Use and perceptions of nutrition information resources in systemic sclerosis: a Scleroderma Patient-centred Intervention Network (SPIN) cohort study

N. Østbø[1,2], E. Yakes Jimenez[3,4], M.-E. Carrier[1], L. Kwakkenbos[5-7], B.D. Thombs[1,8-12] Scleroderma Patient-centred Intervention Network (SPIN) Diet and Nutrition Education Patient Advisory Team, on behalf of the SPIN Investigators

[1]Lady Davis Institute for Medical Research, Jewish General Hospital, Montreal, Quebec, Canada; [2]Department of Health Management and Health Economics, University of Oslo, Norway; [3]Departments of Paediatrics and Internal Medicine and College of Population Health, University of New Mexico Health Sciences Center, Albuquerque, NM, USA; [4]Nutrition Research Network, Academy of Nutrition and Dietetics, Chicago, IL, USA; [5]Department of Clinical Psychology, Radboud University, Nijmegen, the Netherlands; [6]Department of IQ Health care, Radboud University Medical Center, Nijmegen, The Netherlands; [7]Department of Psychiatry, Radboud University Medical Center, Nijmegen, The Netherlands; [8]Department of Psychiatry, McGill University, Montreal, Quebec, Canada; [9]Department of Epidemiology, Biostatistics and Occupational Health, McGill University, Montreal, Quebec, Canada; [10]Department of Medicine, McGill University, Montreal, Quebec, Canada; [11]Department of Psychology, McGill University, Montreal, Quebec, Canada; [12]Biomedical Ethics Unit, McGill University, Montreal, Quebec, Canada.

Abstract

Objective

People with systemic sclerosis (SSc) may find it challenging to obtain high-quality nutrition and diet information. Objectives were to evaluate (i) how commonly different information resources are used and (ii) perceived trustworthiness, accessibility, comprehensibility, and individualisation of resources.

Methods

We administered the Scleroderma Patient-centred Intervention Network Nutrition Information Resources Survey to participants in an international cohort. Participants were asked if they had used 26 informational resources in four categories, including (i) health care providers, (ii) websites or social media, (iii) print materials, and (iv) events, and to rate each resource on trustworthiness, accessibility, comprehensibility, and individualisation (0 = not at all to 10 = completely).

Results

727 participants completed the survey. Most (94%) had sought nutrition or diet information from at least one resource. The most-used category was health care providers (86%), followed by print materials (68%), websites or social media (66%), and events (43%). People who had used a resource generally rated it more favourably across all domains than those who had not. The highest-rated resources across domains were conventional health care providers (doctors, registered dieticians, nurses), SSc patient organisations, SSc support groups, and university or research institution websites.

Conclusion

Respondents used many different diet and nutrition information resources. They preferred resources from conventional health care providers, affiliated with credible institutions (e.g., SSc patient organisations), or with personal connections (e.g., SSc support groups). Future research should address the limited evidence base on nutrition in SSc and assess the quality of information provided by different information resources.

Key words

scleroderma, systemic sclerosis, nutrition, diet, education

Nora Østbø, BA
Elizabeth Yakes Jimenez, PhD, RD
Marie-Eve Carrier, MSc
Linda Kwakkenbos, PhD
Brett D. Thombs, PhD

The Scleroderma Patient-centred Intervention Network (SPIN) Diet and Nutrition Education Patient Advisory Team are listed on page 1613.

The SPIN Investigators are listed in Appendix in the Supplementary file.

Please address correspondence to:
Brett D. Thombs
Jewish General Hospital,
3755 Cote Ste Catherine Road,
Pavilion H4.83
Montréal, Québec H3T 1E4, Canada.
E-mail: brett.thombs@mcgill.ca

Received on September 19, 2023; accepted in revised form on February 28, 2024.

Funding: the Scleroderma Patient-centred Intervention Network (SPIN) has received funding for the SPIN Cohort from the Canadian Institutes of Health Research, the Arthritis Society, the Lady Davis Institute for Medical Research of the Jewish General Hospital, Montreal, Quebec, Canada, the Jewish General Hospital Foundation, Montreal, Quebec, Canada, McGill University, Montreal, Quebec, Canada, the Scleroderma Society of Ontario, Scleroderma Canada, Sclérodermie Québec, Scleroderma Manitoba, Scleroderma Atlantic, the Scleroderma Association of BC, Scleroderma SASK, Scleroderma Australia, Scleroderma New South Wales, Scleroderma Victoria, and Scleroderma Queensland.
B.D. Thombs was supported by a Tier 1 Canada Research Chair.

Competing interests: none declared.

Introduction

Systemic sclerosis (SSc, scleroderma) is a rare autoimmune connective tissue disease characterised by vascular injury, immune dysfunction and abnormal fibrotic processes that can affect multiple organ systems, including the skin, lungs, gastrointestinal tract, and cardiovascular system (1). Gastrointestinal manifestations affect up to 90% of people with SSc and may include microstomia (reduction in mouth aperture), xerostomia (dry mouth), dental disease, dysmotility (impairment to digestive system muscles, including gastroparesis and chronic intestinal pseudo-obstruction), gastroesophageal reflux disease, gastric antral vascular ectasia (fragile blood vessels in the stomach that increase risk of gastrointestinal bleeding), and alterations to the intestinal microbiome (2, 3) and small intestinal bacterial overgrowth, and chronic intestinal pseudo-obstruction (muscle or nerve dysfunction in the intestines) (4, 5). Depending on their disease course, different people with SSc may experience widely varying combinations and severity of gastrointestinal manifestations. Symptoms can include reduced appetite and intake, dysphagia (difficulty swallowing), odynophagia (painful swallowing), early satiety, diarrhoea, constipation, and faecal incontinence, which can result in malabsorption, fat and muscle loss, and moderate to severe malnutrition, along with reduced health-related quality of life and increased mortality (4, 6-11). Malnutrition risk, reduced energy intake, and weight loss can also be caused by other SSc manifestations, including pulmonary arterial hypertension and chronic heart failure (12-14). In some cases, patients with SSc may gain excess weight due to medications they take (*e.g.* antidepressants, beta-blockers, corticosteroids) or other risk factors, such as physical inactivity resulting from functional limitations (15-17).

Evidence to support dietary modifications for the management of gastrointestinal involvement, overall disease course and health, or medication side effects in SSc is limited (12, 13, 18). A recent systematic review and meta-analysis noted that the majority of the evidence available about the effects of diet on rheumatic and musculoskeletal diseases is for people with osteoarthritis and rheumatoid arthritis (19). The authors identified two single arm studies of medical nutrition therapy and three randomised controlled trials (RCTs) of vitamin supplementation for people with SSc (19). The evidence from all five studies was rated very low quality, with no or small effect sizes noted for the medical nutrition therapy studies and large effects on Rodnan skin score noted for two of the vitamin supplementation RCTs (19). One observational study has reported that adherence to a low fermentable oligosaccharides, disaccharides, monosaccharides, and polyols diet had no association with gastrointestinal symptom severity or significant alterations in the intestinal microbiome (3). Thus, people with SSc who seek guidance on dietary strategies and modifications may find it challenging to obtain high-quality, individually tailored information to address their needs. In this context, it is important to understand where they obtain information on nutrition and diet and their perceptions about the characteristics of different information resources.

We previously conducted nominal group technique (NGT) sessions with 15 people with SSc in 4 groups to identify sources they use for information and advice on nutrition and diet to support health and manage gastrointestinal issues as well as perceived advantages and disadvantages of information sources (20). The purpose of the NGT sessions was to support development of the Scleroderma Patient-centred Intervention Network (SPIN) Nutrition Information Resources Survey. The objective of the present study was to administer the survey and evaluate, in a large, international sample of people with SSc (1) how commonly different information resources are used and (2) perceived trustworthiness, accessibility, comprehensibility, and individualisation of information sources.

Methods

This was a cross-sectional study in which survey results from the SPIN Nutrition Information Resources Sur-

vey were deterministically linked using participant email addresses to sociodemographic, medical, and patient-reported outcome measure data from the ongoing SPIN Cohort (21).

Participants and procedures

We recruited participants enrolled in the SPIN Cohort. The SPIN Cohort is a convenience sample of individuals with SSc from 47 sites in 7 countries (Australia, Canada, France, Mexico, Spain, United Kingdom, and United States). Eligible SPIN Cohort participants must be classified as having SSc according to 2013 American College of Rheumatology/European League Against Rheumatism (ACR/EULAR) classification criteria (22); aged ≥18 years; and fluent in English, French or Spanish. Participants are invited to participate in the SPIN Cohort by attending physicians or nurse coordinators at recruiting sites. Written informed consent is obtained, including consent to be contacted about additional studies, and site personnel complete and submit an online medical data form. An automated welcome email is sent to participants with instructions for activating their SPIN account and completing SPIN Cohort measures online. SPIN measures are completed via an online portal upon enrolment and subsequently every three months.

On January 13, 2021, we emailed invitations to all SPIN Cohort participants who complete assessments in English or French to complete the separate SPIN Nutrition Information Resources Survey. We additionally advertised the survey through an announcement presented to SPIN Cohort participants when they logged into the SPIN Cohort portal to complete routine online assessments. To promote participation, we informed participants that ten survey respondents would be randomly selected to win an Amazon gift card of $100 CAD or the equivalent in their country's currency. The email invitation and announcements provided a link to the online survey tool *Qualtrics*® (Provo, Utah) (23). In *Qualtrics*®, participants entered their email address to access and complete the survey. Follow-up reminder emails were sent 6 and 9 weeks after the initial email invitation to Cohort participants who had not completed the survey. The survey was closed on April 9, 2021.

The SPIN Cohort was approved by the Research Ethics Committee of the Centre Intégré Universitaire de Santé et de Services Sociaux du Centre-Ouest-de-l'Île-de-Montréal (no. MP-05-2013-150) and by the ethics committees of all recruiting sites. The present study was approved as an amendment to the SPIN Cohort.

Measures

- Sociodemographic and medical characteristics

Upon enrolment in the SPIN Cohort, participants report sociodemographic and lifestyle information, including sex, age, country, race or ethnicity, civil status, years of education, and occupation status. SPIN physicians provide medical information, including height, weight, gastrointestinal involvement (yes/no for oesophageal, stomach, and intestinal), SSc subtype (limited, diffuse, or sine), date of first non-Raynaud's symptom onset, date of diagnosis, and presence of digital ulcers (distal pulp, anywhere else on the finger), current or past tendon friction rubs, interstitial lung disease, and/or joint contractures (large joints, small joints). Body mass index (BMI) was calculated and categorised as underweight (<18.5), normal weight (18.5 to 24.9), overweight (25.0 to 29.9) or obese (>30) (24).

Race or ethnicity data are self-reported using standard categories that are used in each country. Categories differ across countries, and categories used in one country may not be recognised by participants from other countries. To characterise study participants, they were aggregated as White, Black, and Other. See Supplementary Table S1 for categories used in each country.

Participants in the SPIN Nutrition Information Resources Survey additionally completed the Malnutrition Screening Tool (MST) (25) and the UCLA Scleroderma Clinical Trial Consortium Gastrointestinal Tract 2.0 (UCLA SCTC GIT 2.0) (26) via *Qualtrics*®.

- Malnutrition screening tool (MST)

The MST is a two-item malnutrition screening tool that assesses unintentional weight loss and decreased appetite. Possible total scores range from 0 to 5, and scores ≥2 are used to identify people at risk for malnutrition (25). Although the MST has not been validated in adults with SSc, it has been validated in outpatient and hospitalised adults (27, 28), and self-reported MST scores have been shown to be reliable and valid compared to dietitian-assessed scores (29).

- UCLA SCTC GIT 2.0

The UCLA SCTC GIT 2.0 characterises gastrointestinal tract involvement in people with SSc (26, 30). It consists of 34 items and 7 multi-item scales (reflux, distention or bloating, faecal soilage, emotional well-being, social functioning, diarrhoea, constipation). Items are scored on a 0 to 3 possible range, where 0 indicates better health and 3 indicates worse health (except items number 15 and 31, which are scored on 0 [better health] and 1 [worse health)]). A total score (possible range 0.00 to 2.83) that reflects overall GIT disease severity is calculated by averaging all subscales except constipation (none-to-mild = 0.00 to 0.49, moderate = 0.50 to 1.00, severe-to very severe = 1.01 to 3.00).

- The SPIN Nutrition Information Resources Survey

The SPIN Nutrition Information Resources Survey (See Supplementary Table S2) was developed to investigate how commonly people with SSc use different resources to obtain information and advice on nutrition and diet and their experiences with and perceptions of these resources. Items were developed from four virtual NGT interview sessions with 15 people with SSc from five countries (Canada, USA, UK, the Netherlands, Tunisia) conducted in February 2020 (20). Building on the results of that study, study investigators and the SPIN-DINE Patient Advisory Team, comprised of 7 people with SSc, developed a list of 26 resources, which were grouped into four categories using qualitative content analysis (31): (1) health care providers (9 resources), (2)

print materials (5 resources), (3) websites and other media platforms (7 resources), and (4) events (5 resources). The survey was translated into French using a standard forward–backward translation process.

For each of the 26 resources, participants were asked to respond yes or no to the item: "Please indicate whether you have or have not used the resource for the purpose of getting information and advice related to gastrointestinal symptoms, other SSc-related symptoms, or maintaining a diet that meets your nutritional needs." Survey participants also rated each resource (whether they had used it or not) on four characteristics, which were identified as commonly perceived areas of advantages or disadvantages in the NGT study (12), including: (1) trustworthiness of information (*e.g.* based on scientific evidence, from a credible source), (2) individualisation to the unique needs of each patient, (3) accessibility (*e.g.* cost, need for travel or internet access), and (4) comprehensibility (*e.g.* easy to understand). Participants rated each resource on a scale from 0 (not at all) to 10 (completely), separately for each characteristic. If they had used a type of resource, they were asked to rate it based on their experience; if they had not used the type of resource, they were asked to rate it based on their perception of the resource. Participants were additionally informed that if they had no opinion on an item they could choose "I don't know", but they were encouraged to respond to as many items as they could.

Finally, participants were asked to think globally about all the different resources they have used to get diet or nutrition information related to managing their SSc and to rate how important each characteristic (trustworthiness; individualisation; accessibility; comprehensibility) is to them when selecting a resource (0 = not at all important to 10 = extremely important).

Data analysis

Descriptive statistics were used to characterise study participants on sociodemographic and medical variables, MST risk status, and UCLA SCTC GIT 2.0 scores. Ratings of nutrition resources were non-normal and were thus presented as medians with interquartile ranges (IQR) for the full sample and separately by whether participants had used the resource. For each resource type category, median ratings for trustworthiness, individualisation, accessibility, and comprehensibility were illustrated graphically for each resource in the category. All analyses were conducted using ®SPSS Statistics 27.

Results

Participant characteristics

Among 1434 SPIN Cohort participants who received an invitation to complete the survey and were still actively enrolled in the SPIN Cohort at the final date of data collection, 727 (51%) completed the survey. Table I shows sociodemographic and medical data for survey respondents and non-respondents. Among respondents, the mean age was 59.9 (standard deviation [SD]=12.1), and most respondents were female (n=657, 90%) and white (n = 606, 85%). The majority had limited SSc subtype (n=416, 57%), and mean time since diagnosis was 13.4 years (SD=8.7). At cohort enrolment, when medical data were reported, there were 637 (88%) participants with gastrointestinal involvement (oesophageal, stomach or intestinal), with oesophageal involvement being the most common (n=615, 85%). Based on BMI, 43 (6%) participants were considered underweight, 370 (51%) normal weight, 199 (27%) overweight, and 115 (16%) obese. Mean (SD) time since cohort enrolment was 4.1 years (standard deviation=1.9). Characteristics of SPIN Cohort participants who did not complete the survey were similar to those who did respond.

Mean current UCLA SCTC GIT 2.0 score among people who completed the survey was 0.63 (SD=0.5), which is in the range of moderate severity. Based on MST scores, 117 (16%) participants would be classified as at risk of malnutrition.

Use of information resources

See Table II for data on use of resources for the full sample and by country. Nearly all respondents (n=682, 94%) reported that they had used at least one information resource. The most used category of information resources was health care providers (n=625, 86%), followed by print materials (n=491, 68%), websites and social media (n=482, 66%), and events (n=311, 43%). Resources used by at least 25% of respondents in: (i) the health care provider category included individual consultations with a rheumatologist (65%), gastroenterologist (43%), and internal medicine physician (42%); (ii) the print material category included books or articles by health care professionals (52%), newsletters by SSc patient organisations (48%), academic or scientific journal articles (32%), and books or articles written by a person with SSc (32%); (iii) the websites and other media category included SSc patient organisation websites (49%), university or research institution websites (28%), and lifestyle or food-related social media pages or groups (27%); and (iv) the events category included support groups for people with SSc or autoimmune diseases (30%) and national or regional SSc patient organisation events (26%).

Across all countries included in the study, at least 4 out of 5 participants had seen a health care provider for nutrition or diet guidance, and overall use patterns were similar across countries. Most participants from the USA, Canada, UK, and Australia (74% to 87%) reported consulting with a rheumatologist, while French participants were more likely to have seen an internal medicine physician (68%). Approximately 25% of participants from France, USA, Canada, and Australia reported having consulted a registered dietitian versus 17% in the UK.

Resource ratings

Trustworthiness was the highest-rated consideration when choosing an information resource (median = 9 out of 10, IQR = 6 to 10), followed by comprehensibility (median = 8, IQR = 5 to 10), accessibility (median = 8, IQR = 5 to 10), and individualisation (median= 6, IQR = 4 to 9). Median ratings for resources by characteristic in each resource category for the full sample are shown in Figure 1.

Table I. Characteristics of Scleroderma Patient-centred Intervention Network (SPIN) cohort participants who responded and did not respond to the survey.

Variable[a]	Respondents (n=727)	Non-respondents (n=707)
Sociodemographic		
Age in years, mean (SD)	**59.9 (12.1)[b]**	**58.8 (12.7)**
Female sex, n (%)	**657 (90.4)**	**597 (84.4)**
Race or ethnicity[c], n (%)		
White	**606 (84.6)d**	**553 (81.3)e**
Black	**44 (6.1)d**	**54 (7.9)e**
Other	**66 (9.2d**	**73 (10.7)e**
Years of education, mean (SD)	**15.4 (3.5)f**	**14.7 (3.7)g**
Employed full- or part-time, n (%)	**305 (42.6)h**	**287 (42.2)i**
Married or living as married, n (%)	**525 (73.3)j**	**474 (69.6)k**
Country of recruitment, n (%)		
France	**216 (29.7)l**	**257 (36.4)**
Canada	**215 (29.6)l**	**153 (21.6)**
United States	**193 (26.5)l**	**217 (30.7)**
United Kingdom	**77 (10.6)l**	**64 (9.1)**
Australia	**23 (3.2)l**	**16 (2.3)**
Spain	**1 (0.1)l**	**-**
Medical		
Time in years since first non-Raynaud's phenomenon symptom, mean (SD)	**15.3 (9.4)m**	**14.4 (9.2)n**
Time in years since systemic sclerosis diagnosis, mean (SD)	**13.4 (8.7)o**	**12.9 (8.7)p**
Limited systemic sclerosis subtype, n (%)	416 (57.2)q	413 (58.8)r
Body mass index category, n (%)		
Underweight (< 18.5)	43 (5.9)	37 (5.2)
Normal weight (18.5 to 24.9)	370 (50.9)	353 (49.9)
Overweight (25.0 to 29.9)	199 (27.4)	197 (27.9)
Obese (> 30.0)	115 (15.8)	120 (17.0)
Any gastrointestinal involvement, n (%)	637 (87.6)	611 (87.4)s
Oesophageal	615 (84.6)	591 (84.8)t
Stomach	200 (27.5)	198 (28.0)u
Intestinal	272 (37.4)	251 (35.5)v
Digital ulcers (anywhere on the finger), n (%)	249 (34.3)	289 (40.9)
Tendon friction rubs, n (%)	68 (9.4)w	82 (13.2)x
Moderate or severe contractures of small joints, n (%)	158 (21.8)y	176 (26.5)z
Moderate or severe contractures of large joints, n (%)	75 (10.4)[aa]	80 (23.3)[ab]
Modified Rodnan Skin Score, mean (SD	7.4 (7.9)[ac]	7.7 (8.0)[ad]
Interstitial lung disease, n (%)	215 (29.6)[ae]	249 (35.2)[af]
Malnutrition Screening Tool score ≥ 2 (at risk)	117 (16)	---------
UCLA SCTC GIT 2.0 total score and scale scores[ai]		
Total GIT score, mean (SD)	0.63 (0.50)	----------
Reflux scale, mean (SD)	0.68 (0.61)	----------
Distention/bloating scale, mean (SD)	1.17 (0.86)	----------
Faecal soilage scale, mean (SD)	0.45 (0.80)	----------
Diarrhoea scale, mean (SD)	0.51 (0.62)	----------
Social functioning scale, mean (SD)	0.39 (0.52)	----------
Emotional well-being scale, mean (SD)	0.56 (0.71)	----------
Constipation scale, mean (SD)	0.53 (0.60)	----------

[a]UCLA SCTC GIT 2.0 scores and Malnutrition Screening Tool scores were collected at time of completing the survey. All other variables were collected at time of enrolment into the SPIN Cohort. Participants were enrolled on an ongoing basis from the beginning of the SPIN Cohort in 2013.

[c]Because ethnicity/race information is collected differently across countries, it is aggregated here into the categories "White", "Black", and "Other".

See Supplementary Table S1 for further details about race or ethnicity grouping. Due to missing data: [b]n=725; [d]n=716; [e]n=680; [f]n=716; [g]n=680; [h]n=716; [i]n=726; [j]n=716; [k]n=681; [l]n=725; [m]n=670; [n]n=654; [o]n=698; [p]n=687; [q]n=719; [r]n=702; [s]n=699; [t]n=697 [u]n=674; [v]n=684; [w]n=635; [x]n=635; [y]n=686; [z]n=665; [aa]n=672; [ab]n=653; [ac]n=597; [ad]n=592; [ae]n=708; [af]n=694.

[ai]All domains and Total GIT score are scored from 0 (better health) to 3 (worse health) except diarrhoea and constipation domains with ranges from 0–2 and 0–2.5, respectively. The mean Total GIT score fell under "moderate severity" classification (none-to-mild = 0.00 to 0.49; moderate = 0.50 to 1.00; severe to very severe = 1.01 to 3.00.).

For the full sample, conventional health care providers (medical doctors, registered dietitians, nurses or nurse practitioners) received similarly high ratings (medians ≥8) on trustworthiness, individualisation of information and comprehensibility and slightly lower ratings for accessibility (medians ≥7). Alternative and complementary practitioners (naturopaths, homeopaths, personal trainers) received consistently lower ratings for all four domains (medians ≤7). However, naturopaths and homeopaths were the least-used resources across all the resources included in the survey (used by ≤8% of sample). People who had used alternative and complementary practitioners gave higher ratings on trustworthiness, individualisation, and comprehensibility (medians ≥8) compared to those who had not used these resources (medians ≤6).

Ratings of print materials varied depending on the source of information. Trustworthiness ratings were high for all resources (medians ≥8), including books or articles by health care professionals, newsletters by SSc patient organisations, academic or scientific journal articles, books or articles by people with SSc, except for books or articles by alternative medicine providers (median = 5). Individualisation ratings were low for most resources (medians ≤6), while newsletters provided by SSc patient organisation were rated slightly higher (median = 7). Accessibility and comprehensibility ratings were moderate to high for almost all resources (medians ≥7), except for academic or scientific journal articles with scores slightly lower on comprehensibility (median = 6).

In the websites and other media category, trustworthiness ratings were highly resource-dependent, with SSc patient organisation websites and university or research institution websites scoring highest (medians = 9) and social media pages or groups and generic lifestyle or food websites scoring lower (median ≤6). Individualisation ratings were generally lower (medians ≤7), while accessibility ratings were high (medians ≥8) across resources. Comprehensibility ratings were generally moderate to high (medians ≥7).

All five resources in the events category received high ratings (medians ≥8) on

Table II. Use of nutrition and diet resources for all SPIN nutrition information resources survey respondents and by country.

Resource category	Resource	Full sample (n=727)[a] Used n (%)	France (n=216) Used n (%)	USA (n=215) Used n (%)	Canada (n=193) Used n (%)	UK (n=77) Used n (%)	Australia (n=23) Used n (%)
Health care providers	At least one resource within the category	625 (86)	175 (81)	198 (92)	164 (85)	66 (86)	19 (83)
	Rheumatologist	472 (65)	55 (25)	186 87)	148 (77)	63 (82)	17 (74)
	Gastroenterologist	314 (43)	65 (30)	124 (58)	87 (45)	28 (36)	8 (35)
	Internal medicine physician	303 (42)	147 (68)	104 (48)	38 (20)	10 (13)	2 (9)
	Registered dietitian	171 (24)	50 (23)	55 (26)	47 (24)	11 (17)	6 (26)
	Nurse or nurse practitioner	161 (22)	28 (13)	63 (29)	35 (18)	28 (36)	6 (26)
	Functional medicine physician	84 (12)	29 (13)	16 (7)	33 (17)	4 (5)	2 (7)
	Personal trainer	69 (10)	9 (4)	23 (11)	23 (12)	11 (14)	3 (13)
	Naturopath	60 (8)	16 (7)	10 (5)	30 (16)	1 (1)	3 (13)
	Homeopath	40 (6)	19 (9)	9 (4)	7 (4)	5 (6)	0 (0)
Print material	At least one resource within the category	491 (68)	132 (61)	161 (75)	128 (66)	55 (71)	13 (57)
	Book or article by health care professional	380 (52)	94 (44)	131 (61)	103 (53)	38 (49)	12 (52)
	Newsletter by scleroderma patient organisation	347 (48)	74 (34)	116 (54)	97 (50)	46 (60)	12 (52)
	Academic or scientific journal article	235 (32)	41 (19)	100 (47)	59 (31)	27 (35)	7 (30)
	Book or article by person with scleroderma	233 (32)	45 (21)	80 (37)	66 (34)	34 (44)	7 (30)
	Book or article by alternative medicine provider	139 (19)	39 (18)	45 (21)	36 (17)	17 (22)	2 (9)
Websites and other media platforms	At least one resource within the category	482 (66)	115 (53)	156 (73)	137 (71)	56 (73)	15 (65)
	Scleroderma patient org. website	356 (49)	78 (36)	122 (57)	97 (50)	46 (60)	11 (48)
	Website of a university or research institution	204 (28)	22 (10)	94 (44)	57 (30)	22 (29)	7 (30)
	Lifestyle or food-related social media page or group	195 (27)	35 (16)	68 (32)	66 (34)	20 (26)	3 (13)
	General lifestyle or food-related website	175 (24)	37 (17)	64 (30)	52 (27)	19 (25)	2 (9)
	Facebook group for people with scleroderma	173 (24)	39 (18)	62 (29)	46 (24)	16 (21)	8 (35)
	Website/social media page of a medical professional	111 (15)	18 (8)	46 (21)	27 (14)	14 (18)	5 (22)
	Facebook group for people with autoimmune diseases	78 (11)	17 (8)	32 (15)	19 (10)	8 (10)	1 (4)
Events	At least one resource within the category	311 (43)	53 (25)	116 (54)	99 (51)	32 (42)	9 (39)
	Patient support group	216 (30)	27 (13)	87 (40)	72 (37)	23 (30)	5 (22)
	National or regional scleroderma patient org. event	189 (26)	25 (12)	69 (32)	71 (37)	20 (26)	3 (12)
	Presentation by a health care professional (other than dietitian)	147 (20)	20 (9)	53 (25)	54 (28)	15 (19)	5 (22)
	Presentation by person(s) with scleroderma	103 (14)	20 (9)	29 (13)	44 (23)	7 (9)	2 (9)
	Presentation by a dietitian	72 (10)	11 (5)	23 (11)	28 (15)	7 (9)	3 (13)

[a]One participant was recruited from Spain and is not included in the country breakdown, but is included in the full sample total.

trustworthiness and comprehensibility but were rated lower on individualisation (medians = 7). Accessibility scores were particularly low among participants who had not used the resource (median = 5 for all 5 resources) compared to participants who had used the resource (medians = 8 for 4 of 5 resources).

Supplementary Table S3 shown use of resources in each category and complete ratings for each resource for the full sample and separated by users and non-users of each resource. Across categories and resources within categories, trustworthiness, individualisation, accessibility, and comprehensibility ratings were generally lower among those who had not used resources compared to those who had.

Supplementary Table S4 shows ratings for each resource for participants with none-to-mild or moderate (n=575) *versus* severe or very severe (n=151) gastrointestinal symptoms based on UCLA SCTC GIT 2.0 scores. Ratings were similar across categories regardless of symptom scores.

Discussion

We surveyed 727 people with SSc from the SPIN Cohort and found that nearly all (94%) had sought nutrition or diet information from at least one

informational resource. The most-used category of information resources was health care providers (86%), followed by print materials (68%), websites and social media (66%), and events (43%). People who had used a resource generally rated it more favourably compared to those who had not used the resource. For the full sample, the highest-rated resources across domains were conventional health care providers, SSc patient organisations, patient support groups, and university or research institution websites. The health care provider category received the highest ratings for individualisation, which is likely because of provider ability to consider patient comorbidities, preferences, and social circumstances when providing nutrition information and recommendations.

Resources with the lowest ratings were typically used by a smaller proportion of participants and often had wider rating IQRs by 1 to 2 points compared to resources that had consistently high ratings, suggesting larger discrepancies in perceptions of these resources among participants. This could be in part because publicly accessible resources with few barriers to dissemination, such as websites and social media groups, as well as alternative and complementary practitioners with variable requirements for professional certification, licensure, and credentialing, may have a wider range in credibility and quality (32-35). Experiences with and perceptions of such resources may thus vary more widely. Overall, finding high-quality information online for rare diseases and assessing its quality are substantial challenges (36, 37). Little objective assessment of the quality of online, SSc-specific information has been done (38). One systematic review assessed the quality of online information about Raynaud's phenomenon in SSc and concluded that quality was low (39). Another study assessed 115 YouTube videos with SSc-related content; it found that about 1 in 5 videos was misleading and that the best quality videos were primarily created by academic institutions, professional organisations, and physicians (40). More objective research on the quality and helpfulness of online health information in SSc is needed, including on the topic of nutrition and diet and management of gastrointestinal symptoms. Tools to complete this type of assessment, such as the DISCERN tool and the Global Quality Scale, are available and have been used to assess online health information in written and video form for SSc and other diseases (38, 40-42).

Resources that are provided by generally trusted institutions, such as a university, hospital, or SSc patient organisation, were rated more favourably across domains and particularly high on trustworthiness. Such resources also had a higher proportion of respondents who had used them, suggesting that these are preferred or more easily accessed resources. Lower ratings on trustworthiness were observed when the source of the information cannot easily be verified as credible, such as with many web-based sources or print materials, suggesting that many participants think about possible sources of information critically. This finding may not be generalisable across the SSc patient population, as our survey respondents had on average 15 years of education, which equates to some years of university completed. Higher education levels are often associated with better health and digital literacy (43, 44).

Almost 1 in 3 participants in our study had used a patient support group to seek nutrition information. While some physicians report being concerned about the type of information being shared and the potential for encouragement to utilise unconventional therapies in peer support groups (45, 46), such concerns appear to not be shared with most patients, given their high trust rating (median = 8). Other studies have identified the information-sharing aspect of patient support groups as a patient-perceived benefit of these groups, and this may be particularly important for individuals with rare diseases, who may not have friends or family that understand what they are experiencing (47, 48) Some non-professional resources, including patient organisation material and organised patient support groups, had trustworthiness ratings that were similar to those for professional resources, such as individual consultations with a health care provider. It may be that informational resources that have close links with trusted institutions or are thought to have been vetted by credible patient organisations, such as people facilitating patient support groups, have higher trust among patients. More general nutrition-focused websites and social media-based SSc or autoimmune disease support groups, which are not typically associated with known SSc patient organisations, were rated lower on trust, which is consistent with the idea that organisational affiliation and personal contact may be important factors in trust. However, we have not identified any studies that have examined factors associated with trust in SSc informational resources.

Clinical implications

A 2019 systematic review concluded that clinical practice guidelines for dietary interventions in SSc should take a measured approach due to limited evidence, including the lack of any randomised controlled trials in this area (18). Given this and the findings of our survey, there are several research gaps in the nutritional care of patients with SSc that should be addressed to improve nutrition-related outcomes. First, the evidence-base for nutritional intervention in SSc needs to be bolstered as the current lack of research in this area is a barrier to providing SSc-specific recommendations. Second, actual and perceived barriers to accessing nutrition information resources should be further investigated. In our NGT sessions with 15 patients with SSc, reported barriers to access included high costs, long waiting times for appointments and referrals, and difficulty with comprehending information (20). More research is also needed to identify possible solutions to access barriers. Third, the trustworthiness, comprehensibility and individualisation of online and other resources addressing nutrition and diet and management of gastrointestinal symptoms in SSc need to be objectively evaluated as a first step towards providing people with SSc guidance about the accuracy of information from these resources.

Lastly, further research should assess the impact of accessing different information resources on nutrition-related outcomes (*e.g.* gastrointestinal symptoms, malnutrition status) for people with SSc.

Strengths and limitations

An important strength of this study is that the initial version of the survey was developed based on information provided in NGT groups by people with SSc and then iteratively further developed with input from people with SSc. Although the list of resources that we included may not be exhaustive, we believe that included resources are likely a good representation of the informational resources that people with SSc use in the countries we included in our study, and the domains assessed are likely important to them. Another important strength of this study is its large and international sample. There are also some limitations related to generalisability to consider in interpreting results. First, about half of SPIN Cohort participants did not complete the survey, and we do not know if this group and our study participants differed on interest in nutrition therapies or other important characteristics that we did not measure. However, responders and non-responders did not differ meaningfully on gastrointestinal involvement or BMI scores. In addition, the SPIN Cohort is a convenience sample and may not be representative of the SSc population. Second, we asked participants about preferences and use of information resources, but this did not allow us to assess whether use of information resources influenced gastrointestinal outcomes. Third, we did not collect information on dietary patterns or specific diets followed by participants.

Conclusion

We found that approximately 19 in 20 people with SSc have sought and accessed diet and nutrition information from a variety of resources. The highest-rated resources were linked to organisations or professions with well-established credibility or personal contacts including conventional health care providers, SSc patient organisations, patient support groups, and university or research institution websites. Future research should address the limited evidence-base for nutritional intervention in SSc and objectively assess the quality and impact on patient outcomes for different information resources.

Scleroderma Patient-centred Intervention Network (SPIN) Diet and Nutrition Education Patient Advisory Team members

Deani Baillie, London, UK; Cathleen Dobbs, PhD, Joliet, IL, USA; Amy Gietzen, Scleroderma Foundation, Tri-State Chapter, Buffalo, NY, USA; Geneviève Guillot, PDt, Sclérodermie Québec, Longueuil, Quebec, Canada; Jennifer Johnston, Edmonton, Alberta, Canada; Dorothy M. Kurylo, PhD, Danville, IL, USA; Laura Simmons, Scleroderma Atlantic, Halifax, Nova Scotia, Canada.

References

1. ALLANORE Y, SIMMS R, DISTLER O *et al.*: Systemic sclerosis. *Nat Rev Dis Primers* 2015; 1(1): 1-21. https://doi.org/10.1038/nrdp.2015.2
2. NATALELLO G, BOSELLO SL, PARONI STERBINI F *et al.*: Gut microbiota analysis in systemic sclerosis according to disease characteristics and nutritional status. *Clin Exp Rheumatol* 2020; 38 (Suppl. 125): S73-84.
3. NGUYEN AD, ANDRÉASSON K, MCMAHAN ZH *et al.*: Gastrointestinal tract involvement in systemic sclerosis: The roles of diet and the microbiome. *Semin Arthritis Rheum* 2023; 60: 152185. https://doi.org/10.1016/j.semarthrit
4. NAGARAJA V, MCMAHAN ZH, GETZUG T, KHANNA D: Management of gastrointestinal involvement in scleroderma. *Curr Treatm Opt Rheumatol* 2015; 1(1): 82-105. https://doi.org/10.1007/s40674-014-0005-0
5. VEALE B, JABLONSKI R, FRECH T, PAULING J: Orofacial manifestations of systemic sclerosis. *Br Dent J* 2016; 221(6): 305-10. https://doi.org/10.1038/sj.bdj.2016.678
6. STEEN VD, MEDSGER TA: Changes in causes of death in systemic sclerosis, 1972-2002. *Ann Rheum Dis* 2007; 66(7): 940-4. https://doi.org/10.1136/ard.2006.066068
7. KRAUSE L, BECKER MO, BRUECKNER CS *et al.*: Nutritional status as marker for disease activity and severity predicting mortality in patients with systemic sclerosis. *Ann Rheum Dis* 2010; 69(11): 1951-57. https://doi.org/10.1136/ard.2009.123273
8. CRUZ-DOMÍNGUEZ MP, GARCÍA-COLLINOT G, SAAVEDRA MA *et al.*: Malnutrition is an independent risk factor for mortality in Mexican patients with systemic sclerosis: a cohort study. *Rheumatol Int* 2017; 37(7): 1101-9. https://doi.org/10.1007/s00296-017-3753-y
9. RICHARD N, HUDSON M, WANG M *et al.*: Severe gastrointestinal disease in very early systemic sclerosis is associated with early mortality. *Rheumatology* 2019; 58(4): 636-44. https://doi.org/10.1093/rheumatology/key350
10. DI BATTISTA M, BARSOTTI S, ORLANDI M *et al.*: One year in review 2021: systemic sclerosis. *Clin Exp Rheumatol* 2021; 39 (Suppl. 131): S3-12. https://doi.org/10.55563/clinexprheumatol/izadb8
11. PAOLINO S, PACINI G, SCHENONE C *et al.*: Nutritional status and bone microarchitecture in a cohort of systemic sclerosis patients. *Nutrients* 2020; 12(6): 1632. https://doi.org/10.3390/nu12061632
12. BURLUI AM, CARDONEANU A, MACOVEI LA *et al.*: Diet in scleroderma: is there a need for intervention? *Diagnostics* 2021; 11(11): 2118. https://doi.org/10.3390/diagnostics11112118
13. HOFFMANN-VOLD A, VOLKMANN ER: Gastrointestinal involvement in systemic sclerosis: effects on morbidity and mortality and new therapeutic approaches. *J Scleroderma Relat Disord* 2021; 6(1): 37-43. https://doi.org/10.1177/2397198319891282
14. SARI A, ESME M, AYCICEK GS *et al.*: Evaluating skeletal muscle mass with ultrasound in patients with systemic sclerosis. *Nutrition* 2021; 84: 110999. https://doi.org/10.1016/j.nut.2020.110999
15. DESALERMOS A, RUSSELL B, LEGGETT C *et al.*: Effect of obesogenic medications on weight-loss outcomes in a behavioral weight-management program. *Obesity* 2019; 27(5): 716-23. https://doi.org/10.1002/oby.22444
16. HARB S, PELÁEZ S, CARRIER M *et al.*: Barriers and facilitators to physical activity for people with scleroderma: a scleroderma patient-centered intervention network cohort study. *Arthritis Care Res* (Hoboken) 2022; 74(8): 1300-10. https://doi.org/10.1002/acr.24567
17. FAVA M: Weight gain and antidepressants. *J Clin Psychiatry* 2000; 61 Suppl 11: 37-41.
18. SMITH E, PAULING JD: The efficacy of dietary intervention on gastrointestinal involvement in systemic sclerosis: A systematic literature review. *Semin Arthritis Rheum* 2019; 49(1): 112-18. https://doi.org/10.1016/j.semarthrit.2018.12.001
19. GWINNUTT JM, WIECZOREK M, RODRÍGUEZ-CARRIO J *et al.*: Effects of diet on the outcomes of rheumatic and musculoskeletal diseases (RMDs): systematic review and meta-analyses informing the 2021 EULAR recommendations for lifestyle improvements in people with RMDs. *RMD Open*. 2022; 8: e002167. https://doi.org/10.1136/rmdopen-2021-002167
20. ØSTBØ N, JIMENEZ EY, HARB S *et al.*: Nutrition Information resources used by people with systemic sclerosis and perceived advantages and disadvantages: a nominal group technique study. ACR *Open Rheumatol* 2021; 3(8): 540-49. https://doi.org/10.1002/acr2.11293
21. 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(8). https://doi.org/10.1136/bmjopen-2013-003563
22. VAN DEN HOOGEN F, KHANNA D, FRANSEN J *et al*.: 2013 classification criteria for systemic sclerosis: an American College of Rheumatology/European League against Rheumatism collaborative initiative. *Ann Rheum Dis* 2013; 65(11): 2737-47. https://doi.org/10.1002/art.38098
23. Qualtrics. Provo U. Qualtrics Survey Platform. 2002.
24. WEIR CB, JAN A: BMI classification percentile and cut off points. StatPearls Treasure Island (FL): StatPearls Publishing LLC; 2022.
25. FERGUSON M, CAPRA S, BAUER J, BANKS M: Development of a valid and reliable malnutrition screening tool for adult acute hospital patients. *Nutrition* 1999; 15(6): 458-464. https://doi.org/10.1016/S0899-9007(99)00084-2
26. KHANNA D, HAYS RD, MARANIAN P *et al*.: Reliability and validity of the University of California, Los Angeles Scleroderma Clinical Trial Consortium Gastrointestinal Tract Instrument. *Arthritis Rheum* 2009 September 15; 61(9): 1257-63. https://doi.org/10.1002/art.24730
27. SKIPPER A, COLTMAN A, TOMESKO J *et al*.: Adult malnutrition (undernutrition) screening: an evidence analysis center systematic review. *J Acad Nutr Diet* 2020; 120(4): 669-708. https://doi.org/10.1016/j.jand.2019.09.010
28. CASTRO-VEGA I, VESES MARTIN S, CANTERO LLORCA J *et al*.: Validation of nutritional screening Malnutrition Screening Tool compared to other screening tools and the nutritional assessment in different social and health areas. *Nutr Hosp* 2018; 35(2): 351-58. https://doi.org/10.20960/nh.1619
29. DI BELLA A, CROISIER E, BLAKE C, PELECANOS A, BAUER J, BROWN T: Assessing the concurrent validity and interrater reliability of patient-led screening using the malnutrition screening tool in the ambulatory cancer care outpatient setting. *J Acad Nutr Diet* 2020; 120(7): 1210-15. https://doi.org/10.1016/j.jand.2019.10.015
30. BARON M, HUDSON M, STEELE R, LO E, CANADIAN SCLERODERMA RESEARCH GROUP: Validation of the UCLA Scleroderma Clinica Trial Gastrointestinal Tract Instrument version 2.0 for systemic sclerosis. *J Rheumatol* 2011; 38(9): 1925-30. https://doi.org/10.3899/jrheum.110060
31. MAYRING P: Qualitative content analysis: Theoretical background and procedures. Approaches to qualitative research in mathematics education: *Springer*; 2015. p. 365-80. https://doi.org/10.1007/978-94-017-9181-6_13
32. WIESENER S, FALKENBERG T, HEGYI G, HOK J, ROBERTI DI SARSINA P, FONNEBO V: Legal status and regulation of complementary and alternative medicine in Europe. *Forsch Komplementmed* 2012;19 Suppl. 2: 29-36. https://doi.org/10.1159/000343125
33. COHEN MH, NELSON H: Licensure of complementary and alternative practitioners. *Virtual Mentor* 2011; 13(6): 374-78. https://doi.org/10.1001/virtualmentor.2011.13.6.pfor1-1106
34. COHEN MH: Complementary and integrative medical therapies, the FDA, and the NIH: definitions and regulation. *Dermatol Ther* 2003; 16(2): 77-84. https://doi.org/10.1046/j.1529-8019.2003.01614.x
35. BOON H: Regulation of complementary/alternative medicine: a Canadian perspective. *Complement Ther Med* 2002; 10(1): 14-19. https://doi.org/10.1054/ctim.2002.0499
36. PAUER F, GÖBEL J, STORF H *et al*.: Adopting quality criteria for websites providing medical information about rare diseases. *Interact J Med Res* 2016; 5(3): e5822. https://doi.org/10.2196/ijmr.5822
37. PAUER F, LITZKENDORF S, GÖBEL J, STORF H, ZEIDLER J, GRAF VON DER SCHULENBURG J-M: Rare diseases on the internet: an assessment of the quality of online information. *J Med Internet Res* 2017; 19(1): e23. https://doi.org/10.2196/jmir.7056
38. DEVGIRE V, MARTIN AF, MCKENZIE L, SANDLER RD, HUGHES M: A systematic review of internet-based information for individuals with Raynaud's phenomenon and patients with systemic sclerosis. *Clin Rheumatol* 2020; 39(8): 2363-67. https://doi.org/10.1007/s10067-020-05023-5
39. UNAL-ULUTATAR C, ULUTATAR F: YouTube as a source of information on systemic sclerosis. *Int J Rheum Dis* 2022; 25(8): 887-92. https://doi.org/10.1111/1756-185X.14363
40. CHARNOCK D, SHEPPERD S, NEEDHAM G, GANN R: DISCERN: an instrument for judging the quality of written consumer health information on treatment choices. *J Epidemiol Community Health* 1999; 53(2):105-11. https://doi.org/10.1136/jech.53.2.105
41. SANSEVERE ME, WHITE JD: Quality assessment of online complementary and alternative medicine information resources relevant to cancer. *Integr Cancer Ther* 2021; 20. https://doi.org/10.1177/15347354211066081
42. BERNARD A, LANGILLE M, HUGHES S, ROSE C, LEDDIN D, VAN ZANTEN SV: A systematic review of patient inflammatory bowel disease information resources on the World Wide Web. *Am J Gastroenterol* 2007; 102(9): 2070-77. https://doi.org/10.1111/j.1572-0241.2007.01325.x
43. SØRENSEN K, VAN DEN BROUCKE S, FULLAM J *et al*.: Health literacy and public health: a systematic review and integration of definitions and models. *BMC Public Health* 2012; 12(1): 1-13. https://doi.org/10.1186/1471-2458-12-80
44. NORMAN CD, SKINNER HA: eHealth literacy: essential skills for consumer health in a networked world. *J Med Internet Res* 2006; 8(2): e506. https://doi.org/10.2196/jmir.8.2.e9
45. OWEN JE, GOLDSTEIN MS, LEE JH, BREEN N, ROWLAND JH: Use of health-related and cancer-specific support groups among adult cancer survivors. *Cancer* 2007; 109(12): 2580-89. https://doi.org/10.1002/cncr.22719
46. STEGINGA SK, SMITH DP, PINNOCK C, METCALFE R, GARDINER RA, DUNN J: Clinicians' attitudes to prostate cancer peer-support groups. *BJU Int* 2007; 99(1): 68-71. https://doi.org/10.1111/j.1464-410X.2006.06545.x
47. DOCHERTY A: Experience, functions and benefits of a cancer support group. *Patient Educ Couns* 2004; 55(1): 87-93. https://doi.org/10.1016/j.pec.2003.08.002
48. DELISLE VC, GUMUCHIAN ST, RICE DB *et al*.: Perceived benefits and factors that influence the ability to establish and maintain patient support groups in rare diseases: a scoping review. *Patient* 2017; 10(3): 283-93. https://doi.org/10.1007/s40271-016-0213-9