

Addressing Challenges to Patient-centered Care in a Rare Disease Context: The Scleroderma Patient-centered Intervention Network (SPIN)

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Background

- People with rare diseases face unique challenges.
- Trials are challenging to conduct, and few Canadian centres treat enough patients with any given rare disease to sustain patient-centered intervention programs.
- Systemic sclerosis (SSc, scleroderma) is a rare autoimmune connective tissue disease. Common problems faced in SSc include limitations in hand function and mobility, pain, fatigue, gastrointestinal symptoms, breathing problems, pruritus, depression, and body image distress due to disfigurement.

Objective

The objective of the Scleroderma Patient-centered Intervention Network (SPIN) was to develop an infrastructure that would support observational studies to better understand problems faced by people with SSc and as a framework for large randomized controlled trials (RCTs) of educational, self-management, rehabilitation, and psychosocial programs to support quality of life.

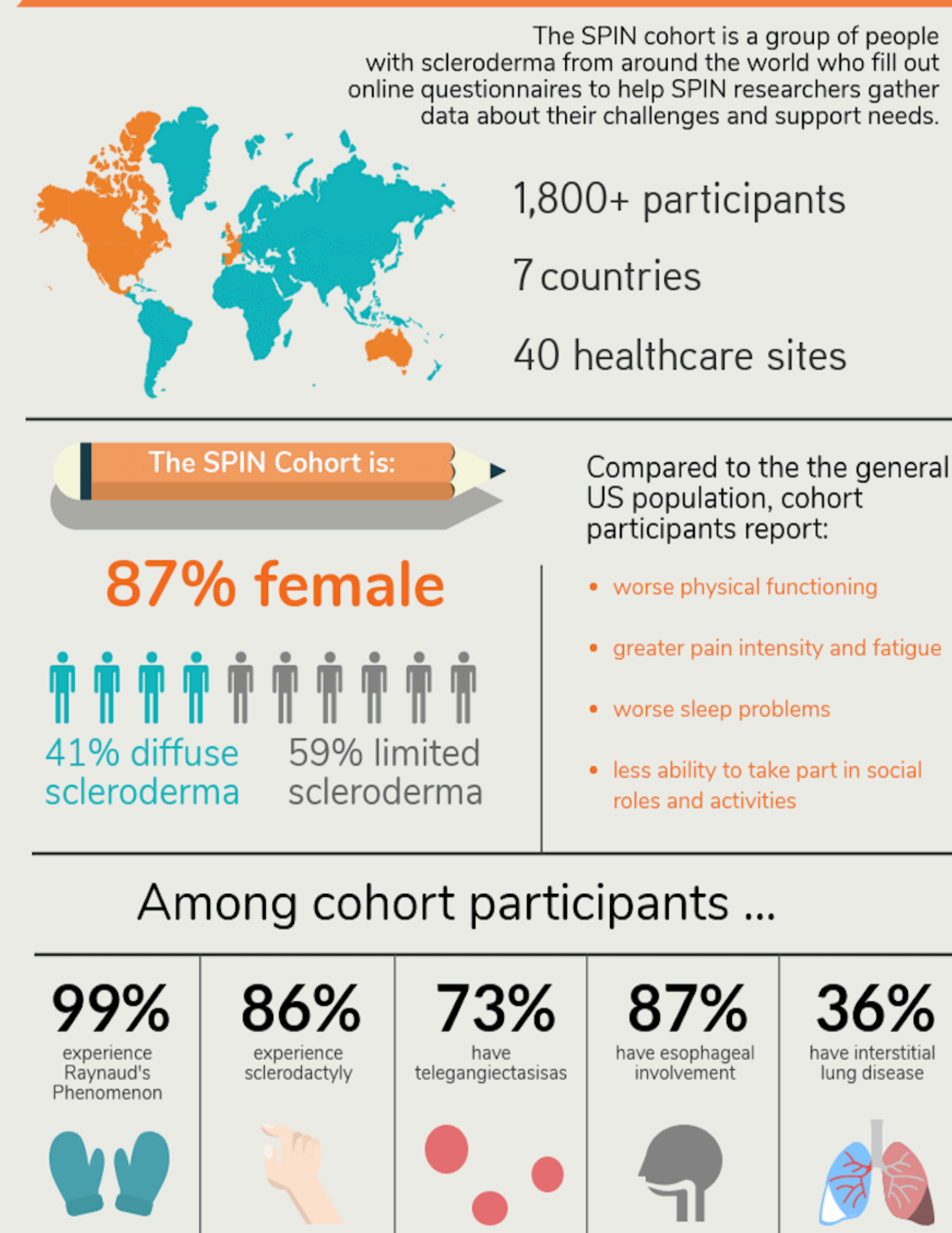
Methods

- SPIN's international team determined that achieving its goals in a rare disease environment would require:
 - (1) strong patient and patient organization partnerships;
 - (2) a global network of SSc care centres and researchers;
 - (3) online patient program delivery;
 - (4) the use of novel cohort-based, economically efficient trial methods.

Key Component 1 – Partnerships

- SPIN was launched in 2011 with seed funding from Scleroderma Canada and the Scleroderma Society of Ontario.
- It received a CIHR Emerging Team Grant in Rare Diseases in 2012.
- People with SSc serve on SPIN's Patient Advisory and several project-specific advisory boards.

Facts about the SPIN Cohort



Key Component 2 – Global Network

- SPIN has brought together over 150 members of the international SSc community and raised over \$3.5 million in funding.
- SPIN has enrolled over 2,400 participants (1,750 active) from 45 sites in 7 countries (Canada, USA, UK, France, Spain, Mexico, Australia) in the SPIN Cohort.

Key Component 3 – Online Delivery

- SPIN Cohort participants complete patient-reported outcome measures via the internet every 3 months and are available to participate in trials of SPIN's internet-based patient programs.

Key Component 4 – Novel Trial Methods

- SPIN adopted the cohort multiple RCT design.
- SPIN has been funded by the CIHR and the Arthritis Society to conduct large (e.g., N = 500 to 600) RCTs of hand exercise and SSc-specific self-management programs.
- Outside of its cohort, SPIN received funding from CIORA and completed a successful feasibility trial of the SPIN Support Group Leader Education (SPIN-SSLED) Program.



Conclusions

- SPIN is an example of international research, clinical, and patient communities working together to overcome barriers to patient-centered intervention research in a rare disease.
- SPIN's approach can be applied in other common and rare rheumatic diseases.

