Understanding coping strategies among people living with scleroderma: A focus group study

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Abstract

Purpose: Systemic sclerosis or scleroderma is a chronic, rare connective tissue disease with negative physical and psychological implications. Coping strategies used by scleroderma patients have not been studied in-depth. The objective of the present study was to gain a greater understanding of the coping strategies employed by people living with scleroderma.

Method: Three semi-structured focus group discussions were conducted with a total of 22 people with scleroderma. Interviews were recorded, transcribed, and analyzed using content analysis. Coping strategies discussed were analyzed through Lazarus and Folkman’s theoretical model of coping, including: 1) problem-focused, 2) emotion-focused, and 3) meaning-focused coping.

Results: Participants reported using a combination of problem-focused (e.g., professional help; seeking disease-related information), emotion-focused (e.g., social support; adaptive distraction techniques), and meaning-focused coping strategies (e.g., benefit finding; goal reappraisal) to help them cope with and manage their disease. However, many patients reported having difficulty accessing support services.

Conclusions: Scleroderma patients use similar coping strategies as patients with more common diseases, but they may not have access to the same level of support services. Accessible interventions, including self-management programs, aimed at improving problem- and emotion-focused coping are needed. Further, increased access to support groups may provide patients with opportunities to obtain social support and enhance coping.

Keywords: systemic sclerosis; coping strategies; qualitative research; patient perspective
People living with chronic medical conditions face challenges that include learning how to manage their medical care and coping with physical and psychological manifestations of their disease.[1] People with rare diseases face the same challenges as patients with any chronic disease, but also typically encounter additional obstacles,[2, 3] including gaps in knowledge about their disease and how to treat it, insufficient access to effective treatments, and a lack of professionally organized supportive care.[2-5] Social implications of rare diseases include stigmatization, social exclusion, and reduced professional opportunities.[3, 6, 7]

Individuals with medical diseases, including rare diseases, develop coping strategies to manage stress from internal and external demands related to their illness.[8] Lazarus and Folkman’s stress-coping model for people with chronic diseases[9, 10] posits that coping strategies can be grouped into: (a) problem-focused coping, (b) emotion-focused coping, and (c) meaning-focused coping.[11, 12] Problem-focused coping refers to actions taken to modify sources of stress,[10] such as obtaining information about one’s disease or seeking advice from others facing similar challenges.[12] Emotion-focused coping involves strategies that seek to reduce, minimize, or prevent negative emotional responses, such as altering thoughts about a situation, using adaptive distraction and avoidance techniques, or seeking out social support.[13] In meaning-focused coping, patients shift their focus onto the positive side of any experienced adverse or stressful situation. This may involve reappraisal of goals to alter the meaning imbued in events or “benefit finding,” in which patients focus on what they have learned or how they have grown in the face of stressors.[14] In the context of rare diseases, in which the etiologies and rates of disease progression are typically unknown, a patients’ evaluation of and response to an unpredictable environment and future is an important piece in the coping process. As such, we
identified Lazarus and Folkman’s model of coping as a well-fitted theoretical model because it emphasizes the importance of coping with stressors that are unpredictable and ongoing.[9,10]

The unique challenges faced by patients living with rare diseases may hinder effective coping. For example, many patients with rare diseases struggle to obtain a definitive diagnosis and a clear understanding of their disease and its trajectory, and this ambiguity makes it more difficult to develop effective coping strategies.[15-17] Further, people with rare diseases often have limited or no access to support resources and other services designed to meet patient needs, and they may struggle to find healthcare practitioners knowledgeable about their disease.[2, 6, 18, 19] These factors present obstacles to developing a clear understanding of the likely course and consequences of disease symptoms, which is an important part of developing effective coping strategies that foster autonomy and well-being.[15, 18] Social learning theories emphasize the importance of observing how others effectively manage stressors.[20] However, the low prevalence of rare diseases and the degree to which course and symptomology vary across patients make it difficult for many rare disease patients to identify peers with similar experiences and to learn from how others have effectively managed their symptoms in the past.[15, 21]

Systemic sclerosis, or scleroderma (SSc), is a rare, chronic, autoimmune disease characterized by fibrosis and internal organ dysfunction that can affect the lungs, heart, and gastrointestinal tract.[21] A recent study estimated the prevalence of SSc in Canada at 44 cases per 100,000 (74 per 100,000 among women, 13 per 100,000 among men).[21] Peak age of onset is between 40-50 years,[21, 22] and median survival time following diagnosis is 7 years in diffuse SSc, which involves extensive skin involvement and rapid initial disease progression, and 15 years in limited SSc, which involves less extensive skin thickening and a more indolent
Common problems that impact health related quality of life and daily functioning for people living with SSc include emotional distress, fatigue, pain, pruritus, sexual dysfunction, and body image distress due to appearance changes.

Little is known about coping in rare diseases, including SSc, and how coping strategies may reflect unique challenges specific to rare disease populations. Several studies have reported on some coping strategies used by SSc patients, but none have examined coping in the context of a theoretical model. The objective of the present study was to use Lazarus and Folkman’s model of coping with a chronic medical disease to explore coping strategies used by SSc patients to manage the emotional, psychosocial, and physical consequences of their illness.

**Methodology**

*Research design and epistemological approach*

Focus groups were conducted to better understand sources of emotional distress and coping strategies used by patients living with SSc. A previous report described sources of emotional distress for people living with SSc. The present study analyzed data from the same focus groups to uncover and identify both shared and unique coping strategies through a social constructionism approach. This approach posits that shared understandings are co-constructed among members of a given social group through the consideration of multiple viewpoints, interactions, exchanges, and through negotiations of meaning. This method of data collection has been widely used in health research and in SSc research. Focus groups are particularly valuable for research when little is known about a phenomenon of interest and when gaining a shared understanding of the experiences of participants concerning a specific phenomenon is sought, as is the case with coping in SSc.
Participants and procedures

Three focus group interviews were conducted. Two English-language focus groups were conducted in Hamilton, Ontario, Canada and one French focus group was conducted in Montréal, Québec, Canada. Eligible participants were men and women diagnosed with SSc who were fluent in English or French. Recruitment of potential participants for the English focus groups was led by the Scleroderma Society of Ontario. For the French focus group potential participants who were enrolled in an ongoing cohort study were recruited by a research nurse coordinator or research assistant.

Prior to each focus group, details about the focus group process were provided, and all participants completed a questionnaire to record basic demographic and disease information, including age, sex, race/ethnicity, and SSc-related information, such as diagnosis subtype and years since diagnosis. The three focus groups ranged in length from 90 to 130 minutes and were conducted between February and July 2013. All three focus groups were held in private meeting rooms and were moderated by a psychologist with experience in qualitative research and a graduate-level trainee in psychology. The focus groups consisted of a series of open-ended questions to encourage open discussion about challenges faced by people with SSc and the coping strategies used to manage the disease. Results related to challenges faced by patients were reported previously.[25]

The group moderators introduced the topic of coping with chronic illness, asked relevant questions (e.g., what are some of the coping strategies that you use to help manage your SSc? What is the best way to teach people positive coping skills?) and then used probes (e.g., Are these strategies used by everyone? Does anyone else want to discuss other strategies that they have tried?) to encourage a greater sharing of experiences, thoughts, and feelings about certain
ideas, and to gain a clearer understanding of the topic under examination (see Supplementary Material).[31, 32] The two English-language focus groups were video and audio recorded, and the French-language focus group was audio recorded only due to technical difficulties. All focus groups were transcribed verbatim to facilitate further recall and analysis.

This study was approved by the Research Ethics Committee of the Jewish General Hospital in Montréal, Québec, Canada, and all participants provided written informed consent. Following each focus group, participants were reimbursed $20 for travel costs.

**Data analysis**

Content analysis was used to analyze raw data and identify overarching themes.[33, 34] This analytic strategy uses existing theoretical perspectives (in this case, Lazarus and Folkman’s model of coping),[10, 12] to explore participants’ viewpoints and provide further knowledge and understanding of our topic.[33-35] This analytic approach requires the investigator to understand the context and environment in which the data is produced, so that appropriate inferences and justifications can be made in support of the phenomenon under question.

Two members of the research team independently analyzed and coded quotations from the three transcribed focus group interviews using Lazarus and Folkman’s coping model.[10, 12] Transcripts were first read numerous times in order to achieve full immersion in the text data and to obtain a sense of it as a whole. Then, the investigators read the documents word-by-word and identified quotations that appeared to fall within each of the three categories. Next, the investigators compared and discussed their findings in order to achieve consensus upon a final coded text and to resolve any discrepancies. In cases where a consensus was not immediately achieved, a third investigator from the research team was consulted to reach an agreement. Coding was supported by the qualitative research software *Atlas.ti.*[36]
To better support the presentation of the findings, we extracted participant’s quotes directly from the transcripts. As the community of Canadians with SSc is limited in size, to protect the confidentiality of the participants, in the present manuscript we purposefully did not present individual demographic characteristics that could serve to identify the participants whose quotes were highlighted. To differentiate the participants, we assigned ‘W’s’ to patients who identified as women and ‘M’s’ to patients who identified as men. Participants in the first focus group (FG1, English) were assigned numbers 1-6, those in the second focus group (FG2, English) 7-11, and those in the third focus group (FG3, French) 12-22.

**Results**

**Participants’ characteristics**

Insert table 1 about here

A total of 22 individuals with SSc (18 females, 4 males) participated in one of the three focus groups (FG1 (English) = 6 females; FG2 (English) = 4 females, 1 male; FG3 (French) = 8 females, 3 males). Sociodemographic characteristics are presented in table 1. The age of participants ranged from 26 to 77 years (mean = 53 years; standard deviation [SD] = 13). Of the 22 participants, 2 (9%) reported having been diagnosed with limited SSc, 10 (45%) reported having diffuse SSc, 5 (23%) reported having CREST symptoms (i.e., tight, hardened skin; Raynaud’s phenomenon), and 5 (23%) were unsure of their disease subtype. Years since diagnosis ranged from 0 to 28 (mean = 10 years; SD = 8). The majority of participants were White (86%), had completed at least some college education (77%), and were either on disability leave (32%) or retired (23%).

**Participants’ coping strategies**

A summary of coping strategies reported by participants is provided in table 2.
Problem-focused coping

Problem-focused coping strategies described by participants included: a) seeking out professional psychological support; b) finding ways to obtain information on symptoms, treatment options, side effects of medications, and methods of coping; and c) organizational strategies such as making lists.

Many participants described that after being diagnosed with SSc they sought professional mental health guidance and support because they felt overwhelmed by feelings of depression and anxiety or when struggling to accept new roles within their families or work places. For example, participant W3 explained: “...my whole family blamed me for having the disease just because they're super religious and they thought God was punishing me,” and when probed about how she chose to cope with this challenge, she said: “...through a psychiatrist; because I can’t talk to my family about my disease.” Participants agreed that some SSc patients benefit from psychological support, but others described that stigma around mental illness and hesitancy about seeking mental health services could be a barrier to coping in this way. Participant W6 explained: “Don’t be scared, don’t think it’s a stigma, a lot of people don’t want to go see help, professional help, because they think that, ‘Well I’m not crazy [...]’. But it’s okay to think in fact you’re depressed or that you’re sad.” Participant W3 replied: “It just makes good sense. You go to the doctor when you get a cold or you get sick or get diagnosed with something bad, so your mind’s the same way. If something is wrong, you need the help to fix it.”

Obtaining valuable disease-related information (e.g., managing symptom flare-ups and side effects, addressing concerns about the progression of the disease) and learning to navigate complex healthcare systems (e.g., trouble with coordination of physician services) was a source...
of distress for many participants. As a result, participants described a number of ways to obtain
disease-relevant information and to feel a sense of autonomy and control over their care,
including: a) joining provincial or national SSc organizations; b) using the internet as a resource;
c) attending SSc information sessions; and d) learning how to ask questions about the disease.

For example, participant W7 described that “…one of the ways I’ve coped over the years
is that since 1988 I have been involved either at the local, the provincial, or the national level of
scleroderma organizations.” She stated that before joining the organization she “…never dealt
with and didn’t know how to deal with [her] own SSc, so [she dealt with] it through being part of
the organization.” Participant W3 commented on the usefulness of online Facebook groups in
helping patients obtain disease-related information, “…what also is very helpful is on Facebook
there was this group called Help Find a Cure for Scleroderma... there were people from all over
the world doing different treatments and people would post a question and then bang! There’s a
bunch of different responses.” Participant W2 also described how: “Attending conferences like,
just that constant journey of education, has helped me to put a little bit back into control. That
loss of control I had initially, I’ve regained it.” This same participant discussed how patients
must “Educate themselves – for example, I’m very much an information seeker by nature so
when I was told I had this – I never heard of the disease, I went right to the internet and
inundated myself” and also that it is important for patients to acknowledge that “…it’s your body
so you know your body best; don’t be afraid to ask questions.” Similarly, participant W7
suggested: “We have to take charge of our wellbeing and ask questions. That’s one of the biggest
things that we should not be afraid to say to the doctor, ‘Is this the medication you are
suggesting? I want to think about it. Or I want more information about it’.”
Participants also discussed the utility of organizational strategies, such as writing out lists, to help them alleviate everyday stressors. One participant (W3) described:

“For me, it’s writing it down and actually seeing what needs to be done; it’s dividing what I’m worrying about because sometimes I’ll have so many different things that stress me out. And then the things that I know I can’t fix because it’s out of my hands, I just toss it to the wind [...]. But when it comes to smaller things; breaking down the problems because sometimes you have so many things on your mind so you have to figure out what needs to be done first, taking that small problem, tackling it and then taking the next small problem and tackling it. And then it gradually releases the stress and the worry because you’re able to, you know, just get a lot of things off your plate and off your mind.”

Emotion-focused coping

Participants discussed how they often experience intense emotions due to the unpredictable nature and diverse symptomology common to SSc. They referred to three main emotion-focused coping techniques to help them reduce or manage the intensity of these emotions, including: a) seeking out social support; b) using adaptive escape-avoidance coping techniques; and c) engaging in relaxation techniques, meditational exercise, or yoga to address physical and emotional stressors.

Many participants sought social support to help minimize or reduce feelings of frustration, anger, fear about the future, concerns about appearance, and sadness. These emotions were associated with being treated differently, including being pitied or judged because of their illness. They described that their well-being was highly influenced by the presence of others who were supportive and could understand the symptoms and severity of their SSc. The social
support participants described came from a variety of sources including: a) their friends and families; and b) support from other SSc patients through participation in SSc support groups.

W11 explained how discussing the disease with a friend helped her cope: “I have an email friend and I’m not complaining to her, but I’ll just say this is what’s going on. And just writing it down, the exercise of writing it down and she’ll write you something back positive. And I find for me that’s a good coping skill as well.” Participant W2 explained how having a friend accompany her to her medical appointments was helpful: “If you’re nervous during these appointments bring a friend or family member for that second set of ears.”

Seeking emotional support from other SSc patients was another main emotion-focused coping strategy employed by participants as well as an important reason why many patients attended SSc support groups. Participant W6 discussed the supportive benefits that can come from others with SSc: “…if you lean on people that you’re with that can help you and if you don’t have family and if you don’t have friends and if you don’t have significant others, you will always have someone else that is going through the same thing.” Participant W2 explained the utility of support groups as the “…biggest significance of these support groups is you’re seen, you’re seeing the face of the disease.” When describing a member of her support group Participant W2 admitted that “…we’ve got this amazing woman in our group, doctor, and she’s been, had it for 40 years. And she’s I think in her 70’s, she’s spry, she’s energetic, she goes dancing,” so basically “you’re seeing that people are living [even with this disease].”

Another participant (W3) stated: “The thing that I found really helpful about these support groups, [...] [is that it] lets you see how well people are doing and how they have overcome or how they’re coping. Or if there are people that are worse than you but they’re just in the best of spirits, you’re just like, ‘Wow! I can do this!’.” Further support for the benefit of
attending a SSc support group was disclosed: “These ladies are part of [my] support group and have been very supportive over the years in listening to all the things that have happened. Thankfully I don’t know what I would do without that group” (W8).

Other emotion-focused coping strategies used by participants included adaptive and avoidant distraction techniques that allowed participants to shift their focus away from their diagnosis or symptoms onto another activities, helping to reduce feelings of stress and anxiety. Among the techniques brought up into discussion, participants mentioned: a) listening to music; b) having a pet; c) watching television and playing games; d) attending the spa; and e) staying busy or focusing on things other than SSc.

For participant W6 “...it’s music, sometimes if I put on certain CDs, and I’ll start singing along, I’ll be like, ‘Oh my gosh, like I’m not stressed anymore. It’s kind of gone,’ or I’ll have like a, you know that running water fountain.” Participant M10 discussed how his dog was an important “coping tool” as:

The stress when you’re not feeling you know all that positive, you just kind of lay with him and he’s you know, he’s just a little guy. He’s only like 12 pounds, a little Shih Tzu poodle cross, so he’s just a little fur ball. So he’s been very good. [...] I don’t know I guess you just look, you pet him and you relax with him and you kind of feel calm and then maybe you don’t get worked up and you know you kind of, I guess forget about things that you should be doing or something.

Participants also mentioned how watching television and playing games helped them to relax or distract themselves from their symptoms. Participants discussed the relaxation benefits that came from attending the spa. Participant W5 explained: “You know what, I went for one last month and I’m telling you, my kids bought it for me for Christmas and you know you’re wrapped
in a big warm bed with light and they’ve got the music playing and they’re massaging you and it was like ah, heaven.”

Some participants focused on staying busy as a way to help them cope with their SSc. Participant W2 described: “I don’t think about it anymore unless I’m symptomatic. That’s the truth. Honestly I get up and I just, I have planned for the day; you know I make, I have a set plan every single day.” However, participant W6 explained how this distraction may lead to the adoption of avoidance techniques: “I think sometimes, I think you could, oh there are so many things you could, you stop eating; you can sleep all day; there’s people that self-medicate. You know there’s so much pain that you know you go through months and months of just self-medicating themselves and they have no hope. Or some people, yeah they’ll sleep all day or they’ll sleep, like they’ll get major, major depression.” However, participant W5 responded: “See I do the opposite; I’ll get busy and do something. […]. I’ll distract myself.”

Lastly, participants described how engaging in exercise and relaxation techniques helped them cope with some of the physical and emotional distress they were experiencing. Participant W5 discussed how practicing yoga helped her relieve some of her physical symptoms. She explained: “…my jaw is killing me, I grind my teeth so much it’s like ow! But I was fortunate to have found Sahaja yoga from my sister-in-law who told me about it! It is very nice and relaxing.” Participant W2 commented: “Deep breathing [from the diaphragm], for me that’s part of it. I took yoga for a year actually and I found that extremely helpful even just learning about deep breathing.” Participant W6 noted: “I have a psychologist that taught me [relaxation techniques]. He tells me just imagine yourself on a beach or wherever you enjoy being. Some people might enjoy being in a totally different place, but [I know] it helps me [relax].”

** Meaning-focused coping
Participants used meaning-focused coping strategies to help them cope with their diagnoses. This was described as a shift in mindset towards moving forwards, self-acceptance, and adopting a new, more positive perspective towards living life with the disease. Participants engaged in positive cognitive reappraisal as a way to re-evaluate feelings held towards disease-related stressors, such as being concerned about the future and progression of their SSc, and learning to accept new roles within their families, among friends, and at work.

Some of the specific meaning-focused strategies discussed by participants included: a) learning to accept their diagnosis by engaging in benefit finding and cognitive appraisal; b) accepting one’s symptoms when they cannot be changed; c) engaging in self-acceptance and forgiveness; d) making the most out of what one has; e) taking it “one day at a time”; f) engaging in affirmations and emphasizing the positives things in life; and g) focusing on what is most important to them.

When benefit finding, participants focused on reflecting over what they had lost through the receiving of their diagnosis, to make way for positivity and growth to occur from this misfortune. For example, patient W4 described how important it is “…to grieve the things that you’ve lost. Whether it’s you know [your] relationships or abilities or the future, things that you like to do and you know you can’t do anymore. Without going through that grieving process it’s really hard to accept [the disease] and move on.” Participant W8 also described how she felt as she: “…began to accept that I couldn’t change what was happening and I couldn’t have done anything any differently than what I did. You know, and I just had to accept that this is how it is. And it’s not so horrible, I still did the very best that I could with what I could do.” Lastly, participant W2 explained:
I was dwelling so much on the 20% of things that I couldn’t do. It was just like, and I kept having that dialogue, like I’d say to my husband, ‘Well I can’t do that, I can’t do this, I can’t.’ Like can’t was so much. So I’ve taken that, that can’t out and now it’s, I hate to use the word redefine, that’s such a buzz word, but focus now on the 80% of what I can do. So you know I can’t do, I’m a passionate gardener for example, like our hobbies, I mean that was one of my most vivid passions. Well I haven’t given that up, it hasn’t stopped, like I’ve redefined what I can do. It’s different but it’s still acceptable, like I’m okay with that.

Many participants reported engaging in self-reflection by drawing awareness to their psychosomatic symptoms, forgiving themselves for any negative thoughts or feelings they may have, and by learning from these experiences. Participant W6 explained:

I think it’s also learning from the mistakes that you make. For me, since I’ve been diagnosed I got married and I had my daughter but then there was the breakdown of my marriage. And so I’ve learned from looking back at it, the mistakes that I did make with anger, with depression, I allowed my depression to get, you know like [...] involved with my marriage. So now that I’m three years out of it and looking back, you can see the mistakes you make and you just try to learn from your mistakes. So I think it’s okay to make mistakes and be depressed and be angry and be all these things that happen when you’re diagnosed with an illness. But I think the most important thing is just realizing that it didn’t help anything. You know being angry is not going to change anything, being, so for me, so it’s experience.

On a similar note, participant W8 explained: “I guess one of my coping skills would be [that] I allow myself to be upset and I allow myself to be angry over what’s happening. Not all the time. Sometimes I’ll say to my husband, ‘I need a couple of days, I’m upset, I need to get over
Participant W7 explained:

*I just wrote the word awareness down because from my experience, sometimes I didn’t know I was down at the bottom of the – and then I would do something ridiculously silly and irritating at home, or anywhere, or myself, I would be annoyed with myself for doing something. And I know after the fact that it was, I was having some problems. Like I was feeling uncomfortable, I was, something was annoying me, you know the pain. And it’s, and I didn’t even, I wasn’t even aware that I was kind of at the bottom of the valley.*

Many patients described experiencing feelings of guilt due to the limitations caused by their symptoms, the inability to do things they could previously do, and because of their reactions towards their family members and loved ones. As a result, patients described the importance of coping with feelings of guilt and distress by engaging in self-acceptance and forgiveness. Participant W2 explained:

*Putting yourself first. I think a lot of people are, they’re used to being the caregivers; they’re used to being the main roles in the family. For myself, a career which I no longer am capable of doing, this disease actually stopped a 23-year career for me. But I was a 911 operator with the police service. So all I knew for that length of time was looking after, like helping others. I didn’t know how to turn that around. So that’s what I found was, you know to put myself first at times and to not feel guilty about, about doing that.*

Further, participant W2 described: “Two steps forward, one step backwards. This is something that I’ve really internalized. I, I used to really beat myself up when I was having a bad day. And I don’t, I don’t think we should. I think we’re entitled to have a really bad day and not
dwell on it. Because I’ve, I found 100% that to every bad day I had; the next day is always without exception a better day. It’s not perfect but it’s a better day.”

This forgiveness and self-acceptance ultimately led participants to adopt a “one day at a time” mindset common to meaning-focused coping, which allowed them to make the most out of their situations and help them manage disease-related limitations. Participant W2 believes patients must: “Embrace the good days. So you’re going to have those bad days – you know it, they’re a given, that’s the reality, um. When you’re having a good day because they are going to happen, like I, that’s what I focus the most on.” However helpful, this change in mindset is not always easy to do as participant W7 explained:

Just knowing that, wow, we all have had similar feelings about some days we’re totally off and we’re cranky and we’re miserable. And we learned, we learned; have to learn to forgive yourself. And at the end of the day I always tell myself if I’ve had, I say, ‘Okay tomorrow I can start again.’ And it’s hard, and we keep falling off. It’s not something that all of a sudden we become, well I’m just great and bubbly all the time, no it’s 28 years later and I’m not great and bubbly all the time.

In response to participant W7’s comment, participant W8 replied: “You know tomorrow is still a new day. Tomorrow’s is always a new day.” The view of several participants on this subject appeared to be summarized by participant W8: “You know what I always say, I do what I can do today and leave tomorrow until tomorrow.”

It became clear that many participants shifted their thinking to include more positive thoughts and affirmations in order to relieve distress. Participant W7 spoke about how keeping a journal helped her stay focused on the positive things in her life:
So I can tell you about a couple of things that were coping strategies for me. I wrote it when I was very, very ill at the beginning, I wrote a journal every day. And the journal, in the journal was only each day one nice thing. And it could be, I remember clearly writing one day, I saw the trilliums in the woods driving to work and that was a beautiful feeling for just that one moment.

Participant W3 adopted a similar strategy: “I used sticky notes, so like for a positive affirmation, just like, oh I feel hot today or sexy to say those things because sometimes you know you have an ulcer on your hand and you’re just like that’s the grossest thing ever. But that doesn’t mean that you’re [gross].” Participant W7 also used a specific technique to emphasize the positive aspects of her life:

As you progress with this disease, so I’m now 28 years later, right, you also um, start looking at positive, what’s positive. And it’s good to have sometimes a piece of paper where you write positives and negatives. I found writing was one of the best things. Not to be a writer. But when I wrote down all the positive things, like I could take 20 things from you already having met you about positive things. And, and its little things. It’s the fact that every day I get to eat 3 meals. Or every day I am able to eat, not all foods, but some foods. Or I’m lucky that I can have nice things to eat. I have children; I have this; I have that, whatever it is. And gradually that becomes part of your psyche.

The final meaning-focused coping strategy described by participants consisted of focusing their attention away from their disease and onto what they felt was important.

Participant W6 explained:

I find that I, I remember that it’s not just about me like I have a daughter. So I see that I’m getting depressed, I’ll sit and think, ‘okay I can’t be depressed because I have to kind of look
after her.' Or show that you know mommy’s going to – because they worry as well or your parents worry or your friends worry; whoever it is that is around you. You might not have to be a mother, but whoever, you kind of just have to remember, ‘Okay I can’t, I can’t be so down because these people care about me.’ And, you know, so that I find that kind of sometimes helps you.

Participant W2 felt similarly: “And that’s what I was saying before about, you know you, yeah you can want to do it for your kids or for husbands or I mean I want to be a better person for my two golden retrievers because someone’s got to look after them and want them.”

Participants agreed that staying strong for their families helped them cope with various challenges and obstacles. Participant W8 explained her initial feelings of frustration about her family: “When raising my children, my two older children had the best of me. We were at the park all the time, we were sledding, we were skating, we were everything always. My little 5 year old did not get that at all.” Despite this frustration, she managed to find positivity within her situation, as she: “…had to turn it to see where there are advantages that she had. [My daughter] just turned five and she reads at a grade one level. Right? There were things that we did [at] mommy school and lots of crafts and stuff because I couldn’t do the other which has benefited her in a very different way that it did my other children.”

Discussion

Consistent with the three-faceted model of coping proposed by Lazarus and Folkman,[10-12], people living with SSc reported that they employ a combination of three main types of coping strategies, namely: problem-focused, emotion-focused, and meaning-focused coping strategies to help them manage their physical and psychosocial stressors. Problem-focused coping strategies included: a) seeking out professional psychological support; b) seeking out
disease-relevant information; and c) organizational strategies such as making lists. Emotion-focused coping included: a) seeking out social support; b) engaging in adaptive distraction techniques; and c) engaging in relaxation techniques, meditational exercise, or yoga. Meaning-focused coping strategies that helped participants develop a more positive perspective towards living life with SSc included: a) engaging in benefit finding and cognitive reappraisal; b) accepting one’s symptoms when they cannot be changed; c) engaging in self-acceptance and forgiveness; d) taking it “one day at a time”; e) engaging in affirmations and emphasizing the positives things in life; and f) focusing on what is most important.

Some of the coping strategies described by participants in the present study have been reported previously in more common chronic disease populations, such as cancer and HIV.[37, 38] Patients in those studies also sought out social support from friends, family, and from other patients with the same disease.[37] Similarly, patients tended to engage in problem-focused coping strategies including taking direct action to seek out information about their disease and using problem-focused strategies as a way of overcoming barriers caused by their symptoms.[37, 38] Meaning-focused coping has also been engaged in previously by other patients and included strategies such as focusing on the positive things in their lives, and engaging in benefit finding and cognitive reappraisal.[38]

Despite having similar coping needs to individuals with common chronic diseases, patients living with rare, relatively poorly understood, and unpredictable diseases may experience unique barriers to effective problem- and emotion-focused coping.[15, 18] Support resources or disease-related information aimed at helping improve quality of life and coping for patients with rare diseases, including SSc, are often not available or are not accessible because of the small number of rare disease patients in any given care centre or region.[6, 15] For instance,
there are challenges to accessing and using some of the resources described in the present study as potential sources of support, including attending annual patient conferences, joining SSc-based organizations, and using the Internet and Facebook as resource centres. National SSc conferences, including meetings hosted by Scleroderma Canada and the Scleroderma Foundation in the United States occur annually, and the international Systemic Sclerosis World Congress is held once every two years. Attendance requires travel that may not be feasible for many patients due to physical disability, time constraints, or financial reasons. Further, disease-related information that can be found on the Internet, such as in Facebook groups, or in online chat forums, may not be accurate or helpful, and patients do not always have the tools to differentiate between credible and less credible information. Lastly, participants stressed the importance and utility of attending SSc support groups as a way to foster feelings of inclusivity, and to facilitate the sharing of emotional and practical health information.[39, 40] These groups, however, tend to be offered only in urban centres, and, even in urban centres, may not be consistently available due to challenges in sustaining grassroots organizations that are managed by people with a serious and chronic disease.

One potential way to improve coping for patients with SSc would be to develop and disseminate accessible tools that support problem-focused and emotion-focused coping. The Scleroderma Patient-centered Intervention Network (SPIN)[6, 41] is an international collaboration of SSc patient organizations, clinicians, and researchers that was formed to develop supportive resources aimed at improving health-related quality of life outcomes among people with SSc. SPIN has developed a series of self-guided programs to support problem-focused disease management strategies and to provide SSc patients with the knowledge, skills, and confidence essential to managing their disease-related limitations.[42] These support tools will
be tested in clinical trials, and subsequently disseminated to people with SSc via partnerships with patient organizations.

Another potential way to support effective coping is to improve access to SSc support groups. There are currently almost 200 SSc support groups listed on the websites of Scleroderma Canada and the Scleroderma Foundation in the United States, most of which are led by people living with the disease.[43, 44] Many people with SSc, however, do not have access to SSc support groups due to location, disability, or because they are unaware that these groups exist. Further, due to the peer-led nature of SSc support groups, many patients experience challenges when trying to establish and sustain these groups. A potential mechanism for improving access to and the effectiveness of support groups would be to offer training programs for patients who are interested in taking on the role of group leaders. Additionally, to provide access to patients in geographically isolated areas or among those experiencing physical disabilities, SSc patient organizations may want to consider offering support groups via teleconferencing or videoconferencing.

Lastly, alternative sources of support may be useful at enhancing coping strategies for patients with SSc. For instance, SSc-patient organizations may consider the use of Facebook groups and online discussion boards or forums, and ensure that they are moderated by patients or staff who are knowledgeable about SSc. Benefits of these online resources include 24-hour availability, easy access, anonymity, and the opportunity to provide information to patients located in rural communities, or among patients whose symptoms are too severe to permit them to leave their homes easily.[45-47]

There are a number of limitations that should be considered when interpreting the results of this study. First, the participants within the present study constituted a convenience sample of
patients with SSc. Recruitment occurred primarily through the Scleroderma Society of Ontario and a single French clinical setting in Montreal. As such, there is the potential that the participants who were able to physically attend and participate in these focus groups represent a subsample of SSc patients who are potentially healthier and more able to engage in coping strategies than the average SSc patient. Further, given that the patients included in this study were both willing and able to attend in-person focus groups, this sample may also be overly representative of individuals with SSc who are comfortable participating in groups. Second, male and female participants were combined in the present study, and it is possible that coping strategies differ between the sexes. Less than 20% of people with SSc are male,[21] and only a small number of men were included in the present study. Future studies should more fully explore whether the specific coping strategies used by men with SSc as these may differ from those used by women. Third, both the number of participants as well as the facilitators in each of the three focus groups differed between groups. Finally, due to concerns about confidentiality in the context of the small scleroderma patient community, we were unable to link specific patient quotes to descriptions of the characteristics of the patients who provided the quotes.

In sum, there are many different physical and psychosocial implications of living with SSc that result in patients requiring unique coping strategies to help them manage life with the disease and improve quality of life and well-being. The present study assessed methods of coping by analyzing focus group discussions through the three-faceted paradigm of coping proposed by Lazarus and Folkman.[10-12] Findings indicated that participants used a combination of problem-focused, emotion-focused, and meaning-focused coping strategies to help them manage aspects of the disease. These results will be used by SPIN and patient organizations to inform the development of online support resources and self-management tools.
and to promote the utility of support groups aimed at improving coping skills among people with SSc.
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Declaration of Interest Statement

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