



# The Scleroderma Patient-centered Intervention Network: SPIN

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# Disclosures

I am not receiving industry funding in relation to the content of this presentation.

# Patient-centered Care

- Patient empowerment
- Shared decision-making
- Care plans taking into account patient preferences and values
- Care that meets the needs of individual patients
- Evidence-based
- In chronic disease:
  - comprehensive care to reduce disability and improve health-related quality of life

# What about rare diseases?

- Typically no access to psychosocial and rehabilitation interventions that are:
  - Specific to needs of people with the disease
  - Adequately tested to determine if useful

# What About Rare Diseases?

- Search for studies across all 6,632 rare diseases listed on NIH Office of Rare Disease Research (July 2011)
- Sought randomized controlled trials (RCT) of psychosocial and rehabilitation interventions with at least 100 patients
- Found only 1 RCT (an exercise intervention for patients recovering from SARS)

# Primer on Clinical Trials of Psychosocial and Rehabilitation Interventions?

- Designed to improve well-being or coping of persons living with a disease, not alter the course of disease or its core symptoms
- Impact of interventions are small compared to a disease-altering medical intervention
- Cost is ideally low for feasible implementation
- Typically need at least 200-300 people in a randomized controlled trial and often more to confidently assess impact of intervention

# Care Options in Scleroderma

EULAR guidelines (Kowal-Bielecka et al. 2009):

*“There are also other treatment options for the management of SSc patients, such as physiotherapy, education, new experimental therapies, etc, which were beyond the scope of this project or could not be included because of the lack of expert consensus”*

# Why?

- Few (specialized) centers treat enough patients with a rare disease to:
  - Develop and sustain disease-specific psychosocial and rehabilitation service
  - Conduct large enough trials of disease-specific interventions
- Health care providers in local settings generally have little or no experience with a rare disease and specific needs of patients

# Challenges

- To develop supportive interventions (psychosocial, rehabilitation) that are:
  - Accessible to people with scleroderma
  - Low cost for feasible implementation
  - Can be delivered on an ongoing basis
- To conduct high-quality trials to confidentially assess impact interventions (including at least 200-300 people)

# SPIN: The Scleroderma Patient-centered Intervention Network



# SPIN

- A collaboration of people living with scleroderma, clinicians and researchers
- Aim: To develop and evaluate psychosocial and rehabilitation interventions that are accessible, low-cost, and can be delivered on an ongoing basis to people living with scleroderma

# SPIN Background

- Canadian Scleroderma Research Group (CSRG)
- Partnering with scleroderma patient organizations
- 2008: Improving psychological health and well-being in scleroderma
  - Bob Buzza, Shirley Haslam, Marion Pacy (Scleroderma Society of Canada)
  - Jennifer Haythornthwaite (John Hopkins, US)
  - Vanessa Malcarne (University of California, US)
  - Wim van Lankveld (Sint Maartenskliniek, the Netherlands)

# Psychological Health and Well-being: A Consensus Research Agenda

- Fatigue
- Pain
- Depressive symptoms
- Pruritus (Itch)
- Body Image
- Sexual function
- Other areas

## Psychological Health and Well-Being in Systemic Sclerosis: State of the Science and Consensus Research Agenda

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### Introduction

Systemic sclerosis (SSc; scleroderma) is a multisystem disorder characterized by disturbance in fibroblast function, microvascular disease, and immune system activation, culminating in fibrosis of the skin and internal organs (1,2). SSc is associated with extensive morbidity, includ-

chronic diarrhea, and renal failure (1,2). The rate of disease onset is highest between 30 and 50 years of age, with the risk for women being 4 to 5 times higher than for men (3,4). Median survival time from diagnosis is ~11 years, and patients are 3.7 times more likely to die within 10 years of diagnosis (44.9% mortality) than age-, sex-, and

# The Canadian Scleroderma Patient Survey of Health Concerns and Healthcare Needs

- Collaboration between Scleroderma Society of Canada and CSRG
- Based on:
  - Existing surveys from other patient groups
  - Patient input
  - Health care professional input
- September 2008 to August 2009
- Over 600 people with scleroderma completed this online or using paper forms distributed by patient support groups across Canada

# SPIN Background

- 2010: Consortium for clinical trials of behavioural, psychological and educational interventions
- 2011: SPIN planning meeting

## *Canadian Institutes for Health Research Team Grant:*

- \$1.5 million (2012 – 2017) for operating costs
- Additional \$300,000 in partner funding
  
- For SPIN in English and French
- Other languages: national funding (e.g. the Netherlands)

# SPIN – Key components

- 1) **Patient organization partnerships throughout the research process and as end user**
- 2) International network of clinical research centers
- 3) Virtually delivered interventions

# Partnering with Scleroderma Patient Organizations



Scleroderma  
Society of Canada

Société Canadienne  
de la Sclérodémie



Scleroderma Society of Ontario



Scleroderma Association of British Columbia



Scleroderma  
Society

Supporting people with Scleroderma for over 25 years



NVLE



Federation of European Scleroderma Associations

# SPIN – Key components

- 1) Patient organization partnerships throughout the research process and as end user
- 2) **International network of clinical research centers around the world**
- 3) Virtually delivered interventions

# SPIN Recruiting Sites

- Canada (CSRG, McGill)
- USA (UCLA, Michigan, Texas, Johns Hopkins, Boston University, Georgetown, Utah, Stanford)
- France
- The Netherlands
- UK
- Australia (Melbourne, Adelaide)
- Spain and Mexico City

# SPIN – Key components

- 1) Patient organization partnerships throughout the research process and as end user
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# Online interventions

- Increasingly common, for instance:
  - Self-management in diabetes (e.g., Lorig et al, 2010)
  - Depressive symptoms (e.g., Gellatly et al, 2007)
  - Anxiety (e.g., Hirai & Clum, 2006)
- Self-guided online interventions were effective in reducing elevated levels of depressive symptoms (Cuijpers et al, 2011)
  - 7 trials (total N = 1,362)
  - $d=0.28$  ( $p<0.001$ )

# Step 1: The SPIN Cohort

Large international study with people with scleroderma (N=1,500-2,000):

- Online questionnaires every 3 months
- Insight in problems important to patients
- How best to measure these outcomes
- Natural history of scleroderma and treatment as usual

## Step 2: Development of interventions

- Specific to living with scleroderma
- Self-guided (no therapist involved)
- Virtual, online
- Engaging (video, animations)
  
- Support patients in coping with their disease (e.g., emotional distress, body image)
- Reduce limitations in daily activities (e.g., exercise, hand function)

## Step 2: Development of interventions

- Fatigue
- Exercises to improve hand function
- Sleep
- Emotional coping
- Social anxiety from visible difference due to disease
- Pain management
- Exercise and fitness
- Nutrition and diet

## Step 3: Feasibility

How well do our study processes and interventions work?

- Research methodology (process and management)
- Acceptability, utility and practicality of intervention to people with scleroderma
- Interviews, online feedback

→ Adjustments if necessary

## Step 4: Randomized Controlled Trials

Does the intervention improve outcomes important to people living with scleroderma?

→ Outcomes of people with scleroderma who received the intervention are compared with outcomes of people who did not receive the intervention (but completed questionnaires in SPIN Cohort)

## Step 5: Dissemination

Making interventions available to people with scleroderma around the world through patient organizations

- Scleroderma Foundation
- Scleroderma Society of Canada
- Federation of European Scleroderma Associations

# SPIN Innovations

- Leveraging technology to deliver care
- Accessible interventions specific to living with scleroderma
- Partnering with patient organizations to develop and deliver interventions not feasibly provided by any single centre
- Network of major clinical research centers across Canada, the US, and Europe
- Novel research methods

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