Exploring the experiences of living with a rare disease: Sources of distress and support group participation among patients with Scleroderma

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ABSTRACT

Background: Systemic sclerosis (scleroderma; SSc) is a rare, chronic, autoimmune disease characterized by multiple physical and psychological symptoms. These symptoms often result in significant disruptions and challenges to daily living, quality of life, and emotional wellbeing. Currently, there is no known cure for SSc and available treatments tend to focus primarily on the management of medical symptoms (e.g., lung disease) and on improving physical functioning. Support interventions aimed at improving the emotionally distressing aspects of the disease are needed. However, research exploring the sources of emotional distress and the impact they may be having on SSc patients is relatively scant. In addition, there is a lack of available professionally-delivered support resources, as such, patients with SSc often turn to peer-led support groups as a way to help them cope with and manage the disease. However, little is known about the benefits of SSc support groups, as well as reasons why patients may not attend these groups. In order to fill a much needed knowledge gap surrounding research on SSc two separate studies were conducted and included in this thesis. The objective of the first study was to use qualitative methods to gain a greater understanding of the sources of emotional distress experienced by patients with SSc. The objective of the second study was to identify the reasons why some patients with SSc do not participate in SSc support groups.

Methods: Within the first study, three semi-structured focus group discussions were conducted with a total of 22 patients with SSc. Participants were recruited through the Scleroderma Society of Ontario and through a SSc clinic in Montreal, Canada. The focus group discussions were recorded, transcribed, and then coded for emerging themes using thematic inductive analysis. Within the second study, a 28-item Scleroderma Support Group Non-Attenders Survey was developed and administered to SSc patients within Canada and the United States. Participants

were recruited through national and provincial SSc organizations including the Scleroderma Society of Canada and the Scleroderma Foundation in the United States. Exploratory factor analysis (EFA) was conducted on 21-items of the 28-item survey assessing reasons for not attending SSc support groups.

Results: Findings from the first study revealed that patients with SSc experience sources of distress prior to receiving a diagnosis and continuing throughout the duration of their lives. Six main themes representing sources of emotional distress were identified, including: (1) facing a new reality; (2) the daily struggle of living with scleroderma; (3) handling work, employment and general financial burden; (4) changing family roles; (5) social interactions; and (6) navigating the health care system. Within the second study, results from the EFA analysis revealed three main themes reflecting reasons for not attending a SSc support group including: (1) Personal Reasons, (2) Practical Reasons, and (3) Beliefs about Support Groups.

Conclusions: Taken altogether the findings from both of these studies allow us to gain a greater understanding of the sources of emotional distress experienced by SSc patients as well as greater insight into why some patients choose to not attend or are unable to attend SSc support groups. These findings will be used by national scleroderma organizations including the Scleroderma Society of Canada and the Scleroderma Foundation in the United States to develop support resources capable of improving the emotionally distressing aspects of the disease, as well as to improve accessibility to and effectiveness of current SSc support groups at meeting the needs of patients.

RÉSUMÉ

Contexte: La sclérose systémique (sclérodermie, ScS) est une maladie chronique auto-immune rare caractérisée par de multiples symptômes physiques et psychologiques. Ces symptômes se manifestent par des défis importants reliés à la vie quotidienne, la qualité de vie et le bien-être émotionnel. À l'heure actuelle, aucune cure n'existe pour la ScS, et les traitements disponibles se concentrent sur la gestion des symptômes et sur l'amélioration du fonctionnement physique. Les interventions de soutien visant à améliorer les aspects émotionnellement difficiles de la maladie sont indispensables. Cependant, la recherche sur les sources de soutien et l'impact du soutien émotionnel sur les patients atteints de ScS est limitée. En effet, on note un manque de ressources de soutien livrés par des professionnels, encourageant les patients à se tourner aux groupes de soutien guidés par un pair atteint de ScS. Les avantages de ces groupes de soutien ainsi que les raisons pour lesquelles les patients ne peuvent assister à ces groupes ont très peu été étudiés. Afin de combler ces lacunes, deux études distinctes ont été menées et inclues dans cette thèse. L'objectif de la première étude était d'utiliser des méthodes qualitatives pour mieux comprendre les sources de détresse émotionnelle vécue par les patients atteints de ScS. L'objectif de la seconde étude était d'identifier les raisons pour lesquelles certains patients atteints de ScS ne participent pas aux groupes de soutien.

Méthodes: Dans la première étude, trois discussions semi-structurées de groupe ont été menées avec un total de 22 patients atteints de ScS. Les participants ont été recrutés par le biais de la Société de Sclérodermie de l'Ontario et dans une clinique de ScS à Montréal, Canada. Les discussions de groupe ont été enregistrées, transcrites, puis codées pour les thèmes émergents en utilisant l'analyse inductive thématique. Dans la deuxième étude, un sondage sur les membres absents des discussions de soutien comportant 28 items (le "Scleroderma Support Group Non-

Attenders Survey") a été développé et administré à des patients atteints de ScS au Canada et aux États-Unis. Les participants ont été recrutés par des organisations de ScS, y compris la Société de sclérodermie du Canada et la fondation américaine de sclérodermie. L'analyse factorielle exploratoire a été menée sur 21 des 28 items évaluant raisons pour lesquelles des patients refusaient de participer à des groupes de soutien de la ScS.

Résultats: Les résultats de la première étude ont révélé que les patients ressentent de la détresse tout au long de leur vie. Les six thèmes principaux suivants représentant des sources de détresse émotionnelle identifiés: (1) faire face à une nouvelle réalité; (2) vivre avec la sclérodermie au quotidien; (3) les travaux de manutention, de l'emploi et de la charge financière générale; (4) l'évolution des rôles de la famille; (5) les interactions sociales; et (6) naviguer dans le système de soins de santé. La deuxième étude démontre que trois thèmes principaux reflètent les raisons pour lesquelles certains patients refusent de participer à un groupe de soutien: (1) des raisons personnelles et (2) pratiques, et (3) des croyances au sujet des groupes de soutien.

Conclusions: Les résultats de ces études illustrent les sources de détresse émotionnelle vécues par les patients de la ScS ainsi que les raisons pour lesquelles certains patients choisissent de ne pas participer ou sont incapables de participer à des groupes de soutien. Ces résultats seront utilisés par les organisations de sclérodermie nationales, y compris la Société de Sclérodermie du Canada et la Fondation de Sclérodermie aux États-Unis pour le développement de ressources de soutien visant à l'amélioration des aspects émotionnellement difficiles de la maladie, ainsi que pour rendre les groupes de soutient actuels plus accessibles et efficaces dans le but de répondre aux besoins des patients.

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CHAPTER 1: Introduction to Systemic Sclerosis

1.1 What is Systemic Sclerosis?

Systemic sclerosis (scleroderma; SSc) is a rare, multisystem, connective tissue disease characterized by three main physiological symptoms including: (1) vascular damage, (2) immunological/inflammatory system activation, and (3) excessive production of collagen [1, 2]. These symptoms often result in a reduction in blood flow to extremities, thickening of the skin, and damage to internal organ systems including the lungs, heart, and gastrointestinal tract [3]. The name "scleroderma" originated from the Greek words *scleros* (hard) and *derma* (skin), which represents the physical changes that commonly occur in SSc as a result of the excessive production and deposits of collagen in the skin [4]. Beyond skin involvement, SSc is associated with significant morbidity including Raynaud's phenomenon, ulcerations, joint contractures, arthritis, gastroesophageal reflux, pulmonary hypertension, pulmonary vascular disease, and interstitial lung disease [5]. Less common but potentially life-threatening manifestations include cardiac disease, lung disease, and renal crisis that can result in chronic renal failure [5, 6].

1.2 Disease Classification and Prevalence

To date, the pathogenesis and aetiology of SSc are complex and not well understood [1]. Both a combination of genetic and environmental influences have been linked to its' development, however, the aetiology of SSc remains unclear [7]. SSc is known to be a heterogeneous disease with varying degrees of symptomology among patients, however it is commonly classified into limited and diffuse clinical subtypes. The limited subtype is characterized by skin involvement that is distal to the elbows and/or knees, whereas diffuse SSc

entails skin involvement proximal to the elbows and/or knees in addition to the trunk and distal extremities [8, 9].

Upon receiving a diagnosis of SSc, patients have a median survival time of approximately 11 years, and are 3.7 times more likely to die within 10 years of diagnosis (44.9% mortality) than age, sex, and race-matched individuals without SSc (12.0% mortality) [8]. Among those diagnosed with SSc, approximately 80-90% are women, and the typical age of onset ranges between 30-50 years [8]. Currently there are over 16,000 Canadians affected by SSc [10], and a recent study in the province of Quebec proposed that prevalence rates are around 443 cases per million adults [3], and vary from less than 0.1 to 4.9 cases per 10,000 [7]. While various treatments for some of the physiological manifestations of SSc are available, there is currently no known cure or true disease modifying agents for the disease as a whole [11].

1.3 Disease Impact on Daily Living

In addition to the many physical symptoms that arise as a result of living with SSc, patients with the disease often experience significant disruptions and challenges to activities of daily living, quality of life, and emotional wellbeing. Many people with SSc are forced to make considerable modifications to their routines and lifestyles, in areas such as employment, homecare, relationships, and social activities [12], imposing a range of social, economic, and psychological burdens on patients. Common problems that impact quality of life and daily functioning for people living with SSc include emotional distress, fatigue, pain, pruritus (itching), sexual dysfunction, and body image distress due to appearance changes [13].

A variety of studies have explored the impact of these secondary symptoms on patients with SSc. One study surveyed SSc patients living in Canada and found that almost 90% of respondents reported experiencing SSc-related fatigue at least some of the time, and that 72% of

respondents reported that fatigue had a moderate to severe impact on their daily living [14]. Additionally, patients with SSc have been found to experience a range of different mental health challenges, including depression, anxiety, and more broadly, psychological distress [1]. One study looked at the prevalence of depression among a sample of SSc patients from France and found that one in five SSc patients met the criteria for a current episode of Major Depressive Disorder [15]. With regards to symptoms of anxiety, many patients with SSc report having fears about the future, including concerns about the extent and speed of disease progression, as well as concerns about being unable to work, becoming dependent on others, facing mortality, and experiencing a major loss of function [16]. Lastly, many patients with SSc experience significant appearance changes, especially to visible and socially relevant body parts, including the face and hands, potentially resulting in social challenges [1]. As such, many patients with SSc report experiencing significant body image distress and difficulty maintaining healthy social interactions [1].

The combination of physical and psychological symptoms that are common in SSc impact many aspects of daily living and quality of life among SSc patients. Despite the clear negative impact that these symptoms have on psychosocial functioning, the majority of available treatments for SSc generally focus on the management of medical symptoms (e.g., lung disease) and on improving physical functioning. Additionally, given that SSc is such a rare and complex disease, it is not well understood by many, including some healthcare professionals who come into contact with and treat patients, for instance those in local settings, who have little or no experience treating SSc and lack knowledge about the specific needs of people with the disease. As a result, resources and support services designed specifically for people with SSc to provide

them with information about their disease, as well as treatments and care options, are very much needed.

CHAPTER 2: Support Needs for Patients with Rare Diseases

2.1 Challenges of Living with a Rare Disease

Rare diseases are defined as conditions that affect fewer than one person in 2,000 [17]. Approximately 1 in 12 Canadians live with one of almost 7,000 known rare diseases [10, 18, 19]. Individuals living with rare diseases face many of the same challenges as people with any serious chronic, medical condition, including physical disability, disruption to social and work environments, and high rates of depression and other forms of psychological distress [20-23]. However, due to the rarity of the condition, people living with rare diseases often face additional unique challenges due to gaps in knowledge about their disease or how to treat it, the unfamiliarity of their disease to others, and often, distinctive and devastating emotional and physical consequences of the disease itself [18, 24]. Many people with rare diseases may feel that they visibly stand out more to others because of changes in appearance from the disease or other notable physical limitations, but at the same time feel unrecognized, because others do not understand their disease or its unique implications [24]. Additionally, it is not uncommon for people with rare diseases to face substantial delays in receiving a diagnosis [18, 24, 25] as well as have limited treatment and support options [18, 26]. Lastly, the financial burden of living with a rare disease is another significant challenge for patients [18, 24], as the majority of rare disease treatments are expensive, and patients (or their caregivers) are commonly unable to work during this time.

2.2 Limitations to Support Services for Patients with Rare Diseases

In common diseases, such as arthritis, diabetes, or heart disease, professional services are routinely provided through the medical system to help individuals with the disease cope with the physical and emotional challenges they face. Examples of professionally delivered supportive

care provided by the health care system may include disease-focused educational and self-management programs or supportive therapy to cope with the emotional aspects of living with a chronic illness. Supportive services designed to meet the specific needs of people with rare diseases are not typically available as part of health care in Canada [27]. As a result, patients with rare diseases, especially those who live in rural areas far from specialized medical centers, may feel that they are left to cope with their illness essentially alone. They may lack even relatively basic information about their disease, and, in many cases, this can result in substantial difficulty coping [28, 29].

As a result, grassroots level, locally formed and organized, peer-led support groups have emerged in rare disease communities, including SSc [26], and have "become embedded in our society as an important part of the health care system" [27]. These support groups follow the self-help concept that rare disease patients enduring the same challenges associated with a disease can help one another simply through social contact [30]. In these groups, people living with a particular rare disease work together to organize a group service capable of providing members with mutual support and opportunities to meet their educational and emotional needs [31]. Additionally, peer-led support groups are typically led by individuals living with the disease who have not undergone professional training for their role as group facilitator.

Support groups can be configured in a variety of ways; they may be online or face-to-face, facilitated by a professional or a peer, or have a structured or unstructured format [32]. These support groups often help reduce feelings of isolation by enhancing members' sense of belongingness, universality [32], and ultimately by providing them with opportunities "to gather the information most valuable to them – stories of similar experience and endurance" [30]. Support groups have also been found to enhance feelings of empowerment and agency towards

one's illness, as well as increase both the giving and receiving of emotional support and practical health knowledge about one's illness [33]. Group activities typically involve educational or information-sharing components, as well as the sharing of experiences and practical disease-relevant support [30, 32].

2.3 Support Groups in Scleroderma

Similar to other rare diseases [26], locally formed and organized support groups that are not connected to the formal medical system are an important resource for many people living with SSc [34]. Currently, there are approximately 30 SSc support groups in Canada and 150 in the United States (US), and all of them are peer-led [35, 36]. Whether local support groups are able to meet the needs of members, however, often depends on factors, such as the health of a single dedicated peer leader or the ability of an untrained peer leader to effectively navigate group dynamics and sometimes complex emotional issues. As a result, some local support groups struggle to meet the needs of members or disband.

To date, leaders of not-for-profit rare disease organizations in Canada and the US, including the Scleroderma Society of Canada and the Scleroderma Foundation in the US, help SSc patients locate local support groups, but provide almost no information regarding how to initiate a support group, nor do they offer formal training or resources and support to the peer facilitators of these groups. These national SSc organizations are committed to developing an infrastructure, including a training and support program for peer facilitators, to improve access to support groups and the ability of these groups to meet members' needs. However, in order to do this, further information about the activities and benefits of support groups, as well as participation behaviours among SSc patients in support groups is needed.

2.4 Limitations of Current Scleroderma Research

To date, the majority of research in SSc focused on understanding the symptoms, prevalence, and challenges of patients living with the disease is quantitative in nature and does not allow for a thorough understanding of the experiences of living with SSc directly from the perspectives of the patients. Before improvements in accessibility to and effectiveness of SSc support groups for patients with the disease can be made, further information must be gathered regarding the challenges and hardships experienced as well as to enhance current understanding of the areas of living with SSc that cause the most distress among patients.

Additionally, despite many people with SSc turning to support groups as a way to help them cope with and manage the symptoms of the disease, little is known about factors that may determine why SSc support groups are formed in some local settings, but not others. Furthermore, the processes by which SSc support groups are conceived, organized, and launched; the factors that influence whether SSc support groups are sustained over time; and the kinds of patient educational and support needs that are more or less effectively met by peer-led SSc support groups, are not well understood. More specifically, little is known about the benefits of current SSc support groups, the organizational factors that influence attendance, the reasons why some SSc patients cannot attend or are unable to attend support groups, and the challenges and support needs of current SSc support group facilitators.

2.5 Objectives

In order to fill the knowledge gaps concerning both the sources of distress experienced by SSc patients and the utility of and accessibility to SSc support groups, two separate studies were conducted and included in this thesis. The objective of the first study was to qualitatively examine the experiences of living with SSc through focus group interviews in order to gain a

better understanding of the sources of distress among patients, experienced directly from their own perspectives. .

Additionally, as described previously, patients with SSc commonly turn to support groups as a way to help them cope with and manage the symptoms and sources of distress experienced. Currently, little is known about the structure of established SSc support groups, why people with SSc either do or do not attend support groups, the role of support groups facilitators, and what facilitators may require in terms of training and support. In order to gain a greater understanding of the experiences of SSc patients with support groups, our research team developed a three-part survey assessing: (1) the benefits of attending support groups for current SSc support group members, (2) the reasons why some SSc patients choose to not attend SSc support groups and, (3) the challenges and support needs of current SSc support group facilitators.

The second manuscript within this thesis reports the results obtained from the second part of the three-part survey assessing reasons why some SSc patients do not attend SSc support groups. Therefore, the objective of this second study was to evaluate reasons why people are unable to attend or choose not to attend SSc support groups through an online survey.

2.6 Connecting Text

Despite experiencing many of the same challenges as other rare disease patients, the diverse symptoms associated with SSc results in a multitude of psychological and psychosocial issues that are currently not well understood. To date, a number of different studies have attempted to explore the sources of emotional distress and understand the ways SSc patients experience life with the disease using an "a priori" approach for how distress and other symptoms are defined and quantified; however, this does not provide patients with the opportunity to freely and openly share their personal experiences with the disease. As such, no study has sought to specifically explore, in-depth, the kinds of emotional challenges faced by people with SSc, in their own words, or examine the daily realities that contribute to these emotional experiences. Therefore, research focused on providing SSc patients with a "voice" is necessary, and will allow us to get a better sense of the challenges faced while living with a rare, fatal, and unpredictable disease.

CHAPTER 3: Manuscript 1

3.1 Exploring Sources of Emotional Distress among People Living with Scleroderma: A Focus Group Study

Published Paper:

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ABSTRACT

Background: Systemic sclerosis, or scleroderma (SSc), is a chronic and rare connective tissue disease with negative physical and psychological implications. Sources of emotional distress and the impact they have on the lives of people with SSc are not well understood.

Objectives: To gain an in-depth understanding of the emotional experiences and sources of emotional distress for women and men living with SSc through focus group discussions.

Methods: Three semi-structured focus group discussions were conducted (two in English, one in French) with a total of 22 people with SSc recruited through the Scleroderma Society of Ontario in Hamilton, Ontario and a SSc clinic in Montreal, Canada. Interviews were recorded, transcribed, and then coded for emerging themes using thematic inductive analysis.

Results: Core themes representing sources of emotional distress were identified, including: (a) facing a new reality; (b) the daily struggle of living with scleroderma; (c) handling work, employment and general financial burden; (d) changing family roles; (e) social interactions; and (f) navigating the health care system. Collectively, these themes refer to the stressful journey of living with SSc including the obstacles faced and the emotional experiences beginning prior to receiving a diagnosis and continuing throughout the participants' lives.

Conclusion: SSc was portrayed as being an unpredictable and overwhelming disease, resulting in many individuals experiencing multiple sources of emotional distress. Interventions and supportive resources need to be developed to help individuals with SSc and people close to them manage and cope with the emotional aspects of the disease.

Keywords: scleroderma; systemic sclerosis; emotional distress; focus groups; qualitative research; patient perspectives

Introduction

Systemic sclerosis (SSc), or scleroderma, is a rare, chronic autoimmune disease characterized by fibrosis and internal organ dysfunction including the lungs, heart, and gastrointestinal tract [1]. Symptoms experienced by people with SSc include disfiguring skin thickening, finger ulcers, joint contractures, chronic diarrhea, and renal failure, among others [2]. Common problems experiences by patients with SSc include pain, fatigue, sexual dysfunction, and reduced physical mobility and hand function [3]. The physical and psychological manifestations of SSc are heterogeneous in nature and can have dramatic biological, psychological, and social implications for people living with the disease [2, 4]. A recent Canadian study estimated the prevalence of SSc to be 44 cases per 100,000, including 74 per 100,000 among women and 13 per 100,000 among men [1]. The peak age of onset lies between 40-50 years [1, 4]. Estimated median survival times following diagnosis are 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 course [5].

Due to the rarity of the condition, people with SSc face additional challenges compared to those with non-rare conditions, including gaps in knowledge about their disease and how best to treat it [6-8]. Many people with SSc wait long periods of time before receiving an accurate diagnosis or explanation for their symptoms, and many experience difficulty accessing appropriate health care [7, 9]. In addition to medical consequences, people with rare diseases, including SSc, often experience social consequences such as stigmatization, exclusion from social communities, and reduced professional opportunities [7]. As a result, some people with SSc may experience significant isolation as they face life with an unpredictable and potentially

fatal disease [7]. Furthermore, supportive resources that are often available to people with common illnesses, such as disease-specific support groups, are often not available or easily accessible for people with rare diseases, including SSc, because of the small number of rare disease patients in any given care center [3].

Studies that have used diagnostic interviews or patient-reported questionnaire data have reported that people with SSc often experience significant emotional distress, including depression, anxiety, fears about disease progression and the future, and body image concerns [10-16]. For example, a Canadian study that assessed 345 SSc patients reported prevalence of major depressive disorder of 4% for the past 30-days, 11% for the past 12-months, and 23% lifetime, which is approximately double the rate in the general Canadian population [16, 17]. Another study, which evaluated 100 French SSc patients found that 49% had at least one current anxiety disorder and that 64% had met criteria for at least one anxiety disorder lifetime [18]. Beyond psychiatric diagnoses, many people with SSc experience significant fear related to disease progression, becoming physically disabled, or becoming dependent on others for help [16, 19, 20]. Body image distress and concerns about appearance are frequent due to changes to highly visible areas of the body, including the face and hands, which can be quite dramatic [16, 21, 22].

Existing quantitative research on emotional distress in SSc has evaluated prevalence, symptom levels, and contributing factors. However, this research has mainly specified *a priori* how distress will be defined and quantified and has not provided participants with the opportunity to freely share their experiences and perspectives concerning the emotional challenges and areas of distress they face [23]. Therefore, research focused on providing people with SSc the opportunity to share their own perspectives is necessary.

We were able to identify five published qualitative studies that have used focus groups or individual interviews to explore the perspectives of people living with SSc [6, 11, 24-26]. Three focus group studies reported major coping challenges for participants and focused mainly on issues related to managing the daily impact of the physical manifestations of the disease [6, 24, 25] or handling the unpredictability of the disease course [6]. A fourth study, which used individual interviews, focused on depression and the label of depression to describe emotional experiences, rather than explore the full range of participants' emotional experiences [11]. A fifth study, which also used individual interviews, explored the daily life experiences of people living with SSc and identified four main themes, including: (a) the physical impact of the disease, (b) the emotional impact of the disease, (c) the social impact of the disease, and (d) steps taken by participants to cope with the disease [26].

Thus, existing studies have explored the nature of the burden of living with SSc and the distress it causes, but they have not explored, in-depth, the range and impact of the emotional experiences of people living with the disease. Therefore, the objective of the present study was to extend previous research and to develop a comprehensive understanding of both the sources of emotional distress and the nature of the emotional experiences of women and men living with SSc, using focus group interviews. Focus group interviews allow us to obtain information directly from patients and may identify previously unknown experiences that would not be discovered using other common research designs, such as closed-ended questionnaires.

Methodology

Research Design and Epistemological Approach

The present study was framed within the social constructionism approach, which is premised on the idea that shared understandings are co-constructed between members of a given

social group through the consideration of multiple viewpoints, interactions, exchanges, and through negotiations of meaning [27-29]. Therefore, to foster the interchange among participants and unveil their experiences of enduring similar or unique challenges related to living with SSc, we conducted focus groups.

Focus groups have been widely used in health research, including SSc, to collect data through participants' discussions and interactions [6, 25, 30-33]. Focus groups are particularly valuable for research when little is known about a phenomenon of interest and gaining a shared understanding of the experiences of participants concerning a specific phenomenon is sought [34].

Participants and Procedures

We conducted a total of three focus group interviews. Two English-language focus groups were conducted in a hotel conference room in Hamilton, Ontario, Canada, and one French-language focus group was conducted at the Jewish General Hospital in Montreal, Quebec, Canada. Eligible participants were men and women diagnosed with SSc who were fluent in English for the Hamilton focus groups and in French for the Montreal focus group. Potential participants for the English focus groups were members of the Scleroderma Society of Ontario, who were contacted about the study by the society. Potential participants for the French focus groups were enrolled in an ongoing cohort study and contacted by either a research nurse coordinator or a research assistant. Participants who expressed interest in the study were contacted by a study investigator, who provided them with details about the study and set up a date and time for them to participate in one of the three focus groups. Prior to each focus group, additional details about the focus group process were provided, and all participants completed a brief demographic questionnaire in order 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 trained psychologist and a graduate-level trainee in psychology. The focus group interviews consisted of a series of open-ended questions aimed at promoting an open discussion about the experiences of emotional distress among people with SSc. After the focus group procedures were explained, the group moderators introduced the topic of emotional distress, asked relevant questions (e.g., With regard to your SSc, what are some things that cause you to experience stress?), and then used probes (e.g., According to you, is the experience you are describing common to all people diagnosed with SSc? Can you elaborate on that?) to encourage a greater sharing of experiences, thoughts, and feelings about certain ideas that arose in the discussions, as well as to gain a clearer understanding of the topic under examination [34, 35] (see S1 Appendix for interview guide). This process of group interviewing allowed the investigator to tease out the strength of participants' beliefs and subtleties about a topic while capturing individuals' ideas, experiences, and attitudes as they developed through group interaction and exchange [36, 37].

Due to technical reasons, the two English focus group interviews were recorded by video and audiotape, and the French focus group interview was recorded by audiotape only. All focus group interviews were transcribed verbatim to facilitate future recall and analysis. To better support the presentation of the findings, we extracted participants' quotes directly from the transcripts. To differentiate the participants, we used F for female and M for male before the numerical identifiers the transcribers arbitrarily attributed to each participant based on the focus

group discussion they were in and the order in which they first spoke while participating within their group. The community of Canadians with SSc is limited in size. Thus, in order to protect the confidentiality of the participants, we purposefully did not present individual demographic characteristics in the present manuscript, that could serve to identify the participants.

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

Data Analysis

A thematic analysis approach [38] was used to analyze data. Thematic analysis is a qualitative research method that supports the identification of new ideas directly from the data, while also relying on existing literature to help integrate novel and predetermined ideas. Thus, this analytic approach allowed the investigators to openly explore the participants' viewpoints and at the same time, to enrich our knowledge and understanding of the phenomenon of interest [38-40].

A professional transcriber fluent in English and French was responsible for the transcription of the three focus groups. One investigator independently initially analyzed the transcriptions of the two English focus group interviews, and a different bilingual investigator initially analyzed the transcriptions of the French focus group interview [41]. These investigators followed the same analysis steps, as follows: First, the investigators read the transcriptions several times to achieve full immersion in the text data and to obtain a sense of it as a whole. Second, different fragments of the text representing participants' discussions were assigned a code depending on their meaning. Third, based on comparisons among different coded groups, similar codes were grouped together. The investigators then created a preliminary coding scheme

based on potential relationships among codes. As such, themes and sub-themes were identified. To assure the consistency of this preliminary coding scheme, the investigators used it to recode the three transcriptions in full. Following this, the investigators re-read the document and highlighted quotations that appeared to capture key thoughts or concepts that meaningfully expressed the core idea of each theme.

Following the development of a preliminary coding scheme, a second bilingual investigator thoroughly reviewed all three coded documents. Then, the investigators discussed their coding in order to achieve consensus upon codes and to resolve any discrepancies in text codings. As a result of these discussions, a sustainable coding scheme was developed. No differences were identified when between-group comparisons were conducted.

To illustrate the themes, we extracted quotes from the transcripts. All patients contributed to the focus group discussions; but, to represent each theme, we chose the most representative and compelling quotes.

As suggested by Poland [42], after agreeing in the meaning of selected quotes, we corrected and adjusted conversational mechanic errors (e.g., incorrect grammar use). Data analysis was supported by the use of the qualitative research software *Atlas.ti* [43].

Results

Participant Characteristics

A total of 22 individuals with SSc (18 females, 4 males) participated in one of the three focus groups (1st English = 6 females; 2nd English = 4 females, 1 male; French = 8 females, 3 males). Sociodemographic characteristics are presented in Table 1. The age of participants ranged from 26 to 77 years with a mean age of 53.3 years (Standard Deviation [SD] = 13.3). Of the 22 participants, there were 2 (9%) who reported that they had been diagnosed with limited SSc, 10

(45%) with diffuse SSc, 5 (23%) with CREST, 5 (23%) who were unsure of their disease subtype. Years since diagnosis ranged from 0 to 28 with a mean of 10 years (SD = 8.0). Of the 22 participants, the majority were White (86%), had completed at least some college education (77%), and were either retired (23%) or on disability leave (32%).

Participants' Perspectives Concerning Sources of Emotional Distress

Six overarching themes related to perceived sources of distress, as described by study participants, were identified: (a) facing a new reality; (b) the daily struggle of living with scleroderma; (c) handling work, employment and general financial burden; (d) changing family roles; (e) social interactions; and (f) navigating the health care system. Code definitions and the coding scheme are provided in the S2 Appendix.

Facing a New Reality

The process of receiving a diagnosis, according to participants, entailed two different stressful events. First, some participants explained that, for a period of time, they endured a multitude of symptoms before being properly diagnosed. Participant M-P11 said, *"my family doctor […] tested me and my blood, said that I carried lupus, and further on they're like no that's not what it is. And it took, like I said, it took a long time to narrow it down."*

Coming to terms with the actual diagnosis of SSc was another emotionally challenging experience due to the unpredictable nature of the disease. For many participants, a major source of emotional distress after receiving a diagnosis related to not knowing how the disease might progress. Participant F-P4 explained, *"after the first year I just fell into a major depressive episode where I was just in bed for like 7 months and that was it."* The same participant said, *"the most frustrating thing about the disease is that once you get to a point where you're comfortable with the level of symptoms and interactions you have, then all of a sudden it*

progresses and then something else goes wrong." Unpredictability was also related to the knowledge that there is no cure for SSc, which was especially difficult for some participants. As participant M-P11 described, *"the hardest part for me was getting my head around the diagnosis. You're not going to be off these drugs, you know, [and] as of right now it's not going to get better."*

Ultimately, this uncertainty about what the future might hold resulted in participants voicing concerns about seeing others with SSc in worse condition. As a result of this, many participants struggled with the decision of whether or not to attend SSc conferences or support groups as they often felt discomfort over the idea of seeing someone with SSc in worse condition than themselves. Participant F-P8 explained, *"the most difficult part was to go to something for the first time, where there would be people with scleroderma, because I was afraid of what I would see."*

The Daily Struggle of Living with SSc

Two major burdens arose within the day-to-day lives of participants, including dealing with symptoms of the disease and undergoing treatments. Participants explained the hardships, frustrations and feelings of hopelessness that arise on a daily basis as they experience and attempt to manage psychological symptoms, such as feelings of depression; and physical symptoms, such as Raynaud's phenomenon, gastrointestinal complications, and breathing problems, simultaneously. Participant F-P1 described experiencing multiple symptoms as *"having your 20th kid and trying to think how are you going to feed that one [when] you've got another one coming along next year you've got to deal with"* and that *"it grows, it's like a cancer; it's going out of control."* In this vein, Participant F-P1 described, *"I can't cope with continual pain that just will not go away. I can't function, I can't do anything."*

For these participants, experiencing fatigue and having problems sleeping made it impossible to consistently meet daily demands, such as keeping their house clean, finishing home renovations, or grocery shopping. Participant F-P4 explained, *"with scleroderma you literally have a bank account of let's just say 1,000 points of energy, but you need, I don't know 20,000 points of energy to get what you need to get done."*

Additionally, many participants experienced feelings of guilt when their symptoms did not permit them to meet expectations that others had for them. Participant F-P9 described, *"it's very hard for everybody else to perceive how hard it is for you to manage through the day. I feel guilty because people have expectations. And, well your house should be clean. Well my house is not clean by any means anymore. And that's guilt – Oh! I should pick this up or I should pick that up. But for me as soon as I go to bend over, I have regurgitation immediately."* Ultimately, this inability to complete daily tasks resulted in some participants changing their priorities and realizing that *"whatever was really, really important that you had to do, all of a sudden [doesn't] matter as much anymore"* (F-P10).

Facing the reality of ongoing appearance changes was also discussed as being a significant daily stressor. Participant F-P9 explained, *"We had family pictures done at Christmas and I couldn't pick out our pictures that had me in it because I didn't look the way I remember looking….my husband actually had to pick out all the pictures because all I could see were all the changes that happened over the last 4 years."*

Experiencing depressive feelings, as well as uncertainty, fear, and anger was common among group members. In this vein, participant F-P2 explained, *"the next day you wake up and you feel really good and all of a sudden those tears start, and you think, holy crap, like what's wrong with you? Am I depressed or not?"* In some cases, the distress and the depressive feelings

were difficult to deal with and had an important negative influence on participants' personal lives, as participant F-P7 described, *"I allowed my depression to get involved with my marriage"* ultimately resulting in its *"breakdown."*

Manifestations of SSc typically include Raynaud's phenomenon, in which cold temperatures or other triggers lead to narrowing of the blood vessels, discoloration, and pain in extremities. As a result, managing and adjusting to cold temperatures or temperature changes was another area identified as being significantly challenging and frustrating for many of the participants on a regular basis. Participant F-P3 illustrated this problem by explaining that while shopping, *"I found a couple of things I wanted to try on. Well I don't try things on, like I don't have that luxury. I mean I can buy something and return it but it's freezing in the room."* Participants also agreed that it was especially upsetting when they were no longer able to participate in the same activities as before their SSc symptoms intensified due to the cold. Participant M-P11 explained, *"I don't even go ice fishing; I don't even go to the cottage in the winter. I just can't handle the cold, you know, like 5 minutes I'm done. I'm absolutely done."*

Undergoing various treatments, medical procedures, and experimenting with different medications were identified as being the second main emotional burden within the daily lives of participants. Participant F-P7 described *"the problem with scleroderma is that there's no treatment; no drug [can] help [every] person."* Not only were participants frustrated with having to take multiple pills without always experiencing significant improvements in their symptoms, but many also struggled to cope with the side-effects of the different medications they were prescribed. For participant F-P4 *"it got to the point where you're just….for me over the summer when they switched my medication and I just kept constantly throwing up and saying, 'It's not working, I'm not functioning.' to the point where you just say, 'I'm not taking medication*

anymore because I got so fed up, like nothing would work'." Through tears, participant F-P3 described her infusion treatments for Raynaud's phenomenon as: *"terrifying, it was beyond painful how symptomatic I was to this infusion. I'd never experienced that in my life."*

Managing Work, Employment, and General Financial Burden

Participants agreed that living with SSc dramatically impacted their ability to work, and as a result many participants have had to stop working or adopt a part time schedule. Participant F-P10 explained, *"I did work full-time, I was a hard worker, carried on 2 full-time jobs at one point and now I'm just a part-time worker. That's all I can do right now."* Additional frustrations arose when participants discussed dealing with their employers. Participant M-P11 described, *"I still work full-time, I work shifts; fatigue has become a real issue. It's unfortunate my employer has not been of any help at all; I thought they would have been. I've requested to reduce my shift work and some other things, and it's almost like, they seem like it's not even a recognized issue that I am going through."*

The inability to work translated into concerns about affording health care, medications, and specialist appointments, as well as having to buy additional items (e.g., gloves, warmer coats), to help participants cope with and manage the disease. In Canada, basic medical care is covered universally; however, prescription medications and other alternative treatments commonly required by people with SSc are typically not covered, resulting in many people the disease having to pay out of pocket in order to receive the care and treatments they need. Participant F-P1 expressed, *"it's all revolved about money. Money will help you be better or you've got to buy all these drugs. And you don't have it. And that is a major stress because it never sinks in […] that you're financially strapped."* Participant F-P1 described, *"the impact in terms of stress that the loss of your job and loss of your income is, if you're married of course*

your income now is minus the income of the female. And I say that because it's more females than males that get scleroderma. But for single people, it's having an income to having zero."

Changing Family Roles

Many participants spoke of their families as being positive and supportive, however some reported that discussing aspects of living with SSc with family members and partners was difficult and stressful. It became clear that some people felt their family members *"forget you actually have something wrong with you"* (F-P2). According to participants, sometimes their family members do not realize the extent to which they experience symptoms or struggle with their SSc, resulting in comments such as, *"you don't look sick"* (F-P4). Participant F-P3 explained the importance of being able to communicate with family members and partners about symptoms as*, "people can't see the fatigue, stress, pain, and the sleep deprivation […] so you've got to tell them."*

Many participants experienced feelings of guilt and worry when they felt they were letting their families down by not being able to maintain their roles as the primary caregivers within their homes. Participant F-P9 discussed her regrets, *"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."* Participant F-P9 admitted, *"I'm always trying to solve it for my kids to make it easy for them and for the people around me. Because this is difficult for me to handle and not that I'm the matriarch, but my kids will tell you I'm the glue that holds the family together, so when I go into the hospital for five days, everybody literally falls apart when I'm not there. So I feel like I have to be that strong person."*

Other participants' felt like having SSc made them a burden to their families. Participant F-P4 explained, *"when the disease first came about, my whole family blamed me for having the*

disease just because they're super religious and they thought God was punishing me." As a result, participant F-P4 said, *"[I] can't talk to my family about my disease because they throw it back in my face. So that's my number one rule, don't talk about my health with my family which makes it difficult because they're family and I'm looking for support."* In a similar vein, participant F-P10 admitted, *"I don't want to be a burden on people, so I haven't told my family things."*

Social Interactions

Social interactions beyond the family, were also identified as a source of emotional distress because many participants felt exposed by their visible differences and, as a result, received unwanted attention. As participant F-P3 explained, *"I was in the grocery store, I went into the freezer section and you know had the [...] same old story, and really painful. And now I'm losing function of my hands and I can't get my coins out of my purse […] and I could feel the anxiety level growing. And a woman you know was very [annoyed], like these are just strangers that feel a need to just comment, and I got so upset."* Participant F-P4 discussed receiving *"the same comments when I'm wearing a sweater or a tuque in the middle of summer, it's like it's not that cold, it's just like you have no idea."*

Participants were also frustrated when others did not understand the severity of the disease as all they see is a *"perfectly, healthy, working individual"* (F-P1). Participant F-P9 described their experience, *"in the summertime my mother-in-law was telling family about how sick I had been. And my husband and I took the kids to go visit these people. And I walked in the door and they were flabbergasted, 'You look so good, you look wonderful!' From all the horror stories they had heard, they had expected me to be dragging myself in. And so it makes it hard for them to believe that you're really sick."* Participant F-P9 admitted, *"you know what my mom*

always says when you go to the doctors? Don't put on any makeup, make sure you're looking all worn so that it's more believable. And you know I said, I said to my mom, 'Why should I have to fake it?' Right? I really am that sick."

Participants also felt frustrated and angry knowing that many people labeled or identified them as the *"individual with scleroderma."* Participants F-P2 and F-P3 admitted that they *"don't want [scleroderma] to be [their] identifier."* This frustration was extended when participants felt they were being given special treatment or receiving pity from their families, friends, or others, because of the disease. In participant F-P9's experience, *"there are some family members who try to butt in and want to take over some of the things that I do, and then there's resentment there because they can do it and it's so hard for me to do it."* As participant F-P8 discussed, *"nothing was more annoying than somebody wanting to come up and open the can of tuna fish for me because I was struggling."*

Navigating the Health Care System

The last source of emotional distress discussed by participants was the health care system, and more specifically, communication with health care professionals and having too many medical appointments. Participant F-P4 explained, *"I was telling [my doctor] about how my symptoms were worsening because I had stomach emptying problems to the point where I couldn't eat anything. Anything I ate it would just come right back up. I had no energy. I was having trouble breathing. Lots of problems over the summer. And, when I was telling him all the problems I was having and the services that I needed, he said, 'Too bad, so sad'."* Participant F-P3 described her experiences with the medical community as being *"dismissive"* as she felt that *"when another body part starts to ache"* doctors *"automatically assume its scleroderma related"* and therefore they do not give her enough time to thoroughly examine her symptoms

and provide her with appropriate explanations. When discussing her specialist, participant F-P3 explained, *"I see mine every three months, but you know we've got a set parameter of things that we discuss and very limited time."*

Participants also expressed frustration with the amount of specialist referrals and appointments they had. For example, participant F-P9 reported having *"seven specialists"* (F-P9). Participant F-P10 explained, *"this is because we keep getting referred. Here's another specialist, here's this, here's that."* When asked about how these appointments directly caused stress in the participants lives, participant F-P10 explained, *"well I've got to take time off work; I've got to go here, got to go there; got to take their medicine for this"* to which participant M-P11 agreed, *"I just can't keep taking time, days off, and using up all my vacation days or my ten government personal days every time I've got to go, but at the same point they're specialists and it's kind of like okay well…"*

Another area causing distress among participants concerns obtaining accurate and helpful information from health care personnel about issues related to intimacy and sexuality with SSc. Participants discussed that information covering aspects of sexuality with SSc is scant, which was particularly discouraging for some participants. Participant F-P9 describes her experience when asking her doctor about ways to address intimacy concerns as follows: *"we went to the Doctor and said, 'Hey give us a hint here, there's a real problem.' And you know what he said? He told my husband, 'You let her relax for the day and you take care of everything and maybe something can happen.' That was their advice. And it's like, 'Are you kidding me?'."*

Lastly, participants expressed great anger and frustration when discussing their health coverage and services, as many felt they were not provided with adequate care or had limited access to satisfactory health services. Participant F-P4 felt that her organization was withholding

services because *"they thought [she] wasn't sick enough"*. She expressed her frustrations with health services: *"does he not understand how severe my disease is and how specific my dietary requirements are and things that I need to manage daily? And when I told him that, you know, I was throwing up and sometimes it would end up on the floor or all over my bathroom and that the PSW [personal support worker] said that they couldn't enter my house it's a dangerous environment. Obviously like that doesn't make any sense because if I can't clean it up because of my energy or I can't bend down or manage water, then what do I do? And he didn't have an answer for me."*

Discussion

The main finding of this study was that sources of emotional distress for people with SSc are multifaceted in nature, begin prior to receiving a diagnosis, and continue throughout their lives in an unpredictable manner. Six main themes related to sources of emotional distress were identified, including: (a) facing a new reality; (b) the daily struggle of living with scleroderma; (c) handling work, employment and general financial burden; (d) changing family roles; (e) social interactions; and (f) navigating the health care system.

Of the six themes identified in the present study, three were consistently reported in all five previous studies [6, 11, 24-26] that explored emotional distress among people with SSc using qualitative methods: Facing a New Reality, the Daily Struggle of Living with SSc, and Social Interactions. In all five studies, participants discussed issues consistent with Facing a New Reality theme, including the process of receiving a diagnosis, experiencing uncertainty about the progression of the disease, and developing feelings of anxiety about the future. Concerning the Daily Struggle of Living with SSc theme, participants in previous studies similarly reported stress due to experiencing and managing the physical and emotional symptoms of SSc, learning

how to manage cold temperatures, experiencing treatment and drug side effects, and maintaining everyday responsibilities including chores and general housework [6, 11, 24-26].

Three of the themes reflecting experiences of sources of distress identified in the present study were not reported in depth in previous qualitative studies: Handling Work, Employment, and Financial Burden; Changing Family Roles as a Potential Source of Stress; and Navigating the Health Care System. Related to Handling Work, Employment, and Financial Burden, participants described the emotional burden related to modifications that they had to make to their occupational or professional roles because of their illness. Many participants discussed having to quit their jobs or reduce the amount of hours they could work. In turn, these reductions in income resulted in participants feeling financially stressed and burdened as many struggled to afford the drugs, treatments, and additional resources needed to be able to effectively cope with and manage their SSc.

The second novel theme was Changing Family Roles. In past research, family members of people with SSc were generally discussed in a positive light, with the family providing support and additional help to the individual with SSc [11, 24, 26]. However, in addition to these positive aspects, many participants in the present study, which focused on stressors, discussed families as an additional source of distress in their lives. Some participants reported feeling overwhelmed and frustrated when discussing their SSc diagnosis with their family members, and some felt they had become burdens to their families as they could no longer maintain their typical roles and responsibilities within their households.

The last unique theme identified and explored in this study concerns the participants' stressful experiences attempting to navigate the health care system. Although this concept has been mentioned previously [24], the extent to which navigating and dealing with the health care

system can cause stress among people with SSc is not well understood. Participants in the present study identified several aspects of their experiences with the health care system as causing significant stress, including having multiple specialists and appointments, experiencing the overgeneralization and dismissive nature of patient-doctor communication, as well as other general problems concerning the organization of the health care system.

Although SSc is a rare disease, some of the sources of emotional distress identified in the present study are also experienced by people with more common chronic diseases, such as cancer and arthritis [6, 11]. Other sources of distress, however, such as difficulty obtaining an accurate diagnosis and specific aspects related to navigating a health care system not designed for people with rare diseases may be unique to the rare disease context [7, 8]. It is common for people with rare diseases to be improperly diagnosed or have to wait long periods of time before receiving an accurate diagnosis [6, 8]. Additionally, people with rare, disfiguring diseases may face additional social consequences such as isolation and stigmatization, compared to those with more common and invisible chronic illnesses. Due to the limited amount of available rare disease-related resources it is not uncommon for people with rare diseases to have to wait long periods of time or travel long distances in order to receive appropriate care and support. For many rare diseases, a lack of scientific knowledge and available information about the disease or how to treat it can make navigating the health care system particularly difficult and lead to having to consult with multiple doctors and specialists simultaneously. This process not only causes financial stress, but can be particularly confusing and exhausting for people with rare diseases, as participants with SSc described within the present study.

Supportive resources to help people with SSc cope with the sources of distress identified in the present study need to be developed. To date, the majority of research on treatments for SSc

focus on the management of medical symptoms (e.g., gastrointestinal disease) and on improvement of physical functioning. Findings from the present study and previous studies [6, 11, 24-26] emphasize that people with SSc experience a significant amount of emotional distress related to their disease, some of which may be specific to people with rare diseases.

Information gathered from the present study will be highly useful to inform the development of resources designed specifically for people with SSc, to help them cope with and manage the six identified areas causing them to experience disease-related emotional distress. Specifically, the Scleroderma Patient-centered Intervention Network (SPIN) [3, 44], an international collaboration of SSc patient organizations, clinicians, and researchers, was formed to develop supportive resources aimed at improving health-related quality of life outcomes among people with SSc. Thus, SPIN will use the information gathered from the present study to develop online supportive resources and tools that can be used by people with SSc to help them learn how to better manage and cope with the emotional distress they face. SPIN has chosen to develop these resources in an online format, in order to reach as many people with SSc as possible, especially patients who live in rural areas far from specialized care who are often left to cope with their disease alone as well as for patients who are concerned about social situations or seeing others with SSc in worse condition than themselves.

There are a number of limitations that should be considered when interpreting the results of this study. First, individuals who participated in this study constitute a convenience sample of SSc patients. Specifically, recruitment occurred through the Scleroderma Society of Ontario and through a research nurse coordinator research assistant at a single hospital site, which may have influenced the characteristics of respondents, and therefore may limit the generalizability of conclusions about the identified areas of distress for people with SSc in other geographical

locations. This is because people with SSc who are able to participate in these studies may be in healthier conditions than those who cannot, therefore our results might be an underestimation of the emotionally distressing experiences faced by people with SSc. Secondly, male and female participants were combined in the present study, and it is possible that the sources of emotional distress differ between the sexes. Only a small number of men were included, and it is possible that the focus group format may not have permitted the identification of stressors specific to men living with SSc. For instance, although we did not have any direct evidence for this, it is possible that some of the male participants may not have felt comfortable disclosing information about their emotional experiences within a female-majority group setting. Third, both the number of participants in each of the three focus groups as well as the facilitators of these groups differed between groups. Because the focus group interviews were semi-structured and led by different researchers, participants within the three groups may not have received precisely the same original research questions or probes. Fourth, the English focus groups were recorded by videotape, whereas the French focus group was recorded by audiotape only. It is possible that there could have been differences between the groups, for instance, in the extent to which participants felt comfortable freely discussing their experiences, because of this difference. Lastly, the participants in each of the three focus groups were only interviewed once, limiting the opportunity for all the participants in the group to have ample time to express their personal experiences. There is a need for future studies that examine the emotional experiences of SSc among men specifically. Although SSc predominantly affects females, men with SSc may experience unique emotional consequences that are currently not well understood.

Conclusion

In conclusion, there are many different aspects of living with SSc that may cause people with the disease to experience emotional distress. The present study identified six core themes related to sources of distress, including: (a) facing a new reality; (b) the daily struggle of living with scleroderma; (c) handling work, employment and general financial burden; (d) changing family roles; (e) social interactions; and (f) navigating the health care system. These six themes represent the main sources of distress experienced by a group of people with SSc. The findings of this study provide insight into the unique emotional experiences and challenges faced by people living with the rare and unpredictable disease, SSc. These findings will be used by SPIN to inform the development of online supportive resources and tools aimed at improving quality of life and coping among people with SSc.

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Table 1. Sociodemographic Characteristics of 22 Focus Group Participants.

Variable	
Female gender, *n (%)*	18 (82%)
Age in years, *mean (SD)*	53.3 (13.3)
White race/ethnicity, *n (%)*	19 (86%)
Level of education, *n (%)*	
Less than high school	1 (5%)
High school graduate	4 (18%)
College/CEGEP[a] graduate	7 (32%)
University graduate	8 (36%)
At least some postgraduate	2 (9%)
Occupational status, *n (%)*	
Homemaker	3 (14%)
Unemployed	2 (9%)
Retired	5 (23%)
On disability leave	7 (32%)
On leave of absence	1 (5%)
Full-time employed	2 (9%)
Part-time employed	2 (9%)
Scleroderma subtype, *n (%)*	
Limited scleroderma	2 (9%)

Diffuse scleroderma	10 (45%)
CREST	5 (23%)
Unsure	5 (23%)
Years since diagnosis, *mean (SD)*	10 (8.0)
Years since Raynaud's onset, *mean (SD)*	21 (13.3)

[a]Collège d'enseignement général et professionnel (CEGEP) is the post-secondary degree equivalent to grade 12 of high school and first year of university in the province of Quebec, Canada

S1 Appendix. Interview Guide (Adapted to Each Participant Group)

Overview:
These are examples of some of the initial questions posed to participants to initiate the discussion. Further questions and probes were used to support the conversation as it progressed naturally.

1. **What are some areas of living with SSc causing you emotional distress?**

    a. What term(s) would you use to describe your emotional experiences related to living with SSc?
    b. Explain to me how these areas you have identified as being distressful cause you to experience disease-related stress?

2. **What are some of the different emotions you have experienced while living with SSc?**

    a. What areas of living with the disease have made you experience anger of frustration?
    b. Are there any emotions we have not covered during this discussion that you experience or have experienced while living with SSc and you would like to share?

S2 Appendix. Identified Themes, Sub-themes, and Corresponding Definitions

Theme and Subthemes	Codes	Definitions
Facing a New Reality	Difficulty with diagnosis	Experiencing distress due to not receiving a timely and accurate diagnosis.
	Receiving a SSc diagnosis	Experiencing distress from the process of receiving a SSc diagnosis.
	Uncertainty about progression of disease	Experiencing distress as a result of feeling uncertain about the progression of their SSc.
	Seeing someone in worse condition	Experiencing distress as a result of seeing someone with SSc in worse condition than themselves.
The Daily Struggle of Living with SSc		
Experiencing and managing psychosomatic symptoms	Comorbidity of symptoms	Experiencing distress caused by having multiple physical or psychological symptoms at the same time.
	Dealing with unique symptoms of SSc	Experiencing distress caused by experiencing unique symptoms or situations caused by their SSc.
	Psychological symptoms	Experiencing distress because of psychological factors or due to having symptoms of depression or not understanding depressive feelings.
	Physiological symptoms	Experiencing distress caused from having constant pain, fatigue, appearance changes, Raynaud's Phenomenon, tight skin, gastrointestinal symptoms, and breathing problems.
	Dealing with the cold and temperature changes	Experiencing distress caused by experiencing cold temperatures or temperature changes.

	Doing general housework	Experiencing distress due to having to do housework.
Treatment complications	Experimenting with medications	Experiencing distress as a result of trying new medications or experiencing medication side-effects.
	Undergoing treatments	Experiencing distress as a result of undergoing various treatments for SSc, including side effects.
Handling Work, Employment, and General Financial Burden	Being unable to work	Experiencing distress as a result of having to quit their jobs, reduce their work hours, or losing their jobs because of SSc.
	Dealing with employers	Experiencing distress caused by their employers not being understanding towards their situations.
	Finances	Experiencing distress due to having a hard time paying their bills.
	Affording health care	Experiencing distress caused by the expenses associated with specialist appointments and other SSc related health care costs.
Changing Family Roles	Talking about SSc with family members	Experiencing distress caused by having to discuss SSc with their families.
	Feeling like a burden in your family	Experiencing distress due to feeling like they are burdens within their families.
	Difficulty maintaining roles and responsibilities within one's family	Experiencing distress caused by having to maintain their roles and responsibilities within their families.
	Lack of family support	Experiencing distress due to not having support from their families.
Social Interactions	General distress related to social interactions and	Experiencing distress due to social interactions and interpersonal relationships.

	interpersonal relationships	
	Comments from strangers	Experiencing distress caused by comments from strangers about their SSc.
	Feeling labeled by the disease	Experiencing distress caused by being defined and labeled by SSc.
	Being treated differently because of SSc	Experiencing distress because they feel they are being treated differently because of their SSc diagnosis.
	People not understanding the severity of the disease	Experiencing distress due to others underestimating or not understanding the severity of their SSc.
	Pushing yourself too hard	Experiencing distress due to pushing themselves past their physical and psychological limits.
	Feeling other people's pity	Experiencing distress because they feel their friends or family members pity them.
	Lying about how you feel	Experiencing distress because they feel they have to lie about or minimize the severity of their symptoms or their SSc.
Navigating the Health Care System	General problems with the health care system	Experiencing distress associated with problems within the health care system.
	Communicating with doctor	Experiencing distress as a result of trying to communicate with their physicians.
	Frequent referrals to specialists	Experiencing distress due to having multiple specialist appointments.
	Discussing sexuality	Experiencing distress as a result of having to discuss sexuality, intimacy, or intercourse with their physicians, friends, families, or partners.

Communication between physician and specialists	Experiencing distress due to being responsible for maintaining communication between physicians and specialists about their SSc.

3.2 Connecting Text

Results from our first study indicated that the sources of psychological distress experienced by patients with SSc are multifaceted in nature, beginning prior to receiving a diagnosis and persisting throughout the rest of their lives. When reviewing the main sources of emotional distress identified by participants, it becomes clear that despite the heterogeneous nature of the disease, many patients face similar disease-related challenges. Some of these obstacles include struggles associated with receiving an accurate diagnosis, developing concerns about the future, having to manage the psychological and physical symptoms, handling work, employment and financial burden, and learning how to discuss the disease with family members and friends.

It is not uncommon for patients living with rare diseases who typically lack available professionally-delivered support services to seek out peer-led support groups as a way to help them connect with others experiencing similar disease-related challenges, and to have an opportunity to give and receive emotional and practical support [26, 27, 31]. SSc is a rare disease where peer-led support groups play an important role [34]. However, despite their known utility, there are still many patients with SSc who choose to not attend SSc support groups. Knowledge of the factors influencing SSc patients to not attend these groups is currently lacking. In order to evaluate and potentially improve support group services for patients with SSc, information about why people choose not to attend or are unable to attend these groups is needed.

To date, no study has systematically examined the various reasons why patients with SSc do not or are unable to attend SSc support groups. As a result, our research team developed the Scleroderma Support Group Non-Attenders Survey, a 28-item survey with the purpose of systematically assessing the reasons why patients with SSc do not or are unable to attend SSc

support groups. The findings generated from this survey as well as future implications of this research are reported and explored within the next section of this thesis.

CHAPTER 4: Manuscript 2

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

Submitted Paper:

Gumuchian ST, Delisle VC, Peláez S, Malcarne VL, El-Baalbaki G, Kwakkenbos L, Jewett LR, Carrier M-E, Pépin M, Thombs BD. (2016). Reasons for Not Participating in Scleroderma Patient Support Groups: A Cross-sectional Study. Manuscript submitted for publication.

ABSTRACT

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

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

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

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

Introduction

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

Many people with chronic diseases join support groups to help them cope with and manage their disease [8, 9]. Support groups may be particularly important for people with rare diseases who are unlikely to have other disease-specific support [3, 5]. Support groups typically include educational or information sharing activities and opportunities for members to give and receive emotional support. They are based on the principle that people enduring similar challenges can help one another via social contact and the sharing of experiences [10, 11]. Support groups for people with chronic diseases may be held face-to-face, online, or via teleconference, may be led by professionals or peers, and may follow structured or unstructured formats [5, 11-13]. Among people with rare diseases, reported benefits of support groups include getting to know others with similar disease-related experiences, gaining information about the disease and treatment options from other patients, having the opportunity to give and receive emotional support, and advocating for improved health care for people with rare diseases [14-23].

Systemic sclerosis (SSc; or scleroderma) is a rare chronic autoimmune disease characterized by fibrosis and internal organ dysfunction, including lung, heart, and

gastrointestinal tract involvement. A recent Canadian study estimated prevalence to be 74 cases per 100,000 for women and 13 per 100,000 for men [24]. People with SSc often face significant physical, emotional, and social consequences, including debilitating symptoms, uncertainty about the future, difficulty accessing appropriate health care, and social exclusion [2, 4]. Support groups play a central role in the lives of many SSc patients, and currently there are estimated to be approximately 30 SSc support groups in Canada and 150 in the United States, almost all of which are peer-led groups that meet in local settings [25, 26].

In order to evaluate and potentially improve support group services, knowledge about why people choose not to attend or are unable to attend these groups is required. Only one study to date [27] has explored reasons why some people with SSc do not participate in support groups. That study reported that the most common reasons for not participating in SSc support groups were not being interested or having no perceived need for support; not having access to a support group; and not being aware that SSc support groups exist. Other reasons included practical barriers, such as not having enough time or being too ill or disabled to attend; emotional factors such as being scared to see patients who are very ill; and having negative perceptions about support groups. An important limitation of the study, however, was that it was based on a single-item that asked respondents to categorize reasons for non-attending as "I'm not interested," "None are easily available," or "Other – (please specify)."

Thus, the objective of the present study was to more systematically evaluate the reasons why people are unable to attend or choose not to attend SSc support groups.

Methodology

Participant Sample

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

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

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

The Scleroderma Support Group Non-Attenders Survey

Initial items for the survey were obtained from items that were used in a similar survey related to cancer support groups [28], generated from published results of a qualitative study on

reasons for not attending cancer support groups [29], and generated from responses to a single item on reasons for not attending SSc support groups, which was administered as part of the Canadian Scleroderma Patient Survey of Health Concerns [27]. In that survey, respondents who indicated that they had not participated in SSc support groups were asked to specify their reasons for not attending from the following response options: (1) I'm not interested; (2) None are easily available; and (3) Other (please specify). Respondents' open-ended responses to the "Other" option were analyzed using thematic analysis [27].

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

The final survey (see Appendix 1) consisted of 21 core items that assessed possible reasons for not attending SSc support groups. In addition, there were three items that assessed reasons for not attending support groups among survey respondents who had previously attended a SSc support group. There were also four additional items that were only relevant to subgroups of survey respondents (respondents with children and respondents who differed from most other patients due to age, sex, or race/ethnicity). Response options for all 28 closed-ended items included Not Important, Somewhat Important, Important, and Very Important (scored 0-3).

Data Analysis

Exploratory factor analysis (EFA) was conducted to group survey items into general themes that reflected reasons for not attending support groups. EFA was done with MPlus 7 [30] using weighted least squares mean with variance adjusted estimation, which accounts for the ordinal nature of the survey items, and geomin oblique rotation. The number of factors was chosen based on the scree plot (eigenvalues), model adequacy, and overall interpretability. Model adequacy was assessed using a chi-square goodness-of-fit test and three fit indices, including the Tucker-Lewis Index (TLI) [31], the Comparative Fit Index (CFI) [32], and the Root Mean Square Error of Approximation (RMSEA) [33]. Since the chi-square test is highly sensitive to sample size and can lead to the rejection of well-fitting models, practical fit indices were emphasized [34]. Models with a TLI and CFI close to 0.95 or higher, and RMSEA close to 0.06 or lower are representative of good fitting models [35]. A CFI of 0.90 or above [36] and a RMSEA of 0.08 or less [37] may also be considered to represent reasonably acceptable model fit.

Since the purpose of the present study was not to develop a measure with scoring properties, but rather to identify general themes, we did not remove items or calculate Cronbach's alpha. Items were included in the theme for which they had the highest factor loading.

Results

Sample Characteristics

There were 276 completed surveys, of which 14 (5%) were excluded because the respondent resided outside of Canada or the United States, 3 (1%) because the respondent reported a diagnosis other than SSc, and 17 (6%) because the respondents were missing some of the 21 survey items (range 7–21). Characteristics of the 242 respondents included in analyses are

shown in Table 1. The majority of respondents were female (84%), White (86%), married (63%), and from the United States (65%). Mean age was 56 years, and mean time since SSc diagnosis was 12 years.

Exploratory Factor Analysis

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

The three identified themes representing reasons for not attending SSc support groups included: (1) Personal Reasons (9 items), (2) Practical Reasons (7 items), and (3) Beliefs about Support Groups (5 items). Factor loadings for items on the three themes ranged from 0.37 to 0.78 for Personal Reasons, 0.37 to 0.80 for Practical Reasons, and 0.48 to 0.98 for Beliefs about Support Groups. Correlations between factors were 0.02 for Personal Reasons and Practical Reasons, 0.43 for Personal Reasons and Beliefs about Support Groups, and 0.07 for Practical Reasons and Beliefs about Support Groups. Item and factor loadings are shown in Table 2. Overall, respondents endorsed a mean of 4.9 (standard deviation (SD) = 3.8; 23% of all items) reasons for not attending as being Important or Very Important (see Figure 1).

The nine items in the Personal Reasons theme (see Table 3) were related to the current emotional and physical health of the respondent, concerns about not having time to attend the meetings, and need for support. Example items included "I do not need a support group because my symptoms are not severe" and "I do not feel comfortable in a group setting." On average, respondents endorsed 2.1 Personal Reasons items (SD = 2.0; 23% of Personal Reasons items) as Important or Very Important to them. The percentage of respondents with ratings of Important or Very Important ranged from 11.1% for item 20 ("I am uncomfortable sharing my experiences with a group;" mean = 0.48; SD = 0.81), to 50.4% for item 6 ("I already have enough support from family, friends, or others;" mean = 1.51; SD = 1.11). The mean item score among the nine Personal Reasons items was 0.78.

The seven items in the Practical Reasons theme reflected scheduling concerns and issues related to having access to a support group. Example items included "I do not know of any scleroderma support groups offered in my area" and "The time of the meetings does not fit in my schedule." The mean number of items endorsed as being Important or Very Important was 1.7 (SD = 1.6; 24% of Practical Reasons items). For Practical Reasons, the percentage of respondents with ratings of Important or Very Important ranged from 8.7% for item 28 ("I am uncomfortable with how I look;" mean = 0.31; SD = 0.68), to 48.3% for item 1 ("I do not know of any scleroderma support groups offered in my area;" mean = 1.46; SD = 1.21). The mean score among the seven Practical Reasons items was 0.76.

The five items in the Beliefs about Support Groups theme reflected concerns about the activities and benefits of support groups. Example items included "I think support groups are too negative" and "I do not think I would learn more about scleroderma than I already know." The mean number of items endorsed as being Important or Very Important was 1.1 (SD = 1.4; 22%

of Beliefs about Support Groups items). The percentage of respondents with ratings of Important or Very Important ranged from 18.2% for item 23 ("I do not think support groups provide educational information that is current and relevant;" mean = 0.57; SD = 0.93), to 25.6% for item 16 ("I do not think I would learn more about scleroderma than I already know;" mean = 0.84; SD = 1.07). The mean score of the five Beliefs about Support Groups items was 0.73.

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

Three items assessed previous support group experiences among a subset of the total respondents (n = 67) who indicated that they had attended a support group in the past. These items included item 25 ("I attended a support group in the past and had a bad experience"), item 26 ("I do not like the current leader of the local support group"), and item 27 ("I do not like the members of the local support group"). As shown in Table 3, almost one quarter of these respondents reported having had a bad experience with a support group in the past as an Important or Very Important reason for non-attendance. Just over 10% indicated that not liking the local support group leader or members of the support group was Important or Very Important to them.

Discussion

The main finding of this study was that reasons for not being able to attend or choosing not to attend SSc support groups could be described by three different themes that reflected personal reasons, practical reasons, and beliefs about support groups. The Personal Reasons and Beliefs about Support Groups latent factors were moderately correlated (0.43), whereas the Practical Reasons factor was not correlated meaningfully with either the Personal Reasons (0.02) or Beliefs about Support Groups factor (0.07). Thus, the three themes captured substantively distinct reasons for non-attendance. On average, patients rated five items as Important or Very Important to them, and endorsement was distributed similarly across items in the three themes.

Based on evaluation of individual survey items, there were two main reasons for non-attendance that were reported as being Important or Very Important to approximately 50% of respondents. These included: (1) already having enough support from family, friends, or others and, (2) not knowing of any local SSc support groups. Additionally, in the Personal Reasons theme, there were two items that were found to be Important or Very Important among at least 25% of respondents, including: (1) being uncomfortable seeing other people with SSc who may be worse off and (2) being too busy with other responsibilities to attend a support group. Within the Practical Reasons theme, reasons for not attending a support group that were Important or Very Important to at least 25% of respondents included: (1) the time of the meetings not fitting their schedules and (2) having problems getting to and from the meetings due to weather, distance, or other factors. In the Beliefs about Support Groups theme, three items were reported as being Important or Very Important to at least 25% of respondents, including beliefs that: (1) support groups are not helpful, (2) support groups are too negative, and (3) that they would not learn more about scleroderma than they already know by attending a support group.

Some of the reasons for not attending support groups that were commonly rated as Important or Very Important in the present study were also found to be important in a previous study that explored reasons for not participating in SSc support groups through open-ended responses to a single question [27]. Some of these reasons included not needing additional support, having symptoms that are not severe, experiencing discomfort about seeing someone in worse condition, and having other demands or being too busy (Personal Reasons); not being aware of support groups generally and locally, and being unable to attend due to symptoms being too severe (Practical Reasons); and perceiving support groups to be too negative or not a helpful resource (Beliefs about Support Groups) [27].

There were also a number of reasons for not attending support groups identified in the present study that had not been identified previously. Examples include not having a reliable way to get to the meetings and being uncomfortable with one's physical appearance (Practical Reasons); not wanting to see oneself as a "scleroderma patient" (Personal Reasons); feeling like one would not learn more about SSc, concerns that discussions during meetings would not focus on SSc, and concerns about privacy (Beliefs about Support Groups).

The knowledge generated in the present study can be used by SSc patient organizations to develop strategies aimed at addressing current limitations of support groups and improving accessibility to and effectiveness of current groups. Respondents rated, on average, five items as Important or Very Important, and, with few exceptions, all survey items were rated as Important or Very Important by at least 15% of respondents. Thus, strategies to improve SSc support groups and increase access to groups should be multifaceted and address the many reasons for non-attendance that were reported by respondents.

Given the predominance of practical reasons for not attending, the implementation of online support groups may be a feasible and economical option for reaching patients without access because of the unavailability of local support groups or due to travel-related barriers. Online support groups have been used successfully in more common disease groups, such as cancer, to improve access. They may also provide an opportunity to reduce potential stressors associated with sharing experiences in a face-to-face context [8] and may reduce travel-related barriers due to disability and limited mobility [38].

Another way to potentially increase local access to SSc support groups would be to provide training to peer facilitators. A training program for peer facilitators could provide SSc patients with the skills and training necessary to successfully establish, sustain, and manage support groups in places where none exist. In addition, many respondents expressed concerns about the utility of support groups and the potential for support groups to be overly negative. Trained peer facilitators could address these concerns by managing group dynamics more effectively and by learning how to tailor the activities and discussion topics of each meeting in order to better meet patients' needs.

Many respondents indicated that they did not know of any local SSc support groups, that they were unsure of the types of activities that occur during SSc support group meetings, or that they did not believe that they needed additional support. SSc patient organizations may be able to increase awareness of existing groups through advertisements at annual conferences, in patient newsletters, or on the websites of SSc patient organizations. If facilitator training and accreditation is made available, this may also increase the ability to disseminate information about the groups through healthcare providers, who are sometimes reluctant to recommend support groups to patients because of uncertainties about the utility of these groups and the

qualifications of the peer facilitators. It may also be the case that patients who have strong support from family or friends may also benefit from interacting with other people who have the disease [15, 21], both in gaining and providing support, and effective dissemination of knowledge about support groups could address this.

There are a number of limitations that should be considered when interpreting the results of this study. First, recruitment occurred through national and provincial SSc organizations, SSc-related chat rooms, and through emails to support group facilitators and members across Canada and the United States, which may have influenced the characteristics of respondents. The majority of survey dissemination and all of the collected responses were done electronically, which may have also influenced the representativeness of the sample. However, the demographic characteristics of respondents were similar to participants in other large SSc patient cohorts [39]. Second, given the self-report nature of the survey, there is no way to be certain that all respondents were accurately diagnosed with SSc. Third, we combined responses from both Canadian and US respondents, and their accessibility to support groups may differ. Fourth, our sample included only small numbers of men, young patients, and non-White patients, so results could not be analyzed separately for subgroups of patients. Nonetheless, the study provides important information that can be used to inform the development of a training program for peer facilitators of SSc support groups and to improve the accessibility to and effectiveness of these groups.

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

and improve their ability to meet members' needs on a sustained basis.

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Table 1. Sociodemographic Characteristics (N = 242)

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

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

Table 2: Exploratory Factor Analysis: Factor Loadings

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

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

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

*Statistically significant, $p < 0.05$.

Table 3. Items and Frequencies (N = 242)

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

17: I feel too depressed or emotionally overwhelmed to attend a support group.	164 (67.8%)	45 (18.6%)	25 (10.3%)	8 (3.3%)	0.49 (0.81)
20: I am uncomfortable sharing my experiences with a group.	163 (67.4%)	52 (21.5%)	16 (6.6%)	11 (4.5%)	0.48 (0.81)
Factor 2: Practical Reasons					
1: I do not know of any scleroderma support groups offered in my area.	76 (31.4%)	49 (20.3%)	47 (19.4%)	70 (28.9%)	1.46 (1.21)
7: My scleroderma symptoms are severe and make it difficult to attend the meetings.	137 (56.6%)	53 (21.9%)	35 (14.5%)	17 (7.0%)	0.72 (0.96)
11: The time of the meetings does not fit in my schedule.	136 (56.2%)	27 (11.2%)	43 (17.8%)	36 (14.9%)	0.91 (1.15)
12: I do not have a reliable way to get to the meetings.	180 (74.4%)	19 (7.9%)	19 (7.9%)	24 (9.9%)	0.53 (1.00)
18: I am worried that my privacy will not be respected.	179 (74.0%)	35 (14.5%)	19 (7.9%)	9 (3.7%)	0.41 (0.79)
21: Getting to and from the meetings is inconvenient due to weather, distance, or other factors.	127 (52.5%)	39 (16.1%)	31 (12.8%)	45 (18.6%)	0.98 (1.18)

28: I am uncomfortable with how I look.	192 (79.3%)	29 (12.0%)	17 (7.0%)	4 (1.7%)	0.31 (0.68)
Factor 3: Beliefs about Support Groups					
5: I think support groups are too negative.	132 (54.5%)	51 (21.1%)	27 (11.2%)	32 (13.2%)	0.83 (1.08)
9: I do not think support groups are helpful.	129 (53.3%)	52 (21.5%)	37 (15.3%)	24 (9.9%)	0.82 (1.03)
16: I do not think I would learn more about scleroderma than I already know now.	130 (53.7%)	50 (20.7%)	32 (13.2%)	30 (12.4%)	0.84 (1.07)
23: I do not think support groups provide educational information that is current and relevant.	163 (67.4%)	35 (14.5%)	29 (12.0%)	15 (6.2%)	0.57 (0.93)
24: I think support groups spend too much time discussing non-scleroderma related topics.	152 (62.8%)	45 (18.6%)	36 (14.9%)	9 (3.7%)	0.60 (0.88)
Additional Items:					
8: I do not think the group would have enough people of a similar cultural background in it.	187 (77.3%)	30 (12.4%)	14 (5.8%)	11 (4.5%)	0.38 (0.79)

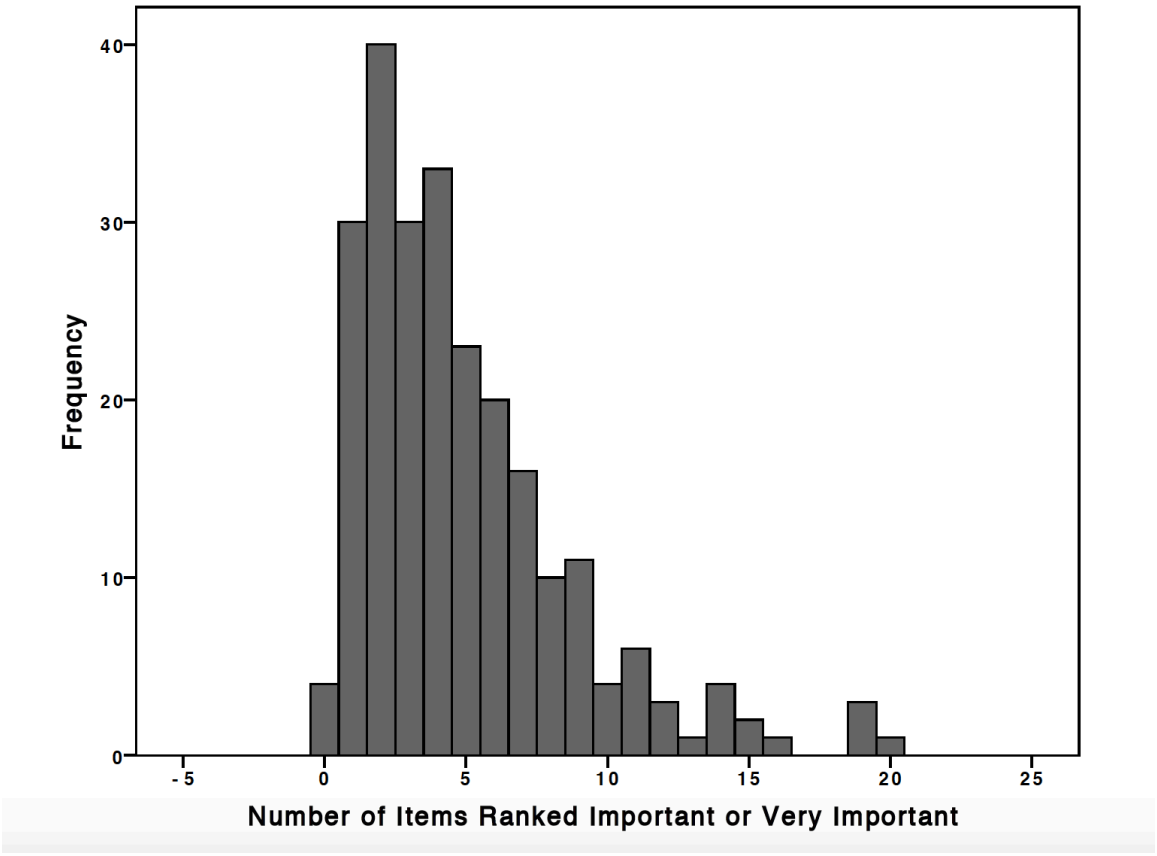
15: I do not think the group would have enough people of a similar age to my age.	176 (72.7%)	41 (16.9%)	16 (6.6%)	9 (3.7%)	0.41 (0.78)
19: I do not think the group would have enough people of the same gender as me in it.	206 (85.1%)	24 (9.9%)	8 (3.3%)	4 (1.7%)	0.22 (0.58)
22: I do not have available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

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

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

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

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



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

DEMOGRAPHICS

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

1. Please indicate your gender: £ Male £ Female
2. Please indicate your age in years: .....
3. In which country do you live?
   - o Canada
   - o United States
   - o Other……
4. In which Canadian province or territory do you live?

o AB	o NS	o QC
o BC	o NT	o SK
o MB	o NU	o YT
o NB	o ON	
o NL	o PE	

5. In which American state do you live?

o AL	o IN	o NE
o AK	o IA	o NV
o AZ	o KS	o NH
o AR	o KY	o NJ
o CA	o LA	o NM
o CO	o ME	o NY
o CT	o MD	o NC
o DE	o MA	o ND
o FL	o MI	o OH
o GA	o MN	o OK
o HI	o MS	o OR
o ID	o MO	o PA
o IL	o MT	o RI

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

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

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

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

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

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

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

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

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

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

Scleroderma Support Group Non-Attenders Survey

Reasons for Not Attending Support Groups

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

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

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

4.2 Connecting Text

Despite support groups playing an important role in the lives of many rare disease patients, including those with SSc, many patients remain unable to or actively choose to not attend these groups. Our findings suggest that a combination of personal reasons, practical reasons, and existing beliefs about support groups, influence SSc support group attendance patterns among patients. These results are important as they provide the information necessary for national SSc organizations, such as the Scleroderma Society of Canada and the Scleroderma Foundation of the US, to develop a framework capable of addressing current limitations in the accessibility to and effectiveness of SSc support groups. The findings generated from this study are unique and will be used to improve the ability of support groups to meet members' needs on a sustained basis.

CHAPTER 5: General Discussion

5.1 Summary of Main Findings

The first study within this thesis employed qualitative methods in the form of focus group interviews to gain a greater understanding of the experiences of living with SSc directly from the patient perspective. The objective of this study was to improve knowledge about the sources of emotional distress experienced by patients with SSc, and to learn more about the potential impact these areas of distress may have on daily living and quality of life. Findings from this study indicated that patients with SSc experience emotional distress from a range of different sources, and that this distress begins prior to receiving a diagnosis and continues throughout the duration of their lives. Six core themes representing sources of emotional distress were identified, including: (1) facing a new reality; (2) the daily struggle of living with scleroderma; (3) handling work, employment and general financial burden; (4) changing family roles; (5) social interactions; and (6) navigating the health care system. Of these six themes, three (facing a new reality; the daily struggle of living with SSc; social interactions) have been previously reported in other studies assessing sources of distress among patients with SSc [12, 37-40]; however, the remaining three identified themes are novel and allowed us to gain a more thorough understanding of what living with SSc might be like for patients. Taken together, these themes provide insight into the unique emotional experiences and challenges faced by people living with SSc.

The second study assessed the reasons why patients with SSc, who typically do not have access to support resources, do not or are unable to attend SSc support groups. Findings from the second study revealed that a combination of personal reasons, practical reasons, and existing beliefs about support groups play a role in influencing patients decision to not attend these

groups. The two most commonly endorsed reasons for not attending SSc support groups included: (1) already having enough support from family, friends, or others; and (2) not knowing of any local SSc support groups. Additional reasons included being uncomfortable seeing other people with SSc in worse condition; being too busy with other responsibilities; the time of the meetings not fitting in one's schedule; believing that support groups are too negative or not helpful; and having problems getting to and from the meetings due to weather, distance, or other factors. Some of these reasons were previously identified in another study that explored reasons for not participating in SSc support groups through open-ended responses to a single question [41]. These included not needing additional support; having other demands or being too busy; not being aware of support groups generally and locally, and being unable to attend due to symptoms being too severe, among others.

5.2 Implications of Findings and Directions for Future Research

The findings from both studies provide researchers, patients, and national SSc organizations, including the Scleroderma Society of Canada and the Scleroderma Foundation in the US, with valuable information that can be used to improve access to and the effectiveness of support services designed to meet the needs of SSc patients. One such organization, the Scleroderma Patient-centered Intervention Network (SPIN), an international collaboration of SSc patient organizations, clinicians, and researchers, will use the findings from the first study to inform the development of an online intervention specifically aimed at alleviating sources of distress among patients with SSc [20]. Additionally, despite the known heterogeneity of the disease, findings from the first study revealed that individual SSc patients experience many of the same sources of distress as one another. As a result, peer-led support groups specifically for people with SSc may be another method of providing support and practical health information

about the disease to SSc patients who do not typically have access to professionally organized support resources.

The second manuscript included in this thesis systematically assessed reasons why people with SSc may choose to or are unable to attend SSc support groups. Currently, there are approximately 30 active SSc support groups in Canada and approximately 150 in the US [35, 36]. All of these groups are locally organized and peer-led. In Canada, the Scleroderma Society of Canada helps people with SSc to find existing support groups but does not organize the support groups, provide formal training or support to peer facilitators, or offer any information on how to structure and initiate a support group. In the US, the Scleroderma Foundation loosely coordinates a network of locally organized support groups and provides some information to support group facilitators, but it does not offer guidance on how to build a support group starting from the grassroots level.

Findings from this second study suggest a number of different ways in which SSc patient organizations may be able to address the current limitations in the accessibility to and effectiveness of SSc support groups. For example, given that many SSc patients do not have access to support groups due to geographical distance or physical disability, implementing online support groups may be an economical and feasible option for delivering support to those with SSc. For many common medical illnesses, such as cancer, online groups have become increasingly popular [42, 43]. Data are not available for Canadian SSc patients, but a recent study found that 85% of Dutch SSc patients use the internet for disease-related purposes [44].

Another possible way to increase the availability and utility of SSc support groups is to provide training for the peer facilitators of these groups. This would provide SSc patients with skills to successfully establish and manage support groups where none exist. Moreover, many

patients in this study indicated they do not participate in support groups because they are afraid to interact or see others with SSc or because they have negative perceptions about these groups. Trained peer facilitators could address these concerns more effectively and, thus, improve the ability of existing groups to meet patients' needs. Finally, a number of participants in this study reported that they were not aware of support groups; it may be possible to improve awareness of existing groups through advertisements at annual conferences, in patient newsletters, or on the websites of SSc patient organizations.

5.3 Limitations

There are several limitations applicable to the current research that need to be considered. First, participants in both of the studies included in this thesis constitute a convenience sample of SSc patients. Specifically, recruitment occurred primarily through national and provincial patient organizations, which may have influenced the characteristics of respondents; therefore limiting the findings of this study to be applicable to all patients with SSc. Additionally, only patients in Canada were recruited to participate in the focus group interviews, and patients from both Canada and the US were recruited to participate in the survey. Although these studies allow us to gain a general understanding of the sources of distress experienced and attitudes towards not attending SSc support groups among patients living in Canada or the US, they do not provide us with knowledge of the experiences of SSc patients living outside of these countries. Additionally, findings generated from the in-person focus group interviews used within the first study may not be representative of all SSc patients as the participants who were able to attend the interviews may be in better health than those who could not. As a result, the sources and extent of emotional distress experienced by these participants may not be entirely representative of the emotionally distressing experiences faced by all people with SSc. Furthermore, a small number

of men were included in the focus group interviews, which may have limited the ability to extract information about sources of distress specific to males living with SSc. Finally, because of the focus group interview format, some of the male participants may have felt outnumbered and not fully comfortable disclosing their experiences with emotional distress.

In regards to the second study, both the self-report nature of the survey and the electronic collection of responses may have influenced the representativeness of the sample. Furthermore, this method of data collection did not allow the researchers to confirm that all participants were, in fact, diagnosed with SSc. Lastly, specific analyses for patient subgroups could not be conducted, as our sample included small numbers of men, younger patients, and non-White patients.

5.4 Conclusion

SSc is a rare, incurable, and unpredictable chronic illness that has marked physical and psychological consequences that impact quality of life and daily living. In addition to the many physiological symptoms that accompany the disease, it is not uncommon for SSc patients to experience significant disease-related emotional distress. However, research focused on gaining a greater understanding of the sources of emotional distress and the impact it may have on daily living for patients with SSc is limited. Additionally, due to the rarity of the condition, it is common for patients with SSc to have limited access to support resources aimed at helping them cope with and ameliorate their experienced symptoms. Furthermore, treatments for SSc generally focus on the management of medical symptoms (e.g., lung disease) and on improvement of physical functioning. As a result, many patients with SSc choose to utilize peer-led SSc support groups as a way of receiving emotional and practical support from other patients facing similar

disease related challenges. However, previous research has not considered reasons why some patients with SSc choose to not attend or are unable to attend SSc support groups.

The present thesis conducted two studies with the purpose of filling a much needed knowledge gap surrounding sources of emotional distress for patients with SSc and on attitudes towards SSc support group participation. Both studies provide an increased understanding of the challenges faced by SSc patients and allow for the modification of support services to better address these distressing disease-related symptoms. The first study found that patients with SSc experience sources of distress from a variety of different areas and that this distress begins prior to receiving a diagnosis and continues throughout the rest of their unpredictable lives. Findings from the second study revealed that a combination of personal reasons, practical reasons, and beliefs about support groups influence patients with SSc to not attend or make them unable to attend SSc support groups.

These findings provide researchers and national SSc organizations with the information necessary to develop and tailor support resources and tools capable of minimizing the sources of emotional distress experienced by patients. Additionally, the Scleroderma Society of Canada and the Scleroderma Foundation in the US can use the knowledge obtained from these studies to develop an infrastructure capable of improving current SSc support group conditions by increasing accessibility to and the effectiveness of these groups at meeting the needs of patients.

5.5 References

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