Support Groups in a Rare Disease Context: Recommendations for Training Scleroderma Support Group Facilitators and Exploration of Factors Affecting Participation in Scleroderma Support Groups

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Abstract

The growing number of Canadians with one or more chronic illnesses, combined with increasingly limited healthcare resources, has made it difficult for healthcare providers and patient organizations to provide the support that people with chronic diseases need in order to deal with their conditions. As such, many patients turn to support groups to help them cope with and manage their disease. In common diseases, support groups are often facilitated by professionals, but in rare diseases, where barriers to providing support are even more pronounced, support groups are typically led by other patients. Scleroderma (or systemic sclerosis, SSc) is an example of a rare disease where peer facilitated support groups play an important role for people living with the disease. The Scleroderma Society of Canada and the Scleroderma Foundation in the United States are committed to increasing the accessibility and effectiveness of SSc support groups. To do this, they have partnered with our research team to develop an educational training program for peer facilitators of SSc support groups. In order to inform the development of this program four independent studies were conducted and included in this thesis. Study 1 was a systematic review examining the existence of training programs for peer facilitators of illness-based support groups and the effect of such programs on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with the support group experience among group members. The findings of the review highlighted the need for training programs for peer facilitators of illness-based support groups and for well-designed and executed trials that evaluate whether these programs improve outcomes among group facilitators and members. Study 2 was a scoping review exploring the 1) benefits of participating in rare disease support groups and 2) facilitators and barriers of initiating and sustaining these groups. The findings of the review suggested that support groups are an important resource for people with rare diseases and that training programs

for peer facilitators of rare disease support groups, such as SSc support groups, could be useful in addressing challenges regarding establishing and maintaining these groups. Studies 3 and 4 employed self-report measures to identify 1) the training and support needs of SSc support group facilitators and 2) the reasons people with SSc do not participate in SSc support groups. These findings underscored the importance of an educational training program for peer facilitators of SSc support group and highlighted areas of focus for this program. Implications and future directions for research are discussed.

Résumé

Il est de plus en plus difficile pour les professionnels des soins de la santé ainsi qu'aux organismes de patients, de fournir les soins requis aux gens atteints de maladies chroniques principalement à cause du nombre croissant de canadiens ayant une ou plusieurs de ces maladies, et des ressources de plus en plus limitées dédiées aux soins de la santé. Par conséquent, plusieurs patients se tournent vers des groupes de soutien pour les aider à affronter et gérer leur maladie. Dans les cas de maladies plus communes, les groupes de soutien des patients sont souvent gérés par des professionnels alors que dans le cas de maladie plus rares, où les obstacles pour fournir de tels services sont plus importants, les groupes de soutien sont typiquement gérés par d'autres patients. La sclérodermie (sclérose systémique, ScS) est un exemple de maladie rare où un groupe de soutien guidé par un pair joue un rôle important pour les gens vivant avec cette maladie. La Société de sclérodermie du Canada et la fondation américaine de sclérodermie se sont engagés à augmenter l'accessibilité et l'efficacité des groupes de soutien ScS. Pour ce faire, ils ont fait un partenariat avec notre équipe de recherche pour développer un programme de formation destiné aux leaders de ces groupes de soutien ScS. Dans le but de supporter le développement de ce programme, quatre études indépendantes ont été réalisées et sont incluses dans cette thèse. L'étude 1 est une revue systématique portant sur l'examen des programmes de formation existants pour les leaders des groupes de soutien et les impacts de tels programmes sur 1) la compétence et l'efficacité des leaders 2) l'efficacité de la gestion de la maladie, les résultats sur la santé et la satisfaction des membres de ces groupes de soutien. Les conclusions de cette première étude ont mis en lumière le besoin de programmes de formation pour les leaders des groupes de soutien, ainsi que la conception minutieuse et l'exécution de tests afin d'évaluer si ces programmes améliorent la performance des leaders et le bien-être des membres. L'étude 2 est une revue de la portée visant 1) les bénéfices reliés à la participation à des groupes de soutien aux

maladies rares, et 2) les facilitateurs et les obstacles reliés à la création et au maintient de ces groupes. Les conclusions de cette deuxième étude indiquent que les groupes de support sont une ressource importante pour les gens avec des maladies rares et que les programmes de formation des leaders des groupes de soutien au maladies rares, tel que le groupe de soutien ScS, pourrait être utile pour confronter les défis reliés à la mise en place et au soutien de ces groupes. Les études 3 et 4 ont employés des méthodes d'auto-évaluation afin d'identifier 1) la formation et les besoins de support des leaders des groupes de soutien ScS et 2) les raisons pour lesquelles les gens avec ScS ne participent pas à des groupes de soutien. Les résultats des ces études soulignent l'importance des programmes de formation pour les leaders des groupes de soutien ScS et mets en lumière les principaux domaines ciblés par ces programmes de formation. Les implications ainsi que des directions futures pour la recherche sont aussi discutées.

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Contribution of Authors

The four manuscripts included in the present thesis are original research. I am the first author on all four papers, as I contributed to the conception and design of each paper; conducted the acquisition, analysis, and interpretation of data; completed the original draft of each paper; incorporated critical feedback from co-authors and reviewers; and prepared and submitted the final draft of each paper. The first manuscript is co-authored by Stephanie Gumuchian, Dr. Lorie Kloda, Jill Boruff, Dr. Ghassan El-Baalbaki, Dr. Annett Körner, Dr. Vanessa Malcarne, Dr. Brett Thombs, and The Scleroderma Support Group Project Advisory Team. Dr. Thombs contributed to the conception and design of the paper; the analysis and interpretation of data; supporting me in drafting and revising the paper; and final approval of the published version. Ms. Gumuchian contributed to the acquisition, analysis, and interpretation of data; revising the paper; and final approval of the published version. Dr. Kloda and Ms. Boruff contributed to the acquisition of data; revising the paper; and final approval of the published version. Dr. El-Baalbaki, Dr. Körner, Dr. Malcarne, and the Scleroderma Support Group Project Advisory Team contributed to the interpretation of data; revising the paper; and final approval of the published version.

The second manuscript is co-authored by Stephanie Gumuchian, Daniel Rice, Alexander Levis, Dr. Lorie Kloda, Dr. Annett Körner, and Dr. Brett Thombs. Dr. Thombs contributed to the conception and design of the paper; the analysis and interpretation of data; supporting me in drafting and revising the paper; and final approval of the published version. Ms. Gumuchian and Ms. Rice contributed to the acquisition, analysis, and interpretation of data; revising the paper; and final approval of the published version. Dr. Kloda and Mr. Levis contributed to the acquisition of data; revising the paper; and final approval of the published version. Dr. Körner contributed to the interpretation of data; revising the paper; and final approval of the published version.

The third manuscript is co-authored by Stephanie Gumuchian, Dr. Sandra Peláez, Dr. Vanessa Malcarne, Dr. Ghassan El-Baalbaki, Dr. Annett Körner, Dr. Marie Hudson, Dr. Murray Baron, Dr. Brett Thombs, and the Scleroderma Support Group Project Advisory Team. Dr. Thombs contributed to the conception and design of the paper; the analysis and interpretation of data; supporting me in drafting and revising the paper; and final approval of the published version. Ms. Gumuchian contributed to the acquisition, analysis, and interpretation of data; revising the paper; and final approval of the published version. Dr. Hudson and Dr. Baron contributed to the acquisition of data; revising the paper; and final approval of the published version. Dr. Peláez, Dr. Malcarne, Dr. El-Baalbaki, Dr. Körner, and the Scleroderma Support Group Project Advisory Team contributed to the interpretation of data; revising the paper; and final approval of the published version.

The fourth manuscript was co-authored by Stephanie Gumuchian, Dr. Ghassan El-Baalbaki, Dr. Annett Körner, Dr. Vanessa Malcarne, Dr. Sandra Peláez, Marie-Eve Carrier, Mia Pépin, Dr. Brett Thombs, and the Scleroderma Support Group Project Advisory Team. Dr. Thombs contributed to the conception and design of the paper; the analysis and interpretation of data; supporting me in drafting and revising the paper; and final approval of the published version. Ms. Gumuchian contributed to the acquisition, analysis, and interpretation of data; revising the paper; and final approval of the published version. Ms. Carrier and Ms. Pépin contributed to the acquisition of data; revising the paper; and final approval of the published version. Dr. El-Baalbaki, Dr. Körner, Dr. Malcarne, Dr. Peláez, and the Scleroderma Support Group Project Advisory Team contributed to the interpretation of data; revising the paper; and final approval of the published version.

Chapter 1

Introduction

Literature Review

Rare diseases are defined as conditions that affect fewer than 1 person in 2,000 (Orphanet, 2012). Approximately 7,000 rare diseases have been identified worldwide and roughly 1 in 12 Canadians have a rare disease (Canadian Organization for Rare Disorders, n.d.; European Organisation for Rare Diseases, 2005, 2009; Orphanet, 2012). People living with rare diseases experience many of the same challenges as people with more common medical diseases, but also face substantial additional challenges due to gaps in knowledge about their disease and how to treat it (European Organisation for Rare Diseases, 2005, 2009).

Many people with rare diseases have difficulty obtaining a correct diagnosis and for some people the process of receiving a diagnosis can take several years (European Organisation for Rare Diseases, 2005, 2009). Because little is known about many rare diseases, some patients need to consult multiple doctors and endure repeated medical tests before receiving a diagnosis (European Organisation for Rare Diseases, 2005, 2009). Furthermore, once diagnosed, many rare disease patients have difficulty obtaining proper treatment (European Organisation for Rare Diseases, 2005, 2009). Not only are clinical research and practice guidelines often limited or non-existent, but treatment and management services are typically scarce, and many patients have to travel long distances or wait long periods of time for care (European Organisation for Rare Diseases, 2005, 2009).

The financial burden of living with a rare disease is yet another challenge for people with rare diseases (European Organisation for Rare Diseases, 2005, 2009). Often, rare disease treatments are expensive, and patients or their caregivers may be forced to stop working (European Organisation for Rare Diseases, 2005, 2009). Other challenges rare disease patients

face include isolation and stigmatization (European Organisation for Rare Diseases, 2005, 2009). Many rare diseases are associated with changes in physical appearance that are uncommon or unfamiliar to the public, and patients may experience unwanted attention or rejection when interacting with other people (European Organisation for Rare Diseases, 2005, 2009).

In order to cope with the emotional and practical challenges of living with a burdensome medical condition, even a common, well-understood condition, many people join support groups (Davison, Pennebaker, & Dickerson, 2000; Newman, Steed, & Mulligan, 2004). Support groups are based on the principal that people who face similar disease-related issues can empower one another through social contact and support (Aymé, Kole, & Groft, 2008). Support groups can be configured in a variety of ways; they may be held face-to-face, online or via teleconference, they may be led by professionals or peers, and they may have a structured or unstructured format (Aymé et al., 2008; Barg & Gullatte, 2001; Davison et al., 2000; Kwakkenbos et al., 2013; Reimann, Bend, & Dembski, 2007). Activities of these groups typically involve an educational or information-sharing component, as well as the giving and receiving of emotional and practical support (Aymé et al., 2008; Barg & Gullatte, 2001; Davison et al., 2000; Kwakkenbos et al., 2013; Reimann et al., 2007).

In the case of many common medical diseases, support groups are offered by the healthcare system and are organized and delivered by professionals who are knowledgeable about the condition (Davison et al., 2000; Newman et al., 2004). In rare diseases, however, professionally led support groups are typically not available or readily accessible (Kwakkenbos et al., 2013). As such, peer-facilitated support groups often emerge through grassroots efforts (Kwakkenbos et al., 2013; Reimann et al., 2007).

About 16,000 Canadians are affected by scleroderma (or systemic sclerosis, SSc) (Bernatsky et al., 2009), which is a rare, chronic, multi-system connective tissue disorder

characterized by abnormal fibrotic processes and excessive collagen production that manifests in thickening of the skin and damage to the internal organs, including the heart, lungs, and gastrointestinal tract (Mayes, 2008; Seibold, 2004). SSc normally occurs between the ages of 30 and 50 years, and approximately 80% of people with the disease are women (Mayes, 2008; The Arthritis Society, n.d.). There is no cure for SSc, and the median survival time from diagnosis is approximately 11 years, with patients 3.7 times more likely to die within 10 years of diagnosis (44.9% mortality) than age, race, and sex-matched persons without the disease (12.0% mortality) (Mayes et al., 2003).

SSc is an example of a rare disease where peer-facilitated support groups play an important role for people with the disease (Kwakkenbos et al., 2015). Currently, there are approximately 200 SSc support groups across Canada and the United States and most of them are facilitated by patients (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.). Previous research has found that support group facilitators of illness-based support groups report a number of challenges in fulfilling their role, including balancing individual and group needs; dealing with complex group dynamics; managing the deteriorating health or death of group members; and a lack of resources and of support from healthcare professionals (Butow et al., 2005; Zordan et al., 2010). These challenges are magnified for facilitators of rare disease support groups, including SSc support groups, who also face logistical problems related to small numbers of potential group members, even in urban settings, and limited support from healthcare and patient organizations, which are not as well-resourced as organizations for people with more common diseases (Kwakkenbos et al., 2013).

The Scleroderma Society of Canada and the Scleroderma Foundation in the United States are committed to enhancing access to SSc support groups and improving the experience of SSc support group facilitators and the ability of SSc support groups to meet the needs of patients. In

order to accomplish this, these organizations have partnered with our research team to develop an educational training program for peer facilitators of SSc support groups. To do this, however, more information is needed regarding the effectiveness of such programs, the training and support needs of facilitators of SSc support groups, and the reasons people with SSc attend or do not attend SSc support groups.

Research Objectives

The aim of the present research is to inform the development of the Scleroderma Support group Leader EDucation (SSLED) Program, an educational training program for peer facilitators of SSc support groups. As such, four independent studies were conducted and included in this thesis. First, a systematic review was conducted to ascertain if training programs for peer facilitators of illness-based support groups were already in existence, as well as to examine the effect of these programs on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with the support group experience among group members. Then, a scoping review was conducted to systematically identify and map evidence on the 1) benefits of participating in rare disease support groups and 2) facilitators and barriers of initiating and sustaining these groups. Finally, two additional studies were performed; the respective objectives of these studies were to 1) identify the training and support needs of SSc support group facilitators and 2) explore the reasons people with SSc do not participate in SSc support groups.

Chapter 2

Manuscript 1 (Paper Published in BMJ Open)

The Effect of Support Group Peer-Facilitator Training Programs on Peer-Facilitator and Support Group Member Outcomes: A Systematic Review

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Abstract

Peer facilitators play an important role in determining the success of many support groups for patients with medical illnesses. However, many facilitators do not receive training for their role and report a number of challenges in fulfilling their responsibilities. The objective of this systematic review was to evaluate the effects of training and support programs for peer facilitators of support groups for people with medical illnesses on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with support groups among group members. Searches included the CENTRAL, CINAHL, EMBASE, MEDLINE, PsycINFO, and Web of Science databases from inception through April 8, 2016; reference list reviews; citation tracking of included articles; and trial registry reviews. Eligible studies were RCTs in any language that evaluated the effects of training programs for peer facilitators compared to no training or alternative training formats on 1) competency or self-efficacy of peer facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with groups of group members. The Cochrane Risk of Bias tool was used to assess risk of bias. There were 9,757 unique titles/abstracts and two full-text publications reviewed. One RCT met inclusion criteria. The study evaluated the confidence and self-efficacy of cancer support group facilitators randomized to 4-months access to a website and discussion forum (N=23; low-resource) versus website, discussion forum, and 2-day training workshop (N=29). There were no significant differences in facilitator confidence (Hedges' g=0.16, 95% confidence interval -0.39 to 0.71) or self-efficacy (Hedges' g=0.31, 95% confidence interval -0.24 to 0.86). Risk of bias was unclear or high for 4 of 6 domains. Well-designed and conducted, adequately powered trials of peer support group facilitator training programs for patients with medical illnesses are needed. Systematic review registration: CRD42014013601.

Keywords: medical illness; peer facilitator; support group; systematic review; training program

The Effect of Support Group Peer-Facilitator Training Programs on Peer-Facilitator and Support Group Member Outcomes: A Systematic Review

Illness-based support groups bring together people who face similar disease-related challenges to give and receive emotional support and exchange disease-related information, sometimes in the form of structured educational activities (Barg & Gullatte, 2001; Davison, Pennebaker, & Dickerson, 2000; Lieberman et al., 2005). These groups can be configured in a variety of ways. They may be held face-to-face, online or via teleconference, they may be led by professionals or peers, and they may have a structured or unstructured format (Barg & Gullatte, 2001; Davison et al., 2000; Lieberman et al., 2005).

Many people with chronic medical illnesses join support groups in order to better cope with the emotional and practical challenges of their disease (Barg & Gullatte, 2001; Davison et al., 2000; Lieberman et al., 2005). A number of studies have assessed the benefits of participating in support groups for common medical diseases, including cancer (Docherty, 2004; Ussher, Kirsten, Butow, & Sandoval, 2006; Yaskowich & Stam, 2003). Participants in these studies have reported a number of benefits, such as obtaining emotional support, receiving information about their disease and treatments, and learning how other patients have coped with the condition. They have also reported that support groups help decrease isolation, foster a sense of community, and instil hope about the future.

In the case of many common medical diseases, support groups are offered by the healthcare system and are organized and delivered by professionals who are knowledgeable about the condition. Peer-led support groups, however, are a less resource-intensive alternative that could potentially reach more patients, with the added benefit of having group leaders who may share important experiences with group members (Gray & Fitch, 2001). In some settings, such as in rare diseases, professionally led support groups are typically not available or readily

accessible, and peer-led support groups often emerge through grassroots efforts (Kwakkenbos et al., 2013; Reimann, Bend, & Dembski, 2007).

Support group facilitators play an important role in determining the success of a group (Butow et al., 2005; Zordan et al., 2010). Many support group facilitators, however, do not receive training for their role and report a number of challenges in fulfilling their responsibilities (Butow et al., 2005; Zordan et al., 2010). These challenges may include a lack of training and other resources; a lack of support from health care professionals; difficulty dealing with problematic group members; difficulty dealing with the worsening health and death of group members; difficulty finding back-up or replacement facilitators, and struggles to sustain the group (Butow et al., 2005; Zordan et al., 2010).

Formal training of peer support group facilitators could potentially address some of these concerns and could improve the experiences of group facilitators and members. No systematic reviews have examined the effects of training and support programs for peer facilitators of support groups for people with medical illness. Thus, the objective of the present systematic review was to evaluate the effect of training and support programs for peer facilitators of support groups for people with medical illnesses on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with the support group experience among group members.

Method

This systematic review was registered in PROSPERO (CRD42014013601), and was conducted and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (Moher, Liberati, Tetzlaff, Altman, & PRISMA Group, 2010).

Search Strategy

The CENTRAL, CINAHL, EMBASE, MEDLINE, PsycINFO, and Web of Science databases were initially searched from inception on September 8, 2014 and updated on April 8, 2016. A medical librarian developed the search strategy and performed the search. No restrictions by date or language or publication were used. The complete search strategy can be found in Appendix A. In addition to database searching, we manually searched the reference lists of included publications and tracked their citations using Google Scholar (Bakkalbasi, Bauer, Glover, & Wang, 2006). We also searched multiple trial registries, including ClinicalTrials.gov (clinicaltrials.gov), ISRCTN (www.isrctn.com), and the World Health Organization registry search portal (apps.who.int/trialsearch) from inception to June 10, 2016.

Publication Selection

The results of the search were downloaded into the citation management database RefWorks (RefWorks-COS, Bethesda, MD, USA), and duplicate references were identified and removed. Following this, references were transferred into the systematic review software DistillerSR (Evidence Partners, Ottawa, ON, Canada). Using this software, we then assessed the eligibility of each publication through a two-stage process. First, two investigators independently reviewed the titles and abstracts of publications that were identified through the search strategy. If either investigator deemed an abstract potentially eligible based on the inclusion criteria, then a full-text review was completed. Disagreements after full-text review were resolved by consensus, with a third investigator consulted if necessary.

Randomized controlled trials (RCTs) of training and support programs for peer facilitators of illness-based support groups reported in any language were eligible for inclusion. For the purpose of this study, peer facilitator training and support programs were defined as formal programs designed to provide facilitators or potential facilitators of support groups with

the knowledge to facilitate a support group, the interpersonal and other skills to do this, or ongoing emotional and practical support. Since the responsibilities of support group facilitators vary, training and support programs could include a number of components aimed at developing the skills related to these responsibilities. The programs could also vary in length, as well as in the instructional methods and techniques used. Training or support programs that included both peer and professional facilitators, who facilitated groups as part of their professional responsibilities, were included. Training programs for only professional group facilitators, however, were not eligible for inclusion.

We only included trials of training and support interventions for peer facilitators of support groups for people with medical illness. To be considered a peer-facilitated support group for people with medical illness, the activities of the group had to include the giving and receiving of emotional and practical support. They could also include educational activities, but a group that only followed a structured learning curriculum with a defined beginning and end (e.g., a self-management program) was not considered a support group. A group that was designed to be ongoing (or could be used in that way), but was time-limited for the purposes of a research study, was considered a support group. Activities of a support group could take place in person, online or via teleconference, but had to include ongoing real-time interaction between group members. Groups that provided psychotherapy were not considered a support group. Second, membership criteria of the group had to include having a particular disease (e.g., diabetes) or type of disease (e.g., a rare disease) or being a survivor of a disease (e.g., cancer). Training programs for peer facilitators of groups designed for people with mental health disorders were not eligible for inclusion.

Eligible comparators included no training comparators or alternative training approaches. Eligible RCTs had to assess outcomes that reflected 1) the competency or self-efficacy of group

facilitators or 2) self-efficacy for disease management, health outcomes, or satisfaction with the support group experience among members of the support groups of the facilitators in the trial.

Data Extraction, Analysis, and Synthesis

Two investigators independently extracted data from included RCTs and entered it into a standardized Excel spreadsheet. A complete list of extracted variables is provided in Appendix B. Disagreements were resolved by consensus, with a third investigator consulted if necessary. The same two investigators also independently assessed risk of bias using the Cochrane Risk of Bias tool (Appendix C) (Higgins, Altman, & Sterne, 2011). The Cochrane Risk of Bias tool was developed by the Cochrane Collaboration in order to assess risk of bias among the following domains: 1) Sequence generation, 2) Allocation concealment; 3) Blinding of participants, personnel, and outcome assessors; 4) Incomplete outcome data; 5) Selective outcome reporting; and 6) Other sources of bias. Again, discrepancies were resolved by consensus, and a third investigator was consulted as necessary. Because only one eligible RCT was identified, there was no pooling of results.

Results

The database search yielded 9,757 unique titles and abstracts. Of these, 9,755 were excluded after title and abstract review, leaving two conference abstracts for full-text review (Zordan, Butow, Batterby, et al., 2009; Zordan, Butow, Kirsten, et al., 2009). Both abstracts reported results from the same RCT. Since we were not able to locate a full report for this RCT at the time of our initial database searches, we contacted the authors. They provided us with a full report, which was subsequently published (Zordan et al., 2015). The RCT met inclusion criteria and was included in the systematic review. No additional eligible studies were identified via manual searching of the reference list of the included publication, by searching trial registries, or by citation tracking. See Figure 1 for the PRISMA flowchart.

Description of Included RCT

The included RCT enrolled individuals who were currently leading a support group for adults with cancer and/or caregivers in New South Wales, Australia (Zordan et al., 2015). Support group facilitators were stratified based on gender, geographical location, and type of group (e.g., general cancer, specific cancer) and block randomized to one of two 4-month long interventions: 1) low-resource, which included access to a website and discussion forum only or 2) high-resource, which included access to the website and discussion forum plus face-to-face training. The website was designed to provide facilitators with basic theoretical and practical information on facilitating a support group. The websites in the low- and high-resource interventions were the same, but the discussion forums were separated in order to prevent contamination. The training for facilitators in the high-resource intervention involved a 2-day group workshop that aimed to "improve confidence in the facilitator role, group facilitation skills, knowledge of group dynamics, understanding of boundaries, and awareness of self-care and burnout". The workshop was led by an academic researcher and two experienced cancer support group facilitators. Participants in the workshop also received a DVD and manual with examples of how they might address a number of possibly challenging support group scenarios.

At the beginning and end of the 4-month intervention period, group facilitators in both trial arms completed a modified version of the Group Leader Self-Efficacy Instrument (GLSI) and the Group Leader Challenges Scale (GLCS). The modified GLSI is a 27-item self-report questionnaire that assesses self-efficacy for performing group facilitator skills on a 6-point Likert scale ranging from 1 (*Strongly Disagree*) to 6 (*Strongly Agree*) (Zordan et al., 2015). Higher scores on the GLSI indicate a higher degree of self-efficacy. The GLCS is a 26-item self-report questionnaire that assesses the confidence of group facilitators in their 1) ability to execute tasks beyond those of a typical group facilitator, 2) self-care, and 3) specific issues related to cancer

support groups (Zordan et al., 2015). It also utilizes a 6-point Likert scale ranging from 1 (*Strongly Disagree*) to 6 (*Strongly Agree*), with higher scores indicating a higher degree of confidence. No outcomes were collected for members of support groups facilitated by participants in the trial.

Altogether, 30 group facilitators were randomized to the low-resource intervention and 35 to the high-resource intervention. Of these, 23 facilitators in the low-resource arm and 29 in the high-resource arm completed post-intervention questionnaires and were included in analyses. Facilitators randomized to the two trial arms included people affected by cancer, as well as healthcare professionals who may or may not have been affected by cancer. Results for facilitators affected by cancer and not affected by cancer were not reported separately. As shown in Table 1, there were no statistically significant differences between patients randomized to the low-resource and high-resource arms, although scores were higher for the high-resource arm for self-efficacy (Hedges' g = 0.31, 95% confidence interval [CI] = -0.24 to 0.86) and confidence (Hedges' g = 0.16, 95% CI = -0.39 to 0.71). A comparison of participants in the high-resource and low-resource groups who provided data pre-intervention (high-resource N = 33; low-resource N = 29) to those who provided data post-intervention (high-resource N = 29; low-resource N = 23) suggested similar and small post-trial minus pre-trial positive differences in confidence in both groups and similar, small negative differences in both groups in self-efficacy.

Risk of Bias

Risk of bias ratings are shown in Table 2. Risk of bias was rated "Unclear" for Sequence generation and Allocation concealment. Risk of bias was rated "High" for Blinding of participants, personnel, and outcome assessors. All other risk of bias domains were rated "Low."

Discussion

The present systematic review did not identify any trials that compared training and support programs for peer facilitators of disease-based support groups to no training comparators. It identified only one RCT that evaluated the effects of alternative training program resources (Zordan et al., 2015). That study evaluated the confidence and self-efficacy of cancer support group facilitators randomized to 4-month long high-resource (website, discussion forum, 2-day face-to-face training) and low-resource (website, discussion forum) interventions. No statistically significant differences were reported between the two groups for either group facilitator self-efficacy or confidence. Outcomes were not collected for members of the support groups of the facilitators.

There are important methodological considerations, however, that limit the ability to confidently draw conclusions about the potential effects of support group facilitator training programs from the trial. One is that the sample size was very small. A total of 65 patients were randomized, and data from only 52 who completed post-intervention assessments were analyzed. To achieve 80% power for a small (Hedges' g = 0.20) or medium (Hedges' g = 0.50) effect size, 788 and 128 patients, respectively, would have been needed. A second is that the study was rated as having unclear risk of bias related to sequence generation, allocation concealment, and incomplete outcome data and high risk of bias related to blinding of participants, personnel, and outcome assessors.

There are also several issues regarding the design of the intervention and the trial that should be considered when conducting trials on training programs for support group facilitators in the future. First, trial participants included both group facilitators affected by cancer and healthcare professionals who may or may not have been affected by cancer. Peer and professional support group facilitators may have different training and support needs, although research on

this topic is limited (Butow et al., 2005; Price, Butow, & Kirsten, 2006). Future studies should consider the possibility of targeting training to peer or professional group leaders.

Second, the information that was provided in the published trial report and a related article (Zordan et al., 2012) did not include important information that would be necessary to attempt to replicate the training program and trial. For example, the authors described that participants in the training arm of the trial participated in one of three 2-day group workshops, but relatively little information was provided on the conduct of the workshops, when they occurred, or how participants were selected for each of the 3 workshops. The authors reported that workshops incorporated "readings, didactic lectures, the DVD and manual, role-plays and group discussion" (Zorda et al., 2012), but no information was provided on how the sessions were structured, and only limited information was provided on the content. The authors did not indicate at what time during the 4-month long intervention period the 2-day workshops occurred, the length of each workshop day, or whether the days took place sequentially or were spaced apart. Learning and retention is improved when learning is repeated and spaced over time, and it is possible that training sessions spaced over time with opportunities to practice new skills could be more effective than one-shot programs (Carpenter, Cepeda, Rohrer, Kang, & Pashler, 2012).

Finally, the trials compared the confidence and self-efficacy of support group facilitators randomized to a low-resource intervention that included only access to a website and chat forum versus the website and chat forum plus the 2-day workshop. However, as many support group facilitators do not receive any training for their role, it would be useful to know how each of these compares to no training at all.

The growing number of people with one or more chronic medical illnesses, combined with increasingly limited healthcare resources, has made it difficult for healthcare providers and patient organizations to provide the support that people with chronic diseases need in order to

deal with their condition. As such, many patients turn to support groups to help them cope with and manage their disease (Barg & Gullatte, 2001; Davison et al., 2000; Lieberman et al., 2005). Peer-led support groups are becoming increasingly popular as a less resource-intensive alternative that could potentially reach more patients and they could free professional resources for patients with more intense psychosocial needs (Gray & Fitch, 2001).

However, there are some important limitations of many current support groups. Many patients are not able to access groups, particularly in rare diseases, and many peer-led groups are not effectively sustained due to factors such as the health of the facilitator or other shortcomings that could potentially addressed via training (Delisle et al., 2016; Goodridge, Hutchinson, Wilson, & Ross, 2011). Training programs for peer facilitators of support groups could improve the availability of support groups by giving people with chronic diseases the skills they need to set up groups where none exist. In addition, these training programs could increase the effectiveness of support groups by teaching support group facilitators how to organize and structure support groups, as well as how to manage group dynamics and difficult group members. More research is needed, however, to better understand options available for providing training to peer facilitators of support groups and to determine if they achieve their desired effects.

In summary, this systematic review found that there is insufficient evidence on the effectiveness of training and support programs for peer facilitators of disease-based support groups and whether they improve the 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with support group experiences among group members. Well-designed and executed trials that assess whether training and support programs for peer facilitators of support groups improve outcomes among group facilitators and group members are needed. Furthermore, as most peer facilitators are

volunteers, any programs that are implemented will need to make sure to address the needs of peer facilitators while not raising substantial barriers to their participation.

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Appendix A

Complete Search Strategy

CENTRAL

1.Self-Help Groups [MeSH descriptor (this term only)]

2. peer* support* or self help group or therapeutic social club* or peer* facilitat* or peer counseling [ti,ab,kw (word variations have been searched)]

3. 1 or 2

CINAHL (via EBSCOhost)

S1 (MH "Support Groups+")

S2 (MH "Peer Counseling")

S3 TX (therapeutic N1 social club*)

S4 TI ((non medical or non professional* or lay or layperson* or peer* or voluntary or patient) N1 (instructor* or tutor* or educator* or consultant* or leader* or expert* or advisor* or facilitat* or deliver* or mentor* or led or guide* or aide* or run))

S5 AB ((non medical or non professional* or lay or layperson* or peer* or voluntary or patient) N1 (instructor* or tutor* or educator* or consultant* or leader* or expert* or advisor* or facilitat* or deliver* or mentor* or led or guide* or aide* or run))

S6 TX randomized

S7 MH treatment outcomes

S8 PT clinical trial

S9 S6 OR S7 OR S8

S10 S1 OR S2 OR S3 OR S4 OR S5

S11 S9 AND S10

EMBASE

1. Self Help/

2. Group Support/

3. support* group*.tw.

4. group* support*.tw.

5. peer* support*.tw.

6. self help group*.tw.

7. (therapeutic adj social club*).tw.

8. ((non medical or non professional* or lay or layperson* or peer* or voluntary or patient) adj2 (instructor* or tutor* or educator* or consultant* or leader* or expert* or advisor* or facilitat* or deliver* or mentor* or led or guide* or aide* or run)).tw.

9. or/1-8

10. random*.tw.

11. clinical trial*.mp.

12. exp Health Care Quality/

13. exp Treatment Outcome/

14. double blind*.mp.

15. placebo*.tw.

16. blind*.tw.

17. or/10-16

18. exp animals/ not humans.sh.

19. 17 not 18

20. 9 and 19

MEDLINE (via OvidSP)

1. Self-Help Groups/

2. support* group*.tw.

3. group* support*.tw.

4. peer* support*.tw.

5. self help group*.tw.

6. (therapeutic adj social club*).tw.

7. ((non medical or non professional* or lay or layperson* or peer* or voluntary or patient) adj2 (instructor* or tutor* or educator* or consultant* or leader* or expert* or advisor* or facilitat* or deliver* or mentor* or led or guide* or aide* or run)).tw.

8. or/1-7

9. Randomized controlled trial.pt.

10. controlled clinical trial.pt.

11. randomi#ed.ab.

12. placebo.ab.

13. (clinical trials as topic or controlled clinical trials as topic or randomized controlled trials as topic).sh.

14. randomly.ab.

15. trial.ti.

16. or/9-15

17. exp animals/ not humans.sh.

18. 16 not 17

19. 8 and 18

PsycINFO

1. Support Groups/

2. Social Group Work/

3. peer* support*.tw.

4. Peer Counseling/

5. self help group*.tw.

6. (therapeutic adj social club*).tw.

7. ((non medical or non professional* or lay or layperson* or peer* or voluntary or patient) adj2 (instructor* or tutor* or educator* or consultant* or leader* or expert* or advisor* or facilitat* or deliver* or mentor* or led or guide* or aide* or run)).tw.

8. or/1-7

9. double-blind.tw.

10. random* assigned.tw.

11. control.tw.

12. or/9-11

13. exp animals/ not humans.sh.

14. 12 not 13

15. 8 and 14

Web of Science (via ISI Web of Knowledge)

1. TOPIC: (peer facilitat* or lay person facilitat* or layperson facilitat* or patient facilitat* or peer led or lay person led or layperson led)

2. TITLE: ((self help group* or support* group* or peer* support* or therapeutic social club* or social group work or peer counseling))

3. #2 AND #1

Appendix B

Data Extraction Variables

First author's last name

Publication year

Country of study

Funding source

Population

Method of recruitment

Age in years

Gender

Ethnicity/race

Disease duration

Intervention group

N randomized to intervention

Control group

N randomized to control

Length of intervention

Setting of intervention

N analyzed – intervention

N analyzed – control

Outcomes

Appendix C

The Cochrane Tool for Assessing Risk of Bias

Sequence Generation

Describe the method used to generate the allocation sequence in sufficient detail to allow an assessment of whether it should produce comparable groups.

Allocation Concealment

Describe the method used to conceal the allocation sequence in sufficient detail to determine whether intervention allocations could have been foreseen in advance of, or during, enrolment.

Blinding of Participants, Personnel, and Outcome Assessors

Assessments should be made for each main outcome (or class of outcomes). Describe all measures used, if any, to blind study participants and personnel from knowledge of which intervention a participant received. Provide any information relating to whether the intended blinding was effective.

Incomplete Outcome Data

Assessments should be made for each main outcome (or class of outcomes). Describe the completeness of outcome data for each main outcome, including attrition and exclusions from the analysis. State whether attrition and exclusions were reported, the numbers in each intervention group (compared with total randomized participants), reasons for attrition/exclusions where reported, and any re-inclusions in analyses performed by the review authors.

Selective Outcome Reporting

State how the possibility of selective outcome reporting was examined by the review authors, and what was found.

Other Sources of Bias

State any important concerns about bias not addressed in the other domains in the tool. If particular questions/entries were pre-specified in the review's protocol, responses should be provided for each question/entry. Was the study apparently free of other problems that could put it at a high risk of bias?

Table 1

Summary of Randomized Controlled Trial Included in Systematic Review

Author Year Country	Funding Source	Population and Recruitment	Comparison	Number of Participants Randomized	Number of Participants Analyzed	Intervention Duration	Outcomes: Hedges' *g* (95% CI)[a]
Zordan et al 2015 Australia	Australian Research Council	Cancer support group facilitators recruited via email through cancer organizations and using a "snowballing technique"	Low-resource: website and discussion forum only High-resource: website, discussion forum, and 2-day training workshop supported by DVD and manual	Low-resource: 30 High-resource: 35	Low-resource: 23 High-resource: 29	4 months	GLSI: Hedges' $g = 0.31$ 95% CI = -0.24 to 0.86 GLCS: Hedges' $g = 0.16$ 95% CI = -0.39 to 0.71

Note. CI=confidence interval; GLSI=Group Leader Self-Efficacy Instrument; GLCS=Group Leader Challenges Scale.

[a]Positive values reflect greater self-efficacy (GLSI) and greater confidence (GLCS) for group facilitators in the high-resource intervention.

Table 2

Assessment of Risk of Bias of Randomized Controlled Trial Included in Systematic Review

Cochrane Risk of Bias Tool Domains	Review Authors' Judgment	Support for Review Authors' Judgment
Sequence generation	Unclear	Authors reported that leaders were stratified based on gender, geographical location and type of group, and block randomized by a statistician. They did not provide information on how the randomization sequence was generated.
Allocation concealment	Unclear	No information provided on allocation concealment method.
Blinding of participants, personnel, and outcome assessors	High	Blinding of participants and personnel was not possible due to nature of the intervention. Outcomes were self-reported by participants, who were not blinded.
Incomplete outcome data	Unclear	Approximately 20% missing data in both trial arms and not included in final analyses.
Selective outcome reporting	Low	Authors reported small, non-significant effect sizes for included outcomes. In addition, although not a pre-specified outcome of the systematic review, they reported emotional distress, which was higher (non-significant) in the high-resource arm.

Other bias	Low	None.

Note. See Appendix C for domain descriptions. Domains are scored as “high,” “low,” or “uncertain” risk of bias. Risk of bias ratings were based only on published information.

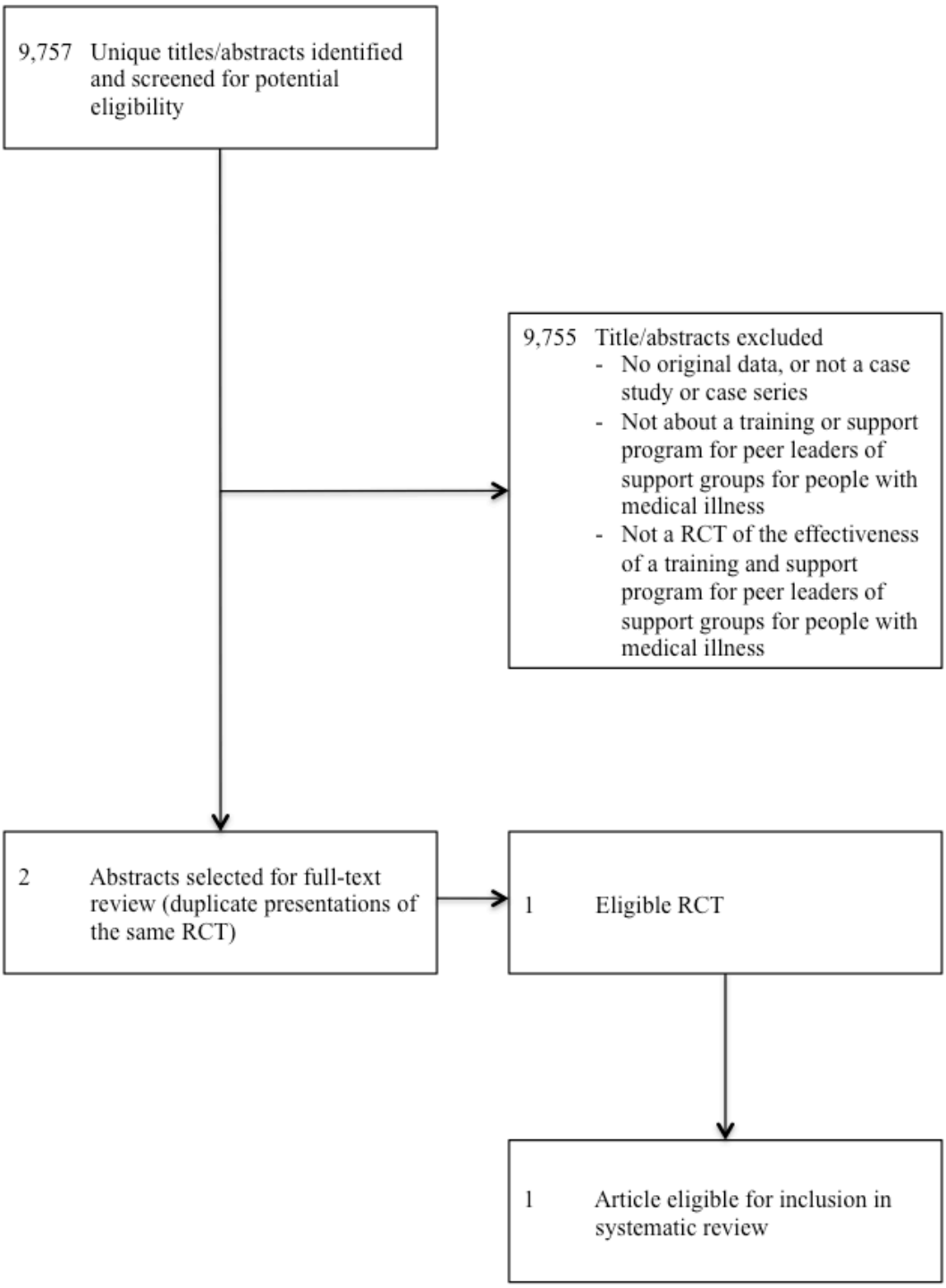

Figure 1. A graphical representation of the flow of publications reviewed in the course of the systematic review.

Linking Manuscripts 1 and 2

Before developing the Scleroderma Support group Leader EDucation (SSLED) Program, an educational training program for peer facilitators of scleroderma (or systemic sclerosis, SSc) support groups, our research team sought to determine if training programs for peer facilitators of other illness-based support groups were already in existence. In addition, we sought to understand the effect of these programs. As such, the first manuscript in the present thesis was a systematic review examining the effect of training programs for peer facilitators of illness-based support groups on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with the support group experience among group members.

While we did not find any randomized controlled trials (RCTs) that compared training programs for peer facilitators of illness-based support groups to no training comparators, we did identify one RCT that evaluated the effects of alternative training program resources (Zordan et al., 2015). That study evaluated the confidence and self-efficacy of facilitators of cancer support groups randomized to 4-month long high-resource (website, discussion forum, 2-day face-to-face training) and low-resource (website, discussion forum) interventions. No statistically significant differences were reported between the two groups for either group facilitator self-efficacy or confidence.

Unfortunately, not enough information regarding the content of the intervention and how it was delivered was provided in that RCT or related publications for our team to attempt to replicate it in the context of SSc. Moreover, the ability to draw conclusions about the effects of training programs for peer facilitators of illness-based support groups from that RCT was limited due to important methodological considerations, including small sample size and unclear or high risk of bias ratings in numerous domains. Altogether, this emphasized the need for the

development of the SSLED Program, as well as for well-designed and executed trials that assess whether training programs for peer facilitators of illness-based support groups improve outcomes among group facilitators and group members. To further inform our work, we then conducted a scoping review exploring the 1) benefits of participating in rare disease support groups and 2) facilitators and barriers of initiating and sustaining these groups.

Chapter 3

Manuscript 2 (Paper Published in The Patient)

Perceived Benefits and Factors that Influence the Ability to Establish and Maintain Patient Support Groups in Rare Diseases: A Scoping Review

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The full reference for Manuscript 2 is as follows:

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Abstract

Support groups are an important resource for many people living with rare diseases. The perceived benefits of participating in support groups for people with rare diseases and factors that may influence the ability to successfully establish and maintain these groups are not well understood. Thus, the objective of this scoping review was to provide a mapping of the available evidence on the 1) benefits or perceived benefits of participating in rare disease support groups and 2) barriers and facilitators of establishing and maintaining these groups. CINAHL and PubMed were searched from January 2000 to August 2015, with no language restrictions. Publications that described the benefits or perceived benefits of participating in rare disease support groups or the barriers and facilitators of establishing and maintaining them were eligible for inclusion. Two investigators independently evaluated titles/abstracts and full-text publications for eligibility, and extracted data from each included publication. Ten publications were included in the scoping review. There was no trial evidence on support group benefits. All 10 publications reported on the perceived benefits of participating in rare disease support groups. Three reported on barriers and facilitators of establishing and maintaining them. Overall, 7 different perceived benefits of participating in rare disease support groups were identified: 1) meeting and befriending other people with the same rare disease and similar experiences; 2) learning about the disease and related treatments; 3) giving and receiving emotional support; 4) having a place to speak openly about the disease and one's feelings; 5) learning coping skills; 6) feeling empowered and hopeful; and 7) advocating to improve healthcare for other rare disease patients. Several facilitators (e.g., meeting via teleconference) and barriers (e.g., getting patients and/or family members to lead the group) of establishing and maintaining these groups were identified. Rare disease support groups are an important source of emotional and practical support for many

patients. There is no trial evidence on the benefits of these groups and limited evidence on the perceived benefits and barriers and facilitators to establishing and maintaining them.

Keywords: barriers; benefits; facilitators; rare diseases; support group; scoping review

Perceived Benefits and Factors that Influence the Ability to Establish and Maintain Patient Support Groups in Rare Diseases: A Scoping Review

Rare diseases are defined as conditions that affect fewer than 1 person in 2,000 (Orphanet, 2012). There are currently approximately 7,000 known rare diseases worldwide, with new diseases being identified each year (Canadian Organization for Rare Disorders, n.d.; European Organisation for Rare Diseases, 2005, 2009; Orphanet, 2012). People living with rare diseases experience many of the same challenges as people with more common medical diseases, including physical and psychological symptoms that require them to modify their family, professional, and social roles (Karasz & Ouellette, 1995; Kralik, 2002). Compared to people with more common diseases, however, those with rare diseases typically face substantial additional challenges due to gaps in knowledge about their disease and how to manage it (European Organisation for Rare Diseases, 2005, 2009).

One major challenge for many rare disease patients is obtaining a correct diagnosis, which can sometimes take several years (European Organisation for Rare Diseases, 2005, 2009). Because little is known about many rare diseases, some patients need to consult multiple doctors and endure repeated medical tests before receiving a diagnosis. Once diagnosed, another major challenge involves obtaining appropriate treatment (European Organisation for Rare Diseases, 2005, 2009). Clinical research and practice guidelines are often limited or non-existent. Treatment and management services are typically scarce, and many patients have to travel long distances or wait long periods of time for care. The financial burden of living with a rare disease is yet another significant challenge for patients (European Organisation for Rare Diseases, 2005, 2009). Often, rare disease treatments are expensive, and patients or their caregivers may be forced to stop working. Other obstacles rare disease patients face include isolation and stigmatization (European Organisation for Rare Diseases, 2005, 2009). Many rare diseases are

associated with changes in physical appearance that are uncommon or unfamiliar to the public, and patients may experience unwanted attention or rejection when interacting with other people.

In order to cope with the challenges of living with a burdensome medical condition, even a common, well-understood condition, many people seek support services (Davison, Pennebaker, & Dickerson, 2000; Newman, Steed, & Mulligan, 2004). In the case of common medical diseases, these services are frequently offered by the healthcare system and are organized and delivered by professionals who are knowledgeable about the condition; in rare diseases, however, they are often not available or readily accessible (Kwakkenbos et al., 2013). In the absence of these services, people with some rare diseases have come together through grassroots efforts to mobilize their own support systems in the form of locally organized support groups (Reimann, Bend, & Dembski, 2007).

The idea that support groups can provide important benefits to people living with a burdensome medical condition is based on the principle that people who face similar disease-related issues can empower one another through social contact and support (Aymé, Kole, & Groft, 2008). Support groups can be configured in a variety of ways; they may be held face-to-face, online or via teleconference, they may be led by professionals or peers, and they may have a structured or unstructured format (Aymé et al., 2008; Barg & Gullatte, 2001; Davison et al., 2000; Kwakkenbos et al., 2013; Reimann et al., 2007). Activities of these groups typically involve an educational or information-sharing component, as well as the giving and receiving of emotional and practical support.

A number of studies have assessed the benefits of participating in support groups for common medical diseases, such as cancer (Docherty, 2004; Ussher, Kirsten, Butow, & Sandoval, 2006; Yaskowich & Stam, 2003). Participants in these studies have reported a number of benefits, such as obtaining emotional support, receiving information about their disease and

treatments, and learning how other patients have coped with the condition. They have also reported that support groups help decrease isolation, foster a sense of community, and instill hope about the future.

Given the challenges faced by people with rare diseases, the perceived benefits of participating in rare disease support groups may differ compared to common medical diseases. Moreover, there are likely challenges that influence the ability to successfully establish and maintain support groups that are unique to a rare disease context. Understanding the benefits or perceived benefits of participating in rare disease support groups and factors that are important for initiating and sustaining these groups is necessary if access to effective support systems is to be increased. There are no studies, however, that have summarized the published academic literature on this topic.

Scleroderma, or systemic sclerosis (SSc), is an example of a rare disease where support groups play an important role for people with the disease (Kwakkenbos et al., 2015). SSc is a chronic multi-system connective tissue disorder characterized by abnormal fibrotic processes and excessive collagen production that manifests in thickening of the skin and damage to the internal organs, including the heart, lungs, and gastrointestinal tract (Mayes, 2008; Seibold, 2004). SSc normally occurs between the ages of 30 and 50 years, and approximately 80% of people with the disease are women (Mayes, 2008; The Arthritis Society, n.d.); there is no cure for SSc (Mayes et al., 2003).

Currently, there are approximately 200 SSc support groups across Canada and the United States, and the majority of these are led by peers (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.). The Scleroderma Society of Canada and the Scleroderma Foundation in the United States help SSc patients locate support groups, but provide almost no information regarding starting a support group or formal training and support to peer facilitators. We are

presently working with these organizations to provide the infrastructure that is required, including a training program for peer facilitators of support groups, in order to enhance access to support groups and the ability of these groups to consistently meet the needs of members. To help inform our work we conducted a scoping review. Specifically, the purpose of our scoping review was to systematically identify and map evidence on the 1) benefits or perceived benefits of participating in support groups for people with rare diseases and 2) facilitators and barriers of establishing and maintaining these groups.

Method

A scoping review is a "form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge" (Colquhoun et al., 2014). A scoping review is rigorous like a systematic review; however, unlike a systematic review, it addresses broader topics and charts all available evidence, regardless of study design or quality (Arksey & O'Malley, 2005). The methods for the present scoping review drew upon those initially recommended by Arksey and O'Malley (2005) and subsequently refined by others (Arksey & O'Malley, 2005; Colquhoun et al., 2014; Levac, Colquhoun, & O'Brien, 2010). As recommended in these publications, we utilized a five-stage methodological framework: 1) Identifying the research question, 2) Identifying relevant studies, 3) Study selection, 4) Charting the data, and 5) Collating, summarizing, and reporting results (Arksey & O'Malley, 2005; Colquhoun et al., 2014; Levac, Colquhoun, & O'Brien, 2010).

Identifying the Research Question

To guide this scoping review, we defined the following research question: What are the 1) benefits or perceived benefits of participating in support groups for people with rare diseases and 2) facilitators and barriers of establishing and maintaining these groups?

Identifying Relevant Studies

In order to identify potentially relevant publications of support groups for people with rare diseases, we searched PubMed (from January 2000 through August 20, 2015) and CINAHL through the EBSCOhost platform (from January 2000 through August 26, 2015). A medical librarian developed the search strategy and performed the search. To develop the search strategy, we extracted all the names of rare diseases listed in Orphanet's Orphadata May 2015 "Rare Disorders and Cross-References" dataset (http://www.orphadata.org/cgi-bin/inc/product1.inc.php). The list included names of disorders, groups of disorders, subtypes, and synonyms and totalled 20,169 unique terms. To manage the size of the search, we excluded names of groups of disorders and synonyms, leaving 6,999 unique rare disorders and subtypes. We then combined these disorder names with terms relevant to support groups and self-help. The complete search strategy can be found in Online Resource 1.

Study Selection

The results of the search were downloaded into the citation management database RefWorks (ProQuest, 2009), and duplicate references were identified and removed. Following this, references were transferred into the systematic review software DistillerSR (Evidence Partners, 2015). We then assessed the eligibility of each publication through a two-stage process. First, two investigators independently reviewed the titles and abstracts of all citations identified through the search strategy. If either investigator deemed a publication potentially eligible based on the scoping review inclusion criteria, then a full-text review was completed, again by two investigators. Disagreements after full-text review were resolved by consensus, with a third investigator consulted as necessary.

Eligible publications were required to describe the benefits or perceived benefits of participating in rare disease support groups or the facilitators and barriers of establishing and

maintaining them. Consistent with standard scoping review methodology, we did not include any study design restrictions in our eligibility criteria (Arksey & O'Malley, 2005; Colquhoun et al., 2014; Levac, Colquhoun, & O'Brien, 2010). Publications about support groups for people with rare diseases in any language were eligible for inclusion.

For the purpose of this study, support groups were defined as an ongoing gathering of individuals who share common experiences. Activities of support groups had to include the giving and receiving of emotional and practical support, and could also include educational activities. Activities of support groups could take place in person, online or via teleconference, and had to include ongoing real-time interaction between group members. Groups that only followed a structured learning curriculum with a defined beginning and end (e.g., self-management programs) were not considered support groups. Groups that provided psychotherapy were not considered support groups.

Eligible rare diseases were diseases listed in Orphanet's "List of rare diseases and synonyms in alphabetical order (May 2015)" and included in the Orphadata dataset (Orphanet, 2015). This list includes diseases that are classified as rare based on their estimated prevalence in Europe. In cases where a publication reported on support groups in a non-European setting where the disease may or may not be rare (e.g., tuberculosis support groups in Africa), we determined the disease prevalence in the country in question using the World Health Organization's website (World Health Organization, 2015). If the support group was conducted in a country where the disease prevalence is less than or equal to 1 person in 2,000, then the publication was included. Publications about support groups that included people with rare diseases and others (e.g., family, friends, and other loved ones) were included if they reported information on people with rare diseases, specifically. Publications about support groups intended for people without rare diseases were excluded, even if some members may have had a rare disease.

Charting the Data, and Collating, Summarizing, and Reporting Results

A descriptive analytical approach was used to chart and summarize the data. Two investigators independently extracted data from included publications and entered it into a standardized Excel spreadsheet. For each publication, we extracted: 1) first author name; 2) publication year; 3) country of study; 4) design of study; 5) rare disease; 6) number of participants; 7) perceived benefits; 8) facilitators; and 9) barriers. Any disagreements were resolved by consensus, and a third investigator was consulted as necessary.

Results

The database search yielded 912 unique titles and abstracts. Of these, 841 were excluded after title and abstract review, leaving 71 publications for full-text review. A total of 10 publications met the inclusion criteria and were included in the scoping review (see Figure 1).

Description of Publications

The majority of included publications (n=8 studies, 80%) were primary research studies that collected qualitative or quantitative patient data (Alderson, Madill, & Balen, 2004; Breau & Norman, 2003; Castro, Mitchell, Rowe, & Jacobs, 2008; Howard et al., 2003; Jalovcic & Pentland, 2009; Moore, Teehan, Cornwall, Ball, & Thomas, 2008; Stewart et al., 2001; Telfair & Gardner, 2000), and two were experiential accounts (Barker, 2009; Vitucci, 2012). Of the eight primary research studies, four collected data using interviews or focus groups (sample sizes 3 to 30) (Alderson et al., 2004; Breau & Norman, 2003; Jalovcic & Pentland, 2009; Stewart et al., 2001), and three used questionnaires (sample sizes 6 to 79) (Howard et al., 2003; Moore et al., 2008; Telfair & Gardner, 2000). One publication, which was published only as a conference abstract, did not indicate the method of data collection (Castro et al., 2008). Of the two experiential accounts, one was a personal reflection that described a woman's experience living with idiopathic pulmonary arterial hypertension and how attending a support group helped her

cope with the disease (Barker, 2009). The other was written by the Chief Executive Officer of the Acoustic Neuroma Association and discussed ways that people with acoustic neuroma may benefit from participating in support groups (Vitucci, 2012).

Seven publications (70%) were from North America (Breau & Norman, 2003; Castro et al., 2008; Howard et al., 2003; Jalovcic & Pentland, 2009; Stewart et al., 2001; Telfair & Gardner, 2000; Vitucci, 2012), and three (30%) were from the United Kingdom (Alderson et al., 2004; Barker, 2009; Moore et al., 2008). All publications were from 2000 or later, and none reported on support groups for the same rare disease. Characteristics of included publications are summarized in Table 1.

Perceived Benefits of Rare Disease Support Groups

All 10 publications reported on the perceived benefits of rare disease support groups (Alderson et al., 2004; Barker, 2009; Breau & Norman, 2003; Castro et al., 2008; Howard et al., 2003; Jalovcic & Pentland, 2009; Moore et al., 2008; Stewart et al., 2001; Telfair & Gardner, 2000; Vitucci, 2012) (see Table 1). Seven overarching categories of perceived benefits were identified (see Table 2): 1) getting to know and befriending other people with the same disease who share similar experiences (n=9 studies, 90%); 2) learning about the disease and related treatments (n=8, 80%); 3) giving and receiving emotional support (n=5, 50%); 4) having a place to speak openly about the disease and one's feelings (n=4, 40%); 5) learning coping skills (n=4, 40%); 6) feeling empowered and hopeful (n=3, 30%); and 7) advocating to improve healthcare for people with the disease (n=1, 10%).

Facilitators and Barriers of Establishing and Maintaining Rare Disease Support Groups

Three publications described facilitators of establishing and maintaining support groups (Castro et al., 2008; Moore et al., 2008; Stewart et al., 2001), and one also described barriers (Moore et al., 2008) (see Table 1). All three publications were primary research studies.

Facilitators reported included: 1) meeting via teleconference; 2) providing leaders with training for their roles; and 3) having more than one leader. Barriers included: 1) finding patients or family members to lead the support group; 2) navigating difficult group discussions; and 3) uncertainty about one's role as a leader.

Discussion

Only ten publications, which used a variety of methods and described 10 different rare diseases, were identified (Alderson et al., 2004; Barker, 2009; Breau & Norman, 2003; Castro et al., 2008; Howard et al., 2003; Jalovcic & Pentland, 2009; Moore et al., 2008; Stewart et al., 2001; Telfair & Gardner, 2000; Vitucci, 2012). Commonly reported perceived benefits of rare disease support groups were getting to know and befriending other people with the same disease and similar experiences, learning about the disease and related treatments, and giving and receiving emotional support. Three publications reported on facilitators and barriers of establishing and maintaining rare disease support groups for people with rare diseases (Castro et al., 2008; Moore et al., 2008; Stewart et al., 2001). Facilitators included: 1) meeting via teleconference; 2) providing leaders with training for their roles; and 3) having more than one leader. Barriers included 1) getting patients and/or family members to lead the support group; 2) navigating difficult group discussions; and 3) uncertainty about one's role as a leader.

A number of studies have examined the benefits of participating in support groups for common medical diseases, including several studies that have interviewed cancer patients about the benefits of attending cancer support groups (Docherty, 2004; Ussher et al., 2006; Yaskowich & Stam, 2003). Patients in those studies reported that support groups provide unconditional acceptance; emotional support; a sense of belonging and community; a place to freely and safely talk about the disease and one's feelings; and information about cancer, its treatment and how other patients have coped with the condition (Docherty, 2004; Ussher et al., 2006; Yaskowich &

Stam, 2003) They also suggested that these groups decrease isolation and instill hope that one can survive the disease (Yaskowich & Stam, 2003).

Although people living with common medical diseases and those living with rare diseases may face different challenges, the findings of the present review and results of studies of cancer patients have a number of similarities. Commonalities include agreement that an important perceived benefit of participating in illness-based support groups is the giving and receiving of emotional support. Similarly, in rare diseases and cancer, people attend support groups to obtain information about the disease and treatment, as well as to meet others going through similar experiences. These latter two points are especially important for people with rare diseases.

The most notable difference between the findings of the present review on rare diseases and the studies of cancer patients is that in the present study, a perceived benefit of participating in support groups was advocating to improve healthcare for other rare disease patients. While this benefit was identified by only one of the studies included in the review, a number of previously published reports have also highlighted the importance of patient advocacy in rare diseases (Aymé et al., 2008; European Organisation for Rare Diseases, 2005, 2009). One possible explanation for this is that compared to common medical diseases, such as cancer, most rare diseases are often overlooked by clinicians, researchers, and politicians (European Organisation for Rare Diseases, 2005, 2009). As such people with rare disease and their families often become involved in trying to raise public awareness; in fact, because they must become their own advocates, rare disease patients and their families may be more proactive than patients with more common illnesses. Some become as knowledgeable or more knowledgeable about their condition as the health professionals who provide their care (European Organisation for Rare Diseases, 2005).

The findings of this scoping review suggest that support groups may be an important resource for many people with rare diseases, but establishing and sustaining support groups involves significant challenges. Rare disease organizations may be able to increase the accessibility of rare disease support groups by helping patients establish support groups, and the findings of our scoping review suggest factors that could potentially facilitate this process. For example, rare disease organizations may be able to implement training programs for potential leaders of support groups. There is not any evidence from well-designed and conducted trials on how to implement such a program or whether such a program would improve the experiences of support group leaders and members (Delisle et al., 2016), but our group is currently in the process of developing a program for patient support group leaders in SSc. Providing ongoing training would be difficult for any single disease-based organization to carry out, but it could be done via umbrella rare disease organizations or partnerships between organizations. Additionally, rare disease organizations might consider providing support groups via teleconferencing or videoconferencing in order to help patients in geographically isolated areas or with physical disabilities. Future research should focus on identifying the potential benefits of participation in support groups and factors associated with participation and non-participation in these groups, as well as factors that can facilitate the establishment and maintenance of successful groups. Well-designed and executed trials that assess the effectiveness of support groups and training programs for peer leaders of these groups are needed. Since rare disease organizations may not have the capacity or resources to conduct such trials, it could be beneficial for them to partner with researchers in the field. For instance, our research team has partnered with key scleroderma patient organizations in order to test the peer leader training program that we are developing. Innovative trial designs, such as the partially nested design (Lohr, Schochet, & Sanders, 2014) and the stepped-wedge design (Brown & Lilford, 2006; Hemming, Haines, Chilton, Girling, &

Lilford, 2015) may help researchers and stakeholders test interventions in a way that is not disruptive to the stakeholders' primary goal of providing support services.

In addition or instead of synchronous support groups, rare disease organizations might also consider alternative sources that have been used by rare disease patients and caregivers for peer support, including Facebook groups, online discussion boards or forums, and patient conferences. Although online support resources facilitate only asynchronous support, they may provide many of the same benefits as the synchronous support groups discussed in this review. Commonly reported perceived benefits of these resources include meeting people with the same disease who have similar experiences, having a space to speak openly about the disease and one's feelings, giving and receiving emotional support, learning about the disease and related treatments, learning coping skills, and feeling hopeful (Campbell, Phaneuf, & Deane, 2004; Doyle, 2015; Heisler, 2010; van Uden-Kraan et al., 2008). Other benefits that are unique to online support resources are that they are available 24 hours a day, easy to disseminate, and less resource-intensive than traditional face-to-face synchronous support groups (Campbell et al., 2004; Heisler, 2010; van Uden-Kraan et al., 2008). They are also accessible to patients who are geographically distant or homebound, such those who are too ill or whose symptoms are too severe to leave their home, as well as to patients who desire anonymity or privacy (Campbell et al., 2004; Heisler, 2010; van Uden-Kraan et al., 2008). There are important limitations that may impact their effectiveness and should be considered. For example, online support resources may not be available to everyone, such as patients without a computer or Internet access and those with limited computer skills (Campbell et al., 2004; Heisler, 2010; van Uden-Kraan et al., 2008). Furthermore, since they often function without a facilitator, there may be little or no control over the quality of the information that is exchanged. There can also be a substantial time lag between when a person asks a question and gets a response, and exchanges may include negative or

inappropriate remarks that may leave patients feeling vulnerable and unsupported (Campbell et al., 2004; Doyle, 2015; van Uden-Kraan et al., 2008). Patient conferences, like online support resources, offer many of the same benefits as synchronous support groups (Doyle, 2015). However, limitations of patient conferences include that they are costly for the organizations running them, they may not be accessible to all patients due to financial or geographical constraints, and they are time-limited and do not provide a mechanism for sustained relationship building and support (Doyle, 2015).

There are a number of limitations that should be considered when interpreting the results of this review. First, because we restricted our search to CINAHL and PubMed, it is possible that we may have missed important articles published in the grey literature, such as information from rare disease organization webpages. Second, although we had data extraction tools and two different investigators extracting data independently from publications, extracting accurate and complete data remained a challenge. There are a number of reasons for this, including that some articles reported methods or results that were incomplete or unclear. Third, by definition, a scoping review does not address the issues of "synthesis," which limits the inferences that be drawn from this review (Seibold, 2004). Most of the included studies were relatively small studies that were done in different rare diseases and used different methodologies; some used questionnaires or patient interviews to obtain data, and some reflected the experience of the writer without collecting data. No trials have been conducted on the benefits and possible harms of support groups for people with rare diseases. We did not believe that we could validly draw conclusions about the frequency or importance of facilitators and barriers or compare disease groups. Fourth, although rare diseases have many similarities, including that they are often degenerative and incurable, difficult to diagnosis and manage, and greatly compromise the quality of life of patients, there is also significant heterogenetity among them (European

Organisation for Rare Diseases, 2005). For example, rare diseases can differ in terms of age of onset (e.g., childhood versus adulthood), prevalence (e.g., rare versus ultra rare diseases), and severity, and it is unclear how these factors may have influenced our findings.

In conclusion, support groups appear to be an important resource for many people living with rare diseases. However, there is limited research on the benefits of participating in support groups for people with rare diseases and the facilitators and barriers of establishing and maintaining these groups. The findings of this review will inform research that is needed on support groups in rare diseases and may inform rare disease organizations, such as the Scleroderma Society of Canada and the Scleroderma Foundation in the United States, on ideas for possibly enhancing access to support groups and improving the ability of support groups to meet members' needs on a sustained basis.

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Table 1

Publications Included in Scoping Review

First Author Year Country	Study Type	Study Aim	Data Source	Sample and Rare Disease	Support Group Characteristics	Perceived Benefits of Support Groups Described in Study	Facilitators of Establishing and Maintaining Support Groups Described in Study	Barriers of Establishing and Maintaining Support Groups Described in Study
Alderson 2004 UK	Primary research study	To provide an understanding of the psychological sequel of androgen insensitivity syndrome (AIS) in phenotypic females in order to begin to inform psychosocial healthcare services	Two interviews with each participant	8 adult women with AIS	Held in person twice a year; leader characteristics not provided	-giving and receiving emotional support -learning about AIS -getting to know other people with AIS -having a place to speak openly about AIS and one's feelings -feeling empowered	Not applicable	Not applicable

Barker 2009 UK	Experiential account	Not applicable	Personal experience	A woman with idiopathic pulmonary arterial hypertension (IPAH)	Not applicable	-getting to know other people with IPAH -having a place to speak openly about one's fears, including death	Not applicable	Not applicable
Breau 2003 Canada	Primary research study	To identify and compare the needs of patients with prostate cancer or interstitial cystitis (IC) and to evaluate the role of self-help groups in meeting those needs	One interview with each participant	30 people with IC	Held in person once a month; led by a nurse practitioner	-receiving emotional support -getting IC- and treatment-related information -learning coping skills	Not applicable	Not applicable
Castro	Primary	To describe the	Methods not	42 people with	Held over the	-learning about	-telephone support	Not applicable

2008 USA	research study	design and evaluation of a telephone support group intervention for people with chronic graft-versus-host disease (cGVHD)	provided (abstract only)	cGVHD	phone once a week for four weeks; led by advanced practice nurses from the National Institutes of Health	cGVHD -being able to talk about the disease with others going through the same experiences	groups	
Howard 2003 USA	Primary research study	To describe the structure, function, and outcomes of The Oklahoma ThrombocytoPenic Purpura-Hemolytic Uremic Syndrome (TTP-HUS) Study Group	Questionnaire	35 former TTP-HUS patients	Held in person three times a year; led by a doctor	-learning about TTP-HUS -having the opportunity to talk to a doctor who is knowledgeable about the TTP-HUS -talking to other people with TTP-HUS and knowing	Not applicable	Not applicable

						that one is not alone		
Jalovcic 2009 Canada	Primary research study	To capture the essence of women's experiences of participation in the Telephone Peer Support Group Program for Women with Spinal Cord Injury	One individual interview and three telephone focus groups with all participants	7 women with spinal cord injury	Held over the phone; leader characteristics not provided	-giving and receiving emotional support -learning up-to-date disease-related information -getting to know other people with the disease and knowing that one is not alone -having a place to speak openly about the disease and one's experiences with it -feeling	Not applicable	Not applicable

						empowered		
Moore 2008 UK	Primary research study	To describe the authors initial experience of establishing a support group for people with mesothelioma	Questionnaire	6 people with mesothelioma	Held in person once a month; led by four healthcare professionals (a patient information officer, a psychotherapist, and two lung cancer nurse specialists) and a peer	-getting to know other people with the disease and hearing about other patients' experiences living with the disease, as well as how they cope and manage it	-having more than one leader -having a collaborative partnership among the leaders	-getting patients and/or family members to take on any organizational responsibility for the group -navigating difficult group discussions -uncertainty about one's role as a leader
Stewart 2001 Canada	Primary research study	To test the feasibility of a telephone support group intervention for men with	One interview with each participant	3 men with hemophilia and HIV/AIDS	Held over the phone once a week for 12 weeks; led by a mental health	-receiving emotional support -getting informational support (e.g.,	-leader training -having a guide of potential discussion topics	Not applicable

		hemophilia and HIV/AIDS and for their family caregivers			nurse and a peer	learning about the disease and treatments) -feeling less alone -learning and employing coping skills		
Telfair 2000 Canada, USA	Primary research study	To explore and describe the experiences of adolescents with sickle cell disease (SCD) who are active members of SCD support groups	Questionnaire	79 adolescents with SCD from 12 support groups	The majority of groups met once a month; all were led by a professional	-learning more about the disease -making new friends -advocating to improve healthcare for other people affected by the disease	Not applicable	Not applicable
Vitucci 2012	Experiential account	Not applicable	Personal experience	Chief Executive Officer of the	Not applicable	-giving and receiving	Not applicable	Not applicable

USA	Acoustic Neuroma Association	emotional support -learning about disease- and treatment-related information -meeting other people who have gone through a similar experience and realizing that one is not alone -learning how other people cope with disease-related problems -feeling hopeful

Note. Alderson =Alderson et al., 2004; Barker=Barker, 2009; Breau=Breau & Norman, 2003; Castro=Castro et al., 2008; Howard=Howard et al., 2003; Jalovcic=Jalovcic & Pentland, 2009; Moore=Moore et al., 2008; Stewart=Stewart et al., 2001; Telfair=Telfair & Gardner, 2000; Vitucci=Vitucci, 2012.

Table 2

Categories of Perceived Benefits of Rare Disease Support Groups and Sources

Perceived Benefit	n Studies (%)	Experiential Account	Original Research Study – Qualitative	Original Research Study – Questionnaire	Published Abstract
Getting to know and befriending other people with the same disease who share similar experiences	9 (90%)	Barker Vitucci	Jalovcic Alderson Stewart	Moore Howard Telfair	Castro
Learning about the disease and related treatments	8 (80%)	Vitucci	Jalovcic Alderson Breau Stewart	Howard Telfair	Castro
Giving and receiving emotional support	5 (50%)	Vitucci	Jalovcic Alderson Breau Stewart	Not applicable	Not applicable
Having a place to speak openly about the disease and	4 (40%)	Barker	Jalovcic	Not applicable	Castro

one's feelings			Alderson		
Learning coping skills	4 (40%)	Vitucci	Breau Stewart	Moore	Not applicable
Feeling empowered and hopeful	3 (30%)	Vitucci	Jalovcic Alderson	Not applicable	Not applicable
Advocating to improving healthcare for other patients	1 (10%)	Not applicable	Not applicable	Telfair	Not applicable

Note. Alderson=Alderson et al., 2004; Barker=Barker, 2009; Breau=Breau & Norman, 2003; Castro=Castro et al., 2008; Howard=Howard et al., 2003; Jalovcic=Jalovcic & Pentland, 2009; Moore=Moore et al., 2008; Stewart=Stewart et al., 2001; Telfair=Telfair & Gardner, 2000; Vitucci=Vitucci, 2012.

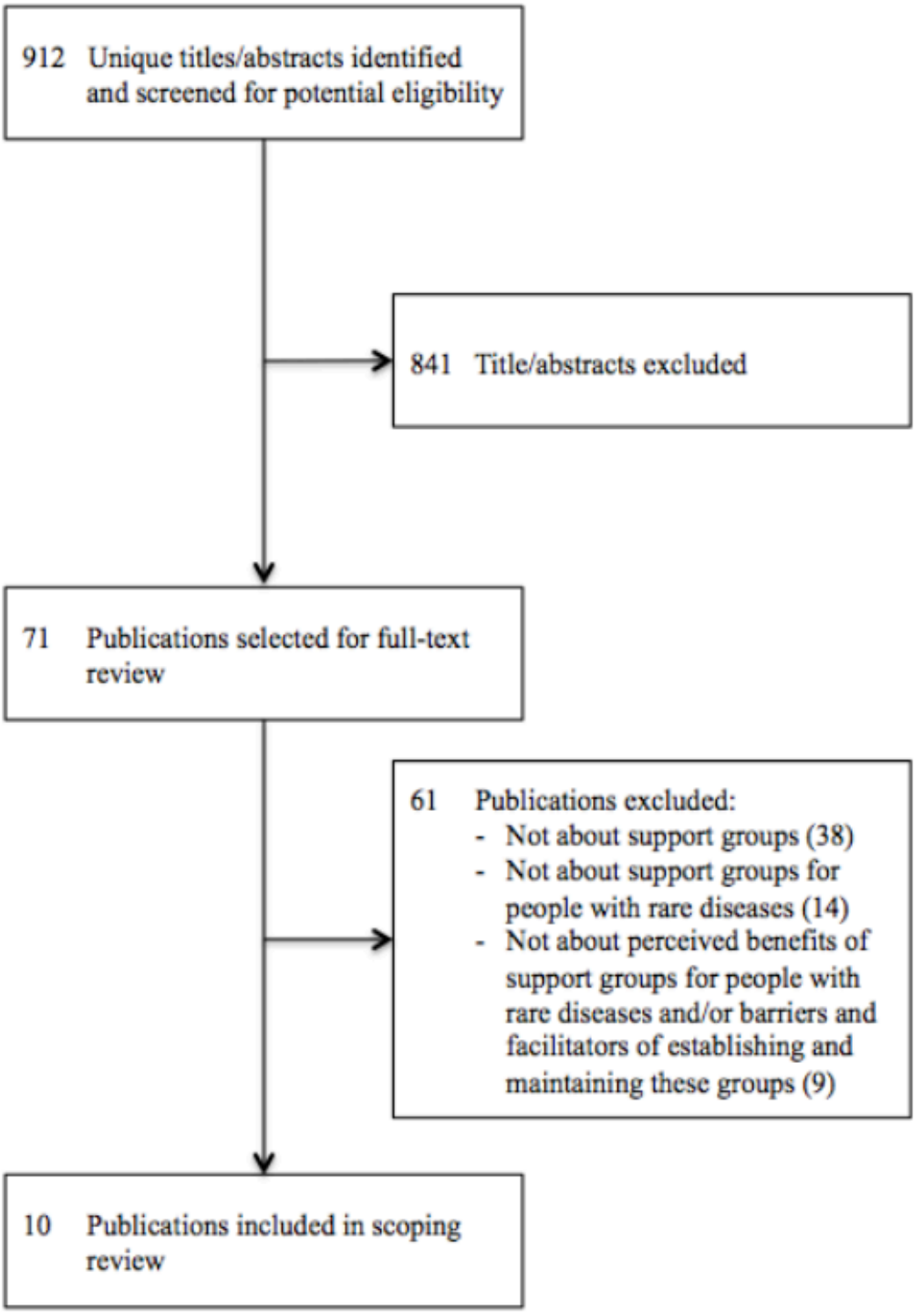

Figure 1. A graphical representation of the flow of publications reviewed in the course of the scoping review.

Linking Manuscripts 2 and 3

The second manuscript in this thesis was a scoping review exploring the 1) benefits of participating in rare disease support groups and 2) facilitators and barriers of initiating and sustaining these groups. A number of perceived benefits of rare disease support groups were identified, including getting to know and befriending other people with the same disease and similar experiences, learning about the disease and related treatments, and giving and receiving emotional support (Alderson, Madill, & Balen, 2004; Barker, 2009; Breau & Norman, 2003; Castro, Mitchell, Rowe, & Jacobs, 2008; Howard et al., 2003; Jalovcic & Pentland, 2009; Moore, Teehan, Cornwall, Ball, & Thomas, 2008; Stewart et al., 2001; Telfair & Gardner, 2000; Vitucci, 2012). These findings suggest that support groups are an important resource for people with rare diseases.

In addition, several facilitators and barriers of initiating and sustaining rare disease support groups were identified (Castro et al., 2008; Moore et al., 2008; Stewart et al., 2001). Facilitators included: 1) meeting via teleconference; 2) providing leaders with training for their roles; and 3) having more than one leader, and barriers included 1) getting patients and/or family members to lead the support group; 2) navigating difficult group discussions; and 3) uncertainty about one's role as a leader. These findings further suggest that educational training programs for peer facilitators of rare disease support groups, such as scleroderma (or systemic sclerosis, SSc) support groups, could be useful in addressing challenges regarding initiating and sustaining these groups. So, to inform the development of the Scleroderma Support group Leader EDucation (SSLED) Program and ensure its pertinence for facilitators of SSc support groups, our research team conducted a third study identifying the training and support needs of SSc support group facilitators.

Chapter 4

Manuscript 3 (Paper Submitted for Publication)

Training and Support Needs of Scleroderma Support Group Facilitators: The North American Scleroderma Support Group Facilitators Survey

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Michelle Richard, Halifax, Nova Scotia, Canada; Robert Riggs, Danvers, Massachusetts, USA; Maureen Sauve, Hamilton, Ontario, Canada; Nancy Stephens, Detroit, Michigan, USA.

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Abstract

Peer-facilitated support groups are an important resource for people living with scleroderma (systemic sclerosis; SSc). Little is known, however, about the challenges that SSc support group facilitators experience in fulfilling their leadership role. The objective of this study was to identify the training and support needs of SSc support group facilitators. A 32-item survey was used to assess confidence to execute tasks necessary for facilitating a support group successfully among SSc support group facilitators in Canada and the United States. To assist in the interpretation of the results, survey items were grouped into seven themes based on content. A total of 80 SSc support group facilitators completed the survey. The percentage of respondents who agreed or strongly agreed that they were confident in their ability to carry out the 32 group facilitator tasks ranged from 41% to 91%. Support group facilitators were generally confident in their ability to complete tasks related to the following themes: 1) Organizing, structuring, and facilitating the group; 2) Addressing individual member needs and diversity of the group; 3) Helping members cope with grief and loss; and 4) Attaining and responding to member feedback. However, they were less confident in their ability to perform tasks related to 1) Managing difficult group dynamics; 2) Promoting and sustaining the group; and 3) Balancing personal and group needs. SSc organizations may be able to increase the accessibility and effectiveness of SSc support groups by implementing an educational training program for peer-facilitators of support groups.

Keywords: facilitators; scleroderma, systemic sclerosis; support groups; self-help groups

Training and Support Needs of Scleroderma Support Group Facilitators: the North American Scleroderma Support Group Facilitators Survey

Scleroderma, or systemic sclerosis (SSc), is a rare, chronic, multi-system connective tissue disorder characterized by abnormal fibrotic processes and excessive collagen production that manifests in thickening of the skin and damage to the internal organs, including the heart, lungs, and gastrointestinal tract (Mayes, 2008; Seibold, 2004). SSc is typically diagnosed between the ages of 30 and 50 years, and approximately 80% of people with the disease are women (Mayes, 2008; The Arthritis Society, n.d.). There is no cure for SSc, and the median survival time from diagnosis is approximately 11 years, with patients 3.7 times more likely to die within 10 years of diagnosis (44.9% mortality) than age, race, and sex-matched persons without the disease (12.0% mortality) (Mayes et al., 2003).

People with rare diseases, including SSc, experience many of the same challenges as those with more common diseases, but also face unique challenges, including gaps in knowledge about their disease, difficulty obtaining an accurate diagnosis, and limited treatment and support options (European Organisation for Rare Diseases, 2005, 2009). Professionally organized support services, which are often provided to people with more common diseases, are typically not available for SSc patients (Kwakkenbos et al., 2013). Thus, many people with SSc have organized peer-facilitated support groups in order to help them cope with and manage their disease (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.).

Illness-based support groups, including SSc support groups, adhere to the principle that people who face similar disease-related challenges can empower one another through social contact and support (Davison, Pennebaker, & Dickerson, 2000). Activities of support groups typically involve an educational or information-sharing component, and the giving and receiving of emotional and practical support. Support groups can be configured in a variety of ways; in

addition to being facilitated by professionals or peers, they may be held face-to-face, online, or via teleconference and have a structured or unstructured format (Aymé, Kole, & Groft, 2008; Barg & Gullatte, 2001).

There are almost 200 SSc support groups across Canada and the United States, the majority of which are facilitated by peers (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.). Many people with SSc, however, do not have access to support groups (Delisle et al., 2016). Where support groups are available, they sometimes have difficulty sustaining themselves due to having a single leader whose health worsens or because of shortcomings related to untrained patient leaders. Furthermore, some patients report that they prefer not to attend support groups because the group in their area is poorly organized or is overly negative and not constructive (Delisle et al., 2016).

Education and training for peer facilitators of support groups could enhance the self-efficacy and improve the ability of peer facilitators to carry out their leader-related tasks and reduce the burden they experience in their leadership roles. This, in turn, could enhance access to support groups and the ability of these groups to consistently meet the needs of members. To develop an education and training program, however, information is needed regarding the training and support needs of SSc support group peer facilitators. The present study was conducted to evaluate those needs.

Method

Participants

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 Scleroderma Canada's and the Scleroderma Foundation's websites, as well as on Canadian provincial SSc society websites; 2) postings on Scleroderma Canada's and

the Scleroderma Foundation's social media venues (e.g., Facebook, Twitter); 3) distribution of flyers at the Scleroderma Foundation's annual conference; 4) announcements in SSc patient newsletters; 5) emails to support group facilitators and members across Canada and the United States; and 6) postings in SSc-related chat rooms.

Respondents who accessed the survey website had the option to 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, that they resided in Canada or the United States, and that they were a facilitator of a SSc support group at the time they completed the survey.

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 Facilitators Survey

Initial items for the Scleroderma Support Group Facilitators Survey were obtained from the Group Leader Self-Efficacy Instrument (GLSI), which is a 37-item self-report questionnaire that assesses self-efficacy for performing group facilitator skills on a 6-point Likert scale ranging from 1 (*Strongly Disagree*) to 6 (*Strongly Agree*) (Page, Pietrzak, & Lewis, 2001). These were complimented by items obtained from a similar questionnaire that was administered to facilitators

of cancer and multiple sclerosis support groups (Zordan et al., 2010), as well as items generated from the published results of a study on the experiences of facilitators of cancer support groups (Butow et al., 2005).

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 by all members of the research team until a consensus on the final item pool was reached. Team members who participated in this process included representatives from Scleroderma Canada, the Scleroderma Foundation, and the Scleroderma Society of Ontario; a Patient Advisory Board that consisted of six current SSc support group peer facilitators from Canada and the United States; and researchers with expertise in SSc.

The final survey consisted of 32 core items that assessed the confidence of SSc support group facilitators to carry out tasks necessary for facilitating a support group successfully. Item response options included *Strongly Disagree, Disagree, Slightly Disagree, Slightly Agree, Agree, and Strongly Agree* (scored 1-6). In addition, there were three questions that asked respondents to report 1) how long they had been a SSc support group facilitator; 2) whether they had received training for their facilitator role; and 3) whether they had been a group member prior to being a facilitator.

Data Analysis

Self-reported sociodemographic and disease-related characteristics were documented. For each item, the number and percentage of participants who responded "*Agree*" and "*Strongly Agree*" were tallied. In addition, the mean and standard deviation of item scores were calculated.

To assist in the interpretation of the results, one investigator independently grouped the items into themes. Next, the item groupings and themes were reviewed iteratively by all members

of the research team until a consensus was reached. The following seven themes were identified: 1) Organizing, structuring, and facilitating the group; 2) Addressing individual member needs and diversity of the group; 3) Managing difficult group dynamics and members; 4) Helping members cope with grief and loss; 5) Promoting and sustaining the group; 6) Attaining and responding to member feedback; and 7) Balancing personal and group needs. We did not conduct a factor analysis to attempt to identify themes because our sample was too small.

Results

Sample Characteristics

Altogether, there were 94 peer facilitators who completed survey. Of these, 1 (1%) was excluded because the respondent resided outside of Canada or the United States, 5 (5%) were excluded because respondents reported a diagnosis other than SSc, and 8 (9%) were excluded because respondents were missing some of the 35 survey items (range 6-33 missing items). The sociodemographic and disease-related characteristics of the 80 respondents included in analyses are presented in Table 1. Most respondents were female (89%), White (93%), from the United States (78%), married (68%), and had some college or university education (45%). The mean age of respondents was 59 years, and the mean number of years since diagnosis of SSc was 15 years.

The mean number of years that respondents had been facilitating a SSc support group was 8 years (SD = 7). Only 30% of respondents had received any training for their role as a group facilitator, and approximately half (51%) had been a group member prior to being a facilitator.

Item Response Frequencies

The frequencies of item responses are shown in Table 2. The percentage of respondents who agreed or strongly agreed that they were confident in their ability to carry out the 32 group facilitator tasks ranged from 41% to 91%.

Organizing, structuring, and facilitating the group. Overall, respondents were confident in their ability to execute tasks related to organizing, structuring, and facilitating the group. More than 80% of respondents were confident in their ability to help the group establish appropriate group rules (e.g., maintaining confidentiality) (91%), facilitate the group meetings so that all members have an opportunity to speak (90%), provide the structure needed for successful group meetings (86%), and help the group stay focused on topics that are relevant to members (85%). Between 70% and 80% of respondents were confident in their ability to keep the group meetings interesting and relevant to both new and returning members (78%) and to organize and plan activities for group members (e.g., having guest speakers) (71%).

Addressing individual member needs and diversity of the group. The majority of respondents were confident in their ability to implement tasks related to addressing individual member needs and diversity of the group. At least 80% of respondents were confident in their ability to help members feel comfortable in the group and relate to one another (90%), help overly shy group members feel comfortable interacting with the group (89%), help group members relate to other members of a different gender (86%), help group members relate to other members of a different age (86%), and help group members relate to other members of a different cultural background (80%). Fewer than 70% of respondents, however, were confident in their ability to address the different needs of group members at varying stages of the disease (65%).

Managing difficult group dynamics and members. The percentages of respondents that were confident in their ability to perform various tasks related to managing difficult group dynamics and members were quite varied. For example, over 80% of respondents were confident in their ability to intervene effectively when group rules are not being followed (81%). Between 70% and 80% of respondents were confident in their ability to manage group members who oversimplify or minimize the concerns of other members (79%), manage group members who are

overly talkative or monopolize the discussion (74%), and manage group members who assume the role of the "know-it-all" (70%). Fewer than 70% of respondents, however, were confident in their ability to manage conflicts and disagreements between group members (63%), talk to a group member about his or her behaviour if it is disruptive to the group (55%), and ask a member to leave the group due to his or her disruptive behaviour (41%).

Helping members cope with grief and loss. Many respondents were confident in their ability to execute tasks related to helping members cope with grief and loss, including supporting members who are grieving (89%) and helping group members cope with difficult events (e.g., death of a member) (78%).

Promoting and sustaining the group. In general, the confidence of respondents in their ability to implement tasks related to promoting and sustaining the group was lower than for other themes. Between 70% and 80% of respondents were confident in their ability to promote the group to health professionals as an important resource for patients (79%). Fewer than 70% of respondents, however, were confident in their ability to effectively publicize the group (55%), obtain financial or other resources needed to run the group (55%), or effectively recruit new members (48%).

Attaining and responding to member feedback. Many respondents were confident in their ability to perform tasks related to attaining and responding to member feedback, such as obtaining feedback from members about the group (80%) and responding constructively to feedback from group members (88%). Obtaining feedback about one's leadership, however, appeared slightly more difficult to do, as between 70% and 80% of respondents felt confident in their ability to carry out this task (71%).

Balancing personal and group needs. Another area where the percentage of respondents that were confident in their ability to perform tasks was wide-ranging was balancing personal and

group needs. For instance, over 80% of respondents were confident in their ability to communicate reasonable boundaries about their availability outside of the group (86%). Between 70% and 80% of respondents were confident in their ability to share responsibilities, including administrative and practical tasks, with a co-facilitator or other group members (76%). Less than 70% of respondents, however, were confident in their ability to recruit a co-facilitator or other group members to help them with leadership responsibilities (66%) and to obtain the support they need to cope with the emotional demands of leading the group (60%).

Discussion

The main finding of this study was that SSc support group facilitators had a high level of confidence in their ability to carry out many support group leader-related tasks; however, they reported lower confidence in their ability to perform tasks in several important areas. SSc support group facilitators were generally confident in their ability to carry out tasks related to the themes 1) Organizing, structuring, and facilitating the group; 2) Addressing individual member needs and diversity of the group; 3) Helping members cope with grief and loss; and 4) Attaining and responding to member feedback. On the other hand, support group facilitators were less confident in their ability to perform tasks related to 1) Managing difficult group dynamics; 2) Promoting and sustaining the group; and 3) Balancing personal and group needs. Many facilitators expressed having lower confidence in their ability to manage conflicts and disagreements between group members, talk to a member about his or her behaviour if it is disruptive to the group, and ask a member to leave the group due to his or her disruptive behaviour. In addition, facilitators indicated having lower confidence in their ability to effectively publicize the group and recruit new members, as well as obtain financial or other resources needed to run the group. They also had lower confidence in their ability to obtain support to cope with the emotional demands of leading the group.

The findings of this study are generally consistent with those of previous studies that have explored the challenges reported by facilitators of illness-based support groups. We identified several studies that sought to understand the difficulties experienced by cancer support group facilitators (Butow et al., 2005; Kirsten, Butow, Price, Hobbs, & Sunquist, 2006; Zordan et al., 2010). The support group facilitators in those studies reported personal challenges, such as maintaining personal balance, preventing burn out, and managing one's own health condition while supporting others; practical challenges, such as access to new and relevant information and guest speakers, lack of resources, and lack of support and referrals from medical professionals; and difficulties with group leadership tasks, including managing complex group dynamics and dealing with the worsening health or death of group members. These challenges are magnified for facilitators of rare disease support groups, including SSc support groups, who often face logistical problems related to small numbers of potential group members, even in urban settings, and limited support from healthcare and patient organizations, which are not as well resourced as organizations for people with more common diseases. This and the finding that SSc support group facilitators report having lower confidence in their ability to carry out a number of important leader-related tasks, underscores the importance of increased education and training for support group facilitators.

The findings of this study will inform the development of an educational and training program to provide information and skills to support the ability of SSc peer support group facilitators to lead effective, sustainable support groups; reduce the emotional and physical toll on leaders; and encourage new leaders to set up support groups where none exist. Scleroderma Canada and the Scleroderma Foundation in the United States are committed to enhancing the accessibility of SSc support groups, improving the experience of SSc support group facilitators, and improving the ability of SSc support groups to meet patient needs. Training peer facilitators

of support groups could help to accomplish this, but no well-conducted trials have evaluated the effectiveness of training programs for peer support group leaders. Our research team has partnered with the Scleroderma Canada and the Scleroderma Foundation to develop the Scleroderma Support group Leader EDucation (SSLED) Program. The SSLED Program will be a 3-month-long, group training program that will be delivered using videoconferencing in order to provide information and skills to improve patient support group leaders' confidence and self-efficacy to carry out their leadership roles.

There are a number of limitations to consider when interpreting the results of this study. First, respondents were recruited through national and provincial SSc organizations, SSc-related chat-rooms and newsletters, and emails to support group facilitators across Canada and the United States. It may have been the case that SSc support group facilitators who were more active within the SSc community and more involved with SSc organizations participated in the study, and these facilitators could differ from other facilitators. Second, the sample size in this study was small. There are fewer than 200 SSc support groups in Canada and the United States (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.), however, so the sample represents close to half of all SSc support group facilitators in these two countries. Third, the grouping of items into themes was done on the basis of content because the sample was too small to conduct a factor analysis. However, the item groupings and themes were reviewed iteratively by all members of the research team, which included representatives from the Scleroderma 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. Thus, we feel confident that thematic groupings provided a reasonable structure for interpreting item responses.

In conclusion, peer-facilitated support groups are an important resource for many people living with SSc. Facilitators of SSc support groups, reported having lower confidence in their ability to carry out a number of important leader-related tasks, including managing difficult group dynamics, promoting and sustaining the group, and balancing personal and group needs. This study provides important information that will be used to inform the development of an education and training program for peer facilitators of SSc support groups and, thus, to improve the accessibility to and effectiveness of these groups.

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Table 1

Sociodemographic and Disease-Related Characteristics (N = 80)

Variable	
Female, *n (%)*	71 (88.8%)
Age in years, *M (SD)*	58.5 (12.2)
Location, *n (%)*	
Canada	18 (22.5%)
United States	62 (77.5%)
Race/ethnicity, *n (%)*	
White	74 (92.5%)
Other	5 (6.3%)
Two or more	1 (1.3%)
Marital status, *n (%)*	
Never married	5 (6.3%)
Living with partner in committed relationship	3 (3.8%)
Married	54 (67.5%)
Separated	0 (0.0%)
Divorced	10 (12.5%)
Widowed	8 (10.0%)
Highest level of education, *n (%)*	
Elementary/primary school	0 (0.0%)
Secondary/high school	8 (10.0%)
Some college/university	36 (45.0%)
University degree	22 (27.5%)
Postgraduate degree	14 (17.5%)
Occupational status, *n (%)*	
Homemaker	5 (6.3%)

Full-time student	0 (0.0%)
Part-time employed	3 (3.8%)
Full-time employed	13 (16.3%)
On leave of absence	0 (0.0%)
On disability	30 (37.5%)
Retired	26 (32.5%)
Unemployed	3 (3.8%)
Scleroderma diagnosis, *n (%)*	
Limited scleroderma	39 (48.8%)
Diffuse scleroderma	36 (45.0%)
Not known	5 (6.3%)
Years since scleroderma diagnosis, *M (SD)*	14.8 (7.8)

Table 2

Item Response Frequencies

Item	Rank	"*Agree*" and "*Strongly Agree*" n (%)	*M (SD)*
I am confident in my ability to…			
Theme 1: Organizing, structuring, and facilitating the group			
…help the group establish appropriate group rules, such as maintaining confidentiality	1	73 (91.3%)	5.26 (0.81)
…facilitate the group meetings so that all members have an opportunity to speak	2	72 (90.0%)	5.30 (0.80)
…provide the structure needed for successful group meetings	7	69 (86.3%)	5.18 (0.76)
…help the group stay focused on topics that are relevant to members	11	68 (85.0%)	5.07 (0.91)
…keep the group meetings interesting and relevant to both new and returning members	17	62 (77.5%)	4.99 (0.82)
…organize and plan activities for group members, such as having guest speakers	21	57 (71.3%)	4.96 (1.08)
Theme 2: Addressing individual member needs and diversity of the group			
…help members feel comfortable in the group and relate to one another	2	72 (90.0%)	5.32 (0.69)
…help overly shy group members feel comfortable interacting with the group	4	71 (88.8%)	5.22 (0.63)

…help group members relate to other members of a different gender	7	69 (86.3%)	5.11 (0.78)
…help group members relate to other members of a different age	7	69 (86.3%)	5.10 (0.69)
…help group members relate to other members of a different cultural background	13	64 (80.0%)	5.02 (0.89)
…address the different needs of group members at varying stages of the disease	25	52 (65.0%)	4.91 (1.01)
Theme 3: Managing difficult group dynamics and members			
…intervene effectively when group rules are not being followed	12	65 (81.3%)	4.93 (0.91)
…manage group members who oversimplify or minimize the concerns of other members	15	63 (78.8%)	4.93 (0.90)
…manage group members who are overly talkative or monopolize the discussion	20	59 (73.8%)	4.83 (0.91)
…manage group members who assume the role of the “know-it-all”	23	56 (70.0%)	4.79 (0.95)
…manage conflicts and disagreements between group members	26	50 (62.5%)	4.65 (1.03)
…talk to a group member about his or her behaviour if it is disruptive to the group	28	44 (55.0%)	4.47 (1.15)
…ask a member to leave the group due to his or her disruptive behaviour	32	33 (41.3%)	4.00 (1.51)
Theme 4: Helping members cope with grief and loss			
…support members who are grieving	4	71 (88.8%)	5.35 (0.78)
…help group members cope with difficult events, such as the death of a	17	62 (77.5%)	4.87 (0.95)

member			
Theme 5: Promoting and sustaining the group			
…promote the group to health professionals as an important resource for patients	15	63 (78.8%)	4.93 (1.19)
…effectively publicize the group	28	44 (55.0%)	4.60 (1.15)
…obtain financial or other resources needed to run the group	28	44 (55.0%)	4.19 (1.41)
…effectively recruit new members	31	38 (47.5%)	4.44 (1.26)
Theme 6: Attaining and responding to member feedback			
…respond constructively to feedback from group members	6	70 (87.5%)	5.14 (0.79)
…obtain feedback from members about the group	13	64 (80.0%)	4.99 (0.95)
…obtain feedback from members about my leadership	21	57 (71.3%)	4.80 (0.92)
Theme 7: Balancing personal and group needs			
…communicate reasonable boundaries about my availability outside of the group	7	69 (86.3%)	5.06 (0.75)
…share responsibilities, including administrative and practical tasks, with a co-facilitator or other group members	19	61 (76.3%)	4.87 (1.28)
…recruit a co-facilitator or other group members to help me with leadership responsibilities	24	53 (66.3%)	4.69 (1.18)
…obtain the support I need to cope with the emotional demands of leading the group	27	48 (60.0%)	4.56 (1.19)

Note. Items are ranked based on the number and percentage of participants that answered "*Agree*" and "*Strongly Agree.*"

Linking Manuscripts 3 and 4

The aim of the third manuscript in this thesis was to identify the training and support needs of scleroderma (or systemic sclerosis, SSc) support group facilitators. Overall, SSc support group facilitators had a high level of confidence in their ability to execute many support group leader-related tasks. However, they also reported lower confidence in their ability to perform several important tasks, including 1) Managing difficult group dynamics (e.g., managing conflicts and disagreements between group members; talking to a member about his or her behaviour if it is disruptive to the group; asking a member to leave the group due to his or her disruptive behavior); 2) Promoting and sustaining the group (e.g., publicizing the group and recruiting new members; obtaining financial or other resources needed to run the group); and 3) Balancing personal and group needs (e.g., obtaining support to cope with the emotional demands of leading the group). These findings underscore the importance of increased education and training for SSc support group facilitators. They also highlight areas of focus for the SSLED Program.

The Scleroderma Society of Canada and the Scleroderma Foundation in the United States are committed to enhancing access to SSc support groups and improving the experience of SSc support group facilitators and the ability of SSc support groups to meet the needs of patients. If the Scleroderma Support group Leader EDucation (SSLED) Program is to address the latter, then more information is needed regarding the factors that influence the decisions of SSc patients to attend SSc support groups. Thus, our research team conducted a fourth study identifying the reasons why people with SSc do not participate in SSc support groups.

Chapter 5

Manuscript 4 (Paper Published in Clinical and Experimental Rheumatology)

Reasons for Non-participation in Scleroderma Support Groups

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Michelle Richard, Halifax, Nova Scotia, Canada; Robert Riggs, Danvers, Massachusetts, USA; Maureen Sauve, Hamilton, Ontario, Canada; Nancy Stephens, Detroit, Michigan, USA.

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Abstract

Peer-led support groups are an important resource for people living with many rare diseases, including scleroderma (systemic sclerosis, SSc). Little is known, however, about the accessibility of SSc support groups and factors that may discourage people from participating in these groups. The objective of this study was to identify reasons why people with SSc do not participate in SSc support groups. Canadians with SSc were recruited to complete the Canadian Scleroderma Patient Survey of Health Concerns and Research Priorities. Data from respondents who answered the question "Have you participated in SSc support groups?" with "No" were analyzed. Frequencies of participants who responded 1) I'm not interested, 2) None are easily available, and 3) Other (please specify) were tallied. A content analysis approach was used to code the open-ended responses to this question. A total of 280 respondents provided a reason for non-participation in SSc support groups. Key reasons for not participating in support groups included: 1) Not interested or no perceived need (36%); 2) No local support group available (35%); 3) Lack of awareness of the existence of SSc support groups (13%); 4) Practical barriers (6%); 5) Emotional factors (4%); 6) Uncertainty about whether to attend (4%); and 7) Negative perceptions about support groups (3%). SSc organizations may be able to address current limitations in the accessibility and effectiveness of SSc support groups by implementing online support groups, as well as by providing support group leaders training to help establish and sustain successful SSc support groups.

Keywords: participation; scleroderma; support groups

Reasons for Non-participation in Scleroderma Support Groups

People with rare diseases, including scleroderma (systemic sclerosis; SSc), experience many of the same challenges as people with common diseases. These include physical and psychological symptoms that require them to modify their family, social, and professional roles (Asbring, 2001; Karasz & Ouellette, 1995; Kralik, 2002; Kwakkenbos et al., 2013). Additionally, due to gaps in knowledge about their disease, people with rare diseases often face substantial delays in diagnosis (Delisle, Hudson, Baron, Thombs, & Canadian Scleroderma Research Group, 2014; European Organisation for Rare Diseases, 2005, 2009) and, limited treatment and support options (European Organisation for Rare Diseases, 2005, 2009; Reimann, Bend, & Dembski, 2007).

In the absence of professionally organized and delivered support services (Kwakkenbos et al., 2013), people with rare diseases have mobilized their own support systems in the form of peer-led support groups (Reimann et al., 2007). Peer-led support groups adhere to the principle that people who face similar disease-related challenges can empower one another through social contact (Aymé, Kole, & Groft, 2008). Group activities typically involve educational or information-sharing components, and the giving and receiving of emotional and practical support (Barg & Gullatte, 2001; Lieberman et al., 2005).

Peer-led support groups are an important resource for many people with SSc (Kwakkenbos et al., 2015). SSc is a rare chronic autoimmune connective tissue disease characterized by abnormal fibrotic processes and excessive collagen production, which manifests itself in skin thickening and internal organ damage, and vascular implications (Seibold, 2004). The prevalence of SSc in Canada is 44 cases per 100,000 (Bernatsky et al., 2009). As with many other rare diseases, people with SSc initially experience debilitating symptoms, and may struggle to obtain a diagnosis (Delisle et al., 2014). Once diagnosed, the future is uncertain, as disease

course is unpredictable (Mayes, 2008). Marked disfigurement from the disease sets many people with SSc apart, and some have described changes in physical appearance so drastic that they are no longer recognized by acquaintances (Jewett, Hudson, & Thombs, 2001; Malcarne, Hansdottir, Greensbergs, Clements, & Weisman, 1999; van Lankveld, Vonk, Teunissen, & van den Hoogen, 2007).

Currently, there are approximately 30 SSc support groups in Canada and 150 in the US, and all of them are peer-led (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.). The Scleroderma Society of Canada and the Scleroderma Foundation in the US help SSc patients locate support groups, but provide almost no information regarding starting a support group or formal training and support to peer facilitators. These 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. To do this, information is needed regarding SSc support group accessibility and factors that may discourage people from utilizing them. We identified only two studies that assessed factors that influence participation in support groups (Ussher, Kirsten, Butow, & Sandoval, 2008; Winefield, Coventry, Lewis, & Harvey, 2003), but both involved cancer patients. No studies have been done with rare disease patients. Thus, the objective of this study was to explore, among SSc patients who do not attend SSc support groups, reasons for non-participation.

Method

Participants and Procedure

Canadians with SSc completed an anonymous online questionnaire, the Canadian Scleroderma Patient Survey of Health Concerns and Research Priorities, between September 2008 and August 2009 (Bassel 2012, 2011). Recruitment occurred through advertisements 1) in Canadian Scleroderma Research Group (CSRG) physicians' offices; 2) at the Scleroderma

Society of Canada's annual conference; 3) in newsletters and on websites of the Scleroderma Society of Canada and Sclérodermie Québec; 4) in Canadian magazines; and via 5) emails to support group facilitators. Participants could complete the survey online or by requesting a paper version. Respondents were included in the present study if they reported a diagnosis of SSc by a healthcare provider, were ≥18 years old, resided in Canada, and answered the survey question "Have you participated in SSc support groups?" with "No". Respondents who selected "No" were asked to specify their reasons for not attending. Response options included: 1) I'm not interested; 2) None are easily available; and 3) Other (please specify). We did not evaluate the proportion of patients who reported participating in SSc support groups because some respondents were recruited through support groups. Thus, this proportion would not provide an accurate reflection of the proportion of Canadians with SSc who attend these groups.

The McGill University Institutional Review Board approved the study. Participants did not provide written informed consent because the survey was anonymous.

Data Analysis

Among participants who answered the question "Have you participated in SSc support groups?" with "No," we documented self-reported sociodemographic and disease-related characteristics. Frequencies of participants who responded 1) I'm not interested, 2) None are easily available, and 3) Other (please specify) were tallied. A content analysis approach was used to code the open-ended responses. This allowed data from the open-ended response option, "Other (please specify)," to be synthesized with data from the closed-ended response options.

Content analysis is a method used to codify interpretation of content from text data (Downe-Wamboldt, 1992; Graneheim & Lundman, 2004; Hsieh & Shannon, 2005). This method is most useful when existing theory or research literature on a phenomenon is limited because researchers can approach the data without preconceived categories, instead allowing categories

and themes to be generated from the data (Downe-Wamboldt, 1992; Graneheim & Lundman, 2004; Hsieh & Shannon, 2005). Since research on why people do not attend support groups is limited, this method was appropriate.

Two investigators independently analyzed participants' open-ended responses. First, responses were coded into categories and then categories were grouped into themes. To identify categories, responses were first read numerous times by each investigator in order to obtain a sense of the data as a whole. Next, one investigator read responses and highlighted statements that appeared to capture key thoughts or concepts. Then, the investigator re-read the responses to develop codes indicative of potentially significant categories and coded the data using these categories.

Following this, a second investigator independently read the responses and coded the data using the same categories. The two investigators discussed their coding in order to achieve consensus on categories and resolve any discrepancies in text codings. In cases where consensus was not reached, a third investigator was included. The two closed-ended response options, "I'm not interested" and "None are easily available," were coded as the categories "Not interested" and "No local support group," respectively. These codes were also used for open-ended responses that reflected category content.

After assigning response categories, the first investigator grouped the categories into themes. This process was repeated by the second investigator. Then, the assignment of categories into themes was discussed to resolve any uncertainties.

Results

Sample Characteristics

Altogether, 856 surveys were completed with most (n=669, 78%) done online. Of the 856 total surveys, 65 (8%) were classified as likely duplicates based on matching demographic data

and were excluded. This generally occurred when respondents began the online survey, submitted part of it, and subsequently started again. Of the 791 people who completed all or part of the survey, 88 (11%) were excluded because they did not report having been diagnosed with SSc by a healthcare provider; 19 (2%) because they answered demographic questions only; 62 (8%) because they were <18 years of age or did not report their age; 59 (7%) because they were not from Canada; 41 (5%) because they did not answer the question "Have you participated in SSc support groups?"; and 225 (28%) because they answered the question "Have you participated in SSc support groups?" with "Yes."

A total of 297 people answered the question "Have you participated in SSc support groups?" with "No". Sociodemographic characteristics of the sample are presented in Table 1.

Reasons for Non-participation

Of the 297 people who indicated they had not participated in SSc support groups, 3 (1%) did not provide a reason for non-participation and 14 (5%) provided a reason that was unclear (e.g., "I don't know"). Of the 280 respondents who provided a reason, 78 (28%) chose the response option "I'm not interested;" 95 (34%) chose the response option "None are easily available;" and 107 (38%) chose the response option "Other (please specify)".

An analysis of the text of the 107 open-ended responses resulted in 96 (90%) respondents receiving one code; 9 (8%) receiving two codes; and 2 (2%) receiving three codes. Thus, 120 coded responses were generated from the open-ended response option "Other (please specify)." Based on content analysis, 16 categories were generated to capture reasons for non-participation, including: 1) Not aware of support groups generally (n=30, 25%); 2) SSc symptoms not severe (n=13, 11%); 3) Other demands or too busy (n=12, 10%); 4) No need for support (n=10, 8%); 5) Newly diagnosed or diagnostic uncertainty (n=7, 6%); 6) Not aware of local support groups (n=7, 6%); 7) Discomfort facing others with SSc (n=6, 5%); 8) No local support group (n=6, 5%); 9)

Support groups too negative (n=6, 5%); 10) Already have alternative source of support (n=5, 4%); 11) Not ready (n=5, 4%); 12) SSc symptoms too severe (n=4, 3%); 13) Support groups not helpful (n=3, 3%); 14) Currently looking for information on support groups (n=2, 2%); 15) Not comfortable (n=2, 2%); and 16) Planning on attending (n=2, 2%).

Themes

Based on the 16 response categories, 7 themes were identified: 1) Not interested or no perceived need; 2) No local support group; 3) Lack of awareness of support groups; 4) Practical barriers; 5) Emotional factors; 6) Uncertainty and contemplation; and 7) Negative perceptions. Thematic groupings and corresponding response categories are provided in Table 2. Frequencies of responses for each theme include the total number of patient responses for included categories, including responses to the closed-ended (n=173) and open-ended (n=120) survey items.

Not interested or no perceived need (n=106, 36%). Seventy-eight (27%) people selected the closed-ended response option "I'm not interested." Thirteen (4%) people indicated they were in good health or experienced minimal symptoms and, therefore, did not feel the need to attend a support group. For example, one respondent mentioned being "very healthy." Ten (3%) people made general statements about not needing support without a more specific reason. Five (2%) respondents reported they were already receiving support through means other than a support group and did not require additional support (e.g., "I keep busy and receive support from my family and friends").

No local support group (n=101, 35%). This included people who selected the closed-ended response option "None are easily available" (n=95, 32%), and those who selected the open-ended response option "Other (please specify)" and then indicated there was no support group in their area (n=6, 2%).

Lack of awareness of support groups (n=37, 13%). Thirty (10%) people indicated they were not aware of support groups generally (e.g., "I didn't know they exist") and 7 (2%) indicated they were not aware of local support groups (e.g., "I don't know of any in my area"). The coding category "Not aware of local support groups" included in this theme and the category "No local support group" included in the previous theme differ in that the former included statements suggesting a lack of awareness about whether or not there may be a group, whereas the latter included statements that indicated a clear lack of availability.

Practical barriers (n=16, 6%). Twelve (4%) people indicated they were too busy with other commitments, such as family or work, to attend a support group (e.g., "I work and don't have time") and 4 (1%) indicated that they were too ill or disabled to participate in a support group.

Emotional factors (n=13, 4%). Six (2%) people indicated they did not attend a support group because they were afraid to interact with or see others with SSc who were worse off. For instance, one respondent mentioned, "It scares me to see myself in others." Five (2%) people made general statements about being emotionally unprepared to attend a support group, but did not elaborate further. Two (1%) respondents reported they were not comfortable attending a support group because they "preferred to deal with the condition privately" or due to concerns about social discomfort not necessarily related to SSc.

Uncertainty and contemplation (n=11, 4%). Seven (2%) people indicated they had been recently diagnosed with SSc or were in the process of being diagnosed with or learning about SSc and, therefore, had not attended a support group. For example, one respondent said, "When I find out more about my condition I will decide." Two (1%) respondents indicated they were attempting to learn more about support groups before deciding whether to attend (e.g., "I am

currently investigating") and 2 others (1%) mentioned they planned to attend a support group in the future.

Negative perceptions (n=9, 3%). The most frequent (n=6, 2%) negative perception mentioned by respondents was that the atmosphere or tone of support groups is negative (e.g., "Too depressing and fixated on the disease"). The second most frequent (n=3, 1%) negative perception reported was that attending a support group would not be helpful (e.g., "I don't feel it would benefit me").

Discussion

Among people with SSc who do not participate in SSc support groups, the most common reason for non-participation was not being interested in SSc support groups or not perceiving a need for additional support because of good health, minimal symptoms, or already receiving support through other means. The second most common reason was lack of availability of local SSc support groups, and the third most common reason was lack of awareness of the existence of SSc support groups. Other reasons for non-participation included: practical barriers, such as alternative commitments or being too ill or disabled; emotional factors, such as being afraid to interact with or see others with SSc; being recently diagnosed or attempting to learn more about SSc support groups before deciding whether to attend; and having negative perceptions about the tone or helpfulness of SSc support groups.

We were unable to identify previous studies examining reasons why people with a rare disease, such as SSc, do not attend illness-based support groups. However, we did identify two studies exploring factors that influence participation in cancer support groups (Ussher et al., 2008; Winefield et al., 2003). The first study (Ussher et al., 2008) used a combination of focus groups and telephone interviews to assess reasons for not attending cancer support groups among 26 patients with any form of cancer who had never attended a support group and who were

recruited through oncology clinics in Sydney, Australia. Reasons for non-attendance were categorized into individual and group factors. Individual factors included: resisting the position of "cancer patient;" personality factors, such as being an introvert or preferring to cope alone; and already having enough support. Group factors included: believing that support groups are negative places; lacking knowledge about what support groups involve; needing more in common with other group members than having cancer; and practical issues, such as the availability of the respondent, and the location and timing of the group.

In the second study (Winefield et al., 2003), authors interviewed 93 women with breast cancer recruited from oncology and radiology clinics at a hospital in Adelaide, Australia, of whom 55 reported that they did not plan to attend a support group. The most common reasons for not planning to attend included: currently having enough information or support; practical issues, such as the location and timing of the group; and not wanting to focus on having cancer. Other less commonly endorsed reasons included: being in good health; being too sick; disliking groups; and worrying about seeing others who are worse off.

Although the nature of SSc and cancer differ significantly, and SSc support groups are almost exclusively peer-led whereas cancer support groups are typically professionally led, the findings from the present study and the two studies of cancer patients have a number of similarities. In particular, all three studies found that many patients report already having good support networks and do not believe that they would benefit from attending a support group. While on one hand this type of response may reflect good existing support networks, it may also be the case that some people who do not attend support groups may not understand that these groups can address needs that other forms of support cannot, such as the ability to share experiences with others undergoing similar disease-related experiences. Additionally, the three

studies found that practical issues, such as the accessibility and timing of the groups, influence participation.

The most notable difference between the findings of the present study and the two studies of cancer patients is that in this study, a common reason for not participating in support groups was not being aware of the existence of SSc support groups in general or the existence of local SSc support groups. Lack of awareness was not identified as a reason for non-participation in cancer support groups. One possible explanation is that because SSc support groups are not available in many locations and are not typically available through healthcare settings, people who live in settings where there are no groups may not be aware that they exist at all. A second possible explanation is that since existing SSc support groups are almost all peer-organized and led, they may not be advertised as frequently or widely as support groups for common diseases, such as cancer, that are professionally led and provided through the healthcare system. A third possible explanation is that because SSc is a rare disease, many patients may not have the opportunity to meet other people with SSc who can tell them about the disease and helpful resources, such as support groups.

Findings from the present study suggest a number of ways in which SSc patient organizations may be able to address current limitations in the accessibility and effectiveness of SSc support groups. 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 (Osei, Lee, Modest, & Pothier, 2013; Winzelberg et al., 2003). 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 (van der Vaart et al., 2013).

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

There are several limitations that should be considered when interpreting the results of this study. First, people who participated in this study constitute a convenience sample of SSc patients. Specifically, recruitment occurred through CSRG physicians, national and provincial SSc organizations, and SSc support groups, which may have influenced the characteristics of respondents. Furthermore, the majority of survey dissemination and response collection was electronic, which may have also influenced the representativeness of the sample. Second, given the self-report nature of the survey, there is no way to be certain that all participants had SSc. Third, a majority of survey respondents did not know their SSc diagnosis subtype, likely because physicians do not often use this terminology with patients. Therefore, we were unable to explore whether the reasons for non-participation may have differed between people with varying degrees of symptom severity. Fourth, female and male participants were combined in this study, and it is possible that the reasons for non-participation may differ between sexes. Given the small number of men in this study, however, it was not possible to examine this. Fifth, most of our sample was White and, as such, our results may not be representative of people from different racial or ethnic

backgrounds. Sixth, we did not ask participants whether their physician had informed them about SSc support groups, which could be useful for understanding non-attending. Seventh, participants who answered the question "Have you participated in SSc support groups?" with "No" were required to choose one of three answer options and the two closed-ended response options did not allow respondents to elaborate or explain their choice. Thus, given the format of the survey, we were able to elicit a broad list of reasons for non-participation, but could not be certain about the relative proportion of reasons. Future studies using more systematic methods for assessing reasons for non-participation, such as survey methods that query all respondents about the importance of possible reasons for non-participation, are needed. The present study provides important background information that could be used to develop such a survey. Finally, this study did not explore the reasons why people do participate in scleroderma support groups. This is also an important topic for future studies.

In conclusion, peer-led support groups are an important resource for many SSc patients. However, there is limited research on the factors that may discourage them from attending these groups. This study found that many patients reported 1) they were not interested in support groups or did not perceive a need for support because they were in good health, experienced minimal symptoms, or already received support through means other than a support group; 2) there was not a support group available locally; or 3) they were not aware of the existence of support groups, generally, or of local groups. Other reasons for non-attendance included being too busy, being afraid to interact with or see others with SSc, being recently diagnosed or in the process of being diagnosed with SSc, and having negative perceptions about SSc support groups. These findings will inform SSc organizations on how they may be able 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=297)

Variable	
Female gender, *n (%)*	254 (85.5%)
Age in years, *M (SD)*	52.3 (13.9)
Race/ethnicity, *n (%)*[a]	
White	191 (84.9%)
Other	30 (13.3%)
White and other[b]	4 (1.8%)
Level of education, *n (%)*	
Less than high school	36 (12.1%)
High school graduate	179 (60.3%)
University graduate	82 (27.6%)
Marital status, *n (%)*	
Single	35 (11.8%)
Married	210 (70.7%)
Separated/divorced/widowed	52 (17.5%)
Primary spoken language, *n (%)*	
English	217 (73.1%)
French	80 (26.9%)
Working (full time or part time), *n (%)*	116 (39.1%)
Treated by rheumatologist, *n (%)*[c]	152 (51.5%)
Years since SSc diagnosis, *M (SD)*[d]	9.5 (10.0)

Note. For variables with data missing, the sample size is indicated in the footnotes.

[a]n=225; [b]Individuals who selected "White," as well as another race/ethnicity; [c]n=295; [d]n=296.

Table 2

Response Categories and Themes

Theme	Response Categories	n (%)	Definitions
Not interested or no perceived need	Not interested	78 (26.6%)	Selected the close-ended response option "I'm not interested."
	SSc symptoms not severe	13 (4.4%)	Any reference to an individual not needing a SSc support group because they are in good health or experience only minimal symptoms.
	No need for support	10 (3.4%)	Any reference to an individual not currently needing support.
	Already have alternative source of support	5 (1.7%)	Any reference to an individual currently receiving support through means other than a support group. Examples might include support from family, friends, and health care professionals.
No local support group	No local support group	101 (34.5%)	Selected the closed-ended response option "None are easily available" or selected the open-ended response option "Other (please specify)" and then indicated that there were no SSc support group in their area.
Lack of awareness of support groups	Not aware of support groups generally	30 (10.2%)	Any reference to an individual not being aware of the existence of SSc support groups in general.
	Not aware of local support groups	7 (2.4%)	Any reference to an individual not being aware of the existence of local SSc support groups.
Practical barriers	Other demands or too busy	12 (4.1%)	Any reference to an individual being too busy with other commitments, such as family or work.

	SSc symptoms too severe	4 (1.4%)	Any reference to an individual being unable to attend a SSc support group due to disability or the severity of their symptoms.
Emotional factors	Discomfort facing others with SSc	6 (2.0%)	Any reference to an individual being afraid to interact with or see others with SSc, such as people with a more severe diagnosis or more severe symptoms. Patients may express being afraid to see how bad the disease can get or what their future may hold.
	Not ready	5 (1.7%)	Any statement indicating that an individual is not yet emotionally prepared to attend a SSc support group.
	Not comfortable	2 (0.7%)	Any reference to an individual not being comfortable attending a SSc support group due to general concerns about social comfort not necessarily related to SSc (e.g., discomfort in groups).
Uncertainty and contemplation	Newly diagnosed or diagnostic uncertainty	7 (2.4%)	Any reference to an individual being recently diagnosed, in the process of being diagnosed, or just learning about SSc.
	Currently looking for information on support groups	2 (0.7%)	Any reference to an individual attempting to learn more about SSc support groups before deciding whether or not to attend.
	Planning on attending	2 (0.7%)	Any reference to an individual planning on attending a SSc support group in the near or distant future.
Negative perceptions	Support groups too negative	6 (2.0%)	Any reference to the tone of SSc support groups being negative.
	Support groups not helpful	3 (1.0%)	Any reference to the idea that an individual does not believe that attending a

SSc support group would be helpful.

Chapter 6

General Discussion

Summary of Main Findings

The aim of the first manuscript in this thesis, a systematic review, was to determine if training programs for peer facilitators of illness-based support groups existed and, if so, to examine the effect of these programs on 1) the competency and self-efficacy of group facilitators and 2) self-efficacy for disease management, health outcomes, and satisfaction with the support group experience among group members. The systematic review did not find any randomized controlled trials (RCTs) that compared training programs for peer facilitators of illness-based support groups to no training comparators, but did identify one trial that evaluated the effects of alternative training program resources (Zordan et al., 2015). That study evaluated the confidence and self-efficacy of facilitators of cancer support groups randomized to 4-month long high-resource (website, discussion forum, 2-day face-to-face training) and low-resource (website, discussion forum) interventions, and did not find evidence that the high-resource intervention was more effective than the low-resource intervention. However, that study had signification limitations, including low power, unclear or high risk of bias ratings in numerous domains, and lack of information about the intervention and its delivery. The findings of the review highlight the need for training programs for peer facilitators of illness-based support groups and for well-designed and executed trials that evaluate whether these programs improve outcomes among group facilitators and members.

The objective of the second manuscript, a scoping review, was to explore the 1) benefits of participating in rare disease support groups and 2) facilitators and barriers of initiating and sustaining these groups. Ten publications were included in the scoping review (Alderson et al., 2004; Barker, 2009; Breau & Norman, 2003; Castro et al., 2008; Howard et al., 2003; Jalovcic &

Pentland, 2009; Moore et al., 2008; Stewart et al., 2001; Telfair & Gardner, 2000; Vitucci, 2012); all ten publications reported on the benefits of participating in rare disease support groups and three of these also reported on barriers and facilitators of initiating and sustaining them (Castro et al., 2008; Moore et al., 2008; Stewart et al., 2001). Commonly reported perceived benefits of rare disease support groups included meeting and befriending other people with the same disease and similar experiences, learning about the disease and related treatments, and giving and receiving emotional support. In addition, several facilitators (e.g., meeting via teleconference; providing leaders with training for their roles; having more than one leader) and barriers (e.g., getting patients and/or family members to lead the support group; navigating difficult group discussions; uncertainty about one's role as a leader) of initiating and sustaining rare disease support groups were identified. The findings of the review suggest that support groups are an important resource for people with rare diseases and that training programs for peer facilitators of rare disease support groups, such as scleroderma (or systemic sclerosis, SSc) support groups, could be useful in addressing challenges and barriers to establishing and maintaining these groups.

The third manuscript sought to identify the training and support needs of SSc support group facilitators. That study found that SSc support group facilitators were generally confident in their ability to complete tasks related to 1) Organizing, structuring, and facilitating the group; 2) Addressing individual member needs and diversity of the group; 3) Helping members cope with grief and loss; and 4) Attaining and responding to member feedback. However, they reported lower confidence in their ability to perform important tasks related to 1) Managing difficult group dynamics; 2) Promoting and sustaining the group; and 3) Balancing personal and group needs. While these findings were consistent with those of previous studies that have explored the challenges reported by facilitators of illness-based support groups, such as cancer support groups (Butow et al., 2005; Kirsten, Butow, Price, Hobbs, & Sunquist, 2006; Zordan et

al., 2010), it is possible that these challenges are magnified for facilitators of rare disease support groups, such as SSc support groups, who often face added logistical problems (e.g., low patient numbers) and limited support from healthcare and patient organizations, which are not as well resourced as organizations for people with more common diseases. Altogether, these findings underscore the importance of educating and training SSc support group facilitators for their group leader roles. Moreover, they highlight areas of focus for the Scleroderma Support group Leader EDucation (SSLED) Program.

Finally, the fourth manuscript sought to identify the reasons why people with SSc do not participate in SSc support groups. That study found that the main reasons for not participating in SSc support groups included 1) Not interested or no perceived need; 2) No local support group available; 3) Lack of awareness of the existence of SSc support groups; 4) Practical barriers; 5) Emotional factors; 6) Uncertainty about whether to attend; and 7) Negative perceptions about support groups. We were unable to find previous studies examining reasons why people with rare diseases, such as SSc, do not attend support groups. The findings of the study suggest a number of possible ways to address current limitations in the accessibility and effectiveness of SSc support groups; for instance, an educational training program for peer facilitators of SSc support groups (i.e., the SSLED Program) could increase the accessibility of these groups by providing SSc patients with the knowledge and skills necessary to successfully establish and manage support groups where none presently exist. Training peer facilitators of SSc support groups could also address many concerns and misperceptions that SSc patients have about support groups (e.g., too negative; unhelpful) and thus, improve the ability of these groups to meet patients' needs.

Implications of Findings and Directions for Future Research

People living with chronic medical illnesses employ numerous coping strategies to help them manage the physical, emotional, and psychosocial consequences of their disease. According

to Lazarus and Folkman's theoretical model of stress and coping (1984; Biggs, Brough, & Drummond 2017), these strategies can be grouped into 1) problem-focused coping, 2) emotion-focused coping, and 3) meaning-focused coping. The aim of these strategies is to modify the source of stress (i.e., problem-focused coping); to minimize or prevent negative emotional responses to the stressor (i.e., emotion-focused coping); or to focus on the positives or look for meaning within the stressor (i.e., meaning-focused coping).

The findings of the present research highlight the importance of rare disease support groups and suggest that attending support groups is one way patients manage the stress of their disease. More specifically, the perceived benefits of participating in support groups are varied, as groups provide patients with opportunities to engage in problem-focused coping strategies (e.g., learning about the disease, its treatment, and symptom management techniques), emotion-focused coping strategies (e.g., meeting and befriending other patients and obtaining support from them), and meaning-focused strategies (e.g., providing emotional and practical support to other patients and advocating to improve healthcare for them). Establishing and maintaining rare disease support groups, however, involves significant challenges. Many people with rare diseases, such as SSc, are not able to access support groups and many support groups that are initiated are not sustained due to a number of shortcomings, some of which are related to untrained patient leaders; for example, some SSc patients report they prefer not to attend support groups because the group in their area is poorly organized or is overly negative and not constructive.

Findings from this research suggest that rare disease organizations may be able to increase the accessibility and effectiveness of rare disease support groups by implementing training programs for facilitators of these groups. Training programs for peer facilitators of rare disease support groups could improve the availability of support groups by giving people with rare diseases the knowledge and skills they need to set up groups where none exist. In addition, such

programs could support the ability of peer facilitators of rare disease support groups to lead effective, sustainable support groups and reduce the emotional and physical toll on leaders.

Our research team has partnered with key SSc patient organizations, including the Scleroderma Society of Canada and the Scleroderma Foundation in the United States, in order to develop the SSLED Program and address limitations of present SSc support groups. The findings from the studies included in this thesis have greatly informed these efforts. The SSLED Program will be a 3-month-long, group training program that will be delivered using videoconferencing in order to provide peer facilitators of SSc support groups with the information and skills necessary to consistently and effectively carry out their leadership roles. The program will utilize a problem-based learning approach. Problem-based learning is a learner-centered approach that integrates theory and practice by providing the necessary knowledge and skills, presenting a complex, real-world problem, and then working to identify an approach to solving the problem (Hmelo-Silver, 2004).

To implement this, the SSLED Program will be comprised of several modules, or learning sessions. Each session will introduce a topic and provide an overview of key information. Example topics include the leader's role; successful support group culture; starting a support group; structuring support group meetings; providing educational material on SSc; grief and crisis; managing group dynamics; resources for leaders and members; and group continuity. This will be followed by videos that show SSc support group facilitators faced with a problem or situation similar to those that training group participants may encounter in their role as a leader. Then, there will be a guided discussion among training group participants about possible approaches and solutions to the problems faced. Given the absence of well-designed and conducted RCTs examining the effect of training programs for peer facilitators of illness-based support groups, future research should consider conducting methodologically robust trials on how

to implement training programs for peer facilitators of illness-based support groups, such as the SSLED Program, and whether such programs improve the experiences of support group facilitators and members.

Limitations

A number of limitations should be considered when interpreting the findings of the present research. For example, both manuscripts 1 and 2 had important limitations that could restrict the inferences that could be drawn from these reviews. First, manuscript 1 did not find any RCTs that compared training programs for peer facilitators of illness-based support groups to no training comparators, and the one trial that was included in the systematic review had significant limitations (e.g., underpowered; unclear or high risk of bias ratings in numerous domains; lack of information about the intervention and its delivery). Similarly, manuscript 2 did not identify any RCTs on the benefits and possible harms of rare disease support groups, and the ten studies that were included in the scoping review were relatively small studies that were done in different rare disease and used different methodologies. Second, although we had data extraction tools and two different investigators extracting data independently for both reviews, extracting accurate and complete data remained a challenge because many of the studies included in the reviews reported methods or results that were incomplete or unclear. Third, because we restricted our search to CINAHL and PubMed in the scoping review (manuscript 2), it is possible that we may have missed important articles published in the grey literature, such as information from rare disease organization webpages.

In addition, manuscripts 3 and 4 also had notable limitations that could influence the generalizability of the results of these studies. First, participants in both of the studies constituted a convenience sample of people living with SSc. Specifically, participants in manuscript 3 were recruited through national and provincial SSc organizations, SSc-related chat-rooms and

newsletters, and emails to support group facilitators across Canada and the United States, and those in manuscript 4 were recruited through Canadian Scleroderma Research Group physicians, national and provincial SSc organizations, and SSc support groups. Since this may have influenced the characteristics of respondents, it is possible that the results of the studies are not applicable to all SSc patients. Furthermore, given the small number of men, younger patients, and non-White patients in the studies, it was not possible to examine if training and support needs or reasons for non-participation in SSc support groups differed between patient subgroups.

Lastly, another limitation of manuscript 3 was that the grouping of items into themes was done on the basis of content because the sample was too small to conduct a factor analysis. However, the item groupings and themes were reviewed iteratively by all members of the research team, which 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. Thus, we felt confident that thematic groupings provided a reasonable structure for interpreting item responses.

Conclusion

People living with rare diseases experience many of the same challenges as people with more common medical diseases, but also face unique challenges, including gaps in knowledge about their disease, difficulty obtaining an accurate diagnosis, and limited treatment and support options (European Organisation for Rare Diseases, 2005, 2009). Professionally organized and delivered support services that are commonly provided to people with more common diseases, are not typically available for people with rare diseases (Kwakkenbos et al., 2013). So, many rare disease patients have organized peer-facilitated support groups to help them cope with and manage their disease (Kwakkenbos et al., 2013; Reimann et al., 2007).

SSc is a rare, chronic, multi-system connective tissue disorder characterized by abnormal fibrotic processes and excessive collagen production (Mayes, 2008; Seibold, 2004). The presentation of SSc can be extremely heterogeneous and its course is highly unpredictable (Mayes, 2008; Seibold, 2004). Thus, many people with SSc join support groups in order to learn how to better manage emotional and physical aspects of living with the disease (Kwakkenbos et al., 2015). There are approximately 200 SSc support groups across Canada and the United States and most of them are facilitated by patients (Scleroderma Canada, n.d.; Scleroderma Foundation, n.d.). In order to improve the accessibility and effectiveness of SSc support groups, the Scleroderma Society of Canada and the Scleroderma Foundation in the United States have partnered with our research team to develop the SSLED Program, an educational training program for peer facilitators of these groups.

The findings of the present research suggest that support groups are an important resource for people with rare diseases like SSc. In addition, they suggest that training programs for peer facilitators of rare disease support groups, such as the SSLED Program for facilitators of SSc support groups, could possibly address challenges and barriers to establishing and sustaining these groups. More specifically, we learned that many people with SSc do not have access to SSc support groups. By providing SSc patients with the knowledge and skills necessary to successfully establish and manage support groups where none exist, the SSLED Program could address this limitation and improve the availability of SSc support groups. In addition, we learned that other barriers to participation in SSc support groups include being afraid to interact or see others with SSc or having negative perceptions about these groups. By training SSc support group facilitators to address these concerns, the SSLED Program could improve the ability of SSc support groups to meet the needs of patients. Finally, we learned that SSc support group facilitators consistently reported lower confidence in their ability to perform tasks related to 1)

Managing difficult group dynamics; 2) Promoting and sustaining the group; and 3) Balancing personal and group needs. This has been taken this into account when developing the SSLED Program and as such, program modules will focus on providing leaders with skills in these and other areas in order to improve the experience of leaders and members.

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