Characteristics

There were 276 completed surveys, of which 14 (5%) were excluded because the respondent resided outside of Canada or the United States, 3 (1%) because the respondent reported a diagnosis other than SSc, and 17 (6%) because the respondents were missing some of the 21 survey items (range 7–21). Characteristics of the 242 respondents included in analyses are shown in Table 1. The majority of respondents were female (84%), White (86%), married (63%), and from the United States (65%). Mean age was 56 years, and mean time since SSc diagnosis was 12 years.

Exploratory Factor Analysis

EFA of the 21 core items yielded six eigenvalues greater than one. Based on examination of the scree plot and of the specific items and factor loadings, we judged that a three-factor solution provided the best interpretable model. The three factors had eigenvalues of 6.32, 2.88, and 1.77 respectively, and explained 52.2% of the total variance. Model fit for the three-factor solution was also reasonably good ($\chi^2(150) = 302.7$, $p<0.001$ CFI = 0.91; TLI = 0.88; RMSEA = 0.07). This model fit substantially better than a two-factor model ($\chi^2(169) = 452.4$, $p < 0.001$; CFI = 0.84; TLI = 0.80; RMSEA = 0.08). A four-factor model did not improve fit substantively ($\chi^2(132) = 233.9$, $p < 0.001$; CFI = 0.94; TLI = 0.91; RMSEA = 0.06), and did not yield clearly interpretable factor themes.

The three identified themes representing reasons for not attending SSc support groups included: (1) Personal Reasons (9 items), (2) Practical Reasons (7 items), and (3) Beliefs about Support Groups (5 items). Factor loadings ranged from 0.37 to 0.78 for

Personal Reasons, 0.37 to 0.80 for Practical Reasons, and 0.48 to 0.98 for Beliefs about Support Groups. Between factor correlations were 0.02 for Personal Reasons and Practical Reasons, 0.43 for Personal Reasons and Beliefs about Support Groups, and 0.07 for Practical Reasons and Beliefs about Support Groups. See Table 2. Overall, respondents endorsed a mean of 4.9 (standard deviation (SD) = 3.8; 23% of all items) reasons for not attending as being Important or Very Important (see Figure 1).

The nine items in the Personal Reasons theme (see Table 3) related to current emotional and physical health, concerns about not having time to attend meetings, and need for support. Example items included “I do not need a support group because my symptoms are not severe” and “I do not feel comfortable in a group setting.” On average, respondents endorsed 2.1 Personal Reasons items (SD = 2.0; 23% of Personal Reasons items) as Important or Very Important. The percentage of respondents with ratings of Important or Very Important ranged from 11.1% for item 20 (“I am uncomfortable sharing my experiences with a group”) to 50.4% for item 6 (“I already have enough support from family, friends, or others”). Mean item score was 0.78.

The seven items in the Practical Reasons theme reflected scheduling concerns and issues related to access. Example items included “I do not know of any scleroderma support groups offered in my area” and “The time of the meetings does not fit in my schedule.” The mean number of items endorsed as being Important or Very Important was 1.7 (SD = 1.6; 24% of Practical Reasons items). The percentage of respondents with ratings of Important or Very Important ranged from 8.7% for item 28 (“I am uncomfortable with how I look”) to 48.3% for item 1 (“I do not know of any scleroderma support groups offered in my area”). Mean item score was 0.76.

The five items in the Beliefs about Support Groups theme reflected concerns about the activities and benefits of support groups. Example items included “I think support groups are too negative” and “I do not think I would learn more about scleroderma than I already know.” The mean number of items endorsed as being Important or Very Important was 1.1 (SD = 1.4; 22% of Beliefs about Support Groups items). The percentage of respondents with ratings of Important or Very Important ranged from 18.2% for item 23 (“I do not think support groups provide educational information that is current and relevant”) to 25.6% for item 16 (“I do not think I would learn more about scleroderma than I already know”). Mean item score was 0.73.

Four items, not included in the EFA, were primarily relevant to a subset of respondents who differed from most potential support group participants based on age, gender, whether they had children, and race or ethnicity. Item frequencies are presented in Table 3, and subgroup analyses by age, gender, and ethnicity groupings are presented in Table 4. Subgroup analyses could not be conducted on item 22 (“I do not have available childcare during the meetings”) because data on number of children were not collected. As shown in Table 4, the percentage of respondents with concerns about group makeup was higher among males compared to females, younger respondents, and non-White respondents. Very small subgroup sizes, however, did not allow statistical analysis.

Three items assessed previous support group experiences among a subset of the total respondents (n = 67) who indicated that they had attended a support group in the past. These items included item 25 (“I attended a support group in the past and had a bad experience”), item 26 (“I do not like the current leader of the local support group”), and item 27 (“I do not like the members of the local support group”). As shown in Table 3, almost one quarter of these respondents reported having had a bad experience with a support group in the past as an Important or Very Important reason for non-attendance. Just over 10% indicated that not liking the local support group leader or members of the support group was Important or Very Important to them. There were no statistically significant or substantive differences in item endorsement by education level.

DISCUSSION

The main finding of this study was that reasons for not attending SSc support groups could be described by three different themes that reflected personal reasons, practical reasons, and beliefs about support groups. Many of the reasons for non-attendance identified in the present study were also found to be important in previous studies in more common disease groups [34-36]. The relative importance of barriers to participation, however, may be different in rare conditions, like SSc, where patients often have difficulty finding information about their disease and may struggle to meet other patients with the same rare disease. None of the previous studies that we identified used a structured survey to rate levels of importance of barriers to support group participation among a large number of non-attenders as was used in the present study. Nonetheless, there do seem to be potential differences. For instance, in one study [28], more than half of 62 cancer support group non-attenders indicated that they did not need more information on cancer, whereas only 25% of SSc patients thought they would not learn more about SSc in support groups. Additionally, no previous studies identified concerns about physical appearance as a barrier for attending support groups. This is an important consideration in SSc because many patients experience extensive facial and body disfigurement [31, 48].

Given the predominance of practical reasons for not attending, making online support groups available may be a feasible and economical option for reaching patients without access, because of the unavailability of local support groups, health-related disability, or travel-related barriers [49]. Online support groups have been used successfully in more common disease groups, such as cancer, to improve access [49]. They may also provide an opportunity to reduce potential stressors associated with sharing experiences in a face-to-face context [8].

Some reasons for not attending, however, will not be addressed by increasing access to these groups. Approximately 50% of patients indicated that they did not attend SSc support groups because they already had enough support. For some patients, it may be the case that their need for support is met. On the other hand, there is a long history of successful incorporation of peer support into efforts to improve healthcare and quality of life of people with chronic medical illnesses, and evidence suggests that peers can better understand some of the informational and emotional needs of people with a condition than “embedded social networks” (e.g., family, friends, co-workers, clergy) [50]. Research is needed to determine if SSc support groups similarly improve overall support for patients with existing support systems. To engage these patients, a focused outreach program that can effectively communicate the potential benefits of peer support beyond existing support from embedded networks is needed.

Improved outreach and pre-engagement preparation for potential participants by support group leaders and patient societies may also allay concerns of patients who would potentially benefit from the groups, but who have concerns about seeing others with more severe symptoms or who doubt that they would be able to provide support to others when they themselves are in need of support. Research suggests that peer support is a communal process with benefits to those who receive and who provide the support [50, 51]. Furthermore, patients who initially engage in support groups to find support, over time, become an important source of support for others [50, 51]. Thus, outreach strategies might include video footage portraying a typical SSc support group and content illustrating how patients experiencing all levels of disease severity and physical disfigurement can contribute to and benefit from support groups.

Pre-engagement outreach can be done by patient organizations via their websites, patient newsletters, or at local and national patient education conferences. Another way to potentially increase local access to SSc support groups would be to provide training to peer facilitators. A training program for peer facilitators could provide SSc patients with the skills and training necessary to successfully establish, sustain, and manage support groups in places where none exist. In addition, many respondents expressed concerns about the utility of support groups and the potential for support groups to be overly negative. Trained peer facilitators could address these concerns by managing group dynamics more effectively and by learning how to tailor the activities and discussion topics of each meeting to better meet patients’ needs.

There are several limitations to consider when interpreting the results of this study. First, recruitment occurred through national and provincial SSc organizations, SSc-related chat rooms, and through emails to support group facilitators and members across Canada and the United States, which may have influenced the characteristics of respondents. The survey was conducted via the internet, which may have also influenced the representativeness of the sample. However, the demographic characteristics of respondents were similar to participants in other large SSc patient cohorts [52]. Second, participants were not required to provide documented proof of their diagnosis. However, this is consistent with actual SSc support groups, which do not require documentation of one's SSc diagnosis in order to participate. Third, comorbid somatic and mental health disorders and disease activity were not assessed. Fourth, we combined responses from Canadian and US respondents, and their accessibility to support groups may differ. Fifth, we did not collect information on the other types of support SSc patients may receive from their spouses, caregivers, or significant others, and as a result were unable to assess whether a patients' extended social support network may play a role in influencing attendance. Nonetheless, the study provides important information that can be used to inform the development of a training program for peer facilitators of SSc support groups and to improve the accessibility to and effectiveness of these groups.

In conclusion, peer-led support groups are an important resource for many SSc patients. Patients reported, on average, five Important or Very Important reasons for not attending, including a combination of personal reasons, practical reasons, and beliefs about support groups. These findings will inform SSc organizations on strategies to enhance access to support groups and improve their ability to meet members' needs on a sustained basis.

REFERENCES

1. Joachim G, Acorn S. Life with a rare chronic disease: the scleroderma experience. J Adv Nurs 2003; 42: 598-606.
2. European Organisation for Rare Diseases. Rare diseases: understanding this public health priority. Available: http://www.eurordis.org/IMG/pdf/princeps_document-EN.pdf
3. Kole A, Faurisson F. The voice of 12,000 patients: experiences and expectations of rare disease patients on diagnosis and care in Europe. Available: http://www.eurordis.org/IMG/pdf/voice_12000_patients/EURORDISCARE_FULLBOOKr.pdf
4. Schieppati A, Henter J-I, Daina E, Aperia A. Why rare diseases are an important medical and social issue. Lancet 2008; 371: 2039-41.
5. Kwakkenbos L, Jewett LR, Baron M, Bartlett SJ, Furst D, Gottesman K, et al. The Scleroderma Patient-centered Intervention Network (SPIN) Cohort: protocol for a cohort multiple randomised controlled trial (cmRCT) design to support trials of psychosocial and rehabilitation interventions in a rare disease context. BMJ Open 2013; 3: e003563.
6. Asbring P. Chronic illness - A disruption in life: identity-transformation among women with chronic fatigue syndrome and fibromyalgia. J Adv Nurs 2001; 34: 312-9.
7. Karasz A, Ouellette SC. Role strain and psychological well-being in women with systemic lupus erythematosus. Women Health 1995; 23: 41-57.
8. Osei DK, Lee JW, Modest NN, Pothier PK. Effects of an online support group for prostate cancer survivors: a randomized trial. Nursing 2013; 33: 3.

9. Vilhauer RP. Perceived benefits of online support groups for women with metastatic breast cancer. Women Health 2009; 49: 381-404.
10. Lieberman MA, Winzelberg A, Golant M, Wakahiro M, DiMinno M, Aminoff M, Christine C. Online support groups for Parkinson's patients: a pilot study of effectiveness. Soc Work Health Care 2006; 42: 23-38.
11. Barg FK, Gullatte MM. Cancer support groups: meeting the needs of African Americans with cancer. Semin Oncol Nurs 2001; 17: 171-8.
12. Davison KP, Pennebaker JW, Dickerson SS. Who talks? The social psychology of illness support groups. Am Psychol 2000; 55: 205.
13. Reimann A, Bend J, Dembski B. [Patient-centred care in rare diseases. A patient organisations' perspective]. Bundesgesundheitsblatt Gesundheitsforschung Gesundheitsschutz 2007; 50: 1484-93.
14. Barker J. Living with a life-limiting illness: a personal account. End of Life Care 2009; 3.
15. Vitucci JB. Importance of local support groups for acoustic neuroma and neurofibromatosis patients. Otolaryngol Clin North Am 2012; 45: 531-5.
16. Jalovcic D, Pentland W. Accessing peers’ and health care experts’ wisdom: a telephone peer support program for women with SCI living in rural and remote areas. Top Spinal Cord Inj Rehabil 2009; 15: 59-74.
17. Castro K, Mitchell S, Rowe T, Jacobs M. A chronic graft-versus-host disease (cGVHD) telephone support group: partnering with an advocacy organization to address an underserved population of cancer survivors. Oncol Nurs Forum 2008; 35: 514.

18. Moore S, Teehan C, Cornwall A, Ball K, Thomas J. ‘Hands of time’: the experience of establishing a support group for people affected by mesothelioma. Eur J Cancer Care 2008; 17: 585-92.
19. Alderson J, Madill A, Balen A. Fear of devaluation: understanding the experience of intersexed women with androgen insensitivity syndrome. Br J Health Psychol 2004; 9: 81-100.
20. Breau R, Norman R. The role of self-help groups in educating and supporting patients with prostate cancer and interstitial cystitis. BJU Int 2003; 92: 602-6.
21. Howard MA, Duvall D, Terrell DR, Christopher AT, Thomas I, Holloway N, et al. A support group for patients who have recovered from thrombotic thrombocytopenic purpura-hemolytic uremic syndrome (TTP-HUS): the six-year experience of the Oklahoma TTP-HUS study group. J Clin Apher 2003; 18: 16-20.
22. Stewart MJ, Hart G, Mann K, Jackson S, Langille L, Reidy M. Telephone support group intervention for persons with hemophilia and HIV/AIDS and family caregivers. Int J Nurs Stud 2001; 38: 209-25.
23. Telfair J, Gardner MM. Adolescents with sickle cell disease: determinants of support group attendance and satisfaction. Health Soc Work 2000; 25: 43-50.
24. Locock L, Brown JB. 'All in the same boat'? Patient and carer attitudes to peer support and social comparison in Motor Neuron Disease (MND). Soc Sci Med 2010; 71: 1498-1505.
25. Bernatsky S, Joseph L, Pineau C, Belisle P, Hudson M, Clarke A. Scleroderma prevalence: demographic variations in a population-based sample. Arthritis Rheum 2009; 61: 400-4.

26. Seibold J. Scleroderma. In: Harris ED, Budd RC, Firestein GS, Genovese MC, Sergent JS, Ruddy S, Sledge CB (eds.). Kelley's textbook of rheumatology. 7th ed. Philadelphia: Elsevier; 2005, p. 1279-1308.
27. Delisle VC, Hudson M, Baron M, et al. Sex and time to diagnosis in systemic sclerosis: an updated analysis of 1,129 patients from the Canadian Scleroderma Research Group Registry. Clin Exp Rheumatol. 2014; 32 Suppl 86: S10-S14
28. Kwakkenbos L, van den Hoogen FH, Custers J, Prins J, Vonk MC, van Lankveld WG, et al. Validity of the Fear of Progression Questionnaire - Short Form in patients with systemic sclerosis. Arthritis Care Res 2012; 64: 930-934.
29. Kwakkenbos L, van Lankveld WG, Vonk MC, Becker ES, van den Hoogen FH, van den Ende CH. Disease-realted and psychosocial factors associated with depressive symptoms in patients with systemic sclerosis, including fear of progression and appearance self-esteem. J Psychosom Res 2012; 72: 199-204.
30. Kwakkenbos L, Willems LM, van den Hoogen FH, van Lankveld WG, Beenackers H, van Helmond TF, et al. Cognitive-behavioural therapy targeting fear of progression in an interdisciplinary care program: a case study in systemic sclerosis. J Clin Psychol Med Settings 2014; 21: 297-312.
31. Jewett LR, Hudson M, Thombs BD. A 38-year-old woman with elevated muscle enzymes, Raynaud’s phenomenon and positive anti-topoisomerase I antibody: Is she depressed? In Silver RM, Denton CP (eds.). Case studies in systemic sclerosis. Chicago: Springer. 2011, p. 229-238.
32. Scleroderma Society of Canada. Find a support group. Available: http://www.scleroderma.ca/Support/Find-A-Support-Group.php

33. Scleroderma Foundation. Support groups. Available: http://www.scleroderma.org/site/PageServer?pagename=patients_supportgroups#.Vbec7RZvdFw
34. Sherman AC, Pennington J, Simonton S, Latif U, Arent L, Farley H. Determinants of participation in cancer support groups: the role of health beliefs. Int J Behav Med 2008; 15: 92-100.
35. Ussher JM, Kirsten L, Butow P, Sandoval M. A qualitative analysis of reasons for leaving, or not attending, a cancer support group. Soc Work Health Care 2008; 47: 14-29.
36. Winefield HR, Coventry BJ, Lewis M, Harvey EJ. Attitudes of patients with breast cancer toward support groups. J Psychosoc Oncol 2003; 21: 39-54.
37. Delisle VC, Gumuchian ST, Peláez S, Malcarne VL, El-Baalbaki G, Körner A, et al. Reasons for non-participation in scleroderma support groups. Clin Exp Rheumatol 2016; 34 Suppl 100: S56-62.
38. Winefield HR, Coventry BJ, Lewis M, Harvey EJ. Attitudes of patients with breast cancer toward support groups. J Psychosoc Oncol 2003; 21: 39-54.
39. Ussher JM, Kirsten L, Butow P, Sandoval M. A qualitative analysis of reasons for leaving, or not attending, a cancer support group. Soc Work Health Care 2008; 47: 14-29.
40. Muthén L, Muthén, BO. Mplus user's guide version 3.11. In: Los Angeles, CA: Muthén & Muthén; 1998-2004.
41. Tucker LR, Lewis C. A reliability coefficient for maximum likelihood factor analysis. Psychometrika 1973; 38: 1-10.

42. Bentler PM. Comparative fit indexes in structural models. Psychol Bull 1990; 107: 238.
43. Steiger JH. Structural model evaluation and modification: an interval estimation approach. Multivariate Behav Res 1990; 25: 173-80.
44. Reise SP, Widaman KF, Pugh RH. Confirmatory factor analysis and item response theory: two approaches for exploring measurement invariance. Psychol Bull 1993; 114: 552.
45. Hu Lt, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives. Struct Equ Modeling 1999; 6: 1-55.
46. Kline RB. Principles and practice of structural equation modeling. In: Little TD, editor. New York (NY): The Guilford Press; 2015.
47. Browne MW, Cudeck R, Bollen KA, Long JS. Alternative ways of assessing model fit. In: Bollen KA & Long JS, editors. Testing structural equation models. Beverly Hills (CA): Sage; 1993. p. 136–162.
48. Kwakkenbos L, Delisle VC, Fox RS, Gholizadeh S, Jewett LR, Levis B, et al. Psychosocial aspects of scleroderma. Rheum Dis Clin North Am 2015;41:519-528.
49. Klemm P. Effects of online support group format (moderated vs peer-led) on depressive symptoms and extent of participation in women with breast cancer. Comput Inform Nurs 2012; 30: 9-18.
50. Dennis C-L. Peer support within a health care context: a concept analysis. Int J Nurs Stud 2003; 40: 321-332.

51. Johnson G, Magee C, Maru M, Furlong-Norman K, Rogers ES, Thompson K. Personal and societal benefits of providing peer support: a survey of peer support specialists. Psychiatr Serv 2014; 65: 678-680.

52. Galluccio F, Walker UA, Nihtyanova S, Moinzadeh P, Hunzelmann N, Krieg T, et al. Registries in systemic sclerosis: a worldwide experience. Rheumatology 2011; 50: 60-68.

Table 1. Sociodemographic Characteristics (N = 242)

Variable	
Female gender, *n (%)*	202 (83.5)
Age in years, *mean (standard deviation)*	55.9 (12.7)
Location, *n (%)*	
United States	156 (64.5)
Canada	86 (35.5)
Race/ethnicity, *n (%)*	
White	209 (86.4)
Other	23 (9.5)
Two or more	10 (4.1)
Marital status, *n (%)*	
Never married	23 (9.5)
Married	153 (63.2)
Living with partner in committed relationship	18 (7.4)
Separated	4 (1.7)
Divorced	34 (14.0)
Widowed	10 (4.1)
Level of education, *n (%)*	
Elementary/primary school	3 (1.2)
Secondary/high school	41 (16.9)
Some college/university	101 (41.7)

University degree	59 (24.4)
Postgraduate degree	38 (15.7)
Occupational status, *n (%)*	
Homemaker	24 (9.9)
Unemployed	9 (3.7)
Retired	67 (27.7)
On disability	55 (22.7)
On leave of absence	4 (1.7)
Full-time employed	49 (20.2)
Part-time employed	24 (9.9)
Full-time student only	10 (4.1)
Scleroderma Diagnosis, *n (%)*	
Limited Scleroderma	126 (52.1)
Diffuse Scleroderma	79 (32.6)
Not known	37 (15.3)
Years since SSc diagnosis, *mean (standard deviation)*	11.5 (7.8)

Table 2: Exploratory Factor Analysis: Factor Loadings

Item and Factor Groupings	Factor 1: Personal Reasons	Factor 2: Practical Reasons	Factor 3: Beliefs about Support Groups
Factor 1: Personal Reasons			
2: I do not need a support group because my symptoms are not very severe.	**0.60***	-0.02	0.01
3: I am too busy with other responsibilities, such as work or children, to attend a support group.	**0.63***	-0.04	-0.07
4: I am uncomfortable seeing other people with scleroderma who may be worse off than me.	**0.78***	0.01	-0.23
6: I already have enough support from family, friends, or others.	**0.37***	-0.06	-0.03
10: I do not feel comfortable in a group environment.	**0.62***	0.26*	0.13
13: I prefer not to see myself as a "scleroderma patient."	**0.57***	-0.02	0.01
14: I do not know enough about what happens at a support group.	**0.44***	0.28*	0.14
17: I feel too depressed or emotionally	**0.60***	0.51*	0.00

overwhelmed to attend a support group.			
20: I am uncomfortable sharing my experiences with a group.	**0.59***	0.39*	-0.02
Factor 2: Practical Reasons			
1: I do not know of any scleroderma support groups offered in my area.	-0.09	**0.37***	0.05
7: My scleroderma symptoms are severe and make it difficult to attend the meetings.	-0.04	**0.58***	0.11
11: The time of the meetings does not fit in my schedule.	0.30*	**0.47***	-0.01
12: I do not have a reliable way to get to the meetings.	0.04	**0.80***	-0.05
18: I am worried that my privacy will not be respected.	0.43*	**0.52***	0.15
21: Getting to and from the meetings is inconvenient due to weather, distance, or other factors.	-0.02	**0.80***	-0.17
28: I am uncomfortable with how I look.	0.26*	**0.58***	-0.02
Factor 3: Beliefs about Support Groups			
5: I think support groups are too negative.	0.21*	-0.10	**0.49***
9: I do not think support groups are helpful.	0.39*	-0.05	**0.48***
16: I do not think I would learn more about scleroderma than I already know now.	0.27*	0.01	**0.59***

23: I do not think support groups provide educational information that is current and relevant.	-0.02	0.14	**0.98***
24: I think support groups spend too much time discussing non-scleroderma related topics.	0.05	0.15	**0.72***

*Statistically significant, $p < 0.05$.

Table 3. Items and Frequencies (N = 242)

	Not Important n (%)	Somewhat Important n (%)	Important n (%)	Very Important n (%)	Item Mean (SD)
Factor 1: Personal Reasons					
2: I do not need a support group because my symptoms are not very severe.	123 (50.8%)	66 (27.3%)	33 (13.6%)	20 (8.3%)	0.79 (0.97)
3: I am too busy with other responsibilities, such as work or children, to attend a support group.	117 (48.3%)	60 (24.8%)	39 (16.1%)	26 (10.7%)	0.89 (1.03)
4: I am uncomfortable seeing other people with scleroderma who may be worse off than me.	129 (53.3%)	39 (16.1%)	30 (12.4%)	44 (18.2%)	0.96 (1.18)
6: I already have enough support from family, friends, or others.	59 (24.4%)	61 (25.2%)	62 (25.6%)	60 (24.8%)	1.51 (1.11)
10: I do not feel comfortable in a group environment.	146 (60.3%)	49 (20.3%)	31 (12.8%)	16 (6.6%)	0.66 (0.94)
13: I prefer not to see myself as a "scleroderma patient."	154 (63.6%)	49 (20.3%)	23 (9.5%)	16 (6.6%)	0.59 (0.91)
14: I do not know enough about	145 (59.9%)	52 (21.5%)	26 (10.7%)	19 (7.9%)	0.67 (0.96)

what happens at a support group.					
17: I feel too depressed or emotionally overwhelmed to attend a support group.	164 (67.8%)	45 (18.6%)	25 (10.3%)	8 (3.3%)	0.49 (0.81)
20: I am uncomfortable sharing my experiences with a group.	163 (67.4%)	52 (21.5%)	16 (6.6%)	11 (4.5%)	0.48 (0.81)
Factor 2: Practical Reasons					
1: I do not know of any scleroderma support groups offered in my area.	76 (31.4%)	49 (20.3%)	47 (19.4%)	70 (28.9%)	1.46 (1.21)
7: My scleroderma symptoms are severe and make it difficult to attend the meetings.	137 (56.6%)	53 (21.9%)	35 (14.5%)	17 (7.0%)	0.72 (0.96)
11: The time of the meetings does not fit in my schedule.	136 (56.2%)	27 (11.2%)	43 (17.8%)	36 (14.9%)	0.91 (1.15)
12: I do not have a reliable way to get to the meetings.	180 (74.4%)	19 (7.9%)	19 (7.9%)	24 (9.9%)	0.53 (1.00)
18: I am worried that my privacy will not be	179 (74.0%)	35 (14.5%)	19 (7.9%)	9 (3.7%)	0.41 (0.79)

respected.					
21: Getting to and from the meetings is inconvenient due to weather, distance, or other factors.	127 (52.5%)	39 (16.1%)	31 (12.8%)	45 (18.6%)	0.98 (1.18)
28: I am uncomfortable with how I look.	192 (79.3%)	29 (12.0%)	17 (7.0%)	4 (1.7%)	0.31 (0.68)
Factor 3: Beliefs about Support Groups					
5: I think support groups are too negative.	132 (54.5%)	51 (21.1%)	27 (11.2%)	32 (13.2%)	0.83 (1.08)
9: I do not think support groups are helpful.	129 (53.3%)	52 (21.5%)	37 (15.3%)	24 (9.9%)	0.82 (1.03)
16: I do not think I would learn more about scleroderma than I already know now.	130 (53.7%)	50 (20.7%)	32 (13.2%)	30 (12.4%)	0.84 (1.07)
23: I do not think support groups provide educational information that is current and relevant.	163 (67.4%)	35 (14.5%)	29 (12.0%)	15 (6.2%)	0.57 (0.93)
24: I think support groups spend too much time discussing non-scleroderma related topics.	152 (62.8%)	45 (18.6%)	36 (14.9%)	9 (3.7%)	0.60 (0.88)

Additional Items:					
8: I do not think the group would have enough people of a similar cultural background in it.	187 (77.3%)	30 (12.4%)	14 (5.8%)	11 (4.5%)	0.38 (0.79)
15: I do not think the group would have enough people of a similar age to my age.	176 (72.7%)	41 (16.9%)	16 (6.6%)	9 (3.7%)	0.41 (0.78)
19: I do not think the group would have enough people of the same gender as me in it.	206 (85.1%)	24 (9.9%)	8 (3.3%)	4 (1.7%)	0.22 (0.58)
22: I do not have available childcare during the meetings.	217 (89.7%)	11 (4.5%)	7 (2.9%)	7 (2.9%)	0.91 (0.62)
Items Assessing Previous Support Group Experiences (n = 67):					
25: I attended a support group in the past and had a bad experience.	35 (52.2%)	16 (23.9%)	9 (13.4%)	7 (10.5%)	0.82 (1.03)
26: I do not like the current leader of the local support group.	55 (82.1%)	4 (6.0%)	5 (7.5%)	3 (4.5%)	0.34 (0.81)
27: I do not like the members of the local support group.	56 (83.6%)	4 (6.0%)	4 (6.0%)	3 (4.5%)	0.31 (0.78)

Table 4. Subgroup Analyses for Subgroup-Relevant Items (N = 242)

	Respondents (n)	Important and Very Important (n, %)
Item #19: I do not think the group would have enough people of the same gender as me in it.		
Gender		
Male	40	6 (15.0%)
Female	202	6 (3.0%)
Item #15: I do not think the group would have enough people of a similar age to my age.		
Age:		
18-29	10	5 (50%)
30-39	13	2 (15.4%)
40-49	37	4 (10.8%)
50-59	90	7 (7.8%)
60-69	58	3 (5.1%)
70-85	34	4 (11.7%)
Item #8: I do not think the group would have enough people of a similar cultural background in it.		
Ethnicity/Race		
White	209	15 (7.1%)
Black	4	1 (25.0%)
Asian	3	0 (0.0%)

Aboriginal/American Indian	5	3 (60.0%)
Two or more	10	2 (20.0%)
Other	11	4 (36.4%)

Figure 1. Frequencies of items ranked Important or Very Important by respondents. N = 242, mean = 4.9, standard deviation = 3.8.

Appendix 1. Demographics and Scleroderma Support Group Non-Attenders Survey

DEMOGRAPHICS

Directions: The following demographic questions are necessary for us to gain a better understanding of who has completed the survey. The items below will not require you to report any information that could lead to your identification.

1. Please indicate your gender: ☐ Male ☐ Female
2. Please indicate your age in years: ....
3. In which country do you live?
    - Canada
    - United States
    - Other……
4. In which Canadian province or territory do you live?
    - AB
    - BC
    - MB
    - NB
    - NL
    - NS
    - NT
    - NU
    - ON
    - PE
    - QC
    - SK
    - YT
5. In which American state do you live?
    - AL
    - AK
    - AZ
    - AR
    - CA
    - CO
    - CT
    - DE
    - FL
    - GA
    - HI
    - ID
    - IL
    - IN
    - IA
    - KS
    - KY
    - LA
    - ME
    - MD
    - MA
    - MI
    - MN
    - MS
    - MO
    - MT
    - NE
    - NV
    - NH
    - NJ
    - NM
    - NY
    - NC
    - ND
    - OH
    - OK
    - OR
    - PA
    - RI

  - SC
  - SD
  - TN
  - TX
  - UT
  - VT
  - VA
  - WA
  - WV
  - WI
  - WY

6. **For Canadians:** What is your racial or ethnic background? (Please check all that apply)
    - White
    - Black
    - Asian
    - Aboriginal
    - Other. Please specify: ______________

7. **For Americans:** What is your racial or ethnic background (Please check all that apply):
    - White
    - Black or African-American
    - American Indian/Alaska Native
    - Asian
    - Native Hawaiian/Other Pacific Islander
    - Other. Please specify: ______________

8. What is your current relationship status?
    - Never married
    - Married
    - Living with partner in committed relationship
    - Separated
    - Divorced
    - Widowed

9. What is the highest level in school that you completed?
    - Elementary/primary school
    - Secondary/high school
    - Some college/university
    - University degree
    - Postgraduate degree

10. What is your current occupational status?
    - Homemaker
    - Unemployed
    - Retired
    - On disability
    - On leave of absence
    - Full-time employed
    - Part-time employed

  - Full-time student only

11. **For Canadians:** What is your family household income (from all sources):
    - Less than $20,000 CAD
    - Between $20,001 and $40,000 CAD
    - Between $40,001 and $60,000 CAD
    - Between $60,001 and $80,000 CAD
    - $80,001 or greater CAD
    - Prefer not to answer

12. **For Americans:** What is your family household income (from all sources):
    - Less than $20,000 USD
    - Between $20,001 and $40,000 USD
    - Between $40,001 and $60,000 USD
    - Between $60,001 and $80,000 USD
    - $80,001 or greater USD
    - Prefer not to answer

13. What is your scleroderma diagnosis?
    - Limited Scleroderma
    - Diffuse Scleroderma
    - CREST
    - I don’t know
    - Other

14. How many years has it been since you first received your scleroderma diagnosis?
    - 0-1
    - 1
    - 2
    - 3
    - 4
    - 5
    - 6
    - 7
    - 8
    - 9
    - 10
    - 11
    - 12
    - 13
    - 14
    - 15
    - 16
    - 17
    - 18
    - 19
    - 20
    - 21
    - 22
    - 23
    - 24
    - 25+

Scleroderma Support Group Non-Attenders Survey

Reasons for Not Attending Support Groups

Directions: We are interested in knowing more about the factors that may influence your decision to not attend a support group. Please indicate the importance of the following reasons.

Response Options:
☐ Very Important ☐ Important ☐ Somewhat Important ☐ Not Important

1. I do not know of any scleroderma support groups offered in my area.
2. I do not need a support group because my symptoms are not very severe.
3. I am too busy with other responsibilities, such as work or children, to attend a support group.
4. I am uncomfortable seeing other people with scleroderma who may be worse off than me.
5. I think support groups are too negative.
6. I already have enough support from family, friends, or others.
7. My scleroderma symptoms are severe and make it difficult to attend the meetings.
8. I do not think the group would have enough people of a similar cultural background in it.
9. I do not think support groups are helpful.
10. I do not feel comfortable in a group environment.
11. The time of the meetings does not fit in my schedule.
12. I do not have a reliable way to get to the meetings.
13. I prefer not to see myself as a "scleroderma patient".
14. I do not know enough about what happens at a support group.
15. I do not think the group would have enough people of a similar age to my age.
16. I do not think I would learn more about scleroderma than I already know now.
17. I feel too depressed or emotionally overwhelmed to attend a support group.
18. I am worried that my privacy will not be respected.
19. I do not think the group would have enough people of the same gender as me in it.
20. I am uncomfortable sharing my experiences with a group.
21. Getting to and from the meetings is inconvenient due to weather, distance, or other factors.
22. I do not have available childcare during the meetings.
23. I do not think support groups provide educational information that is current and relevant.
24. I think support groups spend too much time discussing non-scleroderma related topics.
25. I attended a support group in the past and had a bad experience. (Administered to respondents who indicated attending a support group in the past).
26. I do not like the current leader of the local support group. (Administered to respondents who indicated attending a support group in the past).

27. I do not like the members of the local support group. (Administered to respondents who indicated attending a support group in the past).
28. I am uncomfortable with how I look.