Association of Race/Ethnicity and Sex with Appearance Concerns: A Scleroderma Patient-centered Intervention Network (SPIN) Cohort Study

Short Title: Appearance Concerns in Systemic Sclerosis

Lisa R. Jewett, MSc[1,2]; Linda Kwakkenbos, PhD[1,3-4]; Marie-Eve Carrier, MSc[1]; Vanessa L. Malcarne, PhD[5,6]; Susan J. Bartlett, PhD[7,8]; Daniel E. Furst, MD[9]; Karen Gottesman, BA[10]; Maureen M. Mayes, MD[11]; Shervin Assassi, MD[11]; Diana Harcourt, PhD[12]; Heidi Williamson, PhD[12]; Sindhu R. Johnson[13]; Annett Körner, PhD[1,2]; Virginia Steen, MD[14]; Rina S. Fox, MS[5]; Shadi Gholizadeh, MS[5]; Sarah D. Mills, MS[5]; Jacqueline C. Molnar[1]; Danielle B. Rice, MSc[1,15]; Brett D. Thombs[1-3,8,15-17]; and the SPIN Investigators[17]

[1]Lady Davis Institute for Medical Research, Jewish General Hospital, Montreal, Quebec, Canada; [2]Department of Educational and Counselling Psychology, McGill University, Montreal, Quebec, Canada; [3]Department of Psychiatry, McGill University, Montreal, Quebec, Canada; [4]Behavioural Science Institute, Clinical Psychology, Radboud University, Nijmegen, the Netherlands; [5]Department of Psychology, San Diego State University, San Diego, California, USA; [6]San Diego State University/University of California, San Diego Joint Doctoral Program in Clinical Psychology, San Diego, California, USA; [7]McGill University Health Center, Montreal, Quebec, Canada; [8]Department of Medicine, McGill University, Montreal, Quebec, Canada; [9]Division of Rheumatology, Geffen School of Medicine at the University of California, Los Angeles, Los Angeles, USA; [10]Scleroderma Foundation, USA; [11]Department of Internal Medicine,

Division of Rheumatology, University of Texas Health Science Center, Houston, Texas, USA; [12]Centre for Appearance Research, University of the West of England, Bristol, United Kingdom; [13]Toronto Scleroderma Program, Division of Rheumatology, Department of Medicine, Toronto Western and Mount Sinai Hospitals, Toronto, Ontario, Canada; [14]Department of Medicine, Georgetown University, Washington, DC, USA; Departments of [15]Psychology,[16]Epidemiology, Biostatistics and Occupational Health, and [17]School of Nursing, McGill University, Montreal, Quebec, Canada; [18]SPIN Investigators: Murray Baron, McGill University, Montréal, Québec, Canada; Frank van den Hoogen, Radboud University Medical Center and Sint Maartenskliniek, Nijmegen, The Netherlands; Dinesh Khanna, University of Michigan, Ann Arbor, Michigan, USA; Luc Mouthon, Université Paris Descartes, Paris, France; Warren R. Nielson, St. Joseph's Health Care, London, Ontario, Canada; Serge Poiraudeau, Université Paris Descartes, Paris, France; Robert Riggs, Scleroderma Foundation, Danvers, Massachusetts, USA; Maureen Sauve, Scleroderma Society of Ontario, Hamilton, Ontario; Fredrick Wigley, Johns Hopkins University School of Medicine, Baltimore, Maryland, USA; Isabelle Boutron, Université Paris Descartes, and Assistance Publique-Hôpitaux de Paris, Paris, France; Angela Costa Maia, University of Minho, Braga, Portugal; Ghassan El-Baalbaki, Université du Québec à Montréal, Montréal, Québec, Canada; Carolyn Ells, McGill University, Montréal, Québec, Canada; Cornelia van den Ende, Sint Maartenskliniek, Nijmegen, The Netherlands; Kim Fligelstone, Scleroderma Society, London, UK; Catherine Fortune, Scleroderma Society of Ontario, Hamilton, Ontario, Canada; Tracy Frech, University of Utah, Salt Lake City, Utah, USA; Dominique Godard, Association des Sclérodermiques de France, Sorel-Moussel,

France; Daphna Harel, New York University, New York, New York, USA; Marie Hudson, McGill University, Montréal, Québec, Canada; Ann Impens, Midwestern University, Downers Grove, Illinois, USA; Yeona Jang, McGill University, Montréal, Québec, Canada; Ann Tyrell Kennedy, Federation of European Scleroderma Associations, Dublin, Ireland; Maggie Larche, McMaster University, Hamilton, Ontario, Canada; Catarina Leite, University of Minho, Braga, Portugal; Carlo Marra, Memorial University, St. John's, Newfoundland, Canada; Karen Nielsen, Scleroderma Society of Ontario, Hamilton, Ontario, Canada; Janet L. Poole, University of New Mexico, Albuquerque, New Mexico, USA; Janet Pope, University of Western Ontario, London, Ontario, Canada; Alexandra Portales, Asociación Española de Esclerodermia, Madrid, Spain; Tatiana Sofia Rodriguez Reyna, Instituto Nacional de Ciencias Médicas y Nutrición Salvador Zubirán, Mexico City, Mexico; Anne A. Schouffoer, Leiden University Medical Center, Leiden, The Netherlands; Russell J. Steele, Jewish General Hospital and McGill University, Montréal, Québec, Canada; Maria E. Suarez-Almazor, University of Texas MD Anderson Cancer Center, Houston, Texas, USA; Joep Welling, NVLE Dutch patient organization for systemic autoimmune diseases, Utrecht, The Netherlands; Durhane Wong-Rieger, Canadian Organization for Rare Disorders, Toronto, Ontario, Canada; Alexandra Albert, Université Laval, Québec, Québec, Canada; Guylaine Arsenault, Sherbrooke University, Sherbrooke, Québec, Canada; Lyne Bissonnette, Sherbrooke University, Sherbrooke, Québec, Canada; Gilles Boire, Sherbrooke University, Sherbrooke, Québec, Canada; Alessandra Bruns, Sherbrooke University, Sherbrooke, Québec, Canada; Patricia Carreira, Servicio de Reumatologia del Hospital 12 de Octubre, Madrid, Spain; Lorinda Chung, Stanford University, Stanford, California, USA;

Pierre Dagenais, Sherbrooke University, Sherbrooke, Québec, Canada; Christopher Denton, Royal Free London Hospital, London, UK; Robyn Domsic, University of Pittsburgh, Pittsburgh, Pennsylvania, USA; James V. Dunne, St. Paul's Hospital and University of British Columbia, Vancouver, British Columbia, Canada; Paul Fortin, Université Laval, Québec, Québec, Canada; Anna Gill, Royal Free London Hospital, London, UK; Jessica Gordon, Hospital for Special Surgery, New York City, New York, USA; Genevieve Gyger, Jewish General Hospital and McGill University, Montréal, Québec, Canada; Ariane Herrick, Salford Royal NHS Foundation Trust, Manchester, UK; Monique Hinchcliff, Northwestern University, Chicago, Illinois, USA; Alena Ikic, Université Laval, Québec, Québec, Canada; Niall Jones, University of Alberta, Edmonton, Alberta, Canada; Artur Jose de B. Fernandes, Sherbrooke University, Sherbrooke, Québec, Canada; Suzanne Kafaja, University of California, Los Angeles, California, USA; Nader Khalidi, McMaster University, Hamilton, Ontario, Canada; Benjamin Korman, Northwestern University, Chicago, Illinois, USA; Patrick Liang, Sherbrooke University, Sherbrooke, Québec, Canada; Ariel Masetto, Sherbrooke University, Sherbrooke, Québec, Canada; David Robinson, University of Manitoba, Winnipeg, Manitoba, Canada; Sophie Roux, Sherbrooke University, Sherbrooke, Québec, Canada; Elena Schiopu, University of Michigan, Ann Arbor, Michigan, USA; Doug Smith, University of Ottawa, Ottawa, Ontario, Canada; Robert Spiera, Hospital for Special Surgery, New York, New York, USA; Evelyn Sutton, Dalhousie University, Halifax, Nova Scotia, Canada; Carter Thorne, Southlake Regional Health Centre, Newmarket, Ontario, Canada; John Varga, Northwestern University, Chicago, Illinois, USA; Pearce Wilcox, St. Paul's Hospital and University of British Columbia, Vancouver,

British Columbia, Canada; Vanessa C. Delisle, Jewish General Hospital and McGill University, Montréal, Québec, Canada; Claire Fedoruk, Jewish General Hospital, Montréal, Québec, Canada; Brooke Levis, Jewish General Hospital and McGill University, Montréal, Québec, Canada; Katherine Milette, Jewish General Hospital and McGill University, Montréal, Québec, Canada; Mia R. Pepin, Jewish General Hospital, Montréal, Québec, Canada; Jennifer Persmann, Université du Québec à Montréal, Montréal, Québec, Canada.

Address for Correspondence: Brett D. Thombs, PhD; Jewish General Hospital; 4333 Cote St-Catherine Road, Montréal, Québec, Canada, H3T 1E4; Telephone: (514) 340-8222 ext. 5112; Fax: (514) 340-8124; Email: brett.thombs@mcgill.ca.

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ABSTRACT

Introduction: Appearance concerns are common in systemic sclerosis (SSc), but no study has examined their association with sex or race/ethnicity.

Methods: SSc patients were from the Scleroderma Patient-centered Intervention Network Cohort. Presence or absence of appearance concerns was assessed with a single item. Multivariate logistic regression was used to assess factors associated with appearance concerns, including sex, race/ethnicity, and pre-specified sociodemographic and disease covariates.

Results: Of 644 patients, appearance concerns were present in 72%, including 421 of 565 women (75%) and 42 of 79 men (53%), as well as 392 of 550 patients who identified as White (71%), 35 of 41 identified as Black (85%), and 36 of 53 identified as another race/ethnicity (68%). In multivariate analysis, women had significantly greater odds of reporting appearance concerns than men (odds ratio (OR) = 2.97, 95% confidence interval (CI) = 1.78-4.96, $p < .001$) than men. Black patients had significantly greater odds of appearance concerns than White patients in unadjusted (OR = 2.64, 95% CI = 1.01-6.34, $p = .030$), but not multivariate analysis (OR = 1.71, 95% CI = 0.65-4.48, $p = .279$). Older patients were less likely (OR = 0.98 per year, 95% CI = 0.96-0.99, $p = .009$) and patients with moderate hand contractures were more likely to report appearance concerns (OR = 1.97, 95% CI=1.12-3.46, p=.019).

Conclusion: Women are substantially more likely than men to have appearance concerns. Black patients are more likely than White patients, but this may be due to more severe changes in appearance.

Keywords: appearance; body image; race/ethnicity; scleroderma; sex; systemic sclerosis

INTRODUCTION

Disfiguring appearance changes are common in systemic sclerosis (SSc) andinclude telangiectasias, changes to the texture of the skin, hand contractures, skin pigmentation changes, and altered facial features (2). These visible differences are unique to the disease and distinct from appearance changes in other medical conditions. Furthermore, appearance changes in SSc commonly affect body parts that are highly visible and play a central role in social interactions, such as the face, mouth, and hands (1,3). Treatments can lessen the impact of some SSc symptoms, but do not alleviate manifestations of irreversible tissue damage that affect appearance.

Concerns about appearance are common among people with acquired disfigurements from medical illness or injury (4). In SSc, visual manifestations of the disease are associated with greater body image dissatisfaction and social discomfort and poorer overall psychosocial functioning (5-8). SSc patients with more severe disease symptoms, such as more significant skin changes in the hands, report higher levels of body image dissatisfaction, lower appearance self-esteem, and greater symptoms of depression and anxiety (7-9). Severity of facial disfigurement in SSc, based on observer ratings, has similarly been associated with higher levels of general distress, worry, and feelings of being more noticeable to others (10,11). Compared to older patients, younger people with SSc experience greater social discomfort in relation to their appearance (6, 9). No published studies, however, have investigated the degree to which sex and race/ethnicity may be associated with appearance concerns in SSc.

Among other groups of people with visible differences, women experience more worry about their appearance and greater general distress, social anxiety, and social

avoidance than men (12-15). Women with visible appearance differences are more likely than men to have multiple appearance concerns and tend to experience the impact of their disfigurements on social interactions differently (14). For instance, women are more likely to express embarrassment, self-consciousness, and worry about their visible differences, whereas men may be more inclined to experience feelings of aggression and hostility (14,16). Overall, women are more likely to have difficulty adjusting to visible differences than men (13, 15, 17).

Less is known about the association of race/ethnicity with appearance concerns and body image. One survey of 458 adults with visible disfigurements found that people with non-White racial/ethnic backgrounds experienced significantly greater worry about their appearance and greater concern that their condition was noticeable to others than White respondents (18). One possible reason for this is that visible disfigurements may disproportionately affect non-White individuals. People with darker skin colors are more vulnerable to visible differences due to changes in skin pigmentation compared to individuals with lighter skin tones, and the psychosocial burden of pigmentation changes is greater for individuals with darker skin types (19). In SSc, skin involvement and pigmentation changes are more common among Black patients than White patients, although White patients are more likely to have telangiectasias (20).

The objective of the present study was to examine the association of sex and race/ethnicity with the presence of appearance concerns in a large, international cohort of SSc patients. We hypothesized that female SSc patients would be more likely to have appearance concerns than males and that Black SSc patients would be more likely to

report appearance concerns than White patients, controlling for other sociodemographic and disease variables.

METHODS

Patients and Procedure

The sample consisted of patients enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort (21) who completed baseline study questionnaires from January 2014 through August 2015. Patients were enrolled at 21 SPIN centers from Canada, the USA, and the UK. To be eligible for the SPIN Cohort, patients must have a confirmed diagnosis of SSc according to 2013 American College of Rheumatology/European League Against Rheumatism classification criteria (22), be ≥ 18 years of age, have the ability to give informed consent, be fluent in English or French, and have access and the ability to respond to questionnaires via the Internet. The SPIN sample is a convenience sample. Eligible patients are invited by attending physicians or supervised nurse coordinators from SPIN centers to participate in the SPIN Cohort, and written informed consent is obtained. The local SPIN physician or nurse coordinator completes a medical data form that is submitted online to initiate patient registration. After completion of online registration, an automated welcoming email is sent to participants with instructions for activating their SPIN account and completing SPIN Cohort measures online. SPIN Cohort patients complete outcome measures via the Internet upon enrollment and subsequently every three months. Patients who had complete data at their baseline assessment for all variables necessary for planned multivariate analyses were included in the present study. The SPIN Cohort study was approved by the Research Ethics Committee of the Jewish

General Hospital, Montréal, Canada and by the research ethics committees of each participating center.

Measures

Sociodemographic Characteristics. Patients enrolled in the SPIN Cohort provided sociodemographic data, including age, sex, race/ethnicity, education level, marital status, and employment status. Response options for race/ethnicity differed slightly for patients from Canada, the USA, and the UK samples, consistent with how racial/ethnic status is typically characterized in each country. In the Canadian sample, patients could identify as White, Black, Aboriginal, Asian, Latin American, or Arab. In the USA sample, patients could identify as White (non-Hispanic), African American, Hispanic or Latino, Asian, American Indian/Alaska Native, Native Hawaiian/Other Pacific Islander, or Mixed-race. In the UK sample, patients could identify as White, African, or Asian. In the present study, racial/ethnic status was narrowed to three categories across Canadian, American, and UK samples, consisting of White, Black, or Other. Across countries, responses indicating White racial/ethnic status were combined to create one White race/ethnicity category; responses indicating Black, African American, or African were combined to create one Black race/ethnicity category; and other responses were combined to create one Other race/ethnicity category.

Disease-related Characteristics. SPIN physicians or nurse coordinators provided disease information, including time since onset of the first non-Raynaud's phenomenon symptom (i.e., disease duration), disease subtype (limited or diffuse SSc), telangiectasias, skin pigmentation changes, hand contractures, and skin thickening on both hands. Limited SSc was defined as skin involvement distal to the elbows and

knees only, whereas diffuse SSc was defined as skin involvement proximal to the elbows and knees, and/or the trunk (23). Telangiectasias were defined as the visible dilation of superficial cutaneous blood vessels that collapse upon pressure and fill slowly when pressure is released, excluding normal sun exposure-related telangiectasias (24). In the present study, telangiectasias were coded as present on the body and face, present on the body only, or none. Skin pigmentation changes included either hyper- or hypo-pigmentation of the skin. As with telangiectasias, skin pigmentation changes were coded as present on the body and face, present on the body only, or none. Hand contractures, which entail limitations in the range of motion of a joint, secondary to tightening around the joint, were measured for small joints on the hands (i.e., proximal interphalangeal joints, metacarpals, and/or wrists) and categorized as None/Mild (0-25% limitation in range of motion), Moderate (25-50%), or Severe (>50%). Skin thickening of the fingers was defined as skin thickening or hardening extending proximal to the metacarpophalangeal joints (21).

Presence of Appearance Concerns. The Derriford Appearance Scale (DAS-24) (25,26) is a self-report measure of distress related to problems with appearance and assesses the degree of dysfunction experienced by people living with such body image disturbance, including social anxiety and avoidance related to self-consciousness. The DAS-24 includes an introductory item, which is not scored as part of the scale, that asks, “Is there any aspect of your appearance (however small) that concerns you at all?” (yes/no). This item was used as the primary outcome in the present study.

Data Analysis

Descriptive statistics were calculated for all sociodemographic and disease variables, including means and standard deviations for continuous variables. Chi-square tests were used for categorical variables, and a one-way ANOVA was used for continuous variables to compare patients on sociodemographic and disease characteristics across sex and race/ethnicity categories. For variables with statistically significant overall tests, Bonferroni-corrected comparisons were done to assess statistical significance between pairs of race/ethnicity groups. To maintain the family-wise error rate < .05, the Bonferroni-corrected α for each of the three subgroup comparisons for each variable was .0167.

The associations of sociodemographic variables (age, sex, race/ethnicity, marital status, education level), and disease variables (telangiectasis, skin pigmentation changes, hand contractures, skin thickening of fingers, disease subtype) were assessed using binary logistic regression. All variables included in the regression analysis were selected a priori. Discrimination and calibration of the multivariate model were assessed with the c-index and Hosmer-Lemeshow goodness-of-fit test statistic, respectively (27). The c-index is the percentage of comparisons where patients with appearance concerns had a higher predicted probability of having appearance concerns than patients without appearance concerns for all possible pairs where patients were discrepant on outcome variable status. The Hosmer-Lemeshow goodness-of-fit statistic is a measure of the accuracy of the predicted number of cases of appearance concerns compared to the number of patients who actually reported appearance concerns across the spectrum of probabilities. A relatively large p value indicates reasonably good model fit (27). All

analyses were conducted using SPSS (Version 22), and statistical tests were two-sided with alpha < .05.

RESULTS

Sample Characteristics

In total, 757 SSc patients completed baseline assessments, of which 717 answered the appearance concern item, including 644 with data for all variables included in the logistic regression analysis. Of these, 463 (72%) indicated that there was an aspect of their appearance that caused them concern.

Sociodemographic and disease characteristics are displayed in Table 1. Average age in the total sample was 55.3 years (Standard Deviation [SD] = 12.1), and the majority of patients were White (N = 550, 85%) and married or living as married (N = 471, 73%). Mean time since onset of the first non-Raynaud's symptom was 11.5 (SD = 8.7) years.

The sample included 41 (6%) Black patients and 53 (8%) patients who identified as a member of one of the other racial/ethnic groups. As shown in Table 1, White patients were older and more likely to be married than Black patients, but less likely to have skin thickening on the hands and have diffuse SSc (statistically significant, $p < .0167$). There were also statistically significant differences between White and Black patients in presence of telangiectasias and skin pigmentation changes. White patients were older and less likely to have diffuse SSc than patients from other racial/ethnic groups (statistically significant, $p < .0167$). There were also statistically significant differences in skin pigmentation changes. (see Table 1).

There were a total of 565 (88%) women and 79 (12%) men in the total sample. There were no statistically significant differences between women and men for any sociodemographic or disease variables (see Table 1).

Appearance Concerns

Among women, 75% (521 of 565) reported appearance concerns, compared to 53% (42 of 79) of men ($p < .001$). Across racial/ethnic groups, 85% (35 of 41) of Black patients reported appearance concerns, compared to 71% (392 of 550) of White patients and 68% (36 of 53) from other racial/ethnic groups ($p = .122$).

As shown in Table 2, on an unadjusted basis, the odds of appearance concerns were greater for female patients compared to male patients (OR = 2.70, 95% Confidence Interval [CI] = 1.73-4.22, $p < .001$), and for Black patients versus White patients (OR = 2.64, 95% CI = 1.01-6.34, $p = .030$). Patients with other racial/ethnic status did not have significantly different odds compared to White patients (OR = 0.92, 95% CI = 0.52-1.65, $p = .788$). Other variables that were significantly associated with the presence of appearance concerns on an unadjusted basis included age, disease subtype, hand contractures, and skin thickening of the fingers.

In the multivariate analysis, only sex, age, and presence of moderate hand contractures were significantly associated with appearance concerns. The odds of appearance concerns for female SSc patients were significantly greater than for males (OR = 2.97, 95% CI = 1.78-4.96, $p < .001$). Older patients were also less likely to report appearance concerns than younger patients (OR = 0.98 per year, 95% CI = 0.96-0.99, $p = .009$), equivalent to a reduction of 18% in the OR for every 10-year increase in age. Race/ethnicity was not statistically significant, although the odds of reporting

appearance concerns among Black patients were greater than for White patients (OR = 1.71, 95% CI = 0.65-4.48, p = .279). The odds of reporting appearance concerns for patients with moderate hand contractures were almost twice those of patients with no or only mild hand contractures (OR = 1.97, 95% CI = 1.12-3.46, p = .019) (see Table 2). Model fit for the 11 predictors included was less than ideal based on the Hosmer-Lemeshow test ($\chi^2(8, N = 644) = 17.44$, p = .026), and the c-index statistic was 0.67.

DISCUSSION

The main finding was that women with SSc were substantially more likely than men to report appearance concerns, controlling for sociodemographic and disease variables. Black racial/ethnic group membership was significantly associated with appearance concerns at the bivariate level; however, the association was not statistically significant after accounting for the influences of other sociodemographic and disease variables. Older age was significantly associated with reduced odds of appearance concerns, and moderate hand contractures were significantly associated with greater odds.

The finding that female SSc patients had greater odds of experiencing appearance concerns is consistent with previous research on sex and visible differences, which has highlighted that women tend to experience greater distress, social anxiety, worry, self-consciousness, and difficulty adjusting to disfiguring appearance changes than men (12-15). The finding that younger patients had greater odds of experiencing appearance concerns also aligns with previous research in SSc, which has reported greater social discomfort based on appearance among younger patients (6).

Although 85% of Black patients reported appearance concerns compared to 71% of White patients, race/ethnicity was not independently associated with appearance concerns in multivariate analysis. It is possible that the small number of Black people in the study could explain the non-significant result. This finding may also have occurred because Black patients tended to be younger, have diffuse disease, and have greater pigmentation changes and skin thickening on the fingers compared to White patients. The only other variable that was significantly associated with appearance concerns at the multivariate level was the presence of moderate hand contractures. This finding aligns with previous research demonstrating that hand contractures are linked to more dissatisfaction with appearance in SSc (5). The greater odds of having appearance concerns among individuals with moderate hand contractures compared to those with severe hand contractures is likely due to the small number of patients who had severe hand contractures and not because moderate hand contractures are more likely to create appearance concerns than more severe hand contractures.

The present study is the first to specifically examine the presence of appearance concerns across sex and racial/ethnic groups in SSc, as well as other key sociodemographic and disease characteristics. Results highlight that multiple factors may contribute to appearance concerns in SSc. Overall, almost three of every four patients experience appearance concerns, and no single variable clearly separates patients with and without appearance concerns. For patients with appearance concerns, cognitive behavioural therapy (CBT) (4) and social skills training programs (28, 29) have been recommended as strategies to reduce social avoidance and increase self-esteem in social settings for other patient groups. Changing Faces, a UK not-for-profit

organization (www.changingfaces.org.uk), is an organization that has published a range of educational and self-help resources related to body image concerns due to visible differences that may also be relevant to SSc patients.

The present study has limitations that should be considered in interpreting results. First, the SPIN Cohort constitutes a convenience sample of SSc patients receiving treatment at a SPIN recruiting center, and patients at these centers may differ from those in other settings. Additionally, SSc patients in the SPIN Cohort complete questionnaires online, which may further limit the generalizability of findings. An additional limitation of the present study relates to the nature of disease characteristics included in the analyses. Specifically, the majority of the variables included in the models consisted of fairly crude indicators of either the presence or absence of a particular disease factor and did not provide a measure of severity. This may have reduced the ability to identify any associations, if present, between the severity of visible differences from the disease and the presence of appearance concerns. An additional limitation is that the sample sizes of Black patients and those self-identified as members of another racial/ethnic group were small, which may have limited the ability to detect meaningful differences in the odds of reporting appearance concerns for these groups. These limitations may have been reflected in the model fit statistics, which suggested that the overall model fit was less than ideal (27). It is also possible that the relatively limited information obtained from the predictive model could have been due to the dichotomous outcome variable. Most patients reported the presence of appearance concerns, and measuring appearance concerns may not be something that is easily categorized as present or absent.

In sum, female SSc patients had significantly higher odds of appearance concerns than male patients, as did younger SSc patients compared to older individuals and those with moderate hand contractures compared to those with no/mild hand involvement. Although Black SSc patients had higher odds of experiencing appearance concerns than White patients on an unadjusted basis, this result was not statistically significant when accounting for the influence of other sociodemographic and disease characteristics. It may be the case that greater appearance concerns among Black patients reflect more significant appearance changes. Replications of the current study with larger samples of Black and other racial/ethnic groups are needed. In addition, future studies should include assessment of appearance concerns with a continuously measured outcome variable.

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Table 1. Sociodemographic and Disease Characteristics (N = 644)

	Total	Race/Ethnicity				Sex		
		White	Black	Other		Female	Male	
Variable	**N = 644**	**N = 550**	**N = 41**	**N = 53**	**p value**	**N = 565**	**N = 79**	**p value**
Age (years), *mean ± SD*	55.3 ± 12.1	56.5 ± 11.5[a,b]	47.7 ± 11.5[a]	48.1 ± 14.1[b]	<.001	55.1 ± 12.0	56.2 ± 12.8	.462
Education >12 years, *n (%)*	512 (79.5)	434 (78.9)	33 (80.5)	45 (84.9)	.579	451 (79.8)	61 (77.2)	.591
Currently employed (full or part-time), *n (%)*	268 (41.7)[c]	225 (41.0)[d]	21 (51.2)	22 (41.5)	.439	236 (41.8)	32 (41.0)[e]	.901
Married/living as married, *n (%)*	471 (73.1)	413 (75.1)[a]	21 (51.2)[a]	37 (69.8)	.003	412 (72.9)	59 (74.7)	.741
Time since onset of first non Raynaud's symptoms (years), *mean ± SD*	11.5 ± 8.7[f]	11.6 ± 8.9[g]	8.9 ± 5.6[h]	10.2 ± 7.8[i]	.067	11.7 ± 8.8[j]	10.4 ± (7.8)[k]	.227
Patients with diffuse SSc, *n (%)*	258 (40.1)	200 (36.4)[a,b]	28 (68.3)[a]	30 (56.6)[b]	<.001	226 (40.0)	32 (40.5)	.971
Telangiectasias					<.001			.058
None, *n (%)*	183 (28.4)	137 (24.9)[a]	26 (63.4)[a]	20 (37.7)		153 (27.1)	30 (38.0)	

Body and face, *n (%)*	298 (46.3)	273 (49.6)[a]	7 (17.1)[a]	18 (34.0)		262 (46.4)	36 (45.6)	
Body only, *n (%)*	163 (25.3)	140 (25.5)[a]	8 (19.5)[a]	15 (28.3)		150 (26.5)	13 (16.5)	
Pigmentation changes					<.001			.497
None, *n (%)*	438 (68.0)	396 (72.0)[a,b]	13 (31.7)[a]	29 (54.7)[b]		385 (68.1)	53 (67.1)	
Body and face, *n (%)*	105 (16.3)	71 (12.9)[a,b]	18 (43.9)[a]	16 (30.2)[b]		89 (15.8)	16 (20.3)	
Body only, *n (%)*	101 (15.7)	83 (15.1)[a,b]	10 (24.4)[a]	8 (15.1)[b]		91 (16.1)	10 (12.7)	
Hand contractures					.898			.988
No/mild (0-25%), *n (%)*	488 (75.8)	418 (76.0)	31 (75.6)	39 (73.6)		428 (75.8)	60 (75.9)	
Moderate (25-50%), *n (%)*	121 (18.8)	102 (18.5)	7 (17.1)	12 (22.6)		106 (18.8)	15 (19.0)	
Severe (>50%), *n (%)*	35 (5.4)	30 (5.5)	3 (7.3)	2 (3.8)		31 (5.5)	4 (5.1)	
Skin thickening of fingers, *n (%)*	360 (55.9)	293 (53.3)[a]	31 (75.6)[a]	36 (67.9)	.004	311 (55.0)	49 (62.0)	.242
Presence of appearance concerns, *n (%)*	463 (71.9)	392 (71.3)	35 (85.4)	36 (67.9)	.122	421 (74.5)	42 (53.2)	<.001

SD = standard deviation; [a]White patients statistically significantly different from Black patients; [b]White patients statistically significantly different from patients from another racial/ethnic group; [c]N = 643; [d]N = 549; [e]N = 78; [f]N = 595; [g]N = 510; [h]N = 40; [i]N = 45; [j]N = 519; [k]N = 76.

Table 2. Unadjusted and Adjusted Comparisons of Appearance Concerns across Sociodemographic and Disease Variables

Variable	Unadjusted		Adjusted	
	OR (95% CI)	p value	OR (95% CI)[a]	p value
Race/ethnicity (reference = White)				
Black	2.64 (1.01-6.34)	.030	1.71 (0.65-4.48)	.279
Other	0.92 (0.52-1.65)	.788	0.57 (0.30-1.11)	.098
Age	0.97 (0.96-0.98)	<.001	0.98 (0.96-0.99)	.009
Female sex	2.70 (1.73-4.22)	<.001	2.97 (1.78-4.96)	<.001
Education ≤12 years	0.69 (0.47-1.02)	.063	0.74 (0.48-1.15)	.179
Not married/living as married	1.36 (0.92-2.00)	.121	1.05 (0.68-1.61)	.825
Diffuse disease subtype	1.76 (1.24-2.50)	.002	1.18 (0.76-1.83)	.456
Telangiectasias (reference = none)				
Body and face	0.81 (0.54-1.22)	.313	1.51 (0.90-2.53)	.120
Body only	0.70 (0.44-1.09)	.116	1.21 (0.78-1.88)	.390
Pigmentation changes (reference = none)				

Body and face	1.58 (0.96-2.58)	.071	0.91 (0.53-1.59)	.745
Body only	1.50 (0.92-2.47)	.108	1.01 (0.50-2.03)	.976
Hand contractures (reference = none)				
Moderate	2.33 (1.41-3.86)	.001	1.97 (1.12-3.46)	.019
Severe	2.03 (0.88-4.70)	.099	1.74 (0.66-4.59)	.261
Skin thickening on hands	1.64 (1.18-2.28)	.003	1.40 (0.92-2.11)	.114

OR = Odds ratio; CI = Confidence interval; [a]Adjusted for age, sex, race/ethnicity, education level, marital status, disease subtype, telangiectasis, pigmentation changes, hand contractures, and skin thickening on fingers.