


# The Scleroderma Research Topics Survey for patients and health care professionals: A Scleroderma Patient-centered Intervention Network Project

Journal of Scleroderma and Related Disorders  
2019, Vol. 4(3) 165–172  
© The Author(s) 2019  
Article reuse guidelines:  
sagepub.com/journals-permissions  
DOI: 10.1177/2397198319842969  
journals.sagepub.com/home/jso  


Lydia Tao<sup>1</sup>, Claire Fedoruk<sup>1</sup>, Kimberly A Turner<sup>1,2</sup>, Julie Cumin<sup>1</sup>, Marie-Eve Carrier<sup>1</sup>, Andrea Carboni-Jiménez<sup>1</sup>, Mia R Pépin<sup>1</sup>, Linda Kwakkenbos<sup>3</sup> and Brett D Thombs<sup>1,2,4,5,6,7</sup> ,  
SPIN Patient Advisory Board\*

## Abstract

Involving patients and health care professionals in research has been shown to be a useful strategy to generate program development ideas and establish research priorities. Online surveys can be used to identify patient research priorities. The Scleroderma Patient-centered Intervention Network is an international collaboration dedicated to researching problems important to people with scleroderma and developing, testing, and disseminating educational, self-management, rehabilitation, and psychological tools to support coping within the disease. The objectives of the Scleroderma Patient-centered Intervention Network Scleroderma Research Topics Survey were to (1) obtain input on online programs that Scleroderma Patient-centered Intervention Network could develop, (2) identify research topics important to members of the scleroderma community that could be addressed via the Scleroderma Patient-centered Intervention Network Cohort, and (3) identify potentially understudied groups. Eligible survey respondents included individuals with scleroderma and their caregivers, health care professionals, and patient organization representatives. Responses were evaluated separately for online programs, possible research questions, and understudied groups. In each category, thematic analysis was used to group responses into themes. A total of 125 participants (100 patients, 24 health care professionals, and 1 caregiver) completed the survey, and responses from patients and health care professionals were reported. For online programs, frequent recommendations included Interactive Platforms to facilitate communication between patients, as well as interventions targeting physical activity, and emotions and stress. Common responses for possible research questions were related to calcinosis, accessing health care, and quality of life. The most common suggestions for understudied groups were young patients, minority patients, and patients with calcinosis. Research topic suggestions provided in the survey will inform Scleroderma Patient-centered Intervention Network priorities and can be considered by other researchers.

## Keywords

Scleroderma, systemic sclerosis, survey, patients, health care professionals

Date received: 7 December 2018; accepted: 19 March 2019

<sup>1</sup>Lady Davis Institute, Jewish General Hospital, Montreal, QC, Canada

<sup>2</sup>Department of Psychiatry, McGill University, Montreal, QC, Canada

<sup>3</sup>Behavioural Science Institute, Department of Clinical Psychology, Radboud University, Nijmegen, The Netherlands

<sup>4</sup>Department of Epidemiology, Biostatistics and Occupational Health, McGill University, Montreal, QC, Canada

<sup>5</sup>Department of Medicine, McGill University, Montreal, QC, Canada

<sup>6</sup>Department of Psychology, McGill University, Montreal, QC, Canada

<sup>7</sup>Department of Educational and Counselling Psychology, McGill University, Montreal, QC, Canada

\*The complete list of members of the SPIN Patient Advisory Board is available in the “Acknowledgements” section.

## Corresponding author:

Brett D Thombs, Lady Davis Institute, Jewish General Hospital, 4333 Côte-Sainte-Catherine Road, Montréal, QC H3T 1E4, Canada.  
Email: brett.thombs@mcgill.ca

## Introduction

Systemic sclerosis (SSc; scleroderma) is a rare, chronic autoimmune disease characterized by abnormal fibrotic processes that affect the skin and internal organs, resulting in problems that include limitations in hand function and physical mobility, pain, itch, fatigue, psychological distress, and body image concerns.<sup>1-3</sup> SSc patients face the same challenges as people living with common chronic diseases and also experience additional challenges related to living with a rare disease, such as difficulty accessing specialized medical care, limited treatment options, and few or no evidence-based, disease-specific support resources.<sup>3,4</sup>

Patient engagement is increasingly understood to be a core component of high-quality, patient-centered health care, including research.<sup>5</sup> The potential benefits of involving patients in the research process include the development of more appropriate research questions, methodologies, and outcome measures; greater study enrolment and lower attrition rates; improved knowledge translation (e.g. increased comprehensibility, cultural sensitivity, and credibility with patient communities); and more ethically accountable research.<sup>6-8</sup>

The Scleroderma Patient-centered Intervention Network (SPIN) is an international collaboration of SSc patients, patient organizations, researchers, and health care professionals who work together to develop educational, self-management, psychological, and rehabilitative interventions to improve quality of life for people with SSc.<sup>3,9</sup> Since SPIN's inception, patients have actively contributed to research topic prioritization, intervention development, project oversight, and dissemination of research results. They have done this by providing information via SPIN's ongoing online cohort of >1800 SSc patients;<sup>9,10</sup> by participating in >20 patient focus groups conducted by SPIN investigators; by taking active roles in SPIN's Patient Advisory Board, which consists of 10 patients from 5 countries; and by participating in project-specific patient advisory teams.

Online surveys have been used to identify patient research priorities in a range of medical conditions, including depression,<sup>11</sup> dementia,<sup>12</sup> prostate cancer,<sup>13</sup> asthma,<sup>14</sup> and kidney dialysis,<sup>15</sup> for example. Online surveys offer several advantages that build upon other strategies that SPIN has used, including the ability to collect data from large samples with a global reach, convenience for participants, time efficiency, low administrative costs, and the ability to incorporate diverse response formats within a single survey.<sup>16</sup>

To obtain ideas for areas of research that have not been addressed but may be priorities for SSc patients and health care professionals, SPIN conducted an online survey that asked participants to suggest ideas for future online programs that could be developed by SPIN, to recommend research topics that could be studied via the SPIN Cohort, and to identify understudied SSc groups. The ideas submitted will inform

SPIN priorities and may also be useful for other researchers. The objective of the present report was to describe the results of this survey.

## Methods

### *Participants and procedure*

Eligible participants were individuals living with SSc and their caregivers, health care professionals, and patient organization representatives. Participants were recruited through news items on the SPIN website ([www.spin-sclero.com](http://www.spin-sclero.com)), online announcements to participants in the SPIN Cohort, and an email sent to 158 health care professionals from the over 40 SPIN recruiting sites in Canada, the United States, the United Kingdom, France, Spain, Mexico, and Australia.

The survey was accessible via the online survey tool *Qualtrics* from April to August 2018 (see Appendix 1). The survey was initially developed in English and then translated into French and Spanish by SPIN team members. After clicking on the survey link and choosing their preferred language, participants were prompted to click on an arrow at the bottom of the page to proceed to the survey. The Research Ethics Committee of the Jewish General Hospital determined that ethical approval for this project was not required because the survey was conducted only to gather input on priorities for future research topics and did not collect any information about survey responders beyond whether they were patients, caregivers, or health care providers. Consequently, participants were not required to provide informed consent because the survey was completed anonymously and no personal or identifying information was collected.

### *The survey*

The survey questions were designed based on similar surveys that have been used in rheumatic and other diseases<sup>17,18</sup> and input from team members, including members of the SPIN Patient Advisory Board. The survey first asked participants to categorize themselves as a (1) person living with scleroderma, (2) caregiver of someone living with scleroderma, (3) health care professional, (4) patient organization representative who does not have scleroderma, or (5) other. If participants identified themselves as either a "health care professional" or "other," they were also asked to specify in more detail in a free-text field. Participants were then directed to three questions with free-text field responses. These questions asked participants for (1) suggestions for online programs ("SPIN is dedicated to developing, testing, and disseminating programs to support people living with scleroderma. The types of programs that SPIN develops aim to increase quality of life by teaching coping and disease-management skills. Do you have any suggestions for online programs

that SPIN could develop and provide, which you feel could make a difference in the lives of people living with scleroderma?”), (2) possible research questions (“SPIN also investigates and answers research questions that are important to people living with scleroderma, using questionnaires via the Internet. Please note that we rely on patient questionnaires, thus the research questions that we can best address are related to the experiences of patients. We are not able to evaluate medical treatments. Do you have any specific research questions that you would like SPIN to address?”), and (3) understudied groups (“In your opinion, is there a specific group within the scleroderma community that is especially understudied?”). Participants were not required to respond to all of the questions.

### Data management and analysis

After the end of the survey period, all responses were exported from *Qualtrics* into *Excel*, and all data analysis was performed using *Excel*. French and Spanish responses were translated into English by a SPIN team member and validated by another team member. Vague and blank responses were excluded from further coding and analysis by one team member after verification and agreement by a second team member. If a response to one survey question (e.g. online program suggestions) better addressed a different question (e.g. research questions), it was moved to the appropriate question category.

For each survey question, thematic analysis was used to interpret survey responses. One coder first reviewed all the responses and identified possible response themes for each question (online program suggestions, research questions, understudied groups). Next, a second coder reviewed the identified themes, and the two coders discussed and revised any conflicts. Then, the second coder grouped responses into themes for each question. All identified themes and responses grouped into each theme were reviewed by other members of the research team, and agreement on groupings and any revisions were based on consensus. Suggestions that SPIN could not feasibly address were coded as “Beyond SPIN’s Scope.” The total number of responses for each theme were tabulated and reported.

## Results

### Sample characteristics

A total of 125 eligible participants (100 patients; 24 health care professionals; 1 caregiver) completed the survey. Of the health care professionals, 22 listed their profession (8 physicians, 5 rheumatologists, 3 physician-researchers, 5 research coordinators, and 1 psychologist). Since only 1 caregiver responded, we included responses from only patients and health care professionals. Among the 124

included participants, 69 responded in English (56%), 49 in French (39%), and 6 in Spanish (5%).

### Responses to survey

**Suggestions for online programs.** There were 136 suggestions for online programs, including 107 from patients (79%) and 29 from health care professionals (21%). See Table 1 for program suggestion themes. There were 13 suggestions related to the theme of interactive platforms and the facilitation of communication between patients, but none of these identified an intervention target. The other 123 suggestions recommended intervention targets, with interventions related to physical activity (N = 16) as the most commonly suggested target from patients and interventions to help manage emotions and stress (N = 5) and interventions for skin and wound care (N = 5) as the most common suggestions from health care professionals.

**Suggestions for possible research questions.** As shown in Table 2, there were 65 suggestions for possible research questions, including 44 from patients (68%) and 21 from health care professionals (32%), with the most common topics relating to calcinosis (N = 8), quality of life (N = 7), and accessing health care (N = 7).

**Suggestions for understudied groups.** There were 40 suggestions for understudied groups, including 18 from patients (45%) and 22 from health care professionals (55%; see Table 3). Several patients (N = 4) suggested the need for more research on young patients. The most common suggestions by health care professionals were studies on minority patients (N = 4) and patients with calcinosis (N = 4).

## Discussion

Involving patients and health care professionals in research has been shown to be a useful strategy for generating program development ideas and research priorities.<sup>11–16,19,20</sup> The results of this study indicate that SSc patients and health care professionals have different but overlapping ideas for programs that would be useful to develop and for research questions they would like to see answered. Some of the most common suggestions for online programs are already being developed or tested by SPIN (e.g. hand exercises, emotions and stress, and physical activity). However, other suggestions for patient programs that address pain management, nutrition, mouth exercises, and skin and wound care, for example, provide ideas for future programs that could be developed by SPIN or by other SSc researchers.

Among participants’ suggestions for research questions, several common suggestions (e.g. quality of life, psychological, and caregivers) have been the subjects of

**Table 1.** Patients' and health care professionals' suggestions for online programs.

Themes	Patients		Health care professionals		Total
	No.	Example suggestions	No.	Example suggestions	
<i>Intervention delivery platforms</i>					
Interactive platforms	13	Online forums for patients to share experiences, and chat and receive support from health care professionals; question and answer forum; online support programs with live interactions; workshops for newly diagnosed patients	0	–	13
<i>Intervention targets</i>					
Physical activity	16	Programs helping patients stay active; exercise programs (e.g. yoga, Tai chi, biking, and breathing exercises) targeting flexibility, fatigue, and pain; exercises for mobility and body flow	2	Exercise programs (modified exercises); physical therapy (physiotherapist tutorials)	18
Emotions and stress	11	Programs focusing on mental health; programs on relaxation, positive imagery, meditation, anxiety coping, and stress management that help keep a positive attitude and deal with emotional issues and stress	5	Cognitive behavior therapy programs for depression; positive psychology programs for stress, relaxation, anxiety and depression management, grief, and self-esteem enhancing programs	16
Nutrition	13	Programs offering nutritional tips and eating advice; cooking and dietetics program	2	Programs with diet and food choices to help patients eat properly	15
Skin and wound care	9	Programs providing advice on calcinosis management, care for face and hands, digital ulcers, finger protection, and itch	5	Program offering calcinosis management, advice on itch, digital ulcer treatment and Raynaud's precautions, and protecting hands and feet	14
Hand exercises	9	Programs targeting hand function; rehabilitation exercises for both fingers and hands	4	Hand therapy programs; programs providing advice on how to protect hands from the cold	13
Pain management	10	Relaxation programs to cope with pain and discomfort; programs focusing on exercises and activity pacing for pain management; programs offering tools to manage the impact of pain	0	–	10
Oral health and mouth exercises	7	Mouth and/or face exercise programs (e.g. swallowing exercises); oral health programs (e.g. tooth management); programs offering advice on reducing facial or mouth changes, oral hygiene, and finding a dentist	1	Mouth therapy exercise programs	8
Gastrointestinal involvement	1	Program for managing gastrointestinal involvement	3	Program on gastrointestinal (anti-reflux) precautions; diet/nutrition program for severe gastroesophageal reflux disease and gastroparesis	4
Fatigue management	3	Program offering fatigue management tools and advices on handling fatigue/exhaustion	0	–	3
Body image and social interactions	0	–	2	Programs on how to manage Raynaud's in a social setting; body image program	2
Sleep	1	Program on managing sleeping problems	1	Cognitive behavior therapy program for insomnia	2
Communication with health care providers	2	Program offering strategies on communicating with clinicians who are not familiar with scleroderma	0	–	2
Caregivers	1	Educational and support program for caregivers, especially professional caregivers working in long term care or retirement facilities	0	–	1
Intimacy and sexuality	1	Program providing information on how to maintain intimacy with a spouse	0	–	1

(Continued)

**Table 1.** (Continued)

Themes	Patients		Health care professionals		Total
	No.	Example suggestions	No.	Example suggestions	
Others	7	Program offering advice on useful resources, treatments, and medication; management for eye problems; anal-rectal retention exercise program; social protection program; program helping young individuals with scleroderma find careers	1	Patient education program on current research and treatments	8
Beyond SPIN's scope	5	Programs providing advice on alternative medicine and natural products that help improve quality of life	1	Oral hygiene program providing interventions by involving health professionals (e.g. dentists and oral surgeons)	6

SPIN: Scleroderma Patient-centered Intervention Network.

**Table 2.** Patients' and health care professionals' suggestions for research questions.

Themes	Patients		Health care professionals		Total
	No.	Example suggestions	No.	Example suggestions	
Calcinosis	7	Research on calcinosis/calcification, and reducing or eliminating calcinosis caused by scleroderma; add questions on calcium deposits (calcification) to SPIN questionnaires	1	Effect of calcinosis on quality of life	8
Quality of life	3	Impact of mouth involvement on daily life; impact of pain on daily life; tools to help manage its impact	4	Impact of environmental factors on patients' lives; prevalence of oral complications and their impacts on quality of life; effect of pruritus on quality of life; impact of diet on scleroderma symptoms and quality of life	7
Psychological	4	Relationship between emotional distress and age and types of scleroderma; impact of stress on patients with scleroderma and triggering symptoms (e.g. gastrointestinal symptoms); emotional impact of living with scleroderma on young adults	3	Impact of positive vs negative thinking on the disease; determine the scleroderma type (i.e. diffuse or limited) with the highest risk of developing anxiety and/or depression; determine what aggravates the harmful psychological effects of scleroderma (i.e. the presence of physical changes or changes in interpersonal relationships)	7
Accessing health care	2	Research on the health service utilization patterns for people with scleroderma and the consistency across Canada; clinicians' knowledge of scleroderma	5	Research how health professionals should give diagnoses; patients' access to specific care (including dental care); family doctor relationship with scleroderma patients; ways to improve/regulate the physician-patient relationship; ways to choose a qualified mental health professional	7
Gastrointestinal involvement	6	Research on heartburn, bowel issues, dental issues, and telangiectasia in scleroderma; relationship between scleroderma and gastrointestinal problems; add questions and choices of responses on esophagus problems and ulcers to SPIN questionnaires	0	–	6
Nutrition and environment	4	Importance of food and environment for reducing pain; loss of taste; impact of nutrition and environment on chronic diseases; add diet-related questions to SPIN assessments	1	Impact of ketogenic, gluten-free, paleo diets on scleroderma symptoms; types of food that will make symptoms worse	5

(Continued)

**Table 2.** (Continued)

Themes	Patients		Health care professionals		Total
	No.	Example suggestions	No.	Example suggestions	
Physical activity	4	Research on practical and ergonomic exercises to keep flexibility and strength in muscles; effects of exercises on improving scleroderma symptoms; add exercise-related questions to SPIN assessments; the benefits of exercise and relaxation techniques	0	–	4
Body image and social interactions	1	Changes to physical appearance of the face: thickening of the skin	0	–	1
Caregivers	0	–	1	Creating surveys to understand caregiver concerns and how they think they can be helped	1
Sleep	1	Research on sleep deprivation related to scleroderma	0	–	1
Intimacy and sexuality	0	–	1	Consequences of scleroderma for sexual health	1
Others	4	Research on Sjogren's syndrome and arthritis; research on treatments for joint pain; add choices of responses on supporting materials; research on mobility, dermatology, dryness of the skin	5	Professional consequences of having scleroderma; factors that impair work adherence; common coping patterns among patients with systemic sclerosis, if these patterns change over time, and if they change depending on severity of illness; the role of social support in adjustment/adaptation to illness; research on intercultural perceptions of disability or medication preferences	9
Beyond SPIN's scope	8	Research on treatments and medication issues, alternative medicine and supplements, the origin of scleroderma, neuropathy symptoms and genetic factors in scleroderma, and how cancer treatments can affect the progression of scleroderma (positively or negatively); research on food and dietary advice that help scleroderma patients prolong their lives; research on relationship between leaky gut/allergies/autoimmune responses and onset of scleroderma	0	–	8

SPIN: Scleroderma Patient-centered Intervention Network.

SPIN Cohort studies and research by other SSc research teams. There are few studies on intimacy and sexuality, particularly among male patients. Research questions related to communication with health care providers are currently underaddressed. However, the World Scleroderma Foundation (WSF) and l'Association des Sclérodermiques de France (ASF) have recently funded SPIN researchers (PI Kwakkenbos) to conduct work in this area.

Within understudied groups reported by patients and health care professionals, patients with limited SSc, as well as patients with gastrointestinal involvement, have been frequently studied, whereas there have been few studies related to care for end-of-life patients, male

patients, and minority patients with SSc. Research is needed to identify and address the needs of members of these groups.

There are limitations that need to be considered when interpreting the results of this study. First, participants were recruited from the SPIN Cohort and by emails to health care professionals from SPIN research sites in seven countries. It is possible that responses from patients and health care professionals who are already involved in SPIN research may not be representative. Second, the sample size of this study was small; only 124 participants responded to at least one of the three open-response questions. Third, patients in the SPIN Cohort have confirmed diagnoses of SSc, but we also recruited participants via

**Table 3.** Patients' and health care professionals' suggestions for understudied groups.

Themes	Patients		Health care professionals		Total
	No.	Example suggestions	No.	Example suggestions	
Patients with calcinosis	2	Patients with calcinosis	4	Patients with calcinosis	6
Minority patients	1	Minorities and disadvantaged groups	4	Black patients; Asian patients; Latino patients; patients from Maghreb countries	5
Young patients	4	Young patients; young children; children with morphea	1	Adult patients who were diagnosed as children	5
Patients with gastrointestinal involvement	1	Scleroderma patients with bowel problems	3	Patients with gastrointestinal diseases; patients with fecal incontinence	4
Patients with limited systemic sclerosis	3	Patients with limited scleroderma	1	Patients with mild, limited systemic sclerosis but after years of mild disease, develop pulmonary arterial hypertension	4
Male patients	1	Male patients with excellent health	2	Men with scleroderma	3
End-of-life patients	1	End of life/late stage (palliative care and assisted dying)	2	End of life/late stage	3
Senior patients	1	Senior patients	1	Senior patients	2
Patients with limited access to health care	2	Rural patients who are far from health services; patients with limited access to health care (especially physiotherapists)	0	–	2
Others	2	CREST (calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, and telangiectasia) patients; patients with comorbid disorders (e.g. vasculitis and Sjogren's syndrome)	4	Patients with myopathy resulting in weakness and muscular atrophy; newly diagnosed patients; patients with long-standing disease (10+ years)	6

other methods, and it was not possible to verify the diagnoses of participants. Finally, we did not collect information on participant demographics (e.g. age, sex, and country of residence) or medical information (e.g. type of SSC and other conditions).

In sum, patients and health care professionals provided a variety of suggestions for online programs, research questions, and understudied groups. SPIN will use these suggestions to prioritize future research topics, in conjunction with input from participants in the SPIN Cohort, ongoing focus groups, and input from the SPIN Patient Advisory Board. Research topics suggested by survey participants should also be considered by other researchers and research groups.

### Acknowledgements

SPIN Patient Advisory Board: Maureen Sauvé (co-Chair), Scleroderma Society of Canada, Hamilton, ON, Canada; Karen Gottesman (co-Chair), Scleroderma Foundation, Los Angeles, CA, USA; Catherine Fortuné, Scleroderma Society of Ontario, Hamilton, ON, Canada; Dominique Godard, Association des Sclérodermiques de France [French Scleroderma Association], Sorel-Moussel, France; Geneviève Guillot, Sclérodermie Québec

[Scleroderma Quebec], Longueuil, QC, Canada; Catarina Leite, Associação Portuguesa de Doentes com Esclerodermia [Portuguese Association of Patients with Scleroderma], Senhora da Hora, Portugal; Karen Nielsen, Scleroderma Society of Ontario, Hamilton, ON, Canada; Alexandra Portales, Asociación Española de Esclerodermia [Spanish Scleroderma Association], Madrid, Spain; Joep Welling, Nationale Vereniging voor LUPUS, APS, Sclerodermie en MCTD [Dutch Patient Organization for Systemic Autoimmune Diseases], Utrecht, the Netherlands.

### Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

### Funding

SPIN has been funded by grants from the Canadian Institutes of Health Research (TR3-119192, PJT-148504, and PJT-149073) and the Arthritis Society, as well as contributions from the Jewish General Hospital Foundation and the Lady Davis Institute for Medical Research, Jewish General Hospital, Montreal, QC, Canada; McGill University, Montreal, QC, Canada; Scleroderma Canada; the Scleroderma Society of Ontario; Sclérodermie

Québec; the Scleroderma Society of Nova Scotia; the Scleroderma Society of British Columbia; the Scleroderma Society of Saskatchewan; and the Scleroderma Society of Manitoba. Ms Turner was supported by a Fonds de recherche du Québec–Santé (FRQ-S) masters training award. Dr Thombs was supported by a FRQ-S researcher salary award.

## ORCID iD

Brett D Thombs  <https://orcid.org/0000-0002-5644-8432>

## References

- Seibold J. Scleroderma. In: Harris ED, Budd RC, Firestein GS, et al. (eds) *Kelley's textbook of rheumatology*. 7th ed. Philadelphia, PA: Elsevier, 2005, pp. 1279–1308.
- Mayes M. Systemic sclerosis: clinical features. In: Klippel JH, Stone JH, Craford LJ, et al. (eds) *Primer on the rheumatic diseases*. 13th ed. New York: Springer, 2008, pp. 343–350.
- Kwakkenbos L, Jewett LR, Baron M, 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(8): e003563.
- Kowal-Bielecka O, Landewe R, Avouac J, et al. EULAR recommendations for the treatment of systemic sclerosis: a report from the EULAR Scleroderma Trials and Research group (EUSTAR). *Ann Rheum Dis* 2009; 68(5): 620–628.
- Clancy CM. Patient engagement in health care. *Health Serv Res* 2011; 46: 389–393.
- Brett J, Staniszewska S, Mockford C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. *Health Expect* 2014; 17(5): 637–650.
- Domecq JP, Prutsky G, Elraiyah T, et al. Patient engagement in research: a systematic review. *BMC Health Serv Res* 2014; 14: 89.
- Manafa E, Petermann L, Mason-Lai P, et al. Patient engagement in Canada: a scoping review of the “how” and “what” of patient engagement in health research. *Health Res Policy and Syst* 2018; 16: 5.
- Thombs BD, Jewett LR, Assassi S, et al. New directions for patient-centered care in scleroderma: the scleroderma patient-centered intervention network (SPIN). *Clin Exp Rheumatol* 2012; 30: S23–S29.
- Dougherty DH, Kwakkenbos L, Carrier M-E, et al. The scleroderma patient-centered intervention network (SPIN) cohort: baseline clinical features and comparison to other large scleroderma cohorts. *Rheumatology* 2018; 57: 1623–1631.
- Breault LJ, Rittenbach K, Hartle K, et al. The top research questions asked by people with lived depression experience in Alberta: a survey. *CMAJ Open* 2018; 6(3): E398.
- Kelly S, Lafortune L, Hart N, et al. Dementia priority setting partnership with the James Lind Alliance: using patient and public involvement and the evidence base to inform the research agenda. *Age Ageing* 2015; 44(6): 985–993.
- Lophatananon A, Tyndale-Biscoe S, Malcolm E, et al. The James Lind Alliance approach to priority setting for prostate cancer research: an integrative methodology based on patient and clinician participation. *BJU Int* 2011; 108(7): 1040–1043.
- Elwyn G, Crowe S, Fenton M, et al. Identifying and prioritizing uncertainties: patient and clinician engagement in the identification of research questions. *J Eval Clin Pract* 2010; 16(3): 627–631.
- Manns B, Hemmelgarn B, Lillie E, et al. Setting research priorities for patients on or nearing dialysis. *Clin J Am Soc Nephrol* 2014; 9: 1813–1821.
- Evans JR and Mathur A. The value of online surveys. *Internet Research* 2005; 15: 195–219.
- The Royal Marsden NHS Foundation Trust. Your research suggestions, <https://www.royalmarsden.nhs.uk/our-research/patient-involvement/your-research-suggestions> (accessed 27 November 2018).
- Kjeken I, Ziegler C, Skrolsvik J, et al. How to develop patient-centered research: some perspectives based on surveys among people with rheumatic diseases in Scandinavia. *Phys Ther* 2010; 90(3): 450–460.
- Welsman J, Gibson A, Heaton J, et al. Involving patients and the public in healthcare operational research. *BMJ* 2014; 349: g4903.
- Diffin J, Spence M, Spencer R, et al. Involving healthcare professionals and family carers in setting research priorities for end-of-life care. *Int J Palliat Nurs* 2017; 23(2): 56–59.

## Appendix I

### SPIN Research Topics Survey items

I am a ...

- Person living with scleroderma
- Caregiver of someone living with scleroderma
- Health care professional (please specify)
- Patient organization representative who does not have scleroderma
- Other (please specify)

**Part I: online programs.** The Scleroderma Patient-centered Intervention Network (SPIN) is dedicated to developing, testing, and disseminating programs to support people living with scleroderma. The types of programs that SPIN develops aim to increase quality of life by teaching coping and disease-management skills.

Do you have any suggestions for online programs that SPIN could develop and provide, which you feel could make a difference in the lives of people living with scleroderma?

**Part II: research questions.** SPIN also investigates and answers research questions that are important to people living with scleroderma, using questionnaires sent out via the Internet. Please note that we rely on patient questionnaires, thus the research questions that we can best address are related to the experiences of patients. We are not able to evaluate medical treatments.

Do you have any specific research questions that you would like SPIN to address?

In your opinion, is there a specific group within the scleroderma community that is especially understudied?