

Challenges and Support Service Preferences of Informal Caregivers of People with Systemic Sclerosis: A Cross-sectional Survey

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Abstract

Purpose: To evaluate the importance of different challenges experienced by informal caregivers to persons with systemic sclerosis (SSc) and identify priorities for support services that could be developed.

Materials and Methods: Caregivers of people with SSc from three continents completed an online questionnaire to rate the importance of possible caregiver challenges and likelihood of using different forms of support services. Importance of challenges and likelihood of using support services were rated from 1 (not important; not likely to use) to 4 (very important; very likely to use).

Results: 202 informal caregivers completed the survey (79 women, 123 men). Mean age was 58 years (standard deviation = 13). The most important challenges were related to supporting the care recipient with emotional difficulties and physical discomfort. Caregivers indicated that they would be more likely to use support services that involved online or hard-copy information resources, including those provided soon after diagnosis, compared to support that involved interacting with others.

Conclusions: Supporting the care recipient in managing emotional difficulties and physical discomfort were important challenges among caregivers. Interventions delivered through hardcopy or online resources, including those delivered soon after the care recipient's diagnosis, were rated as being most likely to be used by caregivers.

Key Indexing Terms: Scleroderma, rare disease, caregivers, surveys, questionnaires

Introduction

Systemic sclerosis (SSc), also known as scleroderma, is a rare chronic autoimmune disease. SSc is characterized by abnormal fibrotic processes and excessive collagen production, which results in skin thickening, damage to internal organs including the lungs, kidneys, and gastrointestinal tract, as well as vascular implications [1]. Women comprise more than 80% of cases [2].

Patients with SSc experience diverse challenges that affect their quality of life, including gastrointestinal symptoms, respiratory problems, fatigue, and changes in appearance [3]. In addition to disease manifestations, persons diagnosed with SSc face difficulties related to the rarity of the disease. As compared to more common diseases, challenges include an uncertain prognosis, limited treatment options, difficulty accessing specialists, geographic distance from treatment centres, and a lack of disease-specific support resources [4, 5].

Many persons with rare diseases, including SSc, rely on support from informal caregivers [4, 6, 7]. Informal caregivers are typically family members who do not receive training or payment for their role [8]. Little is known about the experiences of informal caregivers of people with rare diseases, and we identified only one study of informal caregivers of people with SSc [7]. In that study, a doctoral thesis, 13 informal caregivers were interviewed, and the emotional challenges of caregiving of a person with SSc were emphasized [7].

Developing resources that address challenges faced by informal caregivers for persons with SSc could help reduce the negative consequences associated with caregiving. To develop relevant resources, an understanding of the types of challenges that are most important to caregivers of those with SSc and the support services they would be most likely to use is required. Thus, we conducted a series of nominal group technique (NGT) discussions with 13

SSc caregivers to generate survey items that reflected challenges faced by informal SSc caregivers and the caregivers' preferences for types of support services that they believed would be useful [9]. We worked with the caregivers and developed a list of 61 unique challenges in the domains of physical health concerns; financial and work or employment problems; role strain; the need for information, resources, and support; fear, anxiety, and uncertainty; general emotional difficulties; emotional difficulties of the care recipient; changes in social interactions with others; and changes in relationship dynamics with the care recipient. A list of 18 potentially useful support services was also generated, including both online and in-person support methods [9]. The objective of the present study was to assess the frequency and importance of the 61 challenges and of caregivers' preferences for the 18 types of support services identified via the NGT discussions. To do this, we disseminated a survey, developed based on our NGT discussions, to an international sample of caregivers of persons with SSc.

Materials and Methods

Participant Sample and Procedure

Informal caregivers of a person diagnosed with SSc were recruited to anonymously complete an online questionnaire with the survey tool *Qualtrics*, between December 2016 and June 2017. To be eligible for the study, participants had to indicate that they currently or previously provided unpaid care for a friend or family member with SSc. Participants had to be 18 years or older and fluent in English or French. Participants were recruited through SSc patient organizations, including Scleroderma Canada, the Scleroderma Foundation of the United States, Scleroderma and Raynaud's UK, the Association des Sclérodermiques de France, and the Scleroderma Association of New South Wales, Australia. Recruitment also occurred through

emails and posts on SSc-related websites and other social media venues. Advertisements were also emailed to people with SSc participating in an ongoing internet-based cohort [10].

Respondents who accessed the survey website could complete the survey online in English or French. After clicking on the survey link, respondents were shown a consent form that described study objectives and survey instructions. Respondents were given the option to consent by clicking an arrow to continue the survey or to close their browser and not participate. This study was approved by the Research Ethics Committee of the Jewish General Hospital, Quebec, Canada.

Measures

Caregiver, Care Recipient, and Caregiving Characteristics.

Caregivers provided their age, gender, country of origin, race/ethnicity, relationship status, highest level of education achieved, current and past occupational status, and household income. They also provided care recipient information, including age, gender, SSc subtype (diffuse or limited), and years since diagnosis, and caregiving characteristics, including years of caregiving, relationship with the care recipient, length of relationship with the care recipient, hours a week providing care, and types of activities with which they assist or assisted the care recipient.

Challenges Associated with Caregiving

A 61-item questionnaire to measure challenges associated with caregiving was developed from three NGT discussions that involved caregivers to persons with SSc [9]. The use of the NGT allowed SSc caregivers to directly share challenges that they experience. The list of challenges was reviewed and revised by a SSc Caregiver Advisory Team prior to finalizing the questionnaire for the present study [9]. The challenges questionnaire developed from the discussion items includes challenges that were previously [9] grouped thematically into 9

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categories to facilitate ease of reviewing similarly themed items: (1) physical health concerns (3 items); (2) financial problems and work or employment problems (4 items); (3) role strain (8 items); (4) information, resources, and support needs (15 items); (5) fear, anxiety, and uncertainty (3 items); (6) general emotional difficulties (7 items); (7) emotional difficulties of the care recipient (8 items); (8) changes in social interactions with others (3 items); and (9) changes in relationship dynamics with the care recipient (10 items). Caregivers rate each item from 1 (“not important”) to 4 (“very important”) based on the perceived importance of the challenge to them.

Support Service Preferences for Caregivers

An 18-item support services questionnaire was previously developed through NGT discussions at the same time the challenges questionnaire was developed and using the same method. This questionnaire assesses the support services that caregivers to persons with SSc believe they would be most likely to use. Example items include “caregiver-led breakout groups at patient conferences” and “caregiver newsletter”. Caregivers rate items from 1 (“not likely”) to 4 (“very likely”) based on the caregiver’s perceived likeliness of using the support service. Likelihood of using each of the 18 support services was evaluated separately.

Statistical Analyses

Descriptive analyses were performed for demographic variables. Continuous variables were presented as means and standard deviations (SDs), and categorical variables were presented as percentages and counts. Frequencies were presented for all items included in the challenges and support service questionnaires. Since the purpose of the questionnaire was not to develop a measure with scoring properties, but rather to identify important challenges for caregivers, we did not calculate Cronbach’s alpha or conduct analyses of measurement properties. Given

previously identified gender differences for caregiving [11], after having identified study objectives we decided to consider potential gender differences in demographic factors and survey responses using chi-square tests. The Hochberg Sequential Method was used to adjust for multiple comparisons [11] and results are presented with confidence intervals. All statistical analyses were performed using SPSS Statistics, version 22.0 (Chicago, IL).

Results

Sample Characteristics

A total of 262 people who indicated that they were past or current informal caregivers of a person with SSc accessed the survey. Of these, 202 (77%) completed the entire survey and were included in this study (see table 1). The majority of caregivers were from North America (74%) or Europe (23%). The mean age was 57 years (SD = 14 years), and 123 were male (61%). Caregivers were providing care for a partner (72%), parent (12%), child (7%), sibling (4%) or friend (5%). They were employed (49%), retired (40%), students (2%), or homemakers, unemployed, on disability, or on a leave of absence (11%). Twenty-eight percent of caregivers had pursued postgraduate degree, 27% completed a university degree, 24% completed some university, and 21% completed primary to high school education. Caregivers provided a mean of 14 (SD = 13) hours of care per week. The mean age of care recipients was 58 years (SD = 13) and approximately half were diagnosed with diffuse SSc (50%).

[Insert table 1 about here]

Responses to Challenges and Support Services Questionnaires

Challenges Questionnaire

Item Responses. Table 2 shows the percentage of caregivers who rated challenges as “important” or “very important” for the 61 items grouped into the 9 challenge categories.

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Overall, item means ranged from 2.0 to 3.2. The percentage of respondents who rated challenges as “important” or “very important” ranged from 31% to 92%. The item that was rated highest (92%) was “providing emotional support to my care recipient on challenging days”. The item with the lowest percentage (31%) of “important” or “very important” ratings was “feeling ashamed to think about my own well-being or needs”.

Physical health concerns (3 items). The mean of item ratings ranged from 2.2 to 2.9. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 36% to 66%. More than 50% of caregivers rated 1 of the 3 items as “important” or “very important”.

Financial problems and work or employment problems (4 items). The mean of item ratings ranged from 2.0 to 2.5. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 32% to 53%. More than 50% of caregivers rated 1 of the 4 items as “important” or “very important”.

Role strain (8 items). The mean of item ratings ranged from 2.2 to 3.1. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 37% to 84%. More than 50% of caregivers rated 5 of 8 items as “important” or “very important”.

Information, resources, and support needs (15 items). The mean of item ratings ranged from 2.1 to 2.9. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 34% to 69%. More than 50% of caregivers rated 11 of 15 items as “important” or “very important”.

Fear, anxiety, and uncertainty (3 items). The mean of item ratings ranged from 2.5 to 3.0. The percentage of respondents who rated these challenges as “important” or “very important”

was greater than 50% for all 3 items (51% to 72%). More than 50% of caregivers rated all 3 items as “important” or “very important”.

General emotional difficulties (7 items). Seven challenges were included in this category, and the mean of item ratings ranged from 2.1 to 2.9. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 31% to 68%. More than 50% of caregivers rated 6 of 7 items as “important” or “very important”.

Emotional difficulties of the care recipient (8 items). Eight challenges were included in this category, and the mean of item ratings ranged from 2.3 to 3.2. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 41% to 92%. More than 50% of caregivers rated 7 of 8 items as “important” or “very important”.

Changes in social interactions with others (3 items). Three challenges were included in this category, and the mean of item ratings ranged from 2.4 to 2.9. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 41% to 68%. More than 50% of caregivers rated 1 of 3 items as “important” or “very important”.

Changes in relationship dynamics with care recipient (10 items). Ten challenges were included in this category, and the mean of item ratings ranged from 2.4 to 3.1. The percentage of respondents who rated these challenges as “important” or “very important” ranged from 33% to 82%. More than 50% of caregivers rated 9 of 10 items as “important” or “very important”.

Support Services Questionnaire

Table 3 shows the percentage of caregivers who rated each of the 18 items as a support service that they were “likely to use” or “very likely to use”. Overall, the mean of item ratings ranged from 1.6 to 2.7. The percentage of respondents who rated services as “likely to use” or “very likely to use” ranged from 15% to 59%. The item that was rated highest (59%) was a hard-

copy resource for caregivers (“caregiver newsletter”). The item with the lowest percentage (15%) of at least “likely to use” ratings was a telephone-based resource, “caregiver-led telephone-based support group for caregivers”.

[Insert table 2 about here]

Comparisons Between Women and Men

As shown in table 1, there were significant demographic and caregiving differences between women and men for caregiver age, years of caregiving, and years since the care recipient received their diagnosis. Men were significantly older than women, had been caregiving for longer, and had care recipients who had been diagnosed for a longer time. Current occupational status and relationship to care recipient also differed. A greater proportion of men (48%) were retired compared to women (27%), and men were more often caring for a partner (76%) compared to women (29%). Twenty-seven percent of women were caring for a child whereas just 2% of men were caring for a child.

As shown in table 2, the proportion of women who rated challenges as “important” or “very important” were higher than for men on 59 of 61 items. There were statistically significant differences, after adjusting for multiple comparisons, on five challenges, including “finding time for myself”, “not having access to a caregiver support group”, “finding assistance for things that my care recipient use to do”, “feeling ashamed to think about my own well-being or needs”, and “noticing others’ lack of knowledge and awareness about scleroderma” as “important” or “very important”. For each of these items women rated the challenge as being more important than men.

A greater percentage of women than men rated each of the 18 support service items as “likely” or “very likely” to use. These differences were not statistically significant (see table 3).

[Insert table 3 about here]

Discussion

We surveyed 202 informal caregivers of people with SSc to determine the most important challenges and the likelihood that caregivers would use different types of support services, if available. Challenges that were most consistently rated as being “important” or “very important” were related to difficulty addressing the emotional needs of the care recipient (e.g., “providing emotional support to my care recipient on challenging days”) and feeling unable to lessen the care recipients discomfort (e.g., “being unable to help address my care recipient’s pain or discomfort”). Alternatively, items related to financial, work, and employment difficulties were the least likely to be rated as being important.

Among support services that were included in the survey, at least 50% of caregivers rated being “likely” or “very likely” to use information-based support services, including those provided soon after SSc diagnosis (e.g., “information about scleroderma on an online reputable website for caregivers of newly diagnosed patients”). On the other hand, few informal caregivers rated interactive support services such as support groups or peer support as resources that they would “likely” or “very likely” use (e.g., “caregiver-led in-person caregiver support group”).

Only five challenges had statistically significant differences between women and men, controlling for multiple comparisons. Women and men differed substantively and statistically significantly in terms of demographics and the characteristics of their care recipients. For example, the majority of men cared for a partner; whereas, women often cared for a partner, child, or parent. Given the large number of items and the relatively small numbers of women and men in certain characteristics where they differed (e.g., relation to care recipient), we did not conduct multivariable analyses, and it is possible that some of the differences identified may

reflect factors unrelated to gender. In addition to being more likely to rate challenges as at least “important”, women rated being more likely to use the 18 support service items.

Prior to our NGT study, the only previous study on caregivers of people living with SSc that we identified was a doctoral thesis involving individual interviews with 13 caregivers [7]. Consistent with the present study, the thesis emphasized difficulties of caregivers in addressing the emotional difficulties of care recipients.

Caregivers who completed our survey indicated that they preferred information-based supports. We have not identified studies of supportive interventions tested among SSc caregivers, however, there are websites that provide information for informal SSc caregivers [12, 13]. Many different kinds of support services have been developed and tested among caregivers to persons with more common diseases [14, 15, 16], including newsletter resources, information packages, support groups, and psychotherapy tailored for caregivers. In a survey of 188 caregivers to elderly individuals, similar to our findings, caregivers reported being more interested in a newsletter developed for caregivers than interventions that require face-to-face or simultaneous virtual contact, such as support groups or help from a volunteer [17].

The use of information packages delivered online or through hard-copy resources has been previously studied. For example, among caregivers of children with special needs, a “Keeping it Together” (KIT) information package has been tested in Canada and Australia [18, 19]. This package included information about accessing resources and communicating information to care recipients. In Canada, the utility of the KIT was evaluated among 440 parents of children with special needs. After using the KIT, parents’ perceptions of their ability, confidence, and satisfaction in using information in different settings significantly improved [18]. Tailoring

similar interventions to caregivers of persons with SSc may help to alleviate burden associated with the challenges faced in their caregiver role.

Web-based psychoeducation interventions delivered soon after diagnosis have also been tested among informal caregivers to persons with more common health conditions such as cancer. For example, a program that was originally nurse-delivered was adapted to be applied online for patients newly diagnosed with cancer and their family members [20]. The web-based intervention provided information and support tailored to the needs of patients and their caregiver (e.g., communicating with each other). Thirty-eight dyads accessed the online program which included three sessions. After delivery of the intervention, significant reductions in emotional distress were found among patients and caregivers, as well increased quality of life and perceived benefits of caregiving [20]. Participants reported being satisfied with the program usability but wanted additional content. Caregivers in our study rated support services that could be used independently and in their home higher than support services that involved other caregivers or professionals, suggesting that developing web-based psychoeducational content for caregivers of persons with SSc may be an intervention that caregivers would be likely to use.

Important limitations should be considered when interpreting study findings. First, participants were recruited through an ongoing SSc patient cohorts, patient organizations, and social media websites. These recruitment methods could have resulted in an over representation of caregivers who are actively involved in their care recipient's diagnosis. Second, this survey was distributed online among caregivers that were fluent in English or French, therefore only caregivers with a computer and internet access could participate. Lastly, our study surveyed the challenges and support services preferences of caregivers but did not explore why certain challenges were deemed more important than others, or why the support service preferences

were selected as being likely or unlikely to be used. Understanding the rationale for these preferences could help in better tailoring support services for caregivers to persons with SSc.

In sum, caring for a person with SSc can be a challenging role. The most important challenges to caregivers involved supporting their care recipients with their emotional difficulties and physical discomfort. Caregivers indicated that they would be most likely to utilize hardcopy and internet-based information resources. Providing a caregiver information newsletter in addition to reliable information about SSc may help to address caregiver needs. Future research should consider the challenges and support service preferences that caregivers have identified to develop and test interventions that positively impact the caregiving experience.

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Authors' Contribution

DBR, and BDT were responsible for the study conception and design; data acquisition, analysis, and interpretation; and drafting and revising the manuscript. DBR, MCA, and ACJ were responsible for data acquisition, analysis, and interpretation. DBR and BDT drafted and revising the manuscript. MEC and JC were responsible for acquisition of data. VLM, MH, and the Scleroderma Caregiver Advisory Committee were responsible for interpretation of data analysis. All authors reviewed, provided critical input, and approved the final version of this manuscript.

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Table 1. Sociodemographic information among 202 informal caregivers.

Variable	Men	Women
Number, <i>n</i> (%)	123 (60.9)	79 (39.1)
Age, mean (standard deviation)	60.6 (12.5)	51.8 (15.4)
Relationship status, <i>n</i> (%)		
Never Married	3 (2.4)	15 (19.0)
Married	107 (87.0)	47 (59.5)
Living with partner in committed relationship	9 (7.3)	5 (6.3)
Separated or divorced	3 (2.4)	7 (8.9)
Widowed	1 (0.8)	5 (6.3)
Occupational status before caregiving, <i>n</i> (%)		
Employed	90 (73.2)	56 (70.9)
Retired	26 (21.1)	10 (12.7)
Students	1 (0.8)	5 (6.3)
Other	6 (4.9)	8 (10.1)
Current occupational status, <i>n</i> (%)		
Employed	55 (44.7)	40 (50.6)
Retired	59 (48.0)	21 (26.6)
Students	0 (0.0)	4 (5.1)
Other	9 (7.3)	14 (17.7)
Highest level of education obtained, <i>n</i> (%)		
Primary to high school	32 (26.0)	11 (13.9)

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Some college or university	29 (23.6)	19 (24.1)
University degree	26 (21.1)	28 (35.4)
Postgraduate degree	36 (29.3)	21 (26.6)
SSc subtype, <i>n</i> (%)		
Limited scleroderma or CREST	47 (38.2)	21 (26.6)
Diffuse scleroderma	57 (46.3)	43 (54.4)
Unknown or not specified	19 (15.4)	15 (19.0)
Age of person with SSc, <i>mean (standard deviation)</i>	58.8 (12.5)	56.7 (14.6)
Years since care recipient's diagnosis, <i>mean (standard deviation)</i>	13.2 (7.5)	11.0 (6.1)
Years of providing care for care recipient, <i>mean (standard deviation)</i>	31.9 (12.5)	28.0 (13.2)
Relation to person with SSc, <i>n</i> (%)		
Parent	3 (2.4)	12 (15.2)
Child	3 (2.4)	21 (26.6)
Partner	115 (93.5)	31 (39.2)
Sibling	1 (0.8)	6 (7.6)
Friend	1 (0.8)	8 (10.1)
Other	0 (0.0)	1 (1.3)*
Length of relationship with care recipient, <i>mean (standard deviation)</i>	22.8 (6.3)	23.4 (5.7)

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Hours of care per week, <i>mean (standard deviation)</i>	13.9 (12.9)	14.9 (14.2)
Caregiving Tasks, <i>n (%)</i>		
Transportation	83 (67.5)	38 (48.1)
Housework	99 (80.5)	52 (65.8)
Preparing meals	72 (58.5)	47 (59.5)
Managing finances	45 (36.6)	21 (26.6)
Shopping	87 (70.7)	50 (63.3)
Medical tasks	39 (31.7)	30 (38.0)
Arranging other services for care recipient	8 (6.5)	15 (19.0)
Other	31 (25.2)	35 (44.3)

SSc = systemic sclerosis

* Cousin

Table 2. Frequencies for challenge items rated as important and very important among 202 informal caregivers.

	Total Sample		Men versus Women		
		Important or Very Important	Men (N = 123): Important or Very Important	Women (N = 79): Important or Very Important	95% CI Lower Limit – Upper Limit
	Item Mean (SD)	n (%)	n (%)	n (%)	
Physical health concerns					
1. Experiencing fatigue and physical exhaustion	2.3 (1.2)	79 (39.1)	40 (32.5)	39 (49.4)	2.3-30.8
2. Having trouble sleeping	2.2 (1.2)	73 (36.1)	39 (31.7)	34 (43.0)	-2.9-25.4
3. Taking care of my health	2.9 (1.1)	134 (66.3)	75 (61.0)	59 (74.7)	-0.4-26.5
Financial problems and work or employment problems					
4. Balancing caregiving and demands associated with my job	2.3 (1.2)	86 (42.6)	47 (38.2)	39 (49.4)	-3.5-25.4

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5. Having to take days off from work due to caregiving responsibilities	2.0 (1.1)	64 (31.7)	35 (28.5)	29 (36.7)	-5.4-22.2
6. Managing the cost of drugs and medical care	2.5 (1.1)	107 (53.0)	62 (50.4)	45 (57.0)	-8.2-20.8
7. Managing loss of income due to my care recipient's inability to work	2.1 (1.1)	81 (40.1)	43 (35.0)	38 (48.1)	-1.4-27.3

Role strain

8. Balancing caregiving and other family responsibilities	2.6 (1.2)	109 (54.0)	62 (50.4)	47 (59.5)	-5.7-23.2
9. Managing last minute changes due to the unpredictability of the disease	2.5 (1.1)	95 (47.0)	54 (43.9)	41 (51.9)	-6.7-22.3
10. Having to do all of the winter chores alone due to my care recipient's sensitivity to cold temperatures	2.5 (1.1)	103 (51.0)	64 (52.0)	39 (49.4)	-12.0-17.2
11. Having to handle all of the household chores on my own	2.2 (1.2)	74 (36.6)	41 (33.3)	33 (41.8)	-5.7-22.6

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12. Being unable to help address my care recipient's pain or discomfort	3.1 (0.8)	170 (84.2)	100 (81.3)	70 (88.6)	-4.2-17.4
13. Finding time for myself	2.5 (1.2)	103 (51.0)	51 (41.5)	52 (65.8)	9.5-37.7
14. Having to learn new skills and abilities because my care recipient can no longer do certain tasks	2.5 (1.2)	100 (49.5)	60 (48.8)	40 (50.6)	-12.8-16.4
15. Having to make difficult medical decisions	2.7 (1.1)	130 (64.4)	76 (61.8)	54 (68.4)	-7.8-20.0
Information, resources, and support					
16. Not having information about how to be a good caregiver	2.6 (1.2)	101 (50.0)	56 (45.5)	45 (57.0)	-3.4-25.5
17. Not being able to find any answers as to why my care recipient got scleroderma	2.3 (1.1)	85 (42.1)	51 (41.5)	34 (43.0)	-12.7-16.1
18. Not having access to a caregiver support group	2.1 (1.2)	69 (34.2)	30 (24.4)	39 (49.4)	10.7-38.4
19. Not knowing other people who understand what I'm going through	2.2 (1.1)	75 (37.1)	37 (30.1)	38 (48.1)	3.6-31.9

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20. Navigating healthcare issues while travelling	2.9 (1.1)	139 (68.8)	82 (66.7)	57 (72.2)	-8.5-18.5
21. Planning trips and excursions while managing limitations, such as needing wheelchair access or other considerations	2.6 (1.2)	114 (56.4)	70 (56.9)	44 (55.7)	-13.2-15.8
22. Having difficulty finding reliable and accurate information about scleroderma	2.6 (1.1)	102 (50.5)	59 (48.0)	43 (54.4)	-8.3-20.8
23. Having difficulty understanding important information about scleroderma and its treatment	2.6 (1.2)	105 (52.0)	62 (50.4)	43 (54.4)	-10.7-18.4
24. Having difficulty helping my care recipient gain access to knowledgeable health providers	2.5 (1.2)	106 (52.5)	61 (49.6)	45 (57.0)	-7.4-21.6
25. Navigating the medical system	2.8 (1.1)	134 (66.3)	79 (64.2)	55 (69.6)	-8.8-18.7
26. Interacting with medical, insurance, and social service agencies to address the needs of my care recipient	2.7 (1.1)	126 (62.4)	74 (60.2)	52 (65.8)	-8.8-19.3

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27. Interacting with health professionals who are not knowledgeable about scleroderma	2.8 (1.0)	135 (66.8)	76 (61.8)	59 (74.7)	-1.2-25.7
28. Managing rushed, inconsiderate, or insensitive behavior from health professionals	2.6 (1.1)	113 (55.9)	59 (48.0)	54 (68.4)	5.7-33.7
29. Trying to find useful devices to help my care recipient with activities of daily living	2.6 (1.1)	114 (56.4)	65 (52.8)	49 (62.0)	-5.5-23.1
30. Finding assistance for things that my care recipient used to do	2.6 (1.2)	104 (51.5)	51 (41.5)	53 (67.1)	10.8-38.8

Fear, anxiety, and uncertainty

31. Being fearful that I will be left alone	2.5 (1.2)	108 (53.5)	65 (52.8)	43 (54.4)	-13.1-16.0
32. Constantly worrying about my care recipient's limitations	2.6 (1.1)	102 (50.5)	58 (47.2)	44 (55.7)	-6.2-22.8
33. Feeling uncertain about the progression of my care recipient's scleroderma	3.0 (0.9)	145 (71.8)	86 (69.9)	59 (74.7)	-8.9-17.4

General emotional difficulties

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34. Feeling helpless	2.8 (1.1)	128 (63.4)	71 (57.7)	57 (72.2)	0.1-27.5
35. Feeling hopeless	2.5 (1.2)	107 (53.0)	56 (45.5)	51 (64.6)	4.2-32.6
36. Managing my negative emotions towards my care recipient	2.6 (1.1)	118 (58.4)	68 (55.3)	50 (63.3)	-6.6-21.9
37. Managing my stress and relaxing	2.9 (1.0)	138 (68.3)	77 (62.6)	61 (77.2)	0.7-27.1
38. Managing my negative emotions	2.6 (1.1)	111 (55.0)	64 (52.0)	47 (59.5)	-7.3-21.6
39. Guilt about leaving my care recipient alone	2.6 (1.1)	103 (51.0)	58 (47.2)	45 (57.0)	-5.0-24.0
40. Feeling ashamed to think about my own well-being or needs	2.1 (1.1)	63 (31.2)	26 (21.1)	37 (46.8)	11.7-39.0
Emotional difficulties of the care recipient					
41. Understanding the emotional needs of my care recipient	3.2 (0.8)	169 (83.7)	100 (81.3)	69 (87.3)	-5.6-16.3
42. Knowing what to do about my care recipient's guilt	2.9 (1.0)	134 (66.3)	77 (62.6)	57 (72.2)	-4.6-22.6
43. Providing emotional support to my care recipient on challenging days	3.2 (0.7)	185 (91.6)	110 (89.4)	75 (94.9)	-3.8-13.4

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44. Managing resentment from my care recipient towards me	2.3 (1.2)	82 (40.6)	45 (36.6)	37 (46.8)	-4.3-24.5
45. Managing my care recipient's anger about having scleroderma	2.6 (1.1)	118 (58.4)	71 (57.7)	47 (59.5)	-12.8-15.9
46. Managing my care recipient's feelings of depression	2.9 (1.0)	144 (71.3)	86 (69.9)	58 (73.4)	-10.2-16.3
47. Managing my care recipient's thoughts of ending her or his life	2.3 (1.1)	102 (50.5)	57 (46.3)	45 (57.0)	-4.2-24.8
48. Managing the disappointment or frustration of my care recipient when she or he cannot take part in activities	3.0 (1.0)	146 (72.3)	86 (69.9)	60 (75.9)	-7.6-18.5
Changes in social interactions with others					
49. Noticing others' lack of knowledge and awareness about scleroderma	2.9 (1.1)	138 (68.3)	72 (58.5)	66 (83.5)	11.4-36.6
50. Managing social limitations, such as missing events or having to leave events early	2.4 (1.2)	83 (41.1)	45 (36.6)	38 (48.1)	-3.0-25.7

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51. Enjoying myself when spending time with friends without my care recipient	2.5 (1.2)	91 (45.0)	46 (37.4)	45 (57.0)	4.7-33.4
Changes in relationship dynamics with care recipient					
52. Understanding when my help isn't wanted or needed	2.8 (1.1)	117 (57.9)	67 (54.5)	50 (63.3)	-5.9-22.7
53. Helping my care recipient set reasonable limits on activities that have become difficult due to scleroderma	3.1 (0.9)	165 (81.7)	98 (79.7)	67 (84.8)	-7.0-15.9
54. Providing needed help when my care recipient doesn't want it or resists it	2.7 (1.2)	111 (55.0)	63 (51.2)	48 (60.8)	-5.2-23.5
55. Being patient with my care recipient	2.9 (0.9)	152 (75.2)	93 (75.6)	59 (74.7)	-11.5-14.2
56. Finding the balance between interfering and providing care	2.9 (1.1)	127 (62.9)	77 (62.6)	50 (63.3)	-13.7-14.6
57. Helping my care recipient feel useful despite her or his physical limitations	3.0 (0.9)	154 (76.2)	92 (74.8)	62 (78.5)	-9.4-15.7

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58. Feeling a sense of loss because of activities we can no longer do together	2.6 (1.1)	107 (53.0)	55 (44.7)	52 (65.8)	6.3-34.5
59. Accommodating my care recipient's diet restrictions when we eat out	2.4 (1.2)	88 (43.6)	49 (39.8)	39 (49.4)	-5.1-23.8
60. Discussing emotions or worries concerning scleroderma with my care recipient	2.9 (1.0)	127 (62.9)	73 (59.3)	54 (68.4)	-5.4-22.4
61. Dealing with loss of physical intimacy with my care recipient*	2.7 (1.1)	67 (57.3)	53 (56.4)	14 (60.9)	-19.8-26.0

Note: CI = confidence interval; *item was optional.

Table 3. Frequencies of support service items rated as likely and very likely to use among 202 informal caregivers.

	Total sample		Male versus Female Caregivers		
	Item Mean (SD)	Likely to Use or Very Likely to Use n (%)	Male Caregivers (N=123): Likely to Use or Very Likely to Use n (%)	Women (N = 79): Likely to Use or Very Likely to Use n (%)	95% CI Lower Limit – Upper Limit
1. Caregiver internet-based chat group, forum, or social network site without professional moderator	1.8 (1.0)	48 (23.8)	25 (20.3)	23 (29.1)	-3.8-22.0
2. Caregiver internet-based chat group, forum, or social network moderated by a	2.3 (1.1)	86 (42.6)	42 (34.1)	44 (55.7)	6.8-35.3

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	knowledgeable healthcare provider					
3.	Caregiver-led breakout groups at patient conferences	2.0 (1.0)	59 (29.2)	28 (22.8)	31 (39.2)	2.8-30.0
4.	Professionally led breakout groups at patient conferences	2.2 (1.1)	74 (36.6)	36 (29.3)	38 (48.1)	4.4-32.6
5.	Conference caregiver educational sessions and workshops provided by a knowledgeable healthcare provider	2.4 (1.1)	94 (46.5)	54 (43.9)	40 (50.6)	-7.9-21.1
6.	Internet-based psychological and emotional self-help tools	2.1 (1.1)	74 (36.6)	37 (30.1)	37 (46.8)	2.4-30.6
7.	One-to-one peer support (e.g., the ability to call	1.9 (1.0)	54 (26.7)	27 (22.0)	27 (34.2)	-1.0-25.7

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another caregiver on the
phone)

8. Professionally led in-person caregiver support group	2.1 (1.1)	73 (36.1)	41 (33.3)	32 (40.5)	-6.9-21.4
9. Caregiver-led in-person caregiver support group	1.9 (1.0)	53 (26.2)	30 (24.4)	23 (29.1)	-8.1-18.2
10. Professionally led telephone-base support group for caregivers	1.8 (1.0)	41 (20.3)	20 (16.3)	21 (26.6)	-1.7-23.1
11. Caregiver-led telephone-based support group for caregivers	1.6 (0.9)	30 (14.9)	16 (13.0)	14 (17.7)	-5.8-16.5

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12. Professionally led internet-based, live interaction (teleconference, Skype) caregiver support group	1.9 (1.0)	55 (27.2)	25 (20.3)	30 (38.0)	4.2-31.0
13. Caregiver-led internet-based, live interaction (teleconference, Skype) caregiver support group	1.7 (0.9)	43 (21.3)	19 (15.4)	24 (30.4)	2.5-27.8
14. Caregiver newsletter	2.7 (1.0)	118 (58.4)	68 (55.3)	50 (63.3)	-6.6-21.9
15. Retreat for caregivers	1.8 (1.0)	48 (23.8)	24 (19.5)	24 (30.4)	-1.8-24.0
16. Online educational sessions for caregivers to help understand scleroderma and its impact on families	2.4 (1.1)	98 (48.5)	52 (42.3)	46 (58.2)	1.1-29.9

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17. Information package/pamphlet about scleroderma for caregivers of newly diagnosed patients	2.5 (1.1)	109 (54.0)	61 (49.6)	48 (60.8)	-3.6-25.1
18. Information about scleroderma on an online reputable website for caregivers of newly diagnosed patients	2.7 (1.1)	111 (55.0)	60 (48.8)	51 (64.6)	1.0-29.4

Note: CI = confidence interval.