Psychosocial aspects of scleroderma

Linda Kwakkenbos, PhD[1,2]; Vanessa C. Delisle, MSc[1,5]; Rina S. Fox, MS, MPH[9]; Shadi Gholizadeh, MSc[9]; Lisa R. Jewett, MSc[1,5]; Brooke Levis, MSc[1,4]; Katherine Milette, MA[1,5]; Sarah D. Mills, MS[9]; Vanessa L. Malcarne, PhD[8,9]; Brett D. Thombs, PhD[1-7]

[1]Lady Davis Institute for Medical Research, Jewish General Hospital, Montréal, Québec, Canada; [2]Departments of Psychiatry, [3]Medicine, [4]Epidemiology, Biostatistics, and Occupational Health, [5]Educational and Counselling Psychology, [6]Psychology, and [7]School of Nursing, McGill University, Montréal, Québec, Canada; [8]Department of Psychology, San Diego State University, San Diego, California, USA; [9]San Diego State University/University of California, San Diego Joint Doctoral Program in Clinical Psychology, San Diego, California, USA.

Corresponding Author:

Linda Kwakkenbos, PhD

Jewish General Hospital

4333 Cote Ste Catherine Road

Montreal, Quebec H3T 1E4

Tel (514) 340-8222 ext. 8578

Fax (514) 340-8124

E-mail: kwakkenbosl@gmail.com

Disclosure statement: The authors have no conflicts of interest to disclose.

Acknowledgements: This work was supported by a grant from the Canadian Institutes of Health Research (CIHR; #TR3-267681). Dr. Kwakkenbos was supported by a Fonds de la Recherche en Santé Québec (FRSQ) postdoctoral fellowship. Ms. Delisle, Ms. Jewett, and Ms. Milette were supported by CIHR Doctoral Research Awards. Ms. Levis was supported by a FRSQ Doctoral Training Award. Dr. Thombs was supported by an Investigator Salary Award from the Arthritis Society.

KEY WORDS

scleroderma; systemic sclerosis; psychosocial; quality of life; self-management

SYNOPSIS

Patients with systemic sclerosis (SSc; scleroderma) have to cope with not only the physical impacts of the disease, but also the emotional and social consequences of living with the condition. Because there is no cure for SSc, improving quality of life is a primary focus of treatment, and an important clinical challenge. The present review summarizes significant problems faced by patients with SSc, including depression, anxiety, fatigue, sleep disruption, pain, pruritus, body image dissatisfaction, and sexual dysfunction, and describes options to help patients cope with the consequences of the disease.

KEY POINTS BOX

- Symptoms of SSc, including fatigue, pain, pruritus, sleep problems, and sexual impairments negatively influence quality of life in many patients, and may lead to emotional consequences such as depression, anxiety, and body image distress due to appearance changes.
- Providing accessible information to SSc patients regarding problems common to people living with the disease, as well as information regarding useful resources and services to address these problems, can help SSc patients and can be easily implemented by health care professionals.
- In addition to referrals for professional health care interventions, low-intensity strategies such as self-management programs and support groups, may be helpful to some patients with SSc.

- Future research should focus on the development and testing of interventions designed specifically to meet the educational and psychosocial needs of patients with SSc.

INTRODUCTION

Systemic sclerosis (SSc, or scleroderma) has far-reaching consequences for physical health, as well as emotional and social well-being.[1-4] Because there is no known cure for the disease, SSc treatment focuses on reducing symptoms and disability, and improving health-related quality of life (HRQL). The objectives of the present review are to summarize the impact of SSc on common patient-reported problems associated with HRQL and to describe potential interventions to support coping with the consequences of the disease.

Depression

Depression involves symptoms that may include sadness, loss of interest or pleasure, feelings of guilt or low self-esteem, poor concentration, and disturbed sleep or appetite. A study of 345 SSc patients enrolled in a Canadian registry reported the prevalence of major depressive disorder (MDD) was of 4% for the past 30-days, 11% for the past 12-months, and 23% for lifetime.[5] A French study of 50 hospitalized SSc patients and 50 SSc patients who attended a patient organization meeting found that 19% had current MDD and 56% had lifetime MDD, and rates were higher in hospitalized (28% current) versus non-hospitalized (10% current) patients.[6] Depression is substantially more common in SSc than in the general population and may be more prevalent than in other rheumatic diseases.[5] Many patients with SSc and other chronic diseases who meet criteria for MDD at a given time point, however, may not meet criteria consistently. In the Canadian sample, for instance, only 3 of 12 patients with SSc who had a current major depressive episode at baseline met diagnostic criteria one month later.[7] Some episodes may be time-limited and may resolve without targeted intervention or treatment. Others may reflect ongoing moderate symptoms that only variably meet criteria for formal diagnosis. Many SSc patients describe ongoing emotional distress from the burden of living with the disease, but

differentiate this qualitatively from what they would consider clinical depression.[8] Cross-sectionally, factors associated with symptoms of depression in SSc include greater overall disease burden, which may involve degree of gastrointestinal involvement, breathing problems, skin involvement, and tender joints.[9-11]

Anxiety and Fear

Anxiety can be a normal reaction to stress; however, it may also lead to mental health problems when experienced in excess. To date, only one study documented the prevalence of anxiety disorders among SSc patients. In that study, 49% of 50 hospitalized patients and 50 patients who attended a patient meeting had at least one current anxiety disorder, and 64% met criteria for at least one anxiety disorder in their lifetime. Social anxiety and generalized anxiety disorder were the most common.[6] There was no difference in prevalence between hospitalized and non-hospitalized patients.

The course of SSc is highly unpredictable and patients may perceive the future as uncertain. For patients, worry about the future, including fear of disease progression, fear of becoming physically disabled, and fear of being dependent upon others are important sources of stress.[12,13] Because SSc is unpredictable and associated with serious consequences, these concerns are realistic and in themselves do not represent anxiety disorders, for which irrational fear is typically a central component.[13] Nonetheless, fear of progression can impact HRQL substantially. In a cross-sectional study of 215 SSc patients from the Netherlands, for instance, fear of progression was highly associated with symptoms of depression.[13]

Fatigue

Fatigue from a chronic medical disease is characterized by persistent exhaustion that is disproportionate to exertion and not relieved by rest.[14] Fatigue is the most commonly experienced symptom of SSc and has a substantial impact on HRQL, as well as the ability to carry out daily activities, including work.[1,4,15-17] In one Canadian study, 89% of 464 SSc patients reported fatigue at least some of the time, and 81% of these patients indicated that fatigue had at least a moderate impact on their daily function.[15] Levels of fatigue in SSc are similar to those experienced by patients with other rheumatic diseases and cancer patients undergoing active treatment.[18] Cross-sectionally, greater fatigue in SSc is associated with increased medical comorbidities, current smoking, pain, breathing problems, and gastrointestinal symptoms.[14] Longitudinally, fatigue severity has been associated with pain, severity of gastrointestinal involvement, and psychological variables, specifically ineffective coping skills.[19]

Sleep

Significant sleep disruption is common in SSc and has broad implications for patients.[15,20-22] A polysomnography study of 27 SSc patients found that, compared to age-adjusted norms, SSc patients had reduced sleep efficiency and rapid eye movement sleep, as well as increased arousal and slow wave sleep.[20] Sleep disruption was associated with esophageal dyskinesia and dyspnea, which are common complications of SSc, as well as restless legs syndrome.[20] Observational studies have linked dyspnea, pain, fatigue, pruritus, gastrointestinal symptoms, and depressive symptoms with self-reported poor sleep quality and sleep disruption in SSc.[21,22]

Pain

Between 60 and 83% of SSc patients report experiencing pain at any given time, and pain levels in SSc are similar to levels reported in chronic pain and rheumatic conditions.[15,23-24] Pain in

SSc is associated with reduced HRQL, functional disability, work disability, sleep problems, and symptoms of depression.[22-25] Patients with SSc describe their pain as both localized and generalized in quality,[26] and sources can include pain from Raynaud's phenomenon, gastrointestinal pain, joint and musculoskeletal pain, skin pain, and pain due to calcinosis and ulcers.[1,23,26] Pain ratings are higher among patients with diffuse SSc compared to patients with limited disease, although this difference is generally small.[23] Based on patient report, overall pain levels are associated with sleep problems, fatigue, and symptoms of depression, as well as physical function, reduced ability to carry out daily activities, work disability, and poorer HRQL.[22,23,25,27] How patients with SSc describe their pain and the degree to which they believe they can manage it often reflects psychosocial factors, which should be considered in assessment and intervention, especially as related to pharmacologic treatment.[28]

Pruritus

Pruritus, or itch, is common in SSc and is associated with HRQL, even after controlling for sociodemographic and other SSc symptom variables.[29,30] Overall, 43% of 959 SSc patients from a Canadian registry reported pruritus on most days in the last month.[31] This rate was slightly higher, but not statistically significant, among patients with early SSc (< 5 years since onset of non-Raynaud's symptoms; 46%) versus those with longer disease duration (≥ 5 years; 41%).[31] The presence of pruritus is more common among patients with greater skin and gastrointestinal involvement.[31]

Body Image

Acquired disfigurement from an injury or medical illness is often linked to problems with body image, including social avoidance.[32] Appearance changes in highly visible areas of the body,

particularly the face and hands, are common in patients with SSc and contribute to body image distress, which in turn can be associated with symptoms of anxiety and depression.[1,4,32-36] A number of cross-sectional studies have reported that appearance changes of the face, including changes to the mouth, as well as hand involvement, including skin thickening, have been consistently related to body image distress, including dissatisfaction with appearance, decreased appearance self-esteem, and symptoms of anxiety and depression.[32-36] Other appearance changes, including telangiectasias, may also be associated with body image distress.[36,37] Social discomfort due to changes in appearance is also related to age.[36] Younger patients, for whom the importance of meeting new people and developing intimate relationships is more pronounced, may experience a greater negative impact of appearance changes on social relationships.[36]

Sexual Function

Sexual dysfunction is a common problem among women with SSc.[1,38-43] Compared to women in the general population, women with SSc are significantly less likely to be sexually active, and sexually active women with SSc are significantly more likely to be sexually impaired.[39,40,42] Factors that are independently associated with being sexually active include younger age, fewer gastrointestinal symptoms, and less severe Raynaud's phenomenon symptoms.[41] Among women who are sexually active, sexual impairment is associated with older age, as well as more severe skin involvement and breathing problems. Vaginal pain is eight times as common among women with impairment compared to those without.[41]

Among men with SSc, erectile dysfunction (ED) is common with onset typically occurring several years after the manifestation of the first non-Raynaud's symptoms.[44-46] While in the general population, ED is typically associated with atherosclerosis, in SSc, penile blood flow is impaired due to both myointimal proliferation of small arteries and corporal fibrosis.[46] Men with

SSc who have ED are significantly more likely to be older than those without ED and tend to report non-SSc risk factors (e.g., alcohol consumption) at higher rates.[45,46] SSc factors associated with ED include severe cutaneous, muscular, or renal involvement, diffuse disease, elevated pulmonary pressures, restrictive lung disease, endothelial dysfunction, and microvascular damage.[45-47] Most men with SSc who have ED do not receive treatment.[45] Among those who do, sildenafil appears to be commonly used, but its efficacy has not been established in SSc.[44,45]

CLINICAL MANAGEMENT

In addition to the core medical treatment of SSc-related symptoms, providing services and interventions to help manage the psychological, behavioral, and social aspects of living with the disease is an important component of patient-centered care. There are challenges related to the development, testing, and delivery of such patient-centered interventions in SSc, however, including the small number of patients and limited resources available.[2,3] Nonetheless, these types of psychosocial interventions have proven to be effective in reducing disability and improving HRQL in more common conditions, including rheumatic diseases,[48,49] and can be reasonably implemented in SSc.

Stepped-care models for psychological, behavioral, and educational interventions involve matching interventions of differing intensities to patient needs. Generally, stepped-care starts off with the simplest, least intrusive intervention and proceeds to more intense treatment approaches as deemed necessary.[1] Self-help can be a very useful first step towards addressing relatively mild problems associated with psychosocial functioning and HRQL. Providing accessible information to both patients and those who support them regarding issues common to people living with SSc, as well as information regarding useful resources and services to address such problems, is something that can be easily implemented by health care professionals. Health care professionals

should be aware of the important concerns that affect HRQL in order to help patients access appropriate resources and facilitate conversations that address concerns of individual patients. Additionally, links to other sources of information can be provided in clinics regarding self-help programs that are available as a first step in providing psychosocial support.

In the context of stepped care, low-intensity interventions are appropriate for people with less severe psychological concerns. However, referral to professional services is needed for patients with more complex or serious psychological problems such as severe depression, or when lower intensity methods do not work well. For these patients, a focused evaluation of their psychological symptoms and more intense intervention services provided by a mental health professional are often required.

Low-intensity strategies that have been proven effective to improve coping with the consequences of more common chronic diseases, and that may be readily available to many patients with SSc, include self-management programs and support groups.[48,49]

Self-management

Across more prevalent diseases, supportive care programs, such as self-management programs, are increasingly included as core components of patient-centered care. Although there is currently no gold-standard definition, the term "self-management" has been defined as the ability of patients to manage the symptoms, treatment, physical, and psychological consequences, and lifestyle changes inherent in living with a chronic condition. Effective self-management involves the ability to monitor one's condition and to have the cognitive, behavioral, and emotional responses necessary to maintain a satisfactory HRQL.[50]

In more prevalent diseases, such as arthritis, asthma, and diabetes, self-management interventions have been shown to provide benefits to participants in terms of knowledge,

performance of self-management behaviors, self-efficacy, and health status.[50] A Cochrane systematic review (17 trials, $N = 7{,}442$)[49] found that low-cost self-management programs lead to small improvements in participants' self-efficacy to manage their disease, which is the principal target of the programs, as well as improvements in self-rated health status (e.g., pain, fatigue, disability) and some health behaviors (e.g., exercise, cognitive symptom management). No randomized controlled trials (RCTs), however, have investigated the efficacy of self-management interventions for SSc. Two pre-post intervention studies of SSc self-management programs have been conducted.[51,52] One described a mail-delivered self-management program provided to 49 patients with SSc,[51] and the other was a pilot study of an internet-delivered self-management program with 16 SSc patients.[52] The small sample sizes in these studies, however, limit the ability to draw conclusions about effectiveness. In addition, the Scleroderma Patient-centered Intervention Network (SPIN) is developing an online self-management program.[2,3] However, there are currently no SSc-specific programs readily available to patients. An alternative is a general disease self-management program, such as the Chronic Disease Self-Management Program from Stanford University, which is available via the Internet. The Chronic Disease Self-Management Program, which is designed to teach self-care techniques useful to persons with many chronic diseases, improved self-efficacy for disease management and overall health status in an RCT that included patients with heart and lung disease, as well as diabetes.[53]

Support Groups

A large number of patients with chronic medical illnesses, including those with SSc, join support groups in order to better cope with and manage their illness.[54] Activities of support groups include giving and receiving emotional and practical support, as well as providing education and information to patients. The specific activities and focus, as well as facilitator training and

competence, may vary across support groups, which are typically organized locally. Patients may differ in the acceptability of the idea of attending a support group and the degree to which they may benefit from one. Due to their grass-roots nature, support groups can be configured in a variety of ways.[55] For example, some support groups may meet face-to-face whereas others may "meet" online; some groups may be facilitated by a peer whereas others may be facilitated by a professional; and some may include structured educational activities whereas others may not. Research on the effectiveness of support groups is scant, particularly with regard to lay-led groups. However, many people who do attend support groups describe feeling more empowered, more hopeful, and less alone following their group experience.[54] In addition, some patients who attend these groups report feeling more in control of their life, as well as more knowledgeable about their illness, coping strategies, and developments in medical and self-help treatments.

Peer-led support groups have become increasingly popular in recent years.[56] Consistent with this, most SSc support groups are peer-led rather than professionally-led. No studies, however, have examined the effectiveness of these groups on psychosocial or other coping-related outcomes. Given that they may be the sole source of SSc-specific support available to many patients and that they have been effective in other conditions, attending support groups may be beneficial for some patients with SSc. It is important to keep in mind, however, that support groups are meant to complement rather than supplement standard medical care. Medical professionals may want to discuss the possible benefits of attending a support group with their patients, as well as potential pitfalls. For patients who are interested in attending or joining a SSc support group, information can typically be found on local or national organization websites, such as the Scleroderma Society of Canada and the Scleroderma Foundation in the United States.[57,58]

FUTURE CONSIDERATIONS AND SUMMARY

Attention and research regarding the psychological aspects of living with SSc have increased in recent years.[1,4] Furthermore, as the low prevalence of SSc sometimes precludes high-quality research in this area, the substantial progress that has been made in establishing international research networks is promising and provides a solid foundation for future research endeavors.[2,3] Future research should focus on gaining a better understanding of the psychosocial aspects of SSc, their interconnectedness, and the valid measurement of outcomes related to this, as well as on moving forward the development and testing of interventions that help patients with SSc cope with the everyday challenges related to living with their disease.

REFERENCES

1. Thombs BD, van Lankveld W, Bassel M, et al. Psychological health and well-being in systemic sclerosis: State of the science and consensus research agenda. *Arthritis Care Res*. 2010;62:1181-9.
2. Thombs BD, Jewett LR, Assassi S, et al. New directions for patient-centered care in scleroderma: the Scleroderma Patient-centered Intervention Network. *Clin Exp Rheum*. 2012;30:23-9.
3. Kwakkenbos L, Jewett LR, Baron M, et al. The Scleroderma Patient-centered Intervention Network (SPIN) Cohort: Protocol for a cohort multiple randomised controlled trial (cmRCT) design to support trials of psychosocial and rehabilitation interventions in a rare disease context. *BMJ Open*. 2013;3:e003563.
4. Malcarne VL, Fox RS, Mills SD, Gholizadeh S. Psychosocial aspects of systemic sclerosis. *Curr Opin Rheumatol*. 2013;25:707-13.

5. Jewett LR, Razykov I, Hudson M, Baron M, Thombs BD, Canadian Scleroderma Research Group. Prevalence of current, 12-month and lifetime major depressive disorder among patients with systemic sclerosis. *Rheumatology*. 2014;53:1719.
6. Baubet T, Ranque B, Taieb O, et al. Mood and anxiety disorders in systemic sclerosis patients. *Presse Med*. 2011;40:e111-9
7. Thombs BD, Jewett LR, Kwakkenbos L, Hudson M, Baron M, the Canadian Scleroderma Research Group. Major depression diagnoses are often transient among patients with systemic sclerosis: Baseline and 1-month follow-up. *Arthritis Care Res*. 2015. In press.
8. Newton EG, Thombs BD, Groleau D. The experience of emotional distress among women with scleroderma. *Qual Health Res*. 2012;22:1195-1206.
9. Thombs BD, Hudson M, Taillefer SS, et al. Prevalence and clinical correlates of symptoms of depression in patients with systemic sclerosis. *Arthritis Rheum.* 2008;59:504-509.
10. Bodukam V, Hays RD, Maranian P, et al. Association of gastrointestinal involvement and depressive symptoms in patients with systemic sclerosis. *Rheumatology*. 2011;50:330-334.
11. Milette K, Hudson M, Baron M, Thombs BD, Canadian Scleroderma Research Group. Comparison of the PHQ-9 and CES-D depression scales in systemic sclerosis: Internal consistency reliability, convergent validity and clinical correlates. *Rheumatology*. 2010;49:789-796.
12. Van Lankveld WGJM, Vonk MC, Teunissen HA, van den Hoogen FHJ. Appearance self-esteem in systemic sclerosis – subjective experience of skin deformity and its relationship with physician-assessed skin involvement, disease status and psychological variables. *Rheumatology*. 2007;46:972–976.
13. Kwakkenbos L, van Lankveld WGJM, Vonk MC, Becker ES, van den Hoogen FHJ, van den Ende CHM. Disease-related and psychosocial factors associated with depressive symptoms in

patients with systemic sclerosis, including fear of progression and appearance self-esteem. *J Psychosom Res* 2012;72:199-204.

14. Thombs BD, Hudson M, Bassel M, Taillefer SS, Baron M, Canadian Scleroderma Research Group. Sociodemographic, disease, and symptom correlates of fatigue in systemic sclerosis: Evidence from a sample of 659 Canadian Scleroderma Research Group Registry patients. *Arthritis Rheum*. 2009;61:966-973.
15. Bassel M, Hudson M, Taillefer SS, Schieir O, Baron M, Thombs BD. Frequency and impact of symptoms experienced by patients with systemic sclerosis: Results from a Canadian National Survey. *Rheumatology*. 2011;50:762-767.
16. Sandqvist G, Scheja A, Hesselstrand R. Pain, fatigue and hand function closely correlated to work ability and employment status in systemic sclerosis. *Rheumatology*. 2010;49:1739-1746.
17. Sandusky SB, McGuire L, Smith MT, Wigley FM, Haythornthwaite JA. Fatigue: an overlooked determinant of physical function in scleroderma. *Rheumatology, 48,* 165-169.
18. Thombs BD, Bassel M, McGuire L, Smith MT, Hudson M, Haythornthwaite JA. A systematic comparison of fatigue levels in systemic sclerosis with general population, cancer and rheumatic disease samples. *Rheumatology.* 2008;47:1559-1563.
19. Assassi S, Leyva AL, Mayes MD, et al. Predictors of fatigue severity in early systemic sclerosis: A prospective longitudinal study of the GENISOS cohort. *PLOS ONE*. 2011;6:e26061.
20. Prado GF, Allen RP, Trevisani VMF, Toscano VG, Earley CJ. Sleep disruption in systemic sclerosis (scleroderma) patients: Clinical and polysomnographic findings. *Sleep Med.* 2002;3: 341-345.

21. Frech T, Hays RD, Maranian P, Clements PJ, Furst DE, Khanna D. Prevalence and correlates of sleep disturbance in systemic sclerosis – results from the UCLA scleroderma quality of life study. *Rheumatology*. 2011;50:1280-1287.
22. Milette K, Hudson M, Korner A, Baron M, Thombs BD, Canadian Scleroderma Research Group. Sleep disturbances in systemic sclerosis: Evidence for the role of gastrointestinal symptoms, pain, and pruritus. *Rheumatology*. 2013;52;1715-1720.
23. Schieir O, Thombs BD, Hudson M, et al. Prevalence, severity, and clinical correlates of pain in patients with systemic sclerosis. *Arthritis Care Res*. 2010;62:409 –17.
24. El-Baalbaki G, Lober J, Hudson M, et al. Measuring pain in systemic sclerosis: Comparison of the short-form McGill Pain Questionnaire versus a single-item measure of pain. *J Rheumatol*. 2011;38:2581-2587.
25. Hudson M, Steele R, Lu Y, Thombs BD, Canadian Scleroderma Research Group, Baron M. Work disability in systemic sclerosis. *J Rheumatol*. 2009;36:2481-2486.
26. Suarez-Almazor ME, Kallen MA, Roundtree AK, Mayes M. Disease and symptom burden in systemic sclerosis: A patient perspective. *J Rheumatol*. 2007;34:1718-26.
27. Benrud-Larson LM, Haythornthwaite JA, Heinberg LJ et al. The impact of pain and symptoms of depression in scleroderma. *Pain*. 2002;95:267-75.
28. Merz EL, Malcarne VL, Assassi S, et al. Biopsychosocial typologies of pain in a cohort of patients with systemic sclerosis. *Arthritis Care Res*. 2014;66:567-574.
29. Frech TM, Baron M. Understanding itch in systemic sclerosis in order to improve quality of life. *Clin Exp Rheum*. 2013;31:S81-S88.
30. El-Baalbaki G, Razykov I, Hudson M, et al. Association of pruritus with quality of life and disability in systemic sclerosis. *Arthritis Care Res*. 2010;62:1489-1495.

31. Razykov I, Levis B, Hudson M, et al. Prevalence and clinical correlates of pruritus in patients with systemic sclerosis: An updated analysis of 959 patients. *Rheumatology*. 2013;52:2056-2061.
32. Malcarne VL, Hansdottir I, Greenbergs HL, et al. Appearance self-esteem in systemic sclerosis. *Cog Ther Res.* 1999;23:197–208.
33. Benrud-Larson LM, Heinberg LJ, Boling C, et al. Body image dissatisfaction among women with scleroderma: Extent and relationship to psychosocial function. *Health Psychol*. 2003;22:130-139.
34. Amin K, Clarke A, Sivakumar B, et al. The psychological impact of facial changes in scleroderma. *Psychol Health Med*. 2011;16:304-312.
35. Richards H, Herrick A, Griffin K, et al. Psychological adjustment to systemic sclerosis: Exploring the association of disease factors, functional ability, body-related attitudes and fear of negative evaluation. *Psychol Health Med*. 2004;9:29–39.
36. Jewett LR, Hudson M, Malcarne VL, et al. Sociodemographic and disease correlates of body image distress among patients with systemic sclerosis. *PLOS ONE*. 2012;7:e33281.
37. Ennis H, Herrick AL, Cassidy C, et al. A pilot study of body image dissatisfaction and the psychological impact of systemic sclerosis-related telangiectases. *Clin Exp Rheum*. 2013;31:S12-S17.
38. Knafo R, Thombs BD, Jewett L, et al. (Not) talking about sex: A systematic comparison of sexual impairment in women with systemic sclerosis and other chronic disease samples. *Rheumatology*. 2009;48:1300-3.
39. Schouffoer AA, van der Marel J, Ter Kuile MM, et al. Impaired sexual function in women with systemic sclerosis: A cross-sectional study. *Arthritis Rheum*. 2009;61:1601-8.

40. Impens AJ, Rothman J, Schiopu E. Sexual activity and functioning in female scleroderma patients. *Clin Exp Rheum.* 2009;27:38-43.
41. Levis B, Hudson M, Knafo R, et al. Rates and correlates of sexual activity and impairment among women with systemic sclerosis. *Arthritis Care Res*. 2012;64:340-50
42. Levis B, Burri A, Hudson M, et al. Sexual activity and impairment in women with systemic sclerosis compared to women from a general population sample. *PLOS ONE*. 2012;7:e52129
43. Maddali Bongi S, Del Rosso A, Mikhaylova S, et al. Sexual function in Italian women with systemic sclerosis is affected by disease-related and psychological concerns. *J Rheumatol*. 2013;40:1697-705
44. Walker UA, Tyndall A, Ruszat R. Erectile dysfunction in systemic sclerosis. *Ann Rheum Dis*. 2009;68:1083-1085.
45. Foocharoen C, Tyndall A, Hachulla E, et al. Erectile dysfunction is frequent in systemic sclerosis and associated with severe disease: A study of the EULAR Scleroderma Trial and Research group. *Arthritis Res Ther*. 2012;14:R37.
46. Keck AD, Foocharoen C, Rosato E, et al. Nailfold capillary abnormalities in erectile dysfunction of systemic sclerosis: A EUSTAR group analysis. *Rheumatology*. 2014;53:639-643.
47. Rosato E, Barbano B, Gigante A, et al. Erectile dysfunction, endothelium dysfunction, and microvascular damage in patients with systemic sclerosis. *J Sex Med*. 2013;10:1380-8.
48. Iversen MD, Hammond A, Betteridge N. Self-management of rheumatic diseases: State of the art and future perspectives. *Ann Rheum Dis*. 2010;69:955-963.
49. Foster G, Taylor SJ, Eldridge SE, et al. Self-management education programmes by lay leaders for people with chronic conditions. *Cochrane Database Syst Rev*. 2007;4:CD005108.

50. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J. Self-management approaches for people with chronic conditions: A review. *Patient Educ Couns*. 2002;48:177-87.
51. Poole JL, Skipper B, Mendelson C. Evaluation of a mail-delivered, print-format, self-management program for persons with systemic sclerosis. *Clin Rheumatol*. 2013;32:1393-8.
52. Poole JL, Mendelson C, Skipper B, et al. Taking charge of systemic sclerosis: A pilot study to assess the effectiveness of an internet self-management program. *Arthritis Care Res*. 2014;66:778-82.
53. Lorig KR, Ritter PL, Laurent DD, et al. Internet-based chronic disease self-management: A randomized trial. *Med Care*. 2006;44:964-971.
54. Davison KP, Pennebaker JW, Dickerson SS. Who talks? The social psychology of illness support groups. *Am Psychol*. 2000;55:205-217.
55. Uccelli MM, Mohr LM, Battaglia MA, et al. Peer support groups in multiple sclerosis: Current effectiveness and future directions. *Mult Scler*. 2006;10:80-84.
56. Ussher J, Kirsten L, Butow P, et al. What do cancer support groups provide which other supportive relationships do not? The experience of peer support groups for people with cancer. *Soc Sci Med*. 2006;62:2565-2576.
57. http://www.scleroderma.ca/Support/Find-A-Support-Group.php
58. http://www.scleroderma.org/site/PageServer?pagename=patients_supportgroups#.VCeEoSi6pIc