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UNIVERSITY OF CALIFORNIA, SAN DIEGO

SAN DIEGO STATE UNIVERSITY

Latent Profile Analysis-Derived Typologies of Systemic Sclerosis Patients Using Body Image Indicators

A dissertation submitted in partial satisfaction of the requirements for the degree of Doctor of Philosophy

in

Clinical Psychology

by

Shadi Gholizadeh

Committee in charge:

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        Professor Thomas Rutledge

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        Professor Aaron Blashill
        Professor Scott C. Roesch

2018

The Dissertation of Shadi Gholizadeh is approved, and it is acceptable in quality and form for publication on microfilm and electronically:

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Chair

University of California, San Diego

San Diego State University

2018

DEDICATION

"'It doesn't happen all at once,' said the Skin Horse. 'You become. It takes a long time. That's why it doesn't happen often to people who break easily, or have sharp edges, or who have to be carefully kept. Generally, by the time you are Real, most of your hair has been loved off, and your eyes drop out and you get loose in the joints and very shabby. But these things don't matter at all, because once you are Real you can't be ugly, except to people who don't understand.'" –Margery Williams Bianco, <u>The Velveteen Rabbit</u>

I am constantly perplexed, challenged, frustrated, and fascinated with the construct of body image. There are moments where I find myself wishing there was nothing left to study—that the contouring, coloring, shape, disfigurements, and visible differences of the face and body held as much power as, say, the exact specifications of one's spleen in the fostering of human connection and the development of identify, confidence, and worth. However, in this line of research, I am reminded that at present, such is not our state; the outside world can be judgmental and harsh. But, I am also constantly reminded that people can be compassionate, kind, patient, and authentic. This project is dedicated to the humans living with rare diseases, especially those conditions that change the outer masks and exterior armor that adorn us.

TABLE OF CONTENTS

LIST OF FIGURES

LIST OF TABLES

ACKNOWLEDGEMENTS

I feel incredibly fortunate to be completing my doctoral journey under the ever-present wing of Vanessa L. Malcarne, Ph.D. I knew prior to entering the program that I would be very fortunate to grow professionally with the direct mentorship of an outstanding educator, researcher, scholar, and writer; after working alongside Vanessa, I had the pleasant surprise of learning that I would also find a lover of true crime shows, long laughs, good chocolate, and cross-disciplinary thought pieces. This allowed the past five years not only to be academically enriching, but also personally joyous and imbued with fun and creativity. To have such a kind, generous, and dedicated mentor to guide one along the sometimes-sinuous doctoral path is an invaluable gift.

I would like to express my sincere gratitude to Brett D. Thombs, Ph.D., Linda Kwakkenbos, Ph.D., and Marie-Eve Carrier, M.Sc., for bringing me into the wonderful Scleroderma Patient-centered Intervention Network (SPIN) family. As anyone who has worked in the acutely challenging environment of rare diseases can attest, it is truly a blessing to work collaboratively with the researchers and clinicians who devote their lives to rare disease work. The data used for this dissertation come from the SPIN cohort. I am thankful to the people living with scleroderma who contributed to this dataset, and who have been so gracious with their time and spirit.

I would also like to acknowledge the other members of my committee, Niloofar Afari, Ph.D., Aaron Blashhill, Ph.D., Scott C. Roesch, Ph.D., and Thomas Rutledge, Ph.D. Dr. Afari manages to strike that elusive balance between unstoppable academic powerhouse, pushing her mentees to exceed their self-imposed and institutional limits, and genuine nurturer, reminding students of the critical role of values and heart in

scholarship, in a way that is elegant and inspiring. Dr. Blashill has been an interdisciplinary sage, whose in-depth knowledge of the fascinating construct of body image in psychology and public health has been immensely helpful to me, and whose open office door is a welcome respite for laughs and analysis-induced tears alike. To Dr. Roesch I owe so much—he has answered perhaps over 1000 frenetic, Faulknerian emails about statistical queries and anxieties over the past five years; that someone so astoundingly smart can also be kind, accessible, and patient is very humbling. I am also thankful to Dr. Rutledge, who I remembered as supremely engaging and energetic from when he interviewed me for the doctoral program, and who I later learned is also a brilliant public speaker whose presentation on confidence will prevail each time I feel small. I am immensely thankful to my committee for being so generous with their wisdom and time.

I would also like to acknowledge my JDP lab mates and classmates, especially Sarah D. Mills, Rina S. Fox, and Erin L. Merz—what an amazing group of humans; I am grateful to Vanessa for creating this cadre of health obsessed psychologists.

My gratitude is boundless for my wonderful parents, Farzaneh 'Frannie' and Bahram, and my incredible sisters-by-soul Roujin, Megan, Jhalé, and Lily.

And then, how does one thank a friend who not only offers years of encouragement when the pressure is high and heaps genuine understanding when deadlines can dictate day-to-day life, but then ups the ante by asking to forever stand witness to such odd pursuits? I am beyond grateful to my husband, Babak—thank you for giving me the space to do what I love most, even if it sometimes means being away from the one I love most.

The results of this dissertation are being prepared for publication. Publications based on this dissertation will be co-authored Vanessa L. Malcarne, Scott C. Roesch, Marie-Eve Carrier, Linda Kwakkenbos, Sarah D. Mills, Rina S. Fox, and Brett D. Thombs. The dissertation author was the primary investigator and author of this material.

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PUBLICATIONS

Fox, R. S., Kwakkenbos, L., Carrier, M. E., Mills, S. D., **Gholizadeh, S.**, Jewett, L. R., ... & Gottesman, K. (in press). Validation of the Brief Fear of Negative Evaluation Scale-II in patients with systemic sclerosis: A Scleroderma Patient-centered Intervention Network Cohort study. *Arthritis Care & Research.*

Mills, S. D., Kwakkenbos, L., Carrier, M. E., **Gholizadeh, S.**, Fox, R. S., Jewett, L. R., ... & Baron, M. (in press). Validation of the Social Appearance Anxiety Scale in Patients with Systemic Sclerosis: A Scleroderma Patient-centered Intervention Network Cohort Study. *Arthritis Care & Research.*

Mills, S. D., Azizoddin, D., **Gholizadeh, S.**, Racaza, G. Z., & Nicassio, P. M. (in press). The mediational role of helplessness in psychological outcomes in systemic lupus erythematosus. *Lupus.*

Merz, E. L., Kwakkenbos, L., Carrier, M. E., **Gholizadeh, S.**, Mills, S. D., Fox, R. S., ... & Furst, D. E. (2018). Factor structure and convergent validity of the Derriford Appearance Scale-24 using standard scoring versus treating 'not applicable'responses as missing data: a Scleroderma Patient-centered Intervention Network (SPIN) cohort study. *BMJ open*, *8*, e018641.

Gholizadeh, S., Kwakkenbos, L., Carrier, M. E., Mills, S. D., Fox, R. S., Jewett, L. R., ... & SPIN Investigators. (2018). Validation of the Social Interaction Anxiety Scale in scleroderma: A Scleroderma Patient-centered Intervention Network Cohort study. *Journal of Scleroderma and Related Disorders*, *3*, 98-105.

Gholizadeh, S., Rooney, B., Merz, E.L., Malcarne, V.L., Safren, S., & Blashill, A.J. (in press). Body image and condomless anal sex among sexual minority men living with HIV. *AIDS and Behavior, 22,* 658-662.

Mills, S. D., Fox, R. S., **Gholizadeh, S.**, Klonoff, E. A., & Malcarne, V. L. (2017). Acculturation and Health Behaviors Among African Americans: A Systematic Review. *Journal of Cross-Cultural Psychology*, *48*, 1073-1097.

Gholizadeh, S., Mills, S. D., Fox, R. S., Merz, E. L., Roesch, S. C., Clements, P. J., ... & Malcarne, V. L. (2017). Structural Validity of the Rheumatology Attitudes Index in Systemic Sclerosis: Analysis from the UCLA Scleroderma Quality of Life Study. *The Journal of Rheumatology*, *44*, 795-798.

Jewett, L. R., Kwakkenbos, L., Carrier, M. E., Malcarne, V. L., Harcourt, D., Rumsey, N., ... & **Gholizadeh, S.** (2017). Validation of the Body Concealment Scale for Scleroderma (BCSS): Replication in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. *Body Image*, *20*, 99-106.

Gholizadeh, S., Malcarne, V.L., & Schatman, M. (2016). Functioning well in a dysfunctional system: Recommendations for clinical psychologists in workers' compensation settings. *Psychological Injury and Law, 9,* 360-375.

Jewett, L., Kwakkenbos, L., Malcarne, V.L., Bartlett, S., Furst, D. Gottesman, K., Mayes, M., Assassi, S., Harcourt, D., Williamson, H., Johnson, S., Korner, A., Steen, V., Fox, R.S., **Gholizadeh, S.**, Mills, S., Molnar, J., & Thombs, B.D. (2016). Examination of the association of sex and race/ethnicity with appearance concerns: A Scleroderma Patient-centered Intervention Network cohort study. *Clinical and Experimental Rheumatology, 34,* 392-399.

Backhaus, A., **Gholizadeh, S.**, Godrey, K.M., Pittman, J., & Afari, N. (in press). The many wounds of war: The association of service-related and clinical characteristics with problems with the law in Iraq and Afghanistan Veterans. *International Journal of Law and Psychiatry, 49*, 205-213.

Levis, A. W., Harel, D., Kwakkenbos, L., Carrier, M., Mouthon, L., Poiraudeau, S., Bartlett, S. J., Khanna, D., Malcarne, V. L., Sauve, M., van den Ende, C. H. M., Poole, J. L., Schouffoer, A. A., Welling, J., Thombs, B. D., **& the SPIN Investigators** (2016). Development and validation of the Cochin Hand Function Scale Short Form (CHFS-6) using optimal test assembly methods: A Scleroderma Patient-centered Intervention Network (SPIN) cohort study. *Arthritis Care & Research, 68,* 1704-1713.

Riehm, K. E., Kwakkenbos, L., Carrier, M., Bartlett, S. J., Malcarne, V. L., Mouthon, L., Nielson, W. R., Poiraudeau, S., Nielsen, K., Baron, M., Frech, T., Hudson, M., Pope, J., Sauve, M., Suarez-Almazor, M., Wigley, F. M., Thombs, B. D., & **the SPIN Investigators** (in press). Validity of the Self-Efficacy for Managing Chronic Disease (SEMCD) scale: A Scleroderma Patient-centered Intervention Network (SPIN) cohort study. *Arthritis Care & Research.* doi:10.1002/acr.22807

Gholizadeh, S., & Malcarne, V. L. (2015). Professional and ethical challenges in

determinations of causality of psychological disability. *Psychological Injury and Law*, *8*, 334-347.

Gholizadeh, S., Malcarne, V. L., & Schatman, M. E. (2015). Ethical quandaries for psychologists in workers' compensation settings: The GAF gaffe. *Psychological Injury and Law*, *8*, 64-81.

Kwakkenbos, L., Delisle, V. C., Fox, R. S., **Gholizadeh, S.**, Jewett, L. R., Levis, B., ... & Thombs, B. D. (2015). Psychosocial aspects of scleroderma. *Rheumatic Disease Clinics of North America*, *41*, 519-528.

Kwakkenbos, L., Jewett, L. R., Baron, M., Bartlett, S. J., Furst, D., Gottesman, K., Khanna, D., Malcarne, V. L., Mayes, M. D., Mouthon, L., Poiraudeau, S., Sauve, M., Nielson, W. R., Poole, J. L., Assassi, S., Boutron, I., Ells, C., van den Ende, C. H. M, Hudson, M., Impens, A., Körner, A., Leite, C., Costa Maia, A., Mendelson, C., Pope, J., Steele, R. J., Suarez-Almazor, M. E., Ahmed, S., Coronado-Montoya, S., Delisle. V. C., **Gholizadeh, S.**, Jang, Y., Levis, B., Milette, K., Mills, S. D., Razykov, I., Fox, R. S., & Thombs, B. D. (2013). The Scleroderma Patient-centered Intervention Network (SPIN) Cohort: Protocol for a cohort multiple randomized controlled trial (cmRCT) design to support trials of psychosocial and rehabilitation interventions in a rare disease context. *BMJ Open, 3,* e003563.

Malcarne, V. L., Fox, R. S., Mills, S. D., & **Gholizadeh, S.** (2013). Psychosocial aspects of systemic sclerosis: Recent findings. *Current Opinion in Rheumatology, 25*, 707-713.

Beusterien, K., Tsay, S., **Gholizadeh, S.**, & Su, Y. (2013). Real-world experience with colorectal cancer chemotherapies: Patient web forum analysis. *Ecancer Medical Science*, 7, 361-371.

Bygum, A., Aygören-Pürsün, E., Caballero, T., Beusterien, K., **Gholizadeh, S.**, Musingarimi, P., Wait, S., & Boysen, H. (2012). The hereditary angioedema burden of illness study in Europe (HAE-BOIS-Europe): Background and methodology. *BMC Dermatology*, *12*, 4-13.

Asherson, P., Akehurst, R., Kooij, J. J., Huss, M., Beusterien, K., Sasané, R., **Gholizadeh, S.**, & Hodgkins, P. (2012). Under diagnosis of Adult ADHD: Cultural influences and societal burden. *Journal of Attention Disorders*, *16*, 20S-38S.

Gholizadeh, S., & Hook, D. W. (2012). The discursive construction of the 1978–1979 Iranian Revolution in the speeches of Ayatollah Khomeini. *Journal of Community & Applied Social Psychology*, *22*, 74-186.

BOOK CHAPTERS

Gholizadeh, S., Fox, R. S., Mills, S. D., Jewett, L. R., Thombs, B. D., & Malcarne, V. L. (2017). Coping with the disfigurement of scleroderma: Facial, skin, and hand changes (pp. 713-721). In F. M. Wigley (Ed.), *Scleroderma: From pathogenesis to comprehensive management*, 2nd ed. New York: Springer.

ABSTRACT OF THE DISSERTATION

Latent Profile Analysis-Derived Typologies of Systemic Sclerosis Patients Using Body
Image Indicators

by

Shadi Gholizadeh

Doctor of Philosophy in Clinical Psychology

University of California, San Diego, 2018
San Diego State University, 2018

Professor Vanessa L. Malcarne, Chair

Rationale. Systemic Sclerosis (SSc, scleroderma) is a rare and progressive
rheumatic disease of unknown etiology and heterogeneous presentation that results in
fibrosis of the skin and internal organs. A common and distressing symptom of SSc is
disfigurement in visible and socially relevant areas of the body (i.e., face and hands).
Disease-related changes in appearance have been associated with body image
dissatisfaction and social anxiety. Although there have been studies identifying correlates

of body image dissatisfaction, there is a need for an examination that considers the complex relationships among the personal and social aspects of appearance changes. The present study used latent profile analysis (LPA) to identify body image typologies. Identified groups were then compared on key sociodemographic, medical, and psychosocial variables.

Design. The sample consisted of 942 patients with physician-confirmed SSc enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort who completed study questionnaires from April 2014 through October 2016. Patients in the SPIN Cohort were enrolled at 28 centers from Canada, the United States, and the United Kingdom. Prior to running the analysis, the sample was randomized into two different groups that were treated as independent samples ($N = 469$ in Sample 1 and $N = 473$ in Sample 2); this randomization was completed to allow for a replication analysis of the findings. For the first aim, exploratory LPA was used to derive categorical latent variables that signified profiles of similarly scoring individuals using one indicator of objective skin involvement, three indicators of subjective body image, and three indicators of social anxiety. For the second aim, group differences were examined, using the Lanza three-step method for modeling auxiliary variables, for selected sociodemographic, medical, and psychosocial variables.

Results. In both samples, a two-profile solution was derived. These two classes were substantively analyzed for patterns of scores and termed the *Appearance Comfortable* ($n = 334$ and $n = 375$ in Sample 1 and Sample 2, respectively) and *Appearance Distressed* ($n = 135$ and $n = 98$ in Sample 1 and Sample 2, respectively) groups. In both samples, younger age, diffuse disease subtype, and the presence of

xvii

hypo/hyper-pigmentation were associated with membership in the *Appearance Distressed* group. Additionally, patients in the *Appearance Distressed* group had significantly higher scores on measures of depressive and anxious symptoms and physical disability.

Conclusions. The present study was the first to use LPA in the context of body image in SSc, and the first study to identify typologies of patients based on indicators of body image in any disfiguring condition. Two distinct groups were identified distinguishing between an *Appearance Comfortable* group, comprised of patients with lower objective skin involvement, better body image, and lower social anxiety and an *Appearance Distressed* group including patients who had higher objective skin involvement, poorer body image, and higher social anxiety. Interestingly, although the differences in objective skin involvement were relatively small, the differences in body image and social anxiety scores were relatively large. This suggests that the body image experience in SSc is driven by psychosocial factors beyond objective appearance. The results also elucidated variables that can indicate likely group membership and help identify which individuals may be most vulnerable to poorer body image-related outcomes.

CHAPTER 1: INTRODUCTION

The present study was the first to utilize the multivariate statistical technique of latent profile analysis (LPA) in the context of body image in a rare, rheumatic disease, systemic sclerosis (scleroderma, SSc). The use of this methodology enabled the exploration of two questions regarding the experience of body image in SSc. First, LPA allowed the identification of distinct groups that emerged based on body image and social anxiety indicators. Second, once profiles were established, the analysis enabled comparison of the groups to determine whether patients in the identified groups differed on selected sociodemographic, medical, and psychosocial variables.

The present study used baseline data for a total of 942 men and women with SSc who were enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort (Kwakkenbos et al., 2013) in Canada, the United States, and the United Kingdom in order to derive typologies of SSc patients using various body image indicators. The total sample was randomized into two different groups that were treated as independent samples ($N = 469$ in Sample 1 and $N = 473$ in Sample 2) to examine whether the findings could be replicated.

Identifying typologies of patients based on various indicators of body image and social anxiety can enable a more refined understanding of the experience of body image and associated outcomes in SSc patients. The present study was an attempt to unpack the subjective and objective indicators of body image and closely related social impacts in a population in which changes in appearance are prevalent and burdensome (Jewett et al., 2016*a*; Kwakkenbos et al., 2015*a*). In so doing, a multi-dimensional, non-linear, understanding of body image in SSc can be achieved (Bregman, Malik, Page, Makynen,

& Lindahl, 2013). This more refined knowledge of body image in SSc can then identify groups at higher risk for poorer outcomes. It is the goal of this researcher that the results eventually lead to an improved understanding of the broad construct of body image specific to SSc patients to improve overall quality of life and perhaps contribute to disease-specific assessment and intervention development in the future.

Prior to delving into the findings of the present study, a background of the relevant literature in body image will be presented, a brief overview of SSc will be provided, and the existing research examining body image impacts in SSc will be summarized. Gaps in the literature will be highlighted and the role of the present study in filling these gaps will be elucidated.

Chapter 1 is being prepared in part for publication. This publication will be co-authored by Vanessa L. Malcarne, Scott C. Roesch, Marie-Eve Carrier, Linda Kwakkenbos, Sarah D. Mills, Rina S. Fox, and Brett D. Thombs. The dissertation author was the primary investigator and author of this material**.**

CHAPTER 2: BACKGROUND AND SIGNIFICANCE

2.1 The Construct of Body Image

Body image is a complex construct that is notoriously difficult to empirically

tether (Banfield & McCabe, 2002). Broadly, body image refers to perceptions, thoughts,

feelings, and behaviors about one's body. The seeds of body image research took root via

neuropsychological research exploring such phenomena as phantom limb and

hemiasomatognosia over a century ago, and germinated in the late 1960s via

psychoanalytic explorations of body awareness and the body image boundary (Cash,

2004; Fisher, 1986). But it is in recent decades that there has been a booming growth of

body image research, however much of it has been limited to the study of eating disorders

(Cash, 2004; Grogan, 2006). In fact, Grogan (2006) argued that there are biases in

definitions of body image (e.g., the inclusion of body shape and size evaluations) and

populations (e.g., physically healthy girls) shaped by the origins of body image research

in the fields of clinical psychology and psychiatry.

Cash (2004) defined the multidimensional construct of body image as follows,

"Body image refers to the multifaceted psychological experience of embodiment,

especially but not exclusively one's physical appearance" (p. 1). Evaluative (e.g.,

perceptions and attitudes) and investment (e.g., salience or cognitive-behavioral-placed

value) components of body image cohere to create the construct of body image (Cash,

1994, 2002*a*, 2002*b*; Cash, Melnyk, & Hrabosky, 2004*a*). The evaluative component

includes appraisals of and satisfaction with appearance whereas the investment

component includes the value and importance that one affords appearance (Cash, 1994;

Cash, Fleming, Alindogan, Steadman, & Whitehead, 2002). Furthermore, thoughts,

emotions, and behaviors related to one's body can be a product of one's overall appearance or related to a specific body part or quality, such as weight (Cash, Morrow, Hrabosky, & Perry, 2004*b*). The differentiation between body image dissatisfaction and distress is also important to note. Whereas body image dissatisfaction refers to an evaluative process, body image distress (or, disturbance) typically also includes more strongly negative body image emotions and can point to a more extreme, pathological process, for example body dysmorphia in eating disordered populations (Hrabosky et al., 2009).

One of the leading theoretical frameworks in body image research is the sociocultural model, which is based on an understanding that 1) there are socially and culturally constituted beauty ideals; 2) these ideals are societally transmitted to individuals; 3) the ideals are internalized and manifest in [dis]satisfaction within an individual depending on the extent to which the individual meets the prescribed ideals. This model is most commonly referenced in relation to body weight and shape and culturally sanctioned ideals of thinness or beauty (Tiggemann, 2011), and the role of media in perpetuating such ideals (Thompson & Heinbert, 1999). The utility of the model in terms of highlighting the role of one's sociocultural context and products in body image was demonstrated in a study that showed that exposure to Barbie dolls was associated with lower body esteem and thinness desire in young girls (Dittmar, Halliwell, & Ive, 2006). Tiggemann (2011) cautioned, however, that the sociocultural model is overly simplistic and fails to take into account other (e.g., psychological) influences; if the model were sufficient, the great majority of individuals in every society would be suffering from elevated body dissatisfaction and eating disorders.

Although much of the work in body image has centered around eating disorders, there has been an increasing awareness of the construct of body image in other contexts as well, including chronic illness and disfigurement (congenital and acquired; Rumsey & Harcourt, 2004).

2.2 Body Image in Disfigurement

Differentiating disfigurement from physical normality. Rumsey and Harcourt (2004) noted a distinction between the types of concerns about appearance that are common in the general (i.e., non-disfigured) population and the specific concerns of individuals living with visible differences. Of course, the line demarcating disfigurement from physical normality is not always clear and there can be disagreement, especially in cases involving body dysmorphia, about what is considered physically *normal*. There is an entire evolutionary biology literature devoted to such constructs as fluctuating asymmetries facial sexual dimorphism. However, there are numerous health conditions, both congenital (e.g., Treacher-Collins' syndrome) and acquired (e.g., burns) that can contribute to visible differences that can be considered deviations from what is physically typical in one's cultural context. Certain visible differences attributed to medical reasons are common (e.g., approximately one-third of individuals will be affected by skin conditions, such as acne or uticaria, at any given time point; Thompson, 2011), whereas other conditions impacting appearance (e.g., ichthyosis vulgaris) can be quite rare. Although the heterogeneity in visible disfigurement in terms of such characteristics as body areas affected, severity, visibility, and individual characteristics can seem "daunting" (p. 85) in a research context, in fact, the body image challenges tend to most commonly involve 1) negative perceptions of the self and 2) social difficulties (Rumsey

& Harcourt, 2004). In other words, the challenges of living with a visible difference can be divided into personal and the social demands.

Apropos the multidimensional nature of the construct of body image, the severity of disfigurement does not directly predict outcomes such as adjustment (Rumsey, 2002*a*, 2002*b*). Thus, there is not only a distinction between objective severity and subjective severity, but also a host of other variables, such as salience and resilience, that may impact the relationship between severity of disfigurement and various outcomes. Although objective ratings of attractiveness or disfigurement can be challenging to empirically acquire, there have been some studies that examined both objective and subjective disfigurement. For example, Moss (2005) used both objective (i.e., clinician) ratings of severity and subjective (i.e., patient) ratings of severity in order to examine the relationship between severity of disfigurement and psychological adjustment.

Thompson (2011) described three primary categorizations among psycho-dermatological conditions: 1) dermatological issues that are grounded in psychopathology (e.g., trichotillomania); 2) conditions that are psychophysiological (e.g., a subtype of alopecia exacerbated by stress); and 3) medical conditions for which psychological impacts are secondary (e.g., depression and anxiety related to systemic lupus erythematosus [SLE]). Rheumatologic conditions that cause disfigurement, such as scleroderma, would fall into this third category. Jolly (2011) described that body image issues are pervasive across rheumatologic diseases, which include visible and non-visible adverse bodily changes. Jolly (2011) underscored that the detection and treatment of body image dissatisfaction and distress in rheumatologic conditions may improve overall

health outcomes and costs associated with the disease because of the role of body image distress in other psychosocial outcomes and quality of life.

Social impacts. The impacts of a visible difference on social processes can be devastating and individuals with visible differences have reported social challenges such as staring, invasive questions, and rude comments (Rumsey 2002*a*, 2002*b*; Rumsey & Harcourt, 2004). Visible differences have been referred to as a *social disability* by Macgregor (1979) because of the potential for social impacts and noticeability (Rumsey & Harcourt, 2004). Both experimental and observational studies have shown that people tend to react negatively in response to individuals with visible skin disfigurements (Thompson, 2011). Grandfield, Thompson, and Turpin (2005) used the Implicit Association Test to examine implicit attitudes toward individuals with dermatological conditions and identified an implicit preference for clear skin. Across disfiguring diseases, patients with facial disfigurement report fear of negative evaluation and social avoidance and challenges maintaining social interactions (Newell & Marks, 2000; Pruzinsky, 1992). Pruzinsky (1992) reported that the stigmatization of individuals with visible differences can even extend to intimate relationships.

2.3 The Assessment of Body Image

With the growing interest in body image research, there has been a concomitant growth in the assessment of the body image construct. Speaking of this symbiotic relationship, Thompson (2004) explained that, "As it often happens in science, such a phenomenon both stimulates and is fueled by the development of new measurement indices. In the case of body image, the exponential increase in the number of new and revised measures purporting to assess one or more multiple dimensions of body image

has been well documented" (p. 7). The sheer number of terms used to describe the specific body image dimension of interest (e.g., [dis]satisfaction, orientation, quality of life, distress, disturbance, esteem, shame) can be formidable for a researcher, let alone the number of validated measures. Thompson, Roehrig, Cafri, and Heinberg (2005) described some of the most commonly used measures for the assessment of body image in eating disorders and separated these in terms of whether they assessed an affective, behavioral, perceptual, or cognitive dimension of body image. Examples of the most common measures of body image used in general populations included the Body Image Quality of Life Inventory (Cash & Fleming, 2002), which explores how body image affects 19 life domains, the Body Image Ideals Questionnaire (Szymanski & Cash, 1995), which elicits ratings of an individual's specific attributes from their individual viewpoint and that of a romantic partner, and the Multidimensional Body-Self Relations Questionnaire (MBSRQ; Cash, Wintstead, & Janda, 1986), a measure of ten subscales, including Evaluation, Orientation, Fitness, Health/Illness, Overweight Preoccupation, Self-Classified Weight, and the Body Areas Satisfaction Scale (BASS; Cash, 2000). However, there is no "gold standard" assessment of body image. There are also assessments that seek to capture the more pathological experience of body image distress, disturbance, or dysmorphia. For example, the Body Image Disturbance Questionnaire (BIDQ; Cash, Phillips, Santos, & Hrabosky, 2004*c*) measures preoccupation with physical appearance, emotional distress, avoidance behavior, and the functioning burden of body image concerns.

Thompson (2004) warned of common pitfalls in body image research, which are arguably germane in non-eating disorders contexts, given the relative dearth of prior

research and models from which to pull. He noted that the following ten strategies would help to mitigate some of the most common measurement problems:

1) Identify the specific dimension of body image being investigated (e.g., satisfaction with a particular body part);

2) Use several measures of body image to capture multiple dimensions of body image;[1]

3) Ensure that measures have established psychometric properties (e.g., validity);

4) Ensure that measures have demonstrated validity specifically for your population (e.g., rheumatologic disease);

5) Ensure that measure have demonstrated validity for your sample;

6) Consider whether a slight change of wording may better answer your research question if the measure is focused on weight/shape/size; this may be a better alternative to creating a new measure altogether;

7) Consider if a state or trait measure of body image is more appropriate for your research question;

8) Ensure that the instructional protocol is not vague;

9) Consider whether subgroup analyses (e.g., for gender) would be more appropriate;

10) Evaluate data in the context of norms and clinical significance.

The above are meant to signify best practices in assessments for body image research. Although it may not be possible to ensure that each criterion is met, particularly in cases

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[1] Thompson (2004) further added that because many of the body image constructs are orthogonal to each other (e.g., investment and orientation), multicollinearity among measures for certain analyses (e.g., regression) is typically not a concern.

where the researcher is using preexisting data, aiming to meet the criteria and noting as a limitation instances where they cannot be met, can bolster the quality of the research.

There has been a growing interest in the disease-specific assessment of body image in populations reflecting physical illness and visible differences. In fact, Cash and Smolak (2011) referred to body image research in medical contexts as, "one of the new frontiers of scholarship" (p. 8). Body image is a relevant construct in the rare connective tissue disease SSc given that disfigurement is a "central feature" of the disease (Thombs et al., 2010, p. 1183). A brief overview of the epidemiology of SSc and a summary of the extant research on body image in SSc will be provided below.

2.4 Body Image in Systemic Sclerosis

The epidemiology and etiology of SSc. SSc is a multisystem, autoimmune connective tissue disease of unknown etiology characterized by antibody production, vasculopathy of the small vessels, and excessive deposition of collagen (Seibold, 2005; Wigley & Hummers, 2003). Organ involvement most commonly incudes the kidneys, esophagus, heart, and lungs, with lung involvement being the most common cause of death (Gabrielli, Avvedimento, & Krieg, 2009). It is estimated that the prevalence of SSc ranges from 50 to 300 cases per one million persons, with incidence ranging from 2.3 to 22.8 new cases per one million persons annually (Chifflot, Fautrel, Sordet, Chatelus, & Sibilia, 2008). Women are 4.6 time as likely as men to have SSc (Mayes et al., 2003). Black individuals have slightly higher odds (1.15, 95% CI 1.02-1.30) of being affected with SSc than White individuals, and are significantly younger at diagnosis than White individuals (Mayes et al., 2003). To contextualize the rarity of the condition, the prevalence of rheumatoid arthritis (RA), SLE, and SSc among U.S. adults is estimated at

1.3 million, 161,000 to 322,000, and 49,000, respectively (Helmick et al., 2008). SSc is complex and highly heterogeneous in its presentation.

There is no gold-standard measure of severity in SSc, precisely because the disease can present heterogeneously and involve different organ systems (Hudson, Steele, Canadian Scleroderma Research Group, & Baron, 2007). There are, however, commonly used proxies for disease severity, including forced vital lung capacity and skin thickness (Hudson et al., 2007). The classification of such a heterogeneous disease can also be a challenge. As elegantly described by LeRoy and colleagues (1988), whereas disease itself is a "pathobiologic continuum," disease classification is "…a heuristic, compartmental exercise dependent on limited current understanding." They continued, "For those who venture to classify, subsets so defined should make a difference to the patient in outcome, to the health professional in intervention and surveillance, and to the investigator for suitable subjects for study" (p. 202).

There is a widely used, well-accepted, and clinically meaningful disease subtype classification in SSc between limited cutaneous and diffuse cutaneous disease. In limited disease, skin thickening is typically constrained to the, hands, arms, and/or face; if organ involvement is present, it typically has a very delayed (i.e., several decades) onset; in diffuse disease, the skin involvement is widespread, including the trunk, and can include progression and serious organ complications (Gabrielli et al., 2009; Steen, Powell, & Medsger, 1988).

There is no known cause of SSc. A qualitative study (Gholizadeh et al., 2015*a*) explored patient attributions of scleroderma origin and identified that seven themes were most commonly involved in patient theories of etiology: (i) stress; (ii) environment; (iii)

genetics; (iv) medical conditions; (v) diet; (vi) substance use; and (vii) spirituality. There have been theories linking environmental exposures to SSc, with silica exposure being linked to a significantly higher risk of developing in SSc in two studies (Makol, Reilly, & Rosenman, 2011; McCormic et al., 2010), however such environmental disease triggers are associated with a small proportion of total cases (Barnes & Mayes, 2012). Other studies have identified genetic risk factors (e.g., Gorlova et al., 2011), however the cause and underlying mechanisms of the disease remain unknown.

Scleroderma-related visible differences. Scleroderma is derived from the Greek words skleros (hard) and dermos (skin), with the hardening of skin being a manifestation of excessive collagen production (Seibold, 2005). In addition to the hardening of the skin, other appearance changes can include the following (Malcarne et al., 1999):

1. Loss of skin folds;

2. Shiny appearance to the skin;

3. Hypo- or hyper-pigmentation of the skin;

4. Loss of flexibility of the lips and decreased ability to fully open the mouth;

5. Pinched appearance to the nose and eyes;

6. Telangiectasias;

7. Subcutaneous calcinosis, often in the fingers, elbows, and knees;

8. Sclerodactyly;

9. Deformity of the digits;

10. Limited range of motion in the hands caused by contractures and/or non-healing ulcers, or surgical amputation to manage digital ischemia.

Jolly (2011) noted that weight loss is also common and may, "remind patients of their disease, leading to fears that they will visibly deteriorate not be able to recognize themselves" (p. 354).

Although there can be a great deal of heterogeneity in the type, location, and severity of skin involvement in SSc, many patients do experience a similar change in the facial experience involving tightened skin, a pinched nose, and shrunken mouth opening (referred to as a "Mauskopf" appearance in the medical literature; Amin et al., 2011). Amin et al. (2011) used ratings of digital photographs of scleroderma patients' faces by health professionals to examine the relationship between objective ratings of severity and patient perceptions of noticeability. The authors found a high concordance such that as the observer ratings increased in severity, so did individual-reported perceptions of noticeability and scores on a measure of social concerns and anxiety. The authors noted that this finding was interesting because in many other disfiguring conditions, there is greater variance between objective and subjective severity. They posited that, "…there is closer agreement between the subjective rating of appearance and rating by an observer than in other disfiguring conditions; the characteristic 'Mauskopf' appearance for example may focus attention to one feature rather than many, providing essentially a unitary dimension of change and reducing inter-rater variability" (p. 310).

It has been opined that this similarity in disfigurement may also contribute to increased fear of progression among newly diagnosed patients experiencing changes in appearance who observe more severe versions of their own changes in their peers (Gholizadeh, Fox, Mills, Jewett, Thombs, & Malcarne, 2017*a*). Anecdotally, this has been evidenced by patients who have reported avoiding potentially beneficial support

group meetings because of the increased anxiety elicited in seeing other patients with visibly advanced disease. In addition to facial involvement, hand involvement can also be especially challenging for patients. In one study, skin thickening on the fingers of the right hand was identified as the strongest predictor of poor self-esteem (Malcarne et al., 1999). Because the hand and face are "socially relevant" (Benrud-Larson et al., 2003, p. 131) and more readily noticeable areas of the body, it would be expected that disfigurement in these areas would lead to greater distress (Malcarne et al., 1999).

Research in SSc has also demonstrated associations between body image dissatisfaction and poorer interpersonal functioning (Benrud-Larson et al., 2002; Kwakkenbos et al., 2012). The rarity of SSc can contribute to a sense of isolation and stigmatization in patients because others see visible signs that they do not understand but can recognize as different (Joachim & Acorn, 2003). In his seminal work on stigma, Goffman (1963) described physical disfigurement as one form of stigma. Living with a visible chronic illness, one does not have the choice of whether or not to disclose the illness, but rather the disclosure is inherent in the first social interaction. In one of the few studies to qualitatively explore stigmatization and other social experiences of living with SSc, Joachim and Acorn (2003) described, "Although coping with serious life-threatening aspects of scleroderma, several participants stated that their major fear, a fear greater than dying from scleroderma, was being 'ugly' and looking bad to themselves and others. It is possible that being stigmatized, discredited and isolated was their real fear" (p. 603-4).

Psychosocial impacts of SSc. Because a cure for SSc, a chronic and progressive condition, remains elusive, symptom reduction and health-related quality of life

improvement are treatment targets (Kwakkenbos et al., 2015*a*; Thombs et al., 2010).

Thombs and colleagues (2010) reported that although the psychosocial challenges associated with SSc were pervasive and should be areas of clinical focus, the research on psychosocial impacts of the disease was relatively sparse, largely due to the low prevalence of SSc and a lack of validated psychosocial measures in SSc. Malcarne, Fox, Mills, and Gholizadeh (2013) echoed these claims, noting, however, that there had been substantial strides in psychosocial research and especially assessment efforts in scleroderma in even the three years since Thombs and colleagues' important paper.

There are myriad psychosocial impacts of scleroderma, including increased depression and anxiety (Bodukam et al., 2011; Benrud-Larson et al., 2002; Hyphantis et al., 2007; Jewett, Razykov, Hudson, Baron, & Thombs, 2014; Legendre, Allanore, Ferrand, & Kahan, 2005; Richards, Herrick, Griffin, Gwilliam, & Fortune, 2004; Thombs, et al., In Press); fatigue (Assassi et al., 2011; Thombs et al., 2008*a*; Yacoub, Bensabbah, & Hahhah-Hassouni, 2012; Sandusky, McGuire, Smith, Wigley, & Haythornwaite, 2009); pain (Benrud-Larson et al., 2002; Del Rosso et al., 2004; Malcarne et al., 2007; Richards et al., 2003; Schieir et al., 2010); disability (Hudson et al., 2008; Malcarne et al., 2007; Müller, Rehberger, Günther, & Schmitt, 2012; Sharif et al., 2011), and sexual dysfunction (Bongi et al., 2013; Knafo, Haythornthwaite, Heinberg, Wigley, & Thombs, 2011; Levis et al., 2012*a*; Levis et al., 2012*b*; Schouffoer et al., 2009). Additionally, body image impacts, such as dissatisfaction with appearance and lowered appearance-related self-esteem are common, as will be described in the following sections. Benrud-Larson et al. (2003) found that dissatisfaction with

appearance was prevalent in SSc patients, with SSc patients reporting greater body image dissatisfaction than burn injury survivors.

Assessment of body image in SSc. In 2010, Thombs and colleagues highlighted the need for validated measures that explore various aspects of body image in scleroderma, such as body image avoidance. Prior to this call, the existing body image research using validated measures had been limited to the Satisfaction with Appearance Scale or the Adapted Satisfaction with Appearance Scale (e.g., in Benrud-Larson et al., 2003 and Heinberg et al., 2007) and the appearance subscale of the State Self-Esteem Scale (e.g., in Malcarne et al., 1999 and Van Lankveld, Vonk, Teunissen, & van den Hoogen, 2007). In the past few years, however, there has been an increase in scholarship developing and using measures validated specifically in scleroderma to examine various aspects of body image (Malcarne et al., 2013).

At present, the following measures have been validated for use in SSc: the Satisfaction with Appearance Scale (SWAP; Lawrence et al., 1998), the Adapted Satisfaction with Appearance Scale (ASWAP; Heinberg et al., 2007), the Brief Satisfaction with Appearance Scale (Brief-SWAP; Jewett et al., 2010), the Derriford Appearance Scale short-form (DAS-24; Carr, Moss, & Harris, 2005), the Body Concealment Scale for Scleroderma (BCSS; Jewett et al., 2016*b*), and the Appearance Subscale of the State Self-Esteem Scale (SSES; Heatherton & Polivy, 1991).

Presence and Correlates of Body Image (Dis)satisfaction in Scleroderma. Fourteen studies have explored the presence and correlates of some dimension of the body image construct, with a burgeoning of scholarly interest in this area in the last several years (Amin et al., 2011; Benrud-Larson et al., 2003; Ennis, Herrick, Cassidy,

Griffiths, & Richards, 2012; Jewett et al., 2010; Jewett et al., 2012*a*; Jewett et al., 2016*a*; Heinberg et al., 2007; Knafo, Haythornthwaite, Heinberg, Wigley, & Thombs, 2011; Kwakkenbos et al., 2012; Leite & Maia, 2013; Malcarne et al., 1999; Mills et al., 2015*b*; Nusbaum, Gordon, & Steen, 2016; Van Lankveld, Vonk,Teunissen, & van den Hoogen, 2007). Nine of the aforementioned fourteen studies used some variation of the SWAP and two used the appearance self-esteem subscale of the SSES, as their outcomes of interest.

The majority of the research has been limited to (dis)satisfaction with appearance and appearance-related self-esteem as the body image dimension of interest. In previous research examining specific aspects of appearance and body image, face tightening (Benrud-Larson et al., 2003), skin tightening above the elbows (Benrud-Larson et al., 2003), skin tightening above the knees (Benrud-Larson et al., 2003), face involvement (Jewett et al., 2012), finger restriction (Benrud-Larson et al., 2003), skin thickening of the right hand and fingers (Malcarne et al., 1999), and telangiectasias (Ennis et al., 2013) have been significant correlates of body image dissatisfaction and associated psychological distress. Jewett et al. (2012) explored correlates of Dissatisfaction with Appearance (DA) and Social Discomfort (SD) via structural equation modeling, and found that face involvement and fingertip-to-palm distance were significant predictors of DA, whereas age, telangiectasias, and face involvement were significant correlates of SD.

Studies have also examined demographic correlates of body image dissatisfaction. Younger age has been associated with overall body image dissatisfaction (Benrud-Larson et al., 2003). However, two studies disentangling the personal (SWAP-DA) from social components (SWAP-SD) of body image dissatisfaction found that younger age was only significantly associated with SD but not DA (Jewett et al., 2012; Ennis et al., 2013).

Marital status, ethnicity, sex, and education were not associated with body image

dissatisfaction via the Brief-Swap (Jewett et al., 2012*a*). However, Ennis et al. (2013)

found that being single was associated with higher total and subscale ASWAP scores

(i.e., body image distress) and being retired was associated with higher DA scores. In a

recent study examining racial identity and body image dissatisfaction, Nusbaum et al.

(2016) found that African American patients with SSc had significantly higher rates of

body image dissatisfaction than Caucasian participants. Of note, a validated measure of

body image dissatisfaction was not used in the Nusbaum et al. (2016) study; instead,

Likert scales assessed dissatisfaction with specific features (e.g., changes in

pigmentation) and the scores were summed to form a composite dissatisfaction index.

In terms of clinical correlates of body image dissatisfaction, being diagnosed with

the diffuse disease subtype (Benrud-Larson et al., 2003; Ennis et al., 2013; Heniberg et

al., 2008), younger age at first non-Raynaud's[2] symptom (Ennis et al., 2013), higher

modified Rodanan skin scores (mRss, a measure of skin involvement in SSc; Jewett et

al., 2012; Mills et al., 2015*a*), and greater bodily pain (Benrud-Larson et al., 2003;

Heinberg et al., 2008; Jewett et al., 2013) were associated with greater body image

dissatisfaction. The literature has had some discordance, however. For example, Mills et

al. (2015*a*) did not find a significant association for pain and body image and van

Lankfeld et al. (2007) did not find an association between mRss and appearance-related

self-esteem.

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[2] Time from first non-Rayanud's symptom, rather than time since diagnosis, is commonly used as a proxy for disease duration in scleroderma given the often-lengthy time between symptom onset and ultimate diagnosis.

The psychosocial correlates of body image dissatisfaction have been a growing focus of scholarly research in SSc. Previous studies have demonstrated moderate positive associations between depression scores and body image dissatisfaction (Benrud-Larson et al., 2003; Jewett et al., 2010; Kwakkenbos et al., 2013; Leite et al., 2013) and small to moderate positive correlations of body image dissatisfaction with functional deficits/disability (Benrud-Larson et al., 2003; Heinberg et al., 2008; Mills et al., 2015*a*). Other correlates of body image dissatisfaction have been poorer functioning via SF-36 mental and physical composite scores (Jewett et al., 2013; Mills et al., 2015*a*); higher social phobia scores (Leite et al., 2013), higher anxiety scores (Leite et al., 2013); more body concealment behaviors (Jewett et all, 2015); poorer adjustment to illness (Benrud-Larson et al., 2003); and sexual dysfunction (Knafo et al., 2011). Malcarne et al. (1999) also found a significant relationship between lower appearance-related self-esteem and poorer adjustment to illness.

The majority of this research has involved univariate analyses. However, in the aforementioned Jewett et al. (2012) study that examined correlates of body image dissatisfaction, the authors also examined the relationships among the body image dissatisfaction outcome variable and its correlates via structural equation modeling and found that after the inclusion of face involvement into their model, no other variables were significant predictors of body image dissatisfaction. Another study additionally found via path analysis that depression mediated the relationship between body image dissatisfaction and psychosocial functioning (Benrud-Larson et al., 2003); of note, this study was a cross-sectional design, precluding conclusions about true mediation.

In sum, the extant literature on body image in scleroderma has been predominantly based on univariate studies, and the results have not always been consistent. For example, younger age was associated with poorer body image scores in two studies (Benrud-Larson et al., 2003; Jewett et al., 2012); however, Leite et al. (2013) found no relationship with age. Pain had a small-moderate correlation with poorer body image in three studies (Benrud-Larson et al., 2003; Jewett et al., 2015; Heinberg et al., 2007), however Mills et al. (2015*b*) found no relationship with pain. Additionally, the few studies engaging in more advanced statistical methods have found more complex relationships that were not identified using less advanced statistical methods.

State of Scleroderma Body Image Research. Despite the prevalence of and distress associated with changes in appearance in SSc, our understanding of the experience of body image in this disease remains limited (Malcarne, Fox, Mills, & Gholizadeh, 2013; Thombs et al., 2010). While there have been several studies identifying bivariate correlates of body image distress, and a smaller number using more advanced methods, there is a need for an examination that more fully allows the complex relationships among personal and social aspects of bodily changes to be identified. Because previous body image research has demonstrated that there is not always a simple relationship such that more severe disfigurement means more body image distress (Robinson, 1997; Rumsey, Clarke, & Musa, 2002), it cannot be assumed that patients with more severe disease will be the ones experiencing more problems with appearance impacts. There is a need for multivariate research that seeks to better understand the complex construct of body image in SSc in order to best serve the needs of patients.

An important first step is the identification of patient groups based on

multidimensional profiles of variables related to appearance changes and body image distress. This would help to identify distinct patient groups that may exist. For example, there may patients whose disease experience does not include social challenges but who report high levels of personal body image distress, versus other patients for whom both personal and social aspects are problematic. There may be variations in the interplay between body image and social anxiety that define distinct patient groups. Developing a better understanding of the appearance-related experiences of patients with SSc requires using sophisticated data analytic procedures that allow the identification of groups across multiple personal and social aspects of appearance-related distress. A complex construct, such as body image, lends well to a symptom cluster-like approach where clusters are defined as symptoms that are typically interrelated and have a synergistic impact on outcomes that is distinct from the relationship between any one individual symptom and the outcome (Barsevick, Whitmer, Nail, Beck, & Dudley, 2006). A statistical procedure that allows such an approach and would enhance our understanding of how the personal and social aspects of body image relate in SSc is latent profile analysis (LPA).

2.5 Latent Profile Analysis

With greater advancements in and accessibility of multivariate statistical techniques, such as LPA and latent class analysis (LCA), examining more intricate relationships among variables is possible. LPA has become increasingly common in the behavioral and social sciences (Nylund, Asparouhov, & Muthén, 2007). LPA is a useful approach to examining potentially complex interactions given that the interpretation of numerous higher-order interactions via variable-centered analyses can be difficult and statically problematic because of issues such as variance inflation factor elevations

(Cohen, Cohen, West, &, Aiken, 2003; Merz & Roesch, 2012). Contrastingly, a person-centered approach, such as LPA, can enable a more accessible interpretation of the relationships among variables at the level of the *person* rather than the *variable* (Roesch, Villodas, & Villodas, 2010). LPA is a branch of mixture modeling that enables the classification of subgroups of participants that demonstrate similar response patterns to a set of observed variables, called indicator variables. LPA allows for the inclusion of continuous indicator variables, while LCA is limited to categorical indicator variables. The distinction between LPA and LCA is not rigidly demarcated (Hagenaars & McCutcheon, 2002) and the two analyses can be combined (Kongsted & Nielsen, 2016). Via maximum likelihood estimation, LPA uses all observations of the continuous indicator variables to determine classes (Little & Rubin, 1987). Classes are added iteratively to determine optimal model fit; overall model fit and probability of correct individual classification are estimated simultaneously (Hill, Degnan, Calkins, & Keane, 2006).

Barsevick, Whitmer, Nail, Beck, and Dudley (2006) differentiated between a "symptom" as a single indicator of disease, versus "symptom clusters" that are "concurrent and related symptoms…[that] can be related through a common mechanism or etiology, by sharing common variance, or by producing different outcomes than individual symptoms" (p. 86). Barsevick et al. (2006) further noted that that symptom clusters are of diagnostic importance and many psychiatric conditions are diagnosed on the basis of symptoms (e.g., depressed mood; weight gain; fatigue). For the present purposes, the personal and social aspects of body image can be construed as experience symtoms that would lend well to a cluster approach.

Symptom cluster groups can then be used in many ways, for example examining differences among groups by relevant predictors of group membership, reducing the number of required variables and sample size in an analysis, and identifying patients most likely to benefit in an intervention (Clatworthy, Hankins, Buick, Weinman & Horne, 2007). In psychosocial research, cluster analysis is one approach to the derivation of psychological profiles, but there are some limitations to this approach, including 1) lack of clear guidelines to decide on the number of clusters; 2) analysis sensitive to distributions and measurement scales; 3) difficult-to-meet assumptions (e.g., class-invariant classes; Milligan & Cooper, 1987; Morin, Morizot, Boudrias, & Madore, 2011; Speece, 1994). LPA and LCA do not assume that relationships among variables are homogenous for all patients and instead enable a probabilistic approach that gives each individual a probability of belonging to a given subgroup (Kongsted & Nielsen, 2016).

LPA/LCA in the behavioral medicine and body image contexts. LPA and LCA are useful tools in the behavioral medicine and body image contexts given the potential to derive groups of patients with similar characteristics. Despite this, LPA/LCA have been minimally used in the context of body image. Moreover, the studies that do exist tend to focus on eating disordered populations. Jacobs and colleagues (2009) used LPA to identify behavioral profiles of patients with anorexia nervosa (AN), referred to as probands, and their parents with the goal of identifying "cross-generational clusters of behavioral traits" (p. 452) present in the individuals with AN and their parents. Three distinct profiles of families were identified distinguishing among: 1) moderate symptomatology in probands but healthy mothers, 2) the highest symptomatology in probands and moderate symptomatology in mothers, and 3) high symptomatology in

23

probands and high symptomatology in mothers. One-way ANOVAs were conducted as means of external validation to supplement statistical significance of groups with clinical significance, but failed to find significant differences.

Hildebrandt, Schlundt, Langenbucher, and Chung (2006) examined muscle dysmorphic symptoms in male weightlifters ($N = 237$) via an LCA of eight measures of body image and appearance-related anxiety and identified five distinct classes of respondents: 1) *Dysmorphic*, 2) *Muscle Concerned*, 3) *Fat Concerned*, 4) *Normal Behavioral*, and 5) *Normal*. Hildebrandt et al. also conducted one-way ANOVAs to identify between-group "external validators," such as the presence of eating disordered behaviors and steroid use (p. 132). Through the LCA and follow-up ANOVAs, Hildebrandt et al. characterized the groups as qualitatively distinct; for example, the dysmorphic group, "desired to decrease body fat and increase muscle mass, and had the highest scores across dimensions of body image disturbance, associated psychopathology, and appearance-controlling behaviors" (p. 133). Furthermore, this group had higher rates of steroid use compared to the other identified groups.

Lanza, Savage, and Birch (2010) used LCA in sample of women ($N = 197$) to identify classes of weight loss strategies. They identified four distinct subgroups: 1) *No Weight Loss Strategy*, 2) *Dietary Guidelines*, 3) *Guidelines + Macronutrients*, and 4) *Guidelines + Macronutrients + Restrictive*. The groups differed on key psychosocial variables, for example, weight concerns and desire to be thinner were more strongly predictive of membership in the *Guidelines + Macronutrients + Restrictive* group than the *No Weight Loss Strategy* group. Brytek-Matera, Rogoza, Gramaglia, and Zeppegno (2015) also used LCA to examine problematic eating behaviors in a small sample ($N =$

52) of women with eating disorders and used the MBSRQ as one of the measures in the LCA; however, the methodological quality of the study was questionable, and although they identified a two-class solution, it was not clearly described in the paper.

Selected studies in rheumatologic conditions are also provided to further illustrate the utility of LCA/LPA in behavioral medicine contexts. LPA has also been used to classify rheumatologic patients into symptom specific profiles. For example, Sullivan, Smith, and Buchwald (2011) entered over thirty symptoms commonly reported by fibromyalgia and chronic fatigue patients into an LCA and identified a four-class solution where individuals differed not qualitatively in symptomatology but rather on a severity gradient, suggesting a greater similarity between chronic fatigue and fibromyalgia patients than previously considered. Wilson et al. (2001) examined psychological and clinical symptoms in a sample of patients with chronic fatigue syndrome and identified one group in which individuals tended to be younger in age, have a smaller female to male ratio, have a shorter episode duration, and less psychiatric symptomatology and lower functional disability as compared to the second group. The second group also reported a higher prevalence of atypical symptoms (e.g., vision loss) and less responsiveness to factors expected to improve symptoms (e.g., sleep). The second class was therefore conceptualized as more akin to somatoform disorder than chronic fatigue syndrome.

In rheumatoid arthritis (RA) previous research has explored pain and adjustment profiles to identify "*Adaptors*" (i.e., individuals with greater perceptions of control and lower perceptions of pain-related consequences of the disease and disease chronicity) and "*Non-adaptors*" (i.e. individuals with lower perceptions of control and stronger

perceptions of disease chronicity, recurrence, and identity; Hobro, Weinman, & Hankins, 2004). Norton et al. (2014) used LPA in a sample of RA patients ($N = 227$) and also identified two classes (unnamed) corresponding to adaptors and non-adaptors, wherein the non-adaptor-like group had greater negative representations of disease, endorsement of more symptoms, less control, and more severe perceived consequences as compared to the adaptors. The non-adapter-like group's membership predicted higher pain and functional disability.

LPA has been used in SSc in one study. Using LPA to explore pain profiles in SSc, Merz et al. (2014) found that SSc patients could be divided into three biopsychosocial typologies in relation to their pain: 1) *Managing*, 2) *Resilient*, and 3) *Distressed*. The *Managing* group was characterized by relatively less severe disease, better perceived physical health, fewer health worries, better mental health, and more social support. The *Resilient* group was characterized by relatively greater disease severity and poorer perceptions of health, but also fewer health worries, better mental health, and more social support. The *Distressed* group was characterized by relatively less severe disease, but poorer perceptions of health, more health worries, poorer mental health, and lower social support. In other words, individuals in the *Distressed* group had objectively less severe disease than the *Resilient* group, but their subjective reports of pain and medication utilization were higher. This finding is important because it suggests that disease severity alone was not the best way to identify patients who were at greater risk for pain-related problems and medication use; rather, profiles revealed that psychosocial variables, such as mental health and social support, helped to distinguish which patients were most at risk for pain problems. Thus, LPA is a tool that can identify

26

groups of patients with different needs and challenges. This is especially important in a context where severity does not necessarily directly correlate with level of distress, and examination of simple bivariate relationships may obscure more complex relationships among disease parameters. In these cases, LPA can identify profiles of patients across multiple indicators, revealing more complex patterns than can be garnered from other, simpler analyses.

2.6 Summary and Limitations of Prior Research

Changes in appearance are a common and distressing aspect of SSc for many patients (Thombs et al., 2010). Living with SSc presents unique challenges for patients given the increased potential for stigmatization (Joachim & Acorn, 2003) and lack of relevant and accessible resources, especially relating to psychosocial concerns (Thombs et al., 2010). Despite the fact that there has been increased attention to the psychosocial impacts of SSc, there are still many gaps in the understanding of SSc-related body image distress and associated outcomes (Malcarne et al., 2013).

The literature exploring body image in SSc has been inconclusive and few studies have used multivariate methods. Only one previous study has used LPA in SSc (Merz et al., 2014), and this was in the context of deriving pain typologies. As a preferred analytical tool for making complex inferences about individuals (Roesch et al., 2010), LPA is an approach that would enable a more refined understanding of the multidimensional experience of body image in SSc.

2.7 Specific Aims and Hypotheses of the Present Study

The present study aimed to establish typologies of patients with SSc based on multiple indicators of body image and social anxiety, and to examine differences among

identified groups across sociodemographic, medical, and psychosocial variables. Specifically, the present study had the following specific aims:

Specific Aim 1. To establish typologies of patients based on multiple indicators of body image and social anxiety. The first aim of the present study was to establish profiles (i.e., typologies) of patients based on multiple indicators of body image and social anxiety in SSc. Prior to running the analysis, the sample was randomized into two different groups to allow for a replication analysis of the findings. Exploratory LPA was used to derive categorical latent variables that signified profiles of similarly scoring individuals using subjective indicators of body image and social anxiety and one objective indicator of skin involvement. LPA is an exploratory approach, precluding hypotheses regarding which exact classes will emerge from the data. However, it was hypothesized that the analysis would uncover multiple (i.e., greater than two) unique profiles of patients based on the different dimensions of body image and social anxiety. It was also hypothesized that the objective skin involvement indicator would not necessarily be directly associated with uniformly poorer body image and social anxiety scores per the aforementioned research demonstrating that the severity of disfigurement does not directly relate to psychosocial outcomes (Rumsey, 2002*a*, 2002*b*). It was further hypothesized that profiles may emerge differentiating 1) individuals with similar objective skin involvement scores but differing subjective dissatisfaction and social anxiety scores, and 2) similar personal dissatisfaction scores but differing social anxiety scores. Furthermore, it was hypothesized that the number of typologies identified would be replicable between both samples.

Specific Aim 2. To examine the profiles in relation to sociodemographic medical, and psychosocial variables. The second aim of the present study was to evaluate potential differences in sociodemographic (e.g., age) medical (e.g., disease duration) and psychosocial variables (e.g., depression) as a function of profile membership. These group comparison variables are referred to as auxiliary variables (Lanza, Tan, & Bray, 2013). It was hypothesized, per the previous research on correlates of body image, that if groups emerged that indicated greater body image distress and social anxiety, they would be associated with female gender, Black/African American race, lower age, higher disease duration, greater depression, greater anxiety, greater disability, the diffuse disease subtype, and the presence of disease variables reflecting disfigurement in the face and hands. It was hypothesized that the other auxiliary variables (i.e., marital status, year's since first non-Raynaud's symptom, year's since diagnosis, BMI) would not significantly differ across identified groups. Also, it was hypothesized that if resilient-type groups emerged such that there were participants who had relatively high objective skin involvement but had relatively better body image and/or social anxiety scores, patients in this group would have better mental health (e.g., depression, anxiety) as compared to group(s) high in not only objective skin involvement but also dissatisfaction and social anxiety. Furthermore, it was hypothesized that these relationships would be similar in the two randomized samples.

Chapter 2 is being prepared in part for publication. This publication will be co-authored by Vanessa L. Malcarne, Scott C. Roesch, Marie-Eve Carrier, Linda Kwakkenbos, Sarah D. Mills, Rina S. Fox, and Brett D. Thombs.. The dissertation author was the primary investigator and author of this material.

CHAPTER 3: METHODS

3.1 Participants

Patients in the SPIN Cohort were enrolled at 28 centers from Canada, the United States, and the United Kingdom. To be eligible for the SPIN Cohort, patients must have a diagnosis of SSc according to the 2013 ACR/EULAR classification criteria (van den Hoogen et al., 2013) confirmed by a SPIN physician, be at least 18 years of age, have the ability to give informed consent, and be fluent in English or French. Exclusion criteria for participation in the SPIN Cohort include not having access to the internet or otherwise not being able to respond to questionnaires via the internet. Only participants who completed the core indicator measures used in the LPA and had physician-documented limited or diffuse disease were included in the present analysis.

A total of 1343 patients completed study questionnaires from April 2014 through October 2016. One participant who was not an English or French speaker was excluded from the analysis. Participants for whom disease subtype (i.e., limited, diffuse) was not provided or who were classified as the subtype "sine", a relatively rare classification that refers to patients with some of the visceral impacts of SSc but without the skin involvement (Poormoghim, Lucas, Fertig, & Medsger, 2000), were removed ($n = 43$). The research on how to best classify sine patients (i.e., as having an entirely different disease; as constituting a rare, unique disease subtype; or as having an early form of limited SSc; Hachulla & Launay, 2011) is still inconclusive and as such these patients were excluded. One hundred and twenty patients with missing data on one or more of the core body image or social anxiety measure included in the LPA were excluded. Two hundred and

thirty-seven patients without mRss data were also excluded from the present sample. Thus, the study sample consisted of 942 patients enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. Differences between individuals included in the present analysis and those excluded were examined with chi-squared (for categorical variables) and t-test (for continuous variables) analyses. There were no significant differences by age, gender, race, marital status, years of education, or time since diagnosis. There was a significant difference in years since first non-Raynaud's symptom such that excluded participants had a significantly longer duration of experiencing symptoms $t(149) = -2.13$, $p = .008$; M (SD) = 12.29 (9.3) vs. 11.10 (8.4).

The SPIN sample is a convenience sample. Eligible patients are invited by the attending physician or a supervised nurse coordinator to participate in the SPIN Cohort, and written informed consent is obtained. The local SPIN physician or supervised nurse coordinator then completes a medical data form that is submitted online to initiate patient registration in the SPIN Cohort. After completion of online registration, an automated welcome email is sent to participants with instructions to on how to activate their SPIN online account and how to complete the SPIN Cohort patient measures online. SPIN Cohort patients complete outcome measures via the internet upon enrollment and subsequently every 3 months. The SPIN Cohort study was approved by the Research Ethics Committee of the Jewish General Hospital, Montreal, Canada and by the Institutional Reviews Boards of each participating center. The analysis involved archival, de-identified data and was thus exempt from IRB review at SDSU/UC San Diego.

3.2 Study Procedures

Attending physicians or supervised nurse coordinators trained in SPIN

recruitment procedures informed eligible patients about SPIN and enter eligible and

interested patients into the SPIN cohort (see Appendix A for a description of the cohort)

and obtain written informed consent (Kwakkenbos et al., 2013). A medical data form for

the patient was submitted by the associated SPIN physician or nurse coordinator, after

which point participation was initiated and the patient received an email inviting them to

the study with study instructions. Upon enrollment, SPIN cohort patients completed the

baseline questionnaires. Patients were asked to complete a series of measures every three

months as part of participation in the cohort. For the present analysis, all patients with

baseline data for the selected indicator variables were included.

3.3 Measures

Sociodemographic and medical variables. Age, gender, race, marital status,

educational attainment, occupational status, and BMI were assessed via self-report. Given

the multi-country nature of the present study, the various country-specific racial

identifications were re-coded into White/Caucasian, Black/African-American, and All

Other. Disease duration (i.e., date from first non-Raynaud's symptom), disease subset

(limited/diffuse), and various appearance-related variables (dichotomous variables

reporting the presence/absence of sclerodactyly, lower hand thickening, fingertip scars,

telangiectasias, and pigmentation) were physician reported. Sclerodactyly referred to a

localized tightening distal to the four metacarpophalangeal joints but proximal to the

proximal interphalangeal joints. Lower hand thickening referred to skin thickening of the

fingers of both hands extending proximal to the metacarpophalangeal joints. Fingertip

scars specifically referred to fingertip pitting scars that involve depressed areas at digital tips as a result of ischemia, rather than trauma or exogenous causes. Telangiectasias referred to widened blood vessels anywhere on the body. Pigmentation included the presence of either hypo- or hyper-pigmentation anywhere on the body.

Body image and social anxiety variables. *Body Concealment Scale for Scleroderma (BCSS; Jewett et al., 2016b).* The BCSS is a nine-item measure of body concealment and avoidance that was developed from the Body Image Avoidance Questionnaire (Rosen, Srebnik, Saltzberg, & Wendt, 1991). Body concealment can be used as an avoidance strategy by individuals living with visible differences (Jewett et al., 2017). The measure is scored as a total sum of the nine items. Respondents rank on a response scale ranging from 0 (*never*) to 5 (*always*) how often they engage in various concealment behaviors, with higher scores indicating higher frequency of body concealment behaviors. Sample items include, "I wear clothes to hide the changes to my skin," and "I wear gloves to hide my hands." The measure was specifically developed for SSc populations by Jewett et al. (2016*b*) and has shown strong internal consistency and good construct validity in replication with the SPIN SSc population as well (Jewett et al., 2017). In the present sample, the internal consistency for Samples 1 and 2 was $\alpha = 0.89$ and $\alpha = 0.88$, respectively.

Brief-Satisfaction with Appearance Scale (Brief-SWAP; Jewett et al., 2010). The Brief-SWAP is a disease-specific, 6-item measure of body image dissatisfaction derived from the 14-item SWAP (Lawrence, Heinberg, Roca, Munster, Spence, & Fauerbach, 1998). The Brief-Swap has two three-item subscales Dissatisfaction with Appearance and Social Discomfort. Responses are on a Likert-type scale ranging from 0 (*strongly*

disagree) to 6 (*strongly agree*) with each subscale score ranging from 0-18, with higher scores indicating greater dissatisfaction and discomfort. Sample items for Dissatisfaction with Appearance and Social Discomfort scales include, "I am satisfied with the appearance of my hands," (item is reverse scored) and "I feel that my scleroderma is unattractive to others." Psychometric analyses have demonstrated that the measure is reliable and valid in SSc (Jewett et al., 2010; Mills et al., 2015*b*). In the present sample, the internal consistency for the Dissatisfaction with Appearance subscale in Samples 1 and 2 was $\alpha = 0.82$ and $\alpha = 0.81$, respectively; the internal consistency for the Social Discomfort subscale in Samples 1 and 2 was $\alpha = 0.89$ and $\alpha = 0.87$, respectively.

Brief Fear of Negative Evaluation Scale – 8 (BFNE-8; Carleton, Collimire, & Asmundson, 2007). The BFNE-8 is a unidimensional, 8-item revised version of the Brief Fear of Negative Evaluation scale (BFNE; Leary, 1983) that assesses fear of negative evaluation. Items are rated on a 5-point scale ranging from 1 (*not at all characteristic of me*) to 5 (*entirely characteristic of me*). Total scores range from 8 to 40, with higher scores indicating greater fear of negative evaluation. Sample items include, "I am afraid that other people will find fault with me," and "When I am talking to someone, I worry what they may be thinking about me." The BFNE-8 was found to reliable and valid in SSc using the SPIN population (Fox et al., in progress). In the present sample, the internal consistency for Samples 1 and 2 was $\alpha = 0.97$ and $\alpha = 0.96$, respectively.

Social Appearance Anxiety Scale (SAAS; Hart, Flora, Palyo, Fresco, Holle, & Heimberg, 2008). The SAAS is a 16-item, unidimensional assessment of fear in situations where one's appearance may be evaluated. Items are rated on a response scale ranging from 1 (*not at all*) to 5 (*extremely*). Total scores range from 16 to 80, with higher

scores indicating greater fear of situations where one's appearance may be evaluated. The SAAS can be a predictor of poor body image controlling for social anxiety in the general population (Hart et al., 2008). Sample items include, "I am concerned people won't like me because of the way I look," and "I get nervous when talking to people because of the way I look." A psychometric analysis of the measure in the SPIN cohort demonstrated that the unidimensional SAAS is an appropriate measure for use with SSc patients (Mills et al., 2015*a*). In the present sample, the internal consistency for Samples 1 and 2 was α = 0.96 and α = 0.96, respectively.

Social Interaction Anxiety Scale-6 (SIAS-6; Peters, Sunderland, Andrews, Rapee, & Mattick, 2012). The SIAS-6 is a unidimensional, six-item, short-form measure of anxiety resulting from social interactions developed from the 20-item SIAS (Mattick & Clarke, 1989). Response options range from 0 *(not at all characteristic or true of me)* to 4 *(extremely characteristic or true of me)*. There is a total score computed from summing all items, with higher scores indicating greater interactional anxiety. Sample items include, "I have difficulty making eye contact with others" and "I have difficulty talking with other people." A psychometric analysis of the measure using the SPIN cohort demonstrated that the unidimensional SIAS-6 is an appropriate measure for use in both limited and diffuse SSc patients (Gholizadeh et al., 2015*b*). In the present sample, the internal consistency for Samples 1 and 2 was α = 0.89 and α = 0.88, respectively.

Objective skin involvement. Objective skin involvement was assessed via the physician-reported modified Rodnan skin score (mRss; Clements et al., 1990). The mRss is a measure of skin thickening in 17 bodily areas assessed via palpitation and using a four-point scale (0 = uninvolved to 3 = severe thickening). Total scores range from 0 to

51, with higher scores reflecting greater thickening and disease involvement. The measure is widely used as an indicator of disease severity in SSc and frequently used as a primary outcome measure in clinical trials because of its reliability, validity, and responsiveness to change (Clements et al., 1995). The mRss can be analyzed as a single continuous variable in statistical analyses (Amjadi et al., 2009).

Auxiliary psychosocial measures. *Patient Health Questionnaire (PHQ-8;* Kroenke, Spitzer, & William, 2001). The PHQ-8 is a well-validated self-report screener for depression. Patients endorse items on a 0 (*Not at all*) to 3 (*Nearly every day*) response scale. Scores 5, 10, 15, and 20 categorize mild, moderate, moderately severe, and severe depression respectively. The PHQ-8 rather than the PHQ-9 is recommended for use in SSc because the item assessing suicide has poor predictive validity for self-harm (Razykov Hudson, Baron, Thombs, & Canadian Scleroderma Research Group, 2013). The internal consistency for Samples 1 and 2 was $\alpha = 0.88$.

The Patient-Reported Outcomes Measurement Information System (PROMIS®)-29; Cella et al., 2010). PROMIS is a research initiative that develops and psychometrically evaluates item banks for a given construct so that a patient-reported outcomes measure can be developed. The PROMIS-29 is a multi-dimensional, 29-item self-report measure that uses items from the seven domains of physical function, anxiety, depression, fatigue, sleep disturbance, pain interference, and ability to perform social roles; four items from each domain are provided in addition to an 11-point rating scale for pain intensity (ranging from 0-10). Each item has response values ranging from 1 to 5 (except pain intensity) and a raw score can be created for each domain ranging from 4 (lowest possible score) to 20 (highest possible scores). Scores are often standardized

using a T score-metric set to 50 ± 10 (mean $\pm$ SD). Higher scores on the symptom-type domains (i.e., anxiety) indicate worse symptoms. The measure has been validated for use in SSc (Hinchcliff et al., 2011; Kwakkenbos et al., 2015b). Internal consistency for the PROMIS-29 in SSc has not been reported. Previously research has demonstrated that SSc patients scored 0.2 to 0.7 SD lower than the general population across domains except for the physical functioning domain, for which SSc patients scored 1 SD lower (Khanna et al., 2012). For the present analysis, the PROMIS-29 Anxiety domain was used, and the internal consistency for Samples 1 and 2 was $\alpha = 0.92$.

The Scleroderma Health Assessment Questionnaire (SHAQ; Steen & Medsger, 1997) was developed from the Health Assessment Questionnaire (HAQ; Fries et al., 1980). The SHAQ includes the eight domains of the HAQ disability index and scales for pain, patient global assessment, vascular, digital ulcers, lung involvement, and gastrointestinal involvement. The eight HAQ domains are scored from 0 (without difficulty) to 3 (unable to do); the maximum scores from each of the eight domains are added together and divided by the number of completed categories. The added SHAQ items are continuous VAS scales that are transformed into 0-3 scores. The measure has shown good reliability and validity in SSc (*see* Pope, 2011). For the present analysis, the SHAQ-Disability (SHAQ-DI) domain was used. The internal consistency for Samples 1 and 2 was $\alpha = 0.95$.

3.4 Data Analytic Plan

Data preparation. The data were examined and cleaned in order to remove participants as described in the methods. The total sample ($N = 942$) was then

randomized using SPSS Version 24.0 into two random samples: Sample 1 ($N = 469$) and Sample 2 *(N = 473)*.

Analysis for specific Aim 1. For the first aim, exploratory LPA was used to derive categorical latent variables that signified profiles of similarly scoring individuals using three subjective indicators of body image (i.e., the total score of the Body Concealment Scale for Scleroderma and the two subscales of the Brief Satisfaction with Appearance Scale [Dissatisfaction with Appearance and Social Discomfort]; three subjective indicators of social anxiety (total scores from the Brief Fear of Negative Evaluation Scale-8, the Social Appearance Anxiety Scale, and the Social Interaction Anxiety Scale-6); and one objective indicator of skin involvement (the mRss). In order to account for potential gender-specific body image differences (Thompson, 2004), the analysis was run both 1) with all participants, and 2) excluding male participants in order to examine if there was a change in group membership, as described in the exploratory analyses section. Gender was also used as an auxiliary variable for comparison of groups in Aim 2. LPA was conducted using MPlus, version 7.2 (Muthén & Muthén, 1998-2012).

The LPA used all observations of the aforementioned body image and social anxiety constructs in order to identify classes via maximum likelihood estimation. It should be noted that moderate to high correlations among the measures were expected given that various aspects of body image are being assessed, and this does not impede the analysis. LPA was used to identify profiles of individuals scoring similarly on the continuous variables such that complex relationships among these constructs could be summarized in a meaningful and accessible way.

In LPA/LCA, the analysis commences with a model with one subgroup, adding more subgroups until optimal model fit is determined (Kongsted & Nielsen, 2016). The optimal number of classes was derived via evaluation of models per the Lo-Mendell-Rubin Adjusted Likelihood Ratio Test (LMRT; Lo, Mendell, & Rubin, 2001), the Bootstrapped Likelihood Ratio Test (BLRT; Arminger, Stein, & Wittenberg, 1999; McLachlan & Peel, 2000), Akaike information criteria (AIC; Akaike, 1974, sample size-adjusted Bayesian information criteria (sBIC; Schwarz, 1978), and Entropy (Ramaswamy, DeSarbo, Reibstein, & Robinson, 1993). The LMRT is a statistical test to determine whether there is a significant improvement in model fit comparing a model with k profiles to a model with $k-1$ profiles using a log-likelihood (rather than chi-square) distribution (Lo et al., 2001); the test has been identified as a useful indicator in simulation studies (Nylund et al., 2007). The BLRT is also a likelihood-based technique that has performed well in simulation studies (Nylund et al., 2007).

One challenge in mixture modeling is that there can be disagreement among the various indicators. There is not one, universally accepted indicator to determine the number of classes (Nylund et al., 2007). Thus, researchers must decide and prioritize among the various indicators to determine an optimal solution. The justification of classes is akin to the determination of the face validity of the identified solution and cannot be garnered from statistical tests; it is incumbent on the researcher to provide an explanation for why the groups are meaningful and appropriate (Kongsted & Nielsen, 2016). As Jung and Wickrama (2008) argued, "…the number of classes depends on a combination of factors in addition to fit indices, including one's research question, parsimony, theoretical justification, and interpretability. Keeping this in mind, fit indices

and tests of model fit should not be the final word in deciding on the number of classes. However, they are useful in the initial exploratory stages of analyses" (p.311).

In the present study, the BLRT, LMRT, and entropy were first considered to compare the fit of the target model to a comparison model that has one fewer class; the p-values derived from the BLRT and LMRT were examined to determine whether the solution with fewer or greater profiles had superior fit. A significant p-value ($p \leq .05$) would indicate that the solution with one more profile (i.e., $k + 1$) is superior whereas a non-significant p-value ($p > .05$) would indicate that the solution with one fewer profile (k) is appropriate. Entropy is an indicator that examines the uncertainty of classification to determine how accurate the classes are (Celux & Soromenho, 1996); values that are closer to one (i.e., larger) will indicate superior accuracy. Although there are no defined cut-offs, values that fall below 0.80 indicate poor separation between classes; higher values correspond to greater confidence that the individuals in a sample have been placed into the most accurate group given the number of classes being assessed (Ramaswamy et al., 1993). In cases where there is disagreement among BLRT, LMRT, and entropy, the researcher can turn to descriptive indices and interpretability. In cases where there is disagreement among the statistical indicators, the solution with fewer classes would be the more parsimonious solution. Researchers arguing for the merits of adding an additional class in such scenarios should consider the relative improvement of model fit indices and interpretability that would be achieved at the cost of parsimony (de Cock & Shevlin, 2014). The AIC and sBIC were also examined as descriptive fit indices such that smaller values suggested better fit. Furthermore, the number of patients in a given class was considered given that solutions with classes comprising less than 5% of the sample

size are often considered untenable (Hipp & Bauer, 2006). In addition to the statistical indicators of model fit, the interpretability and theoretical justification for the classes was considered, as described in Muthén (2006). Additionally, the replicability of the solution between the two samples was taken into consideration, such that priority was given to the solution that was most defensible in both samples

There is no way to directly calculate power for LPA. However, if a sample is too small, this may produce unstable solutions (Roesch, Villodas, & Villodas, 2010) or limit the ability to identify small but clinically meaningful groups (Kongsted & Nielsen, 2016). Some researchers have argued that, as in structural equation modeling, sample sizes of 200 are considered adequate for LPA (Schumacker & Lomax, 2004).

Analysis for specific Aim 2. Following the identification of the best-fitting models, a three-step approach to examine relationships among the latent profile variables and auxiliary variables was used to evaluate potential differences in sociodemographic, medical, and psychosocial variables as a function of profile group membership as described in Asparouhov and Muthén (2014). This is an alternative to the more commonly used single-step mixture modeling approach, allowing the identification of predictors of the latent profiles using Mplus. This approach by Lanza et al. (2013), also described in Vermunt (2010), been shown to outperform one-step approaches (Asparouhov & Muthén 2014). Traditionally, after the number of classes has been determined, the latent classification is treated as an observed variable and individuals are assigned to the most likely profiles to estimate between-class differences on specified covariates. But, this approach no longer retains an important benefit of latent models, which is taking classification uncertainty into account (Lanza et al., 2013). The Lanza et

al. (2013) approach is an alternative to confront these shortcomings via a simultaneous 1) estimation as a latent variable, and 2) estimation of the difference between estimated classes on specified distal (i.e., independent/predictor) variables. Additionally, Lanza et al.'s (2013) approach is robust to violation of multinomial logistic regression assumptions (Asparouhov & Muthen, 2013).

Thus, the distal variables are treated as latent class predictors simultaneously with the LPA estimation model in an auxiliary model (Perrig-Chiello, Hutchison, Morselli, 2015). To summarize the three-step approach, in the first step, the latent profile model is estimated using the identified indicator variables. Next, a variable is created identifying the most likely profile for each participant using the latent profile distribution in step one. Finally, profile membership is regressed on the predictor or distal variables. In the analysis, the output includes all pairwise comparisons between the two specified classes akin to ANOVA using post-hoc comparisons, but using chi-square tests. Each of these analyses was run separately for Sample 1 and Sample 2.

The Lanza et al. (2013) three-step method was used to empirically evaluate potential differences in sociodemographic, medical, and psychosocial variables as a function of profile group membership after the identification of patient typologies. Specifically, potential differences in sociodemographic (i.e., age, gender, race, education, income, marital status), medical/functioning (i.e., disease duration, disease subtype, disease severity, telangiectasias, skin pigmentation changes, fingertip scars, sclerodactyly, and lower hand thickening) and psychosocial (i.e., depression, SHAQ-Disability domain, and PROMIS-29-Anxiety domain) variables were examined as a function of group membership.

Chapter 3 is being prepared in part for publication. This publication will be co-authored by Vanessa L. Malcarne, Scott C. Roesch, Marie-Eve Carrier, Linda Kwakkenbos, Sarah D. Mills, Rina S. Fox, and Brett D. Thombs. The dissertation author was the primary investigator and author of this material.

CHAPTER 4: RESULTS

4.1 Participant Characteristics and Preliminary Analysis

The sample characteristics for the total sample, Sample 1, and Sample 2 can be found in Table 1. Interdependent t-tests revealed no significant differences across key study variables between Sample 1 and Sample 2. There were 469 and 473 participants in Samples 1 and 2, respectively, with sufficient data to be included in the LPA. The samples were predominantly female, White, married/common law status, and had attained at least some college education, with an average age of approximately 55 years old.

4.2 Body Image Profiles

Primary analysis: Sample 1. Models containing one, two, three, four, and five profiles were fit to the Sample 1 data. Table 2a presents the fit indices for each of the five models. Across the indicators of model fit, the two-profile solution outperformed the one-profile solution. Comparing the three-profile solution to the two-profile solution, there was some disagreement with the model-fit indicators. Although the three-class solution had lower values for the AIC and sBIC, a higher value for entropy, and significant BLRT, the LMRT was not significant ($p = 0.144$). Examination of the four and five profile solutions indicated that the LMRT was again not significant. In undertaking a substantive evaluation of the profiles and examining the sample means and conditional response means, the addition of a profile did not change the pattern of scores; the pattern was consistently all lower or higher scores for the variables included in the LPA regardless of the number of profiles. In other words, a two-profile solution identified a high-low pattern, whereas a three-profile solution identified a high-medium-low pattern. Thus,

44

considering that the two-profile solution was arguably better fitting in the replication sample (see below), the two-profile solution was selected as a more parsimonious solution to describe the observed pattern. The sample means and conditional response means for the two-profile solution have been provided (see Table 3a and Figure 1). Profile 1 comprised 71.2% of the sample ($n = 334$) and included individuals with an average objective skin involvement score of 7.03 ($SD = 9.6$) and subjective body image and social anxiety scores lower than the overall sample mean across all included measures. Profile 1 was labeled the *Appearance Comfortable* group. Profile 2 comprised 28.8% of the sample ($n = 135$), and included individuals with an average objective skin involvement score of 9.87 ($SD = 10.6$) and subjective body-image and society anxiety scores higher than the overall sample mean across all included measures. Profile 2 was labeled the *Appearance Distressed* group.

The analysis was rerun with only the female participants in the sample, but the two-profile solution was still superior. Thus, the male participants were retained in the analysis.

Primary analysis: Sample 2. Models containing one, two, three, four, and five profiles were also fit to the Sample 2 data. Table 2b presents the fit indices for each of the five models. Across the indicators of model fit, the two-profile solution outperformed the one-profile solution. Although the three-profile solution had lower AIC and sBIC values and a significant BLRT value as compared to the two-profile solution, the LMRT was not significant and the entropy was lower, suggesting that the two-profile solution was the better fit. Examining the four and five profile solutions, the LMRT was not significant

and the entropy values were lower than the two-profile solution. Thus, a two-profile

solution was deemed the most appropriate statistical solution in Sample 2.

A substantive analysis was also undertaken via an examination of the sample

means and conditional response means (see Table 3b and Figure 2). Profile 1 comprised

79.3% of the sample ($n = 375$) and included individuals with an average objective skin

involvement score of 7.65 ($SD = 8.8$) and subjective body-image and social anxiety

scores lower than the overall sample mean across all included measures. Profile 1 was

labeled the *Appearance Comfortable* group. Profile 2 comprised 20.7% of the sample ($n

= 98$), and included individuals with an average objective skin involvement score of 9.62

($SD = 10.3$) and subjective body-image and society anxiety scores higher than the overall

sample mean across all included measures. Profile 2 was labeled the *Appearance

Distressed* group.

The analysis was rerun with only the female participants in the sample, but the

two-profile solution was still superior. Thus, the male participants were retained in the

analysis.

Exploratory analyses. A number of exploratory analyses were also undertaken to

examine 1) subjective body image and social anxiety profiles without the objective mRss

measure; 2) subjective body image and social anxiety profiles without the objective mRss

measure with only female participants; 3) all included indicators in the LPA in only

female participants; 4) all included measures in the LPA with the addition of the

psychosocial measures (i.e., the PHQ, HAQ, PROMIS-Anxiety); 5) all included measures

in the LPA with the addition of the psychosocial measures (i.e., the PHQ, HAQ,

PROMIS-Anxiety) in only female participants. Across these exploratory analyses, the

two-profile solution remained the most robust and parsimonious solution when taking both samples into consideration. The patterns in conditional response means remained similar. The model fit indices for these exploratory models are included in Appendix B.

4.3 Associations of Appearance Groups with Sociodemographic, Medical, and Psychosocial Variables

Sociodemographic variables. For Sample 1 participants, significant associations were found between group membership and age ($p < 0.001$), gender ($p < 0.001$), and marital status ($p = 0.003$; see Table 4a). Specifically, participants in the *Appearance Distressed* group were significantly younger and more likely to be single and female. There were no significant relationships between group membership and race or education.

In Sample 2, a significant association was found between group membership and age ($p < 0.001$). Specifically, participants in *Appearance Distressed* group were significantly more likely to be younger. There were no significant relationships in Sample 2 between group membership and gender, marital status, race, or education.

Medical variables. For participants in Sample 1, significant associations were found between group membership and disease subtype ($p < 0.001$); years since 1st non-Raynaud's symptom ($p = 0.033$); sclerodactyly ($p = 0.017$); lower hand thickening ($p < 0.001$); fingertip scars ($p = 0.001$); and pigmentation ($p = 0.029$; see Table 4b). Specifically, participants in the *Appearance Distressed* group were significantly more likely to have diffuse disease, more years since first non-Raynaud's symptom, lower hand thickening, fingertip scars, and pigmentation issues. There were no significant

relationships in Sample 1 between group membership and years since diagnosis, telangiectasia, or BMI.

For participants in Sample 2, significant associations were only found between group membership and disease subtype ($p = 0.020$) and pigmentation ($p = 0.006$; see Table 5b). Specifically, participants in the *Appearance Distressed* group were significantly more likely to have diffuse disease and pigmentation issues. There were no significant relationships in Sample 1 between group membership and years since first non-Raynaud's symptom, years since diagnosis, sclerodactyly, lower hand thickening, fingertip scars, telangiectasia, or BMI.

Psychosocial variables. For participants in Sample 1, significant relationships were found between group membership and all psychosocial measures (all ps < 0.001; see Table 4c). Specifically, participants in the *Appearance Distressed* group were significantly more likely to have higher PHQ-8 (i.e., depressive symptomatology) SHAQ-DI (i.e., disability), and PROMIS-Anxiety (i.e., anxious symptomatology) scores.

For participants in Sample 2, significant relationships were also found between group membership and all psychosocial measures (all ps < 0.001; see Table 5c). Specifically, participants in the *Appearance Distressed* group were significantly more likely to have higher PHQ-8 (i.e., depressive symptomatology) scores, HAQ (i.e., disability), and PROMIS-Anxiety (i.e., anxious symptomatology) scores.

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CHAPTER 5: DISCUSSION

The present study aimed to identify profiles of patients based on variables indicating objective physical changes and subjective body image and social anxiety in the context of SSc, a chronic illness with appearance impacts in socially relevant areas of the body. Once profiles were derived, the second aim was to compare profiles on key sociodemographic, medical, and psychosocial variables.

As will be further explicated below, two profiles emerged after running various iterations of the LPA in both Samples 1 and 2. These two profiles were substantively analyzed for patterns of scores and termed the *Appearance Comfortable* ($n = 334$ and $n = 375$ in Samples 1 and 2, respectively) and *Appearance Distressed* ($n = 135$ and $n = 98$ in Samples 1 and 2, respectively) groups. The *Appearance Comfortable* group was characterized by less skin involvement, less body concealment behaviors, less dissatisfaction with appearance, less social discomfort, less fear of negative evaluation, less social appearance anxiety, and less social interaction anxiety. The *Appearance Distressed* group was characterized by more skin involvement, more body concealment behaviors, more dissatisfaction with appearance, more social discomfort, more fear of negative evaluation, more social appearance anxiety, and more social interaction anxiety. In both samples, younger age, diffuse disease subtype, and the presence of hypo/hyper-pigmentation were associated with membership in the *Appearance Distressed* group. Additionally, patients in the *Appearance Distressed* group had significantly higher scores on measures of depressive and anxious symptoms and physical disability.

5.1 LPA-derived Groups of Objective Skin Involvement, Subjective Body Image, and Subjective Social Anxiety

In the first aim, LPA was utilized to derive profiles of patients in order to optimize homogeneity within groups and heterogeneity between groups (Roesch et al., 2010). It had been hypothesized that multiple (i.e., more than two) profiles would emerge differentiating multiple groups representing individuals with similar objective skin involvement scores but differing subjective dissatisfaction and social anxiety scores and multiple groups representing similar personal dissatisfaction scores but differing social anxiety scores. In the present analysis, the two-profile solution emerged as the most robust and parsimonious model in both samples. Furthermore, across the exploratory analyses that were conducted (see Appendix B), the two-profile solution remained the most defensible solution across the iterations of the analysis per the statistical, descriptive, and substantive interpretations. Examining the conditional response means for each variable can aid in the interpretation of the identified profiles. In addition, comparisons to outside groups, when such comparisons are available in the literature, can further help to characterize the profiles.

In the present results, the *Appearance Comfortable* and *Appearance Distressed* groups differed on objective skin involvement using the mRss. The mRss score is often used as a proxy for disease severity in SSc (Czirjàk, Foeldvari, & Müller-Ladner, 2008). The score can also be considered a proxy for appearance because the mRss score broadly represents degree of skin thickening, which contributes to appearance changes. The *Appearance Distressed* groups in both samples had higher mRss scores, as well as poorer outcomes for all the other included personal and social indicators of body image

compared to the *Appearance Comfortable* group. This could indicate that the two groups are best characterized as simply differentiating a sicker group of patients with worse appearance changes and who are, not unexpectedly, more personally and socially distressed than a less sick group of patients with less severe appearance changes, and less concurrent personal and social distress.

However, although the difference in objective skin scores, as measured by the mRss, was statistically significant, the difference was relatively small. Considering the methodology of the mRss and the range of possible scores, it would appear that the differences in the present sample are likely not substantial in terms of severity or visibility. To derive an mRss score, the clinician palpitates the skin in each of 17 body areas, including the face, upper arms, forearms, hands, fingers, chest, abdomen, thighs, forearms, and feet. A rating is given for each palpitated area per the following scoring criteria: 0 (*No Involvement*), 1 (*Mild Involvement*), 2 (*Moderate Involvement*), or 3 (*Severe Thickening*; Clements et al., 1995), with scores ranging from 0-51. The mean differences between groups were 2.84 and 1.97 in Sample 1 and Sample 2, respectively. Specifically, the *Appearance Comfortable* group in both samples had only slightly lower mRss scores ($M = 7.03$, $SD = 9.6$ in Sample 1; $M = 7.65$, $SD = 8.8$ in Sample 2) compared to the *Appearance Distressed* group ($M = 9.87$, $SD = 10.6$ in Sample 1; $M = 9.62$, $SD = 10.3$ in Sample 2). Comparing the median scores in the two samples, the similarities between groups become more evident. In Sample 1, the overall median was 5.00, whereas the medians for the *Appearance Comfortable* and *Appearance Distressed* groups were 4.00 and 6.00, respectively. In Sample 2, the overall median was 5.00, and the medians for the *Appearance Comfortable* and *Appearance Distressed* groups were

also 5.00. Furthermore, an examination of the frequencies of scores by groups demonstrated relatively similar ranges. In Sample 1, the range of mRss scores in the *Appearance Comfortable* group was 0-47, with 8.1% ($n = 27$) of the sample having an mRss of 0 and 5.7% ($n = 21$) of the sample having an mRss of 20 or greater. The range of mRss scores in the *Appearance Distressed* group was 0-45, with 6.7% ($n = 9$) of the sample having an mRss of 0 and 10.4% ($n = 17$) of the sample having an mRss of 20 or greater. In Sample 2, the range of mRss scores in the *Appearance Comfortable* group was 0-44, with 9.3% ($n = 35$) of the sample having an mRss of 0 and 8.3% ($n = 38$) of the sample having an mRss of 20 or greater; the range of mRss scores in the *Appearance Distressed* group was from 0-48, with 11.0% ($n = 11$) of the sample having an mRss of 0 and 12.2% ($n = 16$) of the sample having an mRss of 20 or greater.

Additionally, for both groups, the mean mRss scores and standard deviations were similar to scores in the literature for other non-SPIN SSc samples (e.g., $M = 9.0$, $SD = 8.7$ in Jewett et al. [2012*a*] with a Canadian sample; $M = 9.5$, $SD = 8.6$ in Jewett et al. [2010] in a U.S. sample; $M = 6.4$, $SD = 5.9$ in Kwakkenbos et al. [2012] in a Dutch sample; $M = 8.70$, $SD = 8.5$ in Mills et al. [2015] in a U.S. sample; $M = 8.6$, $SD = 6.7$ in Tedeschini et al. [2013] in an Italian sample).

Some studies have examined what mRss score differences may be clinically important. In a double-blind, randomized clinical trial (RCT) with 134 SSc patients with diffuse disease that evaluated minimally important difference (MID) in mRss, scores 3.2 to 5.3 (0.40–0.66 effect size; Khanna et al., 2006) were found to indicate MID. However, estimates of MID may depend on baseline scores (Crosby, Kolotkin, & Williams, 2003), and the Khanna et al. (2006) study substantiated this as individuals with higher baseline

mRss scores required larger changes in the score to be considered minimally improved

per physician report. The baseline mRss score in the aforementioned RCT was a mean of

21 (*SD*: 8), and thus a clinically meaningful difference in scores may actually be smaller

in the present sample. However, MID, as assessed in Khanna et al. (2006), was meant to

indicate a change in scores for the purpose of clinical trial outcomes and was anchored by

physician reports of global health. Study physicians were asked to rate the change in the

patient's health over two years and this was the anchor used to determine MID.

Therefore, MID in this setting reflected a clinical change in health status and not

necessarily any direct or meaningful change in appearance. Similarly, skin involvement

severity has been described in terms of overall disease severity in clinical trials, but not in

relation to appearance. In one clinical trial (Khanna et al., 2009), inclusion criteria

included overall mRss scores $\geq$ 20 or scores $\geq$ 16 with truncal involvement. Thus, the

region of skin involvement may also be used clinically to distinguish severity. Amjadi et

al. (2009) noted this cutoff as indicating "moderately severe skin disease" (p. 2492),

however justification was not provided. The Medsger criteria for SSc severity describes

an mRss of 0 as normal, 1-14 as mild, 15-29 as moderate, 30-39 as severe, and $\geq$ 40 as

end stage disease (Medsger et al., 2003). Keeping in mind the variability present in both

profiles, the mean mRss scores for both the *Appearance Comfortable* and *Appearance

Distressed* groups would suggest mild disease per the Medsger criteria. Again, though,

this is in relation to clinical outcomes in SSc generally and not appearance specifically.

　　　To date, there is no known MID in skin scores to determine an objective

difference in appearance. Furthermore, few studies have assessed relationships among

mRss and body image or psychosocial correlates of body image. Two studies examining

mRss as a correlate of depressive symptomatology did not find significant associations (Kwakkenbos et al., 2012; Tedeschini et al., 2013). In one of the few studies directly examining the role of skin involvement in appearance, mRss was not significantly associated with appearance self-esteem (van Lankveld et al., 2007). However, Jewett et al. (2012*a*) found significant correlations between mRss scores and the Brief-SWAP-DA and Brief-SWAP-SD scores ($r = 0.24$, p < 0.001 and $r = 0.23$, $p < 0.001$, respectively). In one other study with SSc patients that considered mRss in the context of an appearance variable, there was no significant difference comparing scores in patients identified as experiencing impacts of disfigurements versus those not (Tedeschini et al., 2013). Of note, a validated assessment was not used to evaluate appearance impacts in this study. A dichotomous variable was derived for impacts of disfigurement based on answers to the question, "Do the changes of your body image induced by disease negatively influence your behavior or relationships with other people?" (p. 189). Overall, in the limited research that exists, there is disagreement regarding the role of mRss in body image.

How clinically meaningful the mRss differences between the two profiles identified in both samples really are in terms of appearance is an empirical question that remains yet to be answered. However, considering the above information, the differences in mRss do not seem substantially different. It is unlikely that there would be a marked appearance change corresponding to the mean differences identified. Therefore, it is of particular interest that, although the differences in objective skin involvement were arguably relatively small, the differences in body image and social anxiety scores were relatively large, suggesting that the experience of living with appearances changes in SSc is driven by factors beyond objective skin involvement.

Although the two identified groups were somewhat similar on the degree of objectively measured skin involvement, they were quite different on both body image and social anxiety indicators. Regarding body image, scores for the BCSS, Brief-SWAP-DA, and Brief-SWAP-SD were markedly higher for patients in the *Appearance Distressed* group. An examination of the BCSS sample and conditional response means (Table 3a and 3b), revealed much higher scores in the *Appearance Distressed* group (M = 16.81 in Sample 1; M = 16.19 in Sample 2) as compared to the *Appearance Comfortable* group (M = 4.52 in Sample 1; M = 4.82 in Sample 2). The BCSS items represent behavioral modifications (e.g., hiding hands, wearing concealing clothing, avoiding shaking hands) made in response to one's appearance. There are no norms or cut-offs for the BCSS, but considering that responses for the nine items range from 0 (*never*) to 5 (*always*), with higher scores indicating more concealment behaviors, the *Appearance Distressed* group appears to either be engaging in some concealment behaviors a significant portion of the time (e.g., a patient with hand concerns may be endorsing a higher frequency of concealment behaviors for the four items referencing hands), or engaging in all of the behaviors at least some of the time. Contrastingly, the *Appearance Comfortable* group seems to be making very few concealment attempts, possibly engaging in only one scenario much of the time (e.g., wearing clothes that he/she does not like) or a few of the scenarios very occasionally. The behavioral responses to appearance changes seem to be quite different between the two groups such that the *Appearance Comfortable* does not seem to be making many changes at all, whereas the *Appearance Distressed* group is clearly engaging in concealment behaviors.

Similarly, in the present study, the *Appearance Distressed* group was more dissatisfied and socially uncomfortable via the Brief-SWAP-DA as compared not only to the *Appearance Comfortable* group but also to the overall sample means for Samples 1 and 2 (see Tables 3a and 3b). An examination of the Brief-SWAP-DA (i.e., personal body image dissatisfaction) sample and conditional response means (Table 3a and 3b), revealed higher scores in the *Appearance Distressed* group ($M = 12.64$ in Sample 1; $M = 12.70$ in Sample 2) as compared to the *Appearance Comfortable* group ($M = 7.72$ in Sample 1; $M = 8.02$ in Sample 2). The *Appearance Distressed* group also scored higher compared to a non-SPIN SSc sample assessed by Jewett et al. (2012) on the Brief-SWAP-DA ($M = 8.4$, $SD = 5.2$); the mean reported in the Jewett et al. study was relatively close to the overall sample means for Samples 1 and 2 and the *Appearance Comfortable* group mean scores. The Brief-SWAP-DA measures self-reported dissatisfaction via three directly worded-items asking about dissatisfaction with one's face, arms, and hands. The response options ranged from 0 (*strongly disagree*) to 6 (*strongly agree*), with higher scores indicating greater dissatisfaction. Thus, it appears that a moderate level of dissatisfaction or at least ambivalence about one's appearance (3 was a response option for *neutral*) was present in the *Appearance Distressed* group. The *Appearance Comfortable* group, however, appears to be at least somewhat satisfied with their appearance. Considering that the Brief-SWAP-DA is capturing the evaluative dimension of the personal aspects of body image, what the difference in scores suggests is that although individuals in both groups are presenting with relatively mild skin involvement, the *Appearance Distressed* group is more personally dissatisfied with these mild bodily changes than the *Appearance Comfortable* group.

Turning to the Brief-SWAP-SD, scores for the *Appearance Distressed* group (*M* = 11.64 in Sample 1; *M* = 11.02 in Sample 2) were much higher than the *Appearance Comfortable* group (*M* = 2.82 in Sample 1; *M* = 3.32 in Sample 2). Similarly, the *Appearance Distressed* group also scored much higher compared to the same Jewett et al. (2012) SSc sample described above (*M* = 5.1, *SD* = 5.1); the mean that Jewett et al. (2012) reported was relatively close to the sample mean scores for Samples 1 and 2 but much higher than *Appearance Comfortable* mean scores. The Brief-SWAP-SD measures social discomfort via three items using the same response scale described above. The items reference discomfort in the presence of others, feeling unattractive to others, and feeling that to others would not want to touch the patient. Although there are no formal norms, the scores in the *Appearance Distressed* group indicate that individuals agree or at least somewhat agree that their condition makes them uncomfortable socially, while scores in the *Appearance Comfortable* group actually suggest little to no social discomfort.

As with the body image scores, the social anxiety indicator scores (i.e., the BFNE-8, SAAS, and SIAS-6) also were much higher in the *Appearance Distressed* group as compared to the *Appearance Comfortable* group. Scores in the *Appearance Distressed* group were also higher than the sample means for Samples 1 and 2, and comparator samples, when available, from the literature. An examination of the BFNE-8 sample and conditional response means (Table 3a and 3b), revealed much higher scores in the *Appearance Distressed* group (*M* = 24.45 in Sample 1; *M* = 25.62 in Sample 2) as compared to the *Appearance Comfortable* group (*M* = 12.71 in Sample 1; *M* = 13.31 in Sample 2). Response options ranged from 1 (*not at all characteristic of me*) to 5 (*entirely*

characteristic of me) for the eight items that assessed fear of being negatively evaluated by social others, for example, worrying about what someone may be thinking about them when they are engaged in a conversation with that person. A different variant of the Fear of Negative Evaluation scale was used in a sample of 49 SSc patients (Richards, Herrick, Griffin, Gwilliam, & Fortune, 2004); in that study although the mean scores fell below the threshold for social anxiety, 27% of patients (*n* = 13) did have scores meeting the social anxiety cutoff. Unfortunately, there is no cutoff for social anxiety using the BFNE-8 version of the measure that was used in the present study. However, in a sample of healthy college students (Coles, Pietrefesa, Schofield, & Cook, 2008), mean BFNE-8 scores were 20.88 (*SD* = 8.01). Scores in the *Appearance Distressed* group were somewhat higher than this mean, indicating that individuals in this group felt that having fears of negative evaluation were more characteristic of them than did healthy college students. Interestingly, however, participants in the *Appearance Comfortable* group seemed to be doing markedly better in terms of fears of negative evaluation than the average college student.

An examination of the SAAS sample and conditional response means (Table 3a and 3b) also revealed relatively much higher scores in the *Appearance Distressed* group (*M* = 43.25 in Sample 1; *M* = 49.0 in Sample 2) as compared to the *Appearance Comfortable* group (*M* = 21.83 in Sample 1; *M* = 22.70 in Sample 2). In studies with physically healthy college students, the mean scores were 30.23 (*SD* = 13.22) in a coed sample of 118 students (Levinson & Rodebaugh, 2011) and 35.64 (*SD* = 12.82) in an all-female sample of 90 students (Titchener & Wong, 2014). The 16 SAAS items include a broad range of concerns, such as feeling nervous about having one's picture taken and

worrying that others will judge them because of their appearance; response options

ranged from 1 (*not at all*) to 5 (*extremely*). In the present sample, scores indicating social

anxiety specifically related to one's appearance were much higher for individuals in the

Appearance Distressed group versus physically healthy college students. However,

scores in the *Appearance Comfortable* group were notably lower compared to physically

healthy college students, suggesting that, even in the presence of disfigurement, these

individuals were less anxious about their appearance in social settings versus non-

disfigured college students.

Finally, considering SIAS-6 sample and conditional response means (Table 3a

and 3b), the results indicated higher scores in the *Appearance Distressed* group ($M = 5.42$

in Sample 1; $M = 6.47$ in Sample 2) versus the *Appearance Comfortable* group ($M = 1.02$

in Sample 1; $M = 1.48$ in Sample 2). The measure has not been used in SSc outside of the

SPIN sample, so comparisons to other SSc populations cannot be made. Because the

SIAS-6 was developed to assess social anxiety in medically healthy populations, the

measure is often used in psychiatric settings (e.g., Peters et al., 2012) where the mean

scores tend to be higher (e.g., $M = 13.2$, $SD = 4.9$ in a social anxiety sample). However,

scores in a non-psychiatric, healthy community setting (Le Blanc et al., 2014) were

markedly lower ($M = 1.5$, $SD = 2.25$). Considering that the response options for the six

items ranges from 0 *(not at all characteristic or true of me)* to 4 *(extremely characteristic

or true of me)*, it seems that the *Appearance Distressed* group had more social interaction

concerns versus non-disfigured controls, but fewer as compared to psychiatric

population; the healthy community scores were comparable to those of the *Appearance

Comfortable* group whereas the psychiatric sample scores were much higher. In a sample

of sexual minority adults (Puckett, Levitt, Horne, & Hayes-Skelton, 2015), the scores were more comparable to those in the *Appearance Distressed* group (*M* = 4.7, *SD* = 4.8) and higher than those in the *Appearance Comfortable* group. A cutoff score of 7 has been identified as identifying between social phobia patients and non-patients using the SIAS-6 (Peters et al., 2012). Thus, the average patient in the *Appearance Distressed* group in the present samples can be conceptualized as nearing the clinical cutoff score for social phobia (renamed Social Anxiety Disorder in the American Psychiatric Associations Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition [DSM-V; APA, 2013]). Specifically, in Sample 1, 33.3% (*n* = 45) of the *Appearance Distressed* and 2.1% (*n* = 7) of the *Appearance Comfortable* group met the cutoff for Social Anxiety Disorder. In Sample 2, 42.9% (*n* = 42) of the *Appearance Distressed* and 3.5% (*n* = 13) of the *Appearance Comfortable* group met the cutoff for Social Anxiety Disorder

The two profiles had scores that were consistently higher or lower across all the indicators of body image and social anxiety, suggesting that the personal and social indicators relate more closely than hypothesized. The hypothesized pattern of finding groups differentiating patients scoring similarly high or low on personal distress but scoring differently on the social anxiety indicators was not identified. A more rigidly demarcated pattern differentiating two distinct group was found such that patients in the *Appearance Comfortable* group were coping well both personally and socially while patients in the *Appearance Distressed* group were struggling with the personal and social aspects of living with visible differences.

In sum, the two identified profiles suggest that, even in a sample with relatively low skin involvement, there are some patients who have responded to their appearance

changes with minimal personal dissatisfaction and relatively low social anxiety, and others who seem quite distressed about their appearance on both personal and social levels. This interpretation should be made with the caveat that we do not know what true MID is for the mRss in terms of appearance. Regardless, two groups of patients who are coping with the personal and social aspects of appearance changes very differently were identified in the present analysis. The associations of the two identified groups with sociodemographic, medical, and psychosocial variables can further unpack the implications of group membership.

5.2 Associations of LPA-derived Groups with Sociodemographic, Medical, and Psychosocial Variables

The second aim of the present study was to identify relationships of group membership to sociodemographic, medical, and psychosocial variables. In both samples, younger age, diffuse disease subtype, and the presence of hypo/hyper-pigmentation were associated with membership in the *Appearance Distressed* group. Additionally, patients in the *Appearance Distressed* group had significantly higher scores on measures of depressive and anxiety symptomatology and disability. The robust findings that existed for both samples will be discussed first, followed by a discussion of the differences between samples.

Significant auxiliary variables in both samples. In both samples, patients in the *Appearance Distressed* group were somewhat younger, as described further below. In the present analysis, age was the only sociodemographic variable to significantly predict group membership in both Sample 1 and Sample 2. Across the variables included in research examining relationships with body image dissatisfaction in SSc, younger age has

61

been one of the most frequently reported variables associated with body image

dissatisfaction (Amin et al., 2011; Benrud-Larson et al., 2003; Ennis et al., 2013; Jewett

et al., 2012). In the broader disfigurement literature, younger age has been associated

with poorer outcomes as well (Fauerbach et al., 1995 cited in Benrud-Larson et al.,

2003). Across previous research in disfigurement, younger individuals (i.e., pediatric

populations) have been described as more appearance-focused with more demanding

body image ideals that can contribute to greater challenges in coping with disfigurement

(Benrud-Larson et al., 2003; Rumsey & Harcourt, 2004). Although the role of age in

adjustment to disfigurement typically refers to the difference between adolescents/teens

and older adults, there can be unique challenges to dealing with appearance changes for

middle aged adults as well (Rumsey & Harcourt, 2004). The present sample was

comprised only of adults, who had an overall mean age of around 55 years old. It is

conceivable that because skin changes associated with aging (e.g., increased wrinkles,

thinner and less elastic skin, pigmentation changes) are also occurring at around this age,

some patients who are older and already experiencing changes in their appearance may

be less distressed than younger patients who have not yet noticed major aging-related

changes to their appearance. Patients in the *Appearance Distressed* group ($M = 50.36$ in

Sample 1; $M = 50.60$) were significantly younger than patients in the *Appearance*

Comfortable group ($M = 58.01$ in Sample 1; $M = 55.90$ in Sample 2), although the sample

could generally be characterized as comprised of middle-aged women and men

(consistent with the epidemiology of the SSc). Another factor to consider, however, is the

age at onset of symptoms (including appearance changes). Although age at onset of

appearance changes was not directly evaluated in the present study, this variable may also

be influencing the present results. Ennis et al. (2003) found that, while disease duration in SSc generally was not associated with poorer body image outcomes, younger age at onset of first non-Raynaud's symptom was associated with body image dissatisfaction. This same type of relationship may hold true specifically for age at onset of first appearance change. There may be a developmental distinction in terms of appearance concerns and coping potential being captured for women who start to experience the changes to their appearance in their late 30s/early 40s versus their 50s, considering that the mean years since first non-Raynaud's symptom was approximately 11.

Regarding medical variables, the results demonstrated that, across both Sample 1 and Sample 2, disease subtype and pigmentation were associated with group membership such that patients with diffuse disease and the presence of pigmentation problems (i.e., hypo/hyper-pigmentation) were more likely to be in the *Appearance Distressed* group. Regarding the distinction between limited and diffuse disease, it is important to consider that in limited disease, skin thickening is typically constrained to the hands, arms, and/or face, whereas in diffuse disease, the skin involvement is widespread, including the trunk (Gabrielli et al., 2009; Steen, Powell, & Medsger, 1988). Thus, although there is more widespread skin involvement in diffuse disease, the additional involved areas that limited disease patients do not experience are in less visible parts of the body (i.e., the trunk). It is possible that truncal involvement, while not socially relevant in day-to-day encounters with strangers or non-intimate acquaintances, may have social impacts in intimate relations, leading to greater associated distress. Moreover, previous research has also related diagnosis of diffuse (versus limited) disease to greater body image dissatisfaction (Benrud-Larson et al., 2003; Ennis et al., 2013; Heniberg et al., 2008; Nusbaum et al.,

2016). The increased severity of illness associated with diffuse disease (e.g., more internal organ involvement) may also be contributing to a more negative embodiment experience contributing to the greater body image distress in diffuse patients.

Regarding the role of specific aspects of appearance in body image outcomes, of the five specific appearance variables that were included in the analysis, only pigmentation problems were significantly associated with group membership in both samples. The other variables (i.e., sclerodactyly, lower hand thickening, fingertip scars, and telangiectasias) that were not associated with group membership in both samples will be discussed further below. The visibility or severity of the appearance variables may have influenced the results; the dichotomous appearance variables did not include information regarding severity of the variables. Pigmentation changes may be more noticeable and impact larger areas of the body than the other assessed variables. However, in the one other study examining pigmentation problems and body image, there was no significant relationship with dissatisfaction with appearance or social discomfort as assessed via the Brief-SWAP (Jewett et al., 2012*a*). One reason for the discrepancy between the Jewett et al. (2012*a*) study and the present study may be the outcome of interest. In the Jewett et al. (2012*a*) study, the Brief-SWAP was the outcome of interest. The latent variables in the present study, however, included both personal and social aspects of body image and captured a distress dimension beyond that which was captured in the Brief-SWAP alone.

Regarding the psychosocial auxiliary variables, in both samples, the analysis demonstrated that group membership was significantly associated with scores on measures of depressive symptomatology, anxiety symptomatology, and disability.

Specifically, patients in the *Appearance Distressed* group were more likely to have higher PHQ-8, PROMIS-Anxiety, and SHAQ-DI scores. Thus, as hypothesized, the group with more appearance-related distress had worse depression, anxiety, and disability outcomes.

Several studies have demonstrated a moderate relationship between poorer body image scores and adverse mood outcomes (Benrud-Larson et al., 2003; Jewett et al., 2010; Jewett et al., 2015; Kwakkenbos et al., 2012; Leite et al., 2013; Mills et al., 2015; Van Lankfelt et al., 2007) or increased disability (Benrud-Larson et al., 2003; Heinberg et al., 2007; Mills et al., 2015; Van Lankfelt et al., 2007). The association of group membership with poorer mood outcomes suggests that individuals in the *Appearance Distressed* group are experiencing general anxiety and depression symptoms in addition to appearance-related distress. Regarding the PHQ-8, considering that scores 5, 10, 15, and 20 categorize mild, moderate, moderately severe, and severe depression respectively, means scores for patients in the *Appearance Distressed* group were in the moderate range, whereas mean scores for patients in the *Appearance Comfortable* group were below even the mild range of depressive symptoms.

The PROMIS-Anxiety measure is scored using T-score metric set to 50 ± 10 ($M \pm$ SD); while mean scores for the *Appearance Comfortable* were just slightly below the mean of 50, scores for the *Appearance Distressed* group were approximately one full standard deviation higher than the mean. Because the PROMIS T-score metric is normed around the general US population (Liu et al., 2010), individuals in the *Appearance Distressed* group are reporting anxiety one standard deviation more severe than the

average U.S. person, while individuals in the *Appearance Comfortable* group are about as anxious as the average U.S. person (Schalet, Cook, Choi, & Cella, 2014).

Regarding disability, items on the SHAQ-DI is scored from 0 (*without difficulty*) to 3 (*unable to do*) and total scores are transformed to the same 0 to 3 range. Mean scores for the *Appearance Distressed* group were around 1, whereas means scores for the *Appearance Comfortable* group were around 0.6, indicating a difference in functioning although both groups are relatively low on disability. The embodied experience in SSc may be different for patients experiencing this slightly greater disability; there may be a greater feeling of detachment from or distrust of one's body promoting greater distress. Previous research examining body esteem in individuals with physical disabilities showed that physical disability adversely impacted body esteem and that social feedback may be a mediator of body esteem for individuals living with physical disabilities (Taleporos & McCabe, 2001). The combination of experiencing appearance changes and physical disabilities may be compounding the body-related distress that patients experience. Additionally, some of the appearance changes, for example hand contractures, may directly relate to both increased disability and poorer social outcomes.

Cash et al. (2004*c*) argued, "While discontent with some aspect of one's appearance increases one's risk for the experience of emotional distress and psychosocial impairment, dissatisfaction per se is not a sufficiently valid index of dysfunction or disorder" (p. 364). There has been a call for a clearer distinction between body image dissatisfaction, which is relative common in community samples, and body image distress or disturbance, which captures a more pathological level of negative evaluation that includes psychological impairment and functioning deficits (Cash et al., 2004*c*;

Thompson, Heinberg, Altabe, & Tantleff-Dunn, 1999). Without a direct measure of the specific construct of body image distress, disturbance, or dysmorphia, one could argue that the moniker *"Appearance Distressed"* for the more impacted profile in the LPA may be inappropriate. However, the strong relationships between group membership and higher depressive and anxious symptomatology suggest that the construct of body image distress is indeed being harnessed in the *Appearance Distressed* group. This is interesting because, traditionally, the construct of body image distress is conceptualized as cohering at the intersection of body image dissatisfaction and appearance investment (Cash, 2002*a)*. Body image disturbance or distress has been differentiated from the more evaluative construct body image dissatisfaction (Cash et al., 2004*c*) via the inclusion of the appearance investment variable. However, even without a direct assessment of appearance investment, the *Appearance Distressed* group is essentially tapping in to the body image distress construct and identifying individuals with elevated psychological distress via anxiety and depression symptomatology.

   Non-significant auxiliary variables in both samples. Regarding the sociodemographic variables, education was not associated with group membership in either group. No significant relationship between education and body image outcomes in SSc has been reported in previous research (Jewett et al., 2012*a*), and thus it had been hypothesized that education would not be associated with group membership in the present study. Because of the varied educational systems in the countries included in the SPIN study, education was a continuous variable describing years of education completed starting with elementary/primary school. The sample was relatively well-educated with a mean of around 15 years of education completed, which would correspond to an average

educational attainment of at least some college/university education. It is possible that a sample with greater educational variance and categorical distinctions between different levels of education would have produced different results.

Race was also not a significant predictor of group membership in either sample. In one previous study, African American race had been associated with higher rates of body image dissatisfaction as compared to Caucasian race (Nusbaum et al., 2016). There has been some research indicating that African Americans with SSc generally have poorer prognoses as compared to White Americans with SSc, even after adjusting for age at disease onset, disease duration, sex, disease subtype, SES, health insurance status, and various medical indicators; this has been hypothesized to suggest a unique disease phenotypic expression in African Americans (Gelber et al., 2013). As described in the study methods, the SPIN sample included different country-specific racial identifications (i.e., in Canada options were Aboriginal, White, Asian, Black, Latin American, Arab, and Other; in the U.S. options were Asian, American Indian/Alaska Native, White, Native Hawaiian/Other Pacific Island, More than one race, and Black or African American; in France, options were White, Asian, African, and Other) that were re-coded in the dataset into White/Caucasian, Black/African-American, and All Other. Although the Nusbaum et al. (2016) study used the same three racial categories in their all U.S. sample, they had a much higher percentage of Black patients (27% versus 5.8% in the present sample). Thus, the lack of variability in the present sample may have precluded significant results. It is also possible that a more nuanced categorization of the specific racial categories (e.g., Asian) would have introduced significant relations to group membership. Such analyses

can be undertaken if future SPIN recruitment produces higher numbers of participants from non-White ethnicities.

Regarding medical variables, BMI and telangiectasias did not predict group membership in either sample. BMI had not been previously assessed as a predictor of body image outcomes in SSc. Although higher BMI can be an important predictor of body image outcomes in other populations (Schwartz & Brownell, 2004), the impacts of the SSc can cause both weight gain (e.g., as a result of medications) and weight loss (e.g., related to gastrointestinal complications). Patients may have challenges associated with weight changes in either direction, and thus the linear relationship of BMI to group membership may not accurately capture the distress associated with weight change. Future research may seek to examine weight in terms of being both underweight and overweight.

Although the presence of telangiectasias (i.e., widened blood vessels that can appear as thread-like lines across the skin) has been associated with body image dissatisfaction in previous research (Ennis et al., 2013; Jewett et al., 2012*a*), there was no association with group membership in either sample. In the total sample and across both *Appearance Distressed* and *Appearance Comfortable* groups in both samples, the presence of telangiectasias was around 70%. Additionally, as with all the appearance symptoms, the severity of the symptom was not assessed. Moreover, this specific symptom is not limited to SSc, and can appear in healthy individuals as well. Thus, it is possible that the noticeability and stigma of this symptom is less acute than more unique symptoms, such as finger thickening.

Differences between groups on associations with auxiliary variables. Two sociodemographic variables, gender and marital status, were associated with group membership in Sample 1 but not Sample 2. Specifically, patients in the *Appearance Distressed* group in Sample 1 were more likely to be female and single. Regarding gender, the majority of the existing research examining body image in SSc has been conducted only with women, given that the disease has a much higher prevalence in women; when men have been included (e.g., Amin et al., 2011; Ennis et al., 2013; Heinberg et al., 2007), analyses have not been conducted to examine differences in body image dissatisfaction by gender. In non-disfigured samples, women tend to have poorer evaluative body image outcomes as well as greater body image investment compared to heterosexual men (e.g., Muth & Cash, 1997). However, Rumsey and Harcourt (2008) reported that the evidence on gender in body image outcomes in visibly disfigured populations is discordant, with some studies showing no gender differences in body image outcomes (e.g., White, 1982) and others finding poorer outcomes in females (e.g., Carr, Harris, & James, 2000).

The association between marital status and body image dissatisfaction has been mixed in the literature, with one study finding a significant association between being single and body image dissatisfaction (Ennis et al., 2013) and one study finding no relationship (Jewett et al., 2012). Another study examining correlates of depression in SSc found that being single was associated with greater depressive symptomatology (Thombs, Hudson, Taillefer, Baron, & Canadian Scleroderma Research Group, 2008*b*). The majority of the overall present sample (around 73%) was married or common law status. In Sample 1 where marital status was significantly associated with group

membership, while around 77% of individuals in the *Appearance Comfortable* group were partnered, only 61.5% in the *Appearance Distressed* group were partnered. Given the cross-sectional nature of the study, however, it could be argued that the couples who are able to remain (or become) a romantic dyad following the interpersonal challenges associated with the diagnosis of an incurable, chronic illness in one partner are already a self-selected, more highly satisfied group, both in terms of body image and relationship quality. Claims as to the directionality of the relationship cannot be made, so it is unclear if being in a relationship promoted better body image and social anxiety outcomes, or if better body image and social anxiety outcomes promoted initiating or maintaining a romantic relationship.

There were also a number of medical variables associated with group membership in Sample 1 but not Sample 2. In Sample 1, more years since first non-Raynaud's symptom and the presence of several appearance variables in addition to pigmentation (i.e., sclerodactyly, lower hand thickening, and fingertip scars) were associated with membership in the *Appearance Distressed* group. Years since first non-Raynaud's symptom is commonly used as a proxy for disease duration in SSc, given that the length of time to achieve an accurate diagnosis can be quite long for some patients. In SSc, the course of the changes in appearance tend to be such that the changes are rapid and progressive early in the course of the disease and then level out (Gholizadeh et al., 2017), so it was hypothesized that longer disease duration would not be associated with better appearance coping or more severe changes in appearance. Additionally, in previous research a significant association between years since first non-Raynaud's symptom and body image dissatisfaction had not been identified (Ennis et al., 2013). In the present

sample, the mean duration since first non-Raynaud's symptom was approximately 11 years; the difference in means for the *Appearance Dissatisfied* and *Appearance Comfortable* was about two years (12.7 versus 10.8, respectively). Thus, individuals in the *Appearance Dissatisfied* group had been living with SSc symptoms, likely including appearance changes, for approximately two years longer than those in the *Appearance Comfortable* in Sample 1. While it may have been conjectured that greater time living with the changes would have enabled improved coping and adjustment to the visible differences, the longer disease duration was actually associated with greater dissatisfaction and distress in Sample 1. As described above relating to age, the longer disease duration may be associated with a younger age at onset of first appearance symptom, contributing to more distress. It is also possible that individuals who have lived longer with the appearance changes have experienced more negative social responses to their disfigurement.

Given the significant associations of appearance variables with body image outcomes in the literature, it was somewhat surprising that all of the appearance variables were not associated with group membership in the present study. Sclerodactyly, lower hand thickening, and fingertip scars were only associated with membership in the *Appearance Distressed* group in Sample 1. In previous literature, face tightening, skin tightening above the elbows, skin tightening above the knees, finger restriction (Benrud-Larson et al., 2003), facial involvement (Jewett et al., 2012*a*), skin thickening of the right hand and fingers (Malcarne et al., 1999), and telangiectasias (Ennis et al., 2013) have been identified as significant correlates of body image dissatisfaction. However, as previously described, the dichotomous assessment of the appearance variables as

72

present/not present, versus a more nuanced description that included severity of each symptom, may have precluded significant findings. Furthermore, the appearance variables were all physician-rated. In line with the results of the present study demonstrating that objective severity does not relate directly to body image distress, it may be that physician ratings of presence of a given appearance symptom would not necessarily correspond to patient perceptions of noticeability and severity of that symptom.

5.3 Implications of the Present Results

The primary implication of the present research is that the subjective personal and social experiences of appearance changes in SSc appear to be driven by factors beyond objective skin involvement; one cannot simply assume degree of distress based on level of skin involvement, or disease severity. In the present analysis, two groups were identified that were generally similar in terms of skin involvement scores, which were relatively low and indicated mild skin thickening, but for whom the experience of living with these appearance changes seemed markedly different.

In addition to appearance dissatisfaction (i.e., the evaluative component of body image), body image behaviors and anxiety were also included in the LPA, arguably coalescing to form a body image distress latent variable. Patients in the *Appearance Distressed* group can be characterized as not only dissatisfied with their appearance, but as truly psychologically distressed. Thus, there exists a group of patients, many of whom have mild disease, or at least mild skin thickening, who may appear physically similar to or "as sick as" other patients, but who are struggling personally and socially with their appearance changes. These are the patients who are most in need of intervention, and

who are arguably most at risk of falling between the referral cracks because they may not look sick enough (or different enough from the patients who do seem to be coping well) to warrant further support.

Based on the present findings, it seems that younger patients with diffuse disease and pigmentation changes are most acutely at risk for adverse appearance-related outcomes, personally and socially. Thus, these are the characteristics that may point to patients, even those with mild skin involvement, at elevated risk for appearance and general psychological distress. Although some of the mechanisms driving these relationships were hypothesized in the discussion, it will be interesting to further examine the ways in which age, diffuse disease, and pigmentation may be impacting distress in future research. For example, including measures on sexual functioning in future research can help examine whether the intimacy impacts of the additional truncal skin involvement that diffuse disease can experience is what is contributing to the additional body image distress in these patients. Longitudinal research can examine the impacts of age throughout the disease course, potentially identifying critical developmental periods where the onset or worsening of appearances changes are more distressing (e.g., for younger patients who experience their first appearance-related symptom in their 30s, who do not yet perceive themselves as having experienced significant aging-related appearance impacts, versus patients who start to experience appearance changes later in life). Because the specific relationship with pigmentation changes was novel, future qualitative research can examine in what ways the experience of living with visible differences in SSc may be unique for patients with pigmentation changes, and why this

specific symptom may be more negatively impactful for patients than other appearance variables (e.g., telangiectasias).

There are also clinical implications of the present study. Gholizadeh et al. (2017) suggested that clinicians foster a more open dialogue around body image concerns by simply asking all patients specifically about appearance-related concerns rather than attempting to elicit distress via a general screening tool or based on the patient's appearance. While mRss or other indicators of severity (e.g., the physician's own assessment of a given patient's facial or hand severity) may be more accessible to physicians, the present results suggest that these factors will not adequately capture the patient experience of living with appearance changes. Moreover, even for patients with relatively mild skin thickening, there can be some patients who seem to cope well and others who seem to be truly distressed. Of course, this introduces a greater challenge to clinicians because the present findings suggest that identifying the patients who are truly suffering is complicated by the fact that these patients may not appear objectively more disfigured. However, the present study indicates that younger patients with diffuse disease who have experienced pigmentation changes may be most acutely at risk. The present results can normalize such concerns by underscoring that research has shown that body image concerns and social difficulties are common in SSc and related to many factors beyond objective skin thickening. This may open the door for distressed patients with relatively less severe disease to feel more validated in broaching topics related to body image and social anxiety.

Unfortunately, clinicians may feel ill-equipped to handle such discussions. Another option for patients is support groups. In-person support groups do exist in many

cities around the world for SSc. These are typically peer-led groups that are established through patient advocacy organizations. A recent study examined the reasons for non-participation among SSc patients ($N = 242$) in these groups, given that they can be an excellent form of psychosocial support (Gumuchian et al., 2017). Reasons for non-participation could be distilled into personal reasons (e.g., not feeling comfortable in a group setting), practical reasons (e.g., no access to local groups), and beliefs about support groups (e.g., beliefs that support groups would be negative settings). A specific practical reason for non-attendance, that 8.7% of the sample endorsed, was discomfort with appearance. Thus, patients who are appearance distressed, who stand to benefit greatly from support, may be avoiding support group-based settings specifically because of their appearance.

Other resources beyond physicians and support groups are needed to help patients struggling with challenges related to appearance changes. One such resource is represented by an online intervention currently in development to specifically address body image concerns in SSc that is extremely relevant to the present sample and could be a useful referral. The SPIN body image intervention is a web-based intervention being developed to address the personal and social challenges of appearances changes in SSc (Kwakkenbos et al., 2013). The online format of the intervention may make the intervention more appealing and relevant to younger patients, who may be at greater risk for body image distress. The web-based format will also increase accessibility for patients with physical disabilities and more advanced (i.e., diffuse) disease that may prohibit in-person support seeking. Given the prevalence of social anxiety, the online intervention may also capture a highly anxious subset of patients who otherwise would

avoid in-person or group-based psychosocial interventions. Although there are certain

factors that a psychosocial intervention cannot remedy (e.g., skin thickening), it is the

subjective personal and social aspects of body image that appear to be driving elevated

body image distress in the present study. These are direct targets of the SPIN

intervention, which is grounded in both cognitive-behavioral skills (e.g., challenging

unrealistic social concerns) and acceptance-based approaches (e.g., coping with the fact

that one's face looks different than it used to). Additionally, the intervention will include

an optional module on camouflage/makeup that can address specific pigmentation

concerns. The intervention is currently being developed and will undergo efficacy testing

via a randomized controlled trial, but the findings of the present study suggest that the

SPIN body image intervention will be an extremely relevant and useful tool for patients

with a wide range of appearance changes in SSc.

A third implication involves possible positive coping mechanisms associated with

the *Appearance Comfortable* group. Across all measures included in the LPA, the

Appearance Comfortable group tended to score more favorably as compared not only to

the overall sample means but also to other comparison groups, often including healthy

community controls, suggesting that for some individuals, appearance changes may be

introducing a heightened coping mechanism or resiliency. Grounded in Lazarus and

Folkman's (1984) Model of Stress and Coping, there is a rich literature examining the

ways that the evaluation of a stressor (i.e., cognitive appraisal) and cognitive and

behavioral attempts at managing a stressor (i.e., coping styles) can impact adjustment and

outcomes related to chronic illness (e.g. Folkman, 2013; Sirois & Gick, 2014; Stanon &

Snider, 1993). For example, the presence of emotion-focused coping (e.g., wishful

thinking, self-blame) has been related to worse psychosocial adjustment and increased depression, even after controlling for physician-rated illness severity (Bombardier, D'Amico, & Jordan, 1990). That patients who overall have mild skin thickening are experiencing the appearance impacts of a disease so differently suggests that differences in appraisals of the physical changes and coping styles related to the visible differences may be influencing outcomes.

There could also be a meaning-making phenomenon taking wherein some individuals who have been faced with the stressors of SSc have been able to reorient their worldviews (including, perhaps, values placed on appearance). Park (2010) described meaning making as comprised of the following tenets: 1) individuals possess a cognitive capacity to orient and find global meaning in response to new experiences; 2) individuals appraise and ascribe meaning to experiences that possess the potential to challenge or place stress upon previously arrived upon global meaning; 3) the discordance between appraised meaning and global meaning is associated with distress; 4) this distress motivates meaning making; 5) this meaning making process has the underlying goal of reducing the discordance such that the world can again be a meaningful place in which the individual life has purpose; and 6) when the process is successful (i.e., the meaning made is achieved via meaning making), individuals are more highly adjusted and cope better.

The diagnosis of the incurable disease SSc and the onset of unpredictable, uncontrollable, and undesirable appearance changes would certainly constitute the type of experience for which the meaning-making model may be applied (Park & Folkman, 1997). Patients who have to face the unavoidable reality of appearance changes may find

78

themselves shifting values about appearance or reframing self-views and identity beliefs such that a changed appearance is no longer a barrier to (and, perhaps, even an impetus toward) living a meaningful life. There has been some criticism of the meaning-making framework, however. Some researchers have argued that better adjustment is not a process of meaning making, but rather the natural course of resiliency in humans; meaning making may actually promote maladaptive ruminative processes rather than recovery (Bonnano, Papa, Lalande, Zhang, & Noll, 2005).

Rumsey and Harcourt (2004) also described that although the research in disfigurement tends to focus on poorer outcomes, there is some evidence for strengths and resilience rather than deficits associated with disfigurement. For example, in a study examining coping patterns in adults with cleft lip and palate, there was a high variability in outcomes, with a smaller subset of patients reporting positive outcomes associated with having a congenital visible difference (Cochrane & Slade, 1999). Regardless of whether it is conceptualized as resilience or meaning making, there appears to be a phenomenon taking place such that for some (the majority, in fact) individuals with SSc, the experience of living with a visible difference may promote better social anxiety outcomes compared to other SSc patients and perhaps, as evidenced by comparisons with general community samples, even compared to the general, non-disfigured public. This is a finding that warrants further research both of a qualitative nature, such that the underlying processes can be explored, and also via quantitative longitudinal designs that promote the tracking of personal and social adjustment to changes in appearance.

5.4 Limitations

It is important to interpret the present results with relevant limitations in mind. The first limitation concerns the model selection process and is present in every LPA analysis. Because there are no gold-standard fit criteria, it is left to the researcher to select the number of classes based on a combination of fit criteria (e.g., entropy, likelihood ratio statistical tests; Tein, Coxe, & Cham, 2013). For the present analysis, LMRT, BLRT, and entropy were first considered, with the descriptive fit indices and substantive interpretation taken into consideration secondarily. Replicability between the two samples was also considered. Using the aforementioned criteria, the two-class solution emerged as the optimal solution. It is possible that focusing on other selection criteria could have changed the results. However, even considering that there was some disagreement across the indicators, the selection of a three (or greater)-profile solution would still not have supported the study hypothesis. An examination of the conditional response means revealed that for the groups that would have emerged, scores would have remained consistently higher or lower for all indicators in any additional groups. In other words, the difference would have been between a high-medium-low pattern versus a high-low pattern; the additional profiles in the three, four, or five solutions would not have introduced groups like the ones hypothesized, with some relatively higher and some relatively lower indicator values.

Additionally, the SPIN Cohort is a convenience sample of SSc patients who are receiving treatment at SPIN recruiting centers, which tend to be large, university-based hospitals or centers with rheumatologists with expertise in SSc. Patients at these centers may differ from SSc patients in other settings. SPIN Cohort patients also complete all study questionnaires online, potentially further limiting the generalizability of findings.

Of note, however, patients who did not have access to the Internet or a means to complete the questionnaires but wanted to participate were provided access at the recruitment centers. Additionally, the majority of the present sample was White; given that aspects of body image may be socio-culturally bound (Fallon, 1990), results may not be generalizable to the body image experiences of participants from other racial/ethnic backgrounds, and the ability to make racial/ethnic group comparisons was limited. The cross-sectional nature of the study also precludes claims of causality or directionality. For example, it is not possible to know whether the *Appearance Distressed* patients were a more psychosocially distressed group prior to the SSc diagnosis.

Another important limitation of the present research involves the conceptual underpinnings of body image as a construct. Body image is a multidimensional construct (Cash & Pruzinsky, 1990). Researchers have many options of specific dimensions of body image and related measures to utilize in order to capture a particular aspect of the construct (Thompson, 2011). An LPA (and most exploratory research) is only as good the measures that are included in the analysis; the groups are developed based on the selected indicators. It is conceivable that for each of the selected dimensions of body image, different measures could have been selected or entirely different dimensions of body image may have been targeted, potentially leading to different groups.

5.5 Summary and Conclusions

The results of the present study, even interpreted within the context of the aforementioned limitations, contribute to the growing literature on body image in SSc. The present study was the first to use LPA in the context of body image in SSc, and the first study to identify typologies of patients based on indicators of body image in any

81

disfiguring condition. The present study proposed to examine the role of various body image and social anxiety indicators in SSc. The results demonstrated that LPA can be used to distinguish between profiles of patients based on body image and social anxiety indicators. However, rather than a more complex, multi-group solution demonstrating variability in patients' relationships with objective and subjective body image evaluations and personal and social aspects of body image, two distinct profiles were identified. The profiles distinguished between an *Appearance Comfortable* group, comprised of patients with lower objective skin involvement, better body image, and lower social anxiety and an *Appearance Distressed* group including patients who had higher objective skin involvement, poorer body image, and higher social anxiety. The results also elucidated variables that were associated with group membership and may be used to help identify patients who may be in need of intervention and who are at risk for poorer outcomes due to significant problems with personal and social facets of body image. Younger patients with diffuse disease and pigmentation changes seem to be particularly at risk for adverse appearance-related outcomes.

An important implication of the present study is that the subjective experience of appearance changes in SSc appears to be driven by factors beyond objective skin involvement, and thus one cannot simply assume degree of distress based on level of skin thickening. Although this can make the identification of patients at risk for distress more challenging, it can also normalize and validate personal and social challenges of living with appearance changes even in patients with relatively mild disease. An SSc-specific body image intervention currently in development of the SPIN team that will target personal and social aspects of body image may be especially relevant for the *Appearance*

Distressed patients. Of interest, the subjective body image and social anxiety scores for the *Appearance Comfortable* group suggested that patients in this group not only fared better than their *Appearance Distressed* peers, but also had better outcomes than general, healthy community samples, possibly pointing to a meaning-making or resilience process.

Chapter 5 is being prepared in part for publication. This publication will be co-authored by Vanessa L. Malcarne, Scott C. Roesch, Marie-Eve Carrier, Linda Kwakkenbos, Sarah D. Mills, Rina S. Fox, and Brett D. Thombs.. The dissertation author was the primary investigator and author of this material**.**

REFERENCES

Akaike, H. (1974). A new look at the statistical model identification. *IEEE Transactions on Automatic Control*, *19*, 716-723.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Amin, K., Clarke, A., Sivakumar, B., Puri, A., Fox, Z., Brough, V., ... & Butler, M. D. (2011). The psychological impact of facial changes in scleroderma. *Psychology, Health & Medicine*, *16*, 304-312.

Amjadi, S., Maranian, P., Furst, D. E., Clements, P. J., Wong, W. K., Postlethwaite, A. E., ... & Khanna, D. (2009). Course of the modified Rodnan skin thickness score in systemic sclerosis clinical trials: Analysis of three large multicenter, double-blind, randomized controlled trials. *Arthritis & Rheumatism*, *60*, 2490-2498.

Arminger, G., Stein, P., & Wittenberg, J. (1999). Mixtures of conditional mean- and covariance-structure models. *Psychometrika*, *64*, 475-494.

Asparouhov, T., & Muthén, B. (2014). Auxiliary variables in mixture modeling: Three-step approaches using M plus. *Structural Equation Modeling: A Multidisciplinary Journal*, *21*, 329-341.

Assassi S., Leyva A. L., Mayes M. D., Sharif R., Nair D. K., Fischbach, M., ... McNearney, T. A. (2011) Predictors of Fatigue Severity in Early Systemic Sclerosis: A Prospective Longitudinal Study of the GENISOS Cohort. *PLoS ONE, 6*: e26061.

Banfield, S. S., & McCabe, M. P. (2002). An evaluation of the construct of body image. *Adolescence*, *37*, 373-394.

Barnes, J., & Mayes, M. D. (2012). Epidemiology of systemic sclerosis: incidence, prevalence, survival, risk factors, malignancy, and environmental triggers. *Current opinion in Rheumatology*, *24*, 165-170.

Barsevick, A. M., Whitmer, K., Nail, L. M., Beck, S. L., & Dudley, W. N. (2006). Symptom cluster research: conceptual, design, measurement, and analysis issues. *Journal of Pain and Symptom Management*, *31*, 85-95.

Benrud-Larson, L.M., Haythornwaite, J.A., Heinberg, L.J., Boling, C., Reed, J., White, B., & Wigley, F. M. (2002). The impact of pain and symptoms of depression in scleroderma. *Pain*, 95, 267-275.

Benrud-Larson, L. M., Heinberg, L. J., Boling, C., Reed, J., White, B., Wigley, F. M., &

Haythornthwaite, J. A. (2003). Body image dissatisfaction among women with scleroderma: Extent and relationship to psychosocial function. *Health Psychology*, 22, 130-139.

Bodukam, V., Hays, R. D., Maranian, P., Furst, D. E., Seibold, J. R., Impens, A., ... & Khanna, D. (2010). Association of gastrointestinal involvement and depressive symptoms in patients with systemic sclerosis. *Rheumatology, 50*, 330-334.

Bombardier, C. H., D'Amico, C., & Jordan, J. S. (1990). The relationship of appraisal and coping to chronic illness adjustment. *Behaviour Research and Therapy*, *28*, 297-304.

Bongi, S.M., Del Rosso, A., Mikhaylova, S., Baccini, M., & Cerinic, M.M. (2013). Sexual function in Italian women with systemic sclerosis is affected by disease-related and psychological concerns. *The Journal of Rheumatology*, *40,* 1697-170.

Bonanno, G. A., Papa, A., Lalande, K., Zhang, N., & Noll, J. G. (2005). Grief processing and deliberate grief avoidance: A prospective comparison of bereaved spouses and parents in the United States and the People's Republic of China. *Journal of Consulting and Clinical Psychology*, 73, 86 –98.

Brytek-Matera, A., Rogoza, R., Gramaglia, C., & Zeppegno, P. (2015). Predictors of orthorexic behaviours in patients with eating disorders: a preliminary study. *BMC Psychiatry*, *15*, 252-260.

Carleton, R. N., Collimore, K. C., & Asmundson, G. J. (2007). Social anxiety and fear of negative evaluation: Construct validity of the BFNE-II. *Journal of Anxiety Disorders*, *21*, 131-141.

Carr, T., Harris, D., & James, C. (2000). The Derriford Appearance Scale (DAS59): A new scale to measure individual responses to living with problems of appearance. *British Journal of Health Psychology, 5,* 201–215.

Carr, T., Moss, T., & Harris, D. (2005). The DAS24: A short form of the Derriford Appearance Scale DAS59 to measure individual responses to living with problems of appearance. *British Journal of Health Psychology, 10*, 285–298.

Cash, T. F. (1994). Body-image attitudes: Evaluation, investment, and affect. *Perceptual and Motor Skills, 78*, 1168–1170.

Cash, T. F. (2000). Manual for the Multidimensional Body-Self Relations Questionnaire (3rd rev.). Document available for purchase at the author's Web site at http://www.body-images.com.

Cash, T.F. (2002a). Cognitive behavioral perspectives on body image. In T.F. Cash & T.

Pruzinsky (Eds.), *Body image: A handbook of theory, research, and clinical practice* (pp. 38–46). New York: Guilford Press.

Cash, T.F. (2002b). The management of body image problems. In C.G. Fairburn & K.D. Brownell (Eds.), *Eating disorders and obesity: A comprehensive handbook* (2nd ed., pp. 599–603). New York: Guilford Press.

Cash, T. F. (2004). Body image: Past, present, and future. *Body Image*, *1*, 1-5.

Cash, T. F., & Fleming, E. C. (2002). The impact of body image experiences: development of the body image quality of life inventory. *International Journal of Eating Disorders*, *31*, 455-460.

Cash, T. F., Fleming, E. C., Alindogan, J., Steadman, L., & Whitehead, A. (2002). Beyond body image as a trait: The development and validation of the Body Image States Scale. *Eating Disorders*, *10*, 103-113.

Cash, T. F., Melnyk, S. E., & Hrabosky, J. I. (2004*a*). The assessment of body image investment: An extensive revision of the Appearance Schemas Inventory. *International Journal of Eating Disorders*, *35*, 305-316.

Cash, T. F., Morrow, J. A., losky, J. I., & Perry, A. A. (2004*b*). How has body image changed? A cross-sectional investigation of college women and men from 1983 to 2001. *Journal of Consulting and Clinical Psychology*, *72*, 1081-1089.

Cash, T. F., Phillips, K. A., Santos, M. T., & Hrabosky, J. I. (2004*c*). Measuring "negative body image": validation of the Body Image Disturbance Questionnaire in a nonclinical population. *Body Image*, *1*, 363-372.

Cash, T. F., & Pruzinsky, T. (Eds.) (1990). *Body images: Development, deviance, and change*. New York: Guilford Press.

Cash T.F. & Smolak, L. (2011). In Cash, T. F., & Smolak, L. (Eds.). (2011). *Body image: A handbook of science, practice, and prevention* (2nd ed.). New York, NY: Guilford Press.

Cash, T. F., Winstead, B. W., & Janda, L. H. (1986). The great American shape-up: Body image survey report. *Psychology Today*, *20,* 30–37.

Cella, D., Riley, W., Stone, A., Rothrock, N., Reeve, B., Yount, S., ... & Cook, K. (2010). The Patient-Reported Outcomes Measurement Information System (PROMIS) developed and tested its first wave of adult self-reported health outcome item banks: 2005–2008. *Journal of Clinical Epidemiology*, *63*, 1179-1194.

Celeux, G., & Soromenho, G. (1996). An entropy criterion for assessing the number of

clusters in a mixture model. *Journal of Classification*, *13*, 195-212.

Chifflot, H., Fautrel, B., Sordet, C., Chatelus, E., & Sibilia, J. (2008). Incidence and prevalence of systemic sclerosis: a systematic literature review. *Seminars in Arthritis and Rheumatism, 37,* 223-235.

Clatworthy, J., Hankins, M., Buick, D., Weinman, J., & Horne, R. (2007). Cluster analysis in illness perception research: A Monte Carlo study to identify the most appropriate method. *Psychology and Health*, *22*, 123-142.

Clements, P. J., Lachenbruch, P. A., Ng, S. C., Simmons, M., Sterz, M., & Furst, D. E. (1990). Skin score: a semiquantitative measure of cutaneous involvement that improves prediction of prognosis in systemic sclerosis. *Arthritis & Rheumatism*, *33*, 1256-1263.

Clements, P. H., Lachenbruch, P., Siebold, J., White, B., Weiner, S., Martin, R., ... & Collier, D. (1995). Inter and intraobserver variability of total skin thickness score (modified Rodnan TSS) in systemic sclerosis. *The Journal of Rheumatology*, *22* 1281-1285.

Cochrane, V. M., & Slade, P. (1999). Appraisal and coping in adults with cleft lip: Associations with well-being and social anxiety. *British Journal of Medical Psychology, 72*, 485– 503.

Cohen, J., Cohen, P., West, S. G., & Aiken, L. S. (2003). *Applied multiple regression/correlation analysis for the behavioral sciences* (3rd ed.). Mahwah, NJ: Erlbaum.

Coles, M. E., Pietrefesa, A. S., Schofield, C. A., & Cook, L. M. (2008). Predicting changes in obsessive compulsive symptoms over a six-month follow-up: A prospective test of cognitive models of obsessive compulsive disorder. *Cognitive Therapy and Research*, *32*, 657-675.

Czirják, L., Foeldvari, I., & Müller-Ladner, U. (2008). Skin involvement in systemic sclerosis. *Rheumatology*, *47*, v44-v45.

Crosby, R. D., Kolotkin, R. L., & Williams, G. R. (2003). Defining clinically meaningful change in health-related quality of life. *Journal of Clinical Epidemiology*, *56*, 395-407.

de Cock, T. P., & Shevlin, M. (2014). Parental Bonding: A Typology of the Parent–Child Relationship in a Population Sample. *SAGE Open*, *4*, 1-13.

Del Rosso, A., Boldrini, M., D'Agostino, D., Placidi, G. P., Scarpato, A., Pignone, A., ... Matucci-Cerinic, M. (2004). Health-related quality of life in systemic sclerosis as

measured by the Short Form 36: Relationship with clinical and biologic markers. *Arthritis and Rheumatism, 51*, 475-481.

Dittmar, H., Halliwell, E., & Ive, S. (2006). Does Barbie make girls want to be thin? The effect of experimental exposure to images of dolls on the body image of 5-to 8-year-old girls. *Developmental Psychology*, *42*, 283-292.

Ennis, H., Herrick, A. L., Cassidy, C., Griffiths, C. E., & Richards, H. L. (2012). A pilot study of body image dissatisfaction and the psychological impact of systemic sclerosis-related telangiectases. *Clinical and Experimental Rheumatology*, *31*, 12-17.

Fallon, A. (1990). Culture in the mirror: Sociocultural determinants of body image. In T.F. Cash & T. Pruzinsky (Eds.), *Body images: Development, deviance, and change* , (pp. 80-109). New York: Guilford Press.

Fisher, S. (1986). *Development and structure of the body image*, Hillsdale, NJ: Erlbaum.

Fox, R. S., Mills, S. D., Gholizadeh, **S.,** Malcarne, V. L., Kwakkenbos, L., Jewett, L. R., Roesch, S. C., Gottesman, K., Thombs, B. D., & the SPIN Investigators. (in press). Psychometric evaluation of the Brief Fear of Negative Evaluation Scale in patients with systemic sclerosis: Results from the Scleroderma Patient-Centered Intervention Network study. *Arthritis Care and Research*.

Fries, J. F., Spitz, P., Kraines, R. G., & Holman, H. R. (1980). Measurement of patient outcome in arthritis. *Arthritis & Rheumatism*, *23*, 137-145.

Gabrielli, A., Avvedimento, E. V., & Krieg, T. (2009). Scleroderma. *New England Journal of Medicine*, *360*, 1989-2003.

Gholizadeh, S., Drizin, J., Hansdottir, I., Weisman, M.H., Clements, P.J., Furst, D.E., & Malcarne, V. L. (2015*a*, April). Patient Attributions of Causality for a Disease of Unknown Etiology. Poster presented at the annual meeting of the Society of Behavioral Medicine, San Antonio, TX.

Gholizadeh, S., Fox, R. S., Mills, S. D., Jewett, L. R., Thombs, B. D., & Malcarne, V. L. (2017*a*). Coping with the disfigurement of scleroderma: Facial, skin, and hand changes. In F. M. Wigley (Ed.), *Scleroderma: From pathogenesis to comprehensive management*, 2nd ed. New York: Springer.

Gholizadeh, S., Mills, S.D., Fox, R.S., Jewett, L.R., Carrier, M.E., Kwakkenbos, L., Gottesman, K., Roesch, S.C., Thombs, B.D., Malcarne, V.L., and SPIN Investigators. (2015*b*, November). A Psychometric Analysis of the Social Interaction Anxiety Scale (SIAS-6) in Systemic Sclerosis: Preliminary results from the Scleroderma Patient-centered Intervention Network (SPIN) Cohort.

Paper Presented at the Annual Meeting of the American College of Rheumatology/Association of Rheumatology Health Professionals, San Francisco, CA.

Goffman E. (1963) *Stigma: Notes on the management of spoiled identity*. Prentice-Hall, Englewood Cliffs, New Jersey.

Gorlova, O., Martin, J. E., Rueda, B., Koeleman, B. P., Ying, J., Teruel, M., ... & Alizadeh, B. Z. (2011). Identification of novel genetic markers associated with clinical phenotypes of systemic sclerosis through a genome-wide association strategy. *PLoS Genet*, *7*, e1002178.

Grogan, S. (2006). Body image and health: Contemporary perspectives. *Journal of Health Psychology*, *11*, 523-530.

Grandfield, T. A., Thompson, A. R., & Turpin, G. (2005). An attitudinal study of responses to a range of dermatological conditions using the implicit association test. *Journal of Health Psychology*, *10*, 821-829.

Gumuchian, S. T., Delisle, V. C., Peláez, S., Malcarne, V. L., El-Baalbaki, G., Kwakkenbos, L., ... & Scleroderma Support Group Project Advisory Team. (2018). Reasons for Not Participating in Scleroderma Patient Support Groups: A Cross-Sectional Study. *Arthritis care & research*, *70*, 275-283.

Hagenaars, J. A., & McCutcheon, A. L. (Eds.). (2002). *Applied latent class analysis*. Cambridge University Press.

Hart, T. A., Flora, D. B., Palyo, S. A., Fresco, D. M., Holle, C., & Heimberg, R. G. (2008). Development and examination of the social appearance anxiety scale. *Assessment*, *15*, 48-59.

Heatherton, T. F., & Polivy, J. (1991). Development and validation of a scale for measuring state self- esteem. *Journal of Personality and Social Psychology, 60,* 895-910.

Heinberg, L. J., Kudel, I., White, B., Kwan, A., Medley, K., Wigley, F., & Haythornthwaite, J. (2007). Assessing body image in patients with systemic sclerosis (scleroderma): validation of the adapted Satisfaction with Appearance Scale. *Body Image*, *4*, 79-86.

Helmick, C. G., Felson, D. T., Lawrence, R. C., Gabriel, S., Hirsch, R., Kwoh, C. K., ... & Pillemer, S. R. (2008). Estimates of the prevalence of arthritis and other rheumatic conditions in the United States: Part I. *Arthritis & Rheumatism*, *58*, 15-25.

Hildebrandt, T., Schlundt, D., Langenbucher, J., & Chung, T. (2006). Presence of muscle dysmorphia symptomology among male weightlifters. *Comprehensive Psychiatry*, *47*, 127-135.

Hill, A. L., Degnan, K. A., Calkins, S. D., & Keane, S. P. (2006). Profiles of externalizing behavior problems for boys and girls across preschool: the roles of emotion regulation and inattention. *Developmental Psychology*, *42*, 913-928.

Hipp, J. R., & Bauer, D. J. (2006). Local solutions in the estimation of growth mixture models. *Psychological Methods, 11*, 36-53.

Hinchcliff, M., Beaumont, J. L., Thavarajah, K., Varga, J., Chung, A., Podlusky, S., ... & Cella, D. (2011). Validity of two new patient-reported outcome measures in systemic sclerosis: Patient-reported outcomes measurement information system 29-item health profile and functional assessment of chronic illness therapy– dyspnea short form. *Arthritis Care & Research*, *63*, 1620-1628.

Hrabosky, J. I., Cash, T. F., Veale, D., Neziroglu, F., Soll, E. A., Garner, D. M., ... & Phillips, K. A. (2009). Multidimensional body image comparisons among patients with eating disorders, body dysmorphic disorder, and clinical controls: A multisite study. *Body Image*, *6*, 155-163.

Hobro, N., Weinman, J., & Hankins, M. (2004). Using the self-regulatory model to cluster chronic pain patients: the first step towards identifying relevant treatments? *Pain*, *108*, 276-283.

Hudson, M., Thombs, B. D., Steele, R., Watterson, R., Taillefer, S., Baron, M., & the Canadian Scleroderma Research Group. (2008). Clinical correlates of quality of life in systemic sclerosis measured with the World Health Organization Disability Assessment Schedule II. *Arthritis and Rheumatism, 59, 279*-284.

Hudson, M., Steele, R., Canadian Scleroderma Research Group, & Baron, M. (2007). Update on indices of disease activity in systemic sclerosis. *Seminars in Arthritis and Rheumatism, 37*, 93-98.

Hyphantis, T. N., Tsifetaki, N., Siafaka, V., Voulgari, P. V., Pappa, C., Bai, M., ... Drosos, A. A. (2007). The impact of psychological functioning upon systemic sclerosis patients' quality of life. *Seminars in Arthritis and Rheumatism, 37*, 81-92.

Jacobs, M. J., Roesch, S., Wonderlich, S. A., Crosby, R., Thornton, L., Wilfley, D. E., ... & Halmi, K. A. (2009). Anorexia nervosa trios: behavioral profiles of individuals with anorexia nervosa and their parents. *Psychological Medicine*, *39*, 451-461.

Jewett, L.R., Hudson M., Haythornthwaite, J.A., Heinberg, L., Wigley, F., et al. (2010) Development and validation of the Brief-Satisfaction with Appearance Scale for systemic sclerosis. *Arthritis Care and Research, 62,* 1779–86.

Jewett, L. R., Hudson, M., Malcarne, V. L., Baron, M., Thombs, B. D., & Canadian Scleroderma Research Group. (2012*a*). Sociodemographic and disease correlates of body image distress among patients with systemic sclerosis. *PloS One*, *7*, e33281.

Jewett, L. R., Kwakkenbos, L., Carrier, M. E., Malcarne, V. L., Bartlett, S. J., Furst, D. E., ... & Williamson, H. (2016*a*). Examination of the association of sex and race/ethnicity with appearance concerns: A Scleroderma Patient-centered Intervention Network (SPIN) Cohort study. *Clinical and Experimental Rheumatology*, *34,* (Suppl. 100): S92-9.

Jewett, L. R., Malcarne, V. L., Kwakkenbos, L., Harcourt, D., Rumsey, N., Körner, A., ... & Heinberg, L. (2016*b*). Development and Validation of the Body Concealment Scale for Scleroderma. *Arthritis Care & Research*, *68*, 1158-1165.

Jewett, L. R., Kwakkenbos, L., Carrier, M. E., Malcarne, V. L., Harcourt, D., Rumsey, N., ... & Gholizadeh, S. (2017). Validation of the Body Concealment Scale for Scleroderma (BCSS): Replication in the Scleroderma Patient-centered Intervention Network (SPIN) cohort. *Body Image*, *20*, 99-106.

Jewett, L. R., Razykov, I., Hudson, M., Baron, M., Thombs, B. D., & Canadian Scleroderma Research Group. (2012*b*). Prevalence of current, 12-month and lifetime major depressive disorder among patients with systemic sclerosis. *Rheumatology, 52,* 669-675.

Joachim, G., & Acorn, S. (2003). Life with a rare chronic disease: the scleroderma experience. *Journal of Advanced Nursing*, *42*, 598-606.

Jolly, M. (2011). Body Image Issues in Rheumatology In Cash, T. F., & Smolak, L. (Eds.). (2011). *Body image: A handbook of science, practice, and prevention* (2nd ed., pp. 350-357). New York, NY: Guilford Press.

Jung, T., & Wickrama, K. A. S. (2008). An introduction to latent class growth analysis and growth mixture modeling. *Social and Personality Psychology Compass*, *2*, 302-317.

Khanna, D., Clements, P. J., Furst, D. E., Korn, J. H., Ellman, M., Rothfield, N., ... & Steen, V. D. (2009). Recombinant human relaxin in the treatment of systemic sclerosis with diffuse cutaneous involvement: A randomized, double-blind, placebo-controlled trial. *Arthritis & Rheumatism*, *60*, 1102-1111.

Khanna, D., Furst, D. E., Hays, R. D., Park, G. S., Wong, W. K., Seibold, J. R., ... & Barr, W. (2006). Minimally important difference in diffuse systemic sclerosis: results from the D-penicillamine study. *Annals of the Rheumatic Diseases*, *65*, 1325-1329.

Khanna, D., Maranian, P., Rothrock, N., Cella, D., Gershon, R., Khanna, P. P., ... & Hays, R. D. (2012). Feasibility and construct validity of PROMIS and "legacy" instruments in an academic scleroderma clinic. *Value in Health*, *15*, 128-134.

Knafo, R., Haythornthwaite, J. A., Heinberg, L., Wigley, F. M., & Thombs, B. D. (2011). The association of body image dissatisfaction and pain with reduced sexual function in women with systemic sclerosis. *Rheumatology, 50*, 1125-1130.

Kongsted, A., & Nielsen, A. M. (2016). An introduction to Latent Class Analysis in health research. *Journal of Physiotherapy*. In Press, accessed from http://dx.doi.org/10.1016/j.jphys.2016.05.018.

Kwakkenbos, L., Delisle, V. C., Fox, R. S., Gholizadeh, S., Jewett, L. R., Levis, B., ... & Thombs, B. D. (2015*a*). Psychosocial aspects of scleroderma. *Rheumatic Disease Clinics of North America*, *41*, 519-528.

Kwakkenbos, L., Jewett, L. R., Baron, M., Bartlett, S. J., Furst, D., Gottesman, K., ... & Poiraudeau, S. (2013). 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. *British Medical Journal Open*, *3*, e003563.

Kwakkenbos, L., Thombs, B. D., Bartlett, S., Hudson, M., Mouthon, L., Malcarne, V., ... & Khanna, D. (2015*b*). SAT0465 Construct Validity of the PROMIS-29 in Systemic Sclerosis: Preliminary Results from the Scleroderma Patient-Centered Intervention Network (Spin) Cohort. *Annals of the Rheumatic Diseases*, *74*(Suppl 2), 829-829.

Kwakkenbos, L., van Lankveld, W. G., Vonk, M. C., Becker, E. S., van den Hoogen, F. H., & van den Ende, C. H. (2012). Disease-related and psychosocial factors associated with depressive symptoms in patients with systemic sclerosis, including fear of progression and appearance self-esteem. *Journal of Psychosomatic Research*, *72*, 199-204.

Kroenke, K., Spitzer, R. L., & Williams, J. B. (2001). The Phq-9. *Journal of General Internal Medicine*, *16*, 606-613.

Lanza S., Flaherty B.P., Collins L.M. (2003). Latent class and latent transition analysis. In: Schinka JA, Velicer WF (eds, pp. 663-685). *Handbook of Psychology, Vol 2, Research Methods in Psychology*. Hoboken, NJ: Wiley.

Lanza, S. T., Savage, J. S., & Birch, L. L. (2010). Identification and prediction of latent classes of weight-loss strategies among women. *Obesity*, *18*, 833-840.

Lanza, S. T., Tan, X., & Bray, B. C. (2013). Latent class analysis with distal outcomes: A flexible model-based approach. *Structural Equation Modeling, 20*, 1–26.

Lawrence, J. W., Heinberg, L. J., Roca, R., Munster, A., Spence, R., & Fauerbach, J. A. (1998). Development and validation of the Satisfaction with Appearance Scale: Assessing body image among burn-injured patients. *Psychological Assessment, 10,* 64–70.

Lazaraus, RS, Folkman, S. (1984). *Stress, appraisal, and coping*. Springer Publishing Company: New York.

Le Blanc, A. L., Bruce, L. C., Heimberg, R. G., Hope, D. A., Blanco, C., Schneier, F. R., & Liebowitz, M. R. (2014). Evaluation of the psychometric properties of two short forms of the social interaction anxiety scale and the social phobia scale. *Assessment*, *21*, 312-323.

Leary, M. R. (1983). A brief version of the Fear of Negative Evaluation Scale. *Personality and Social Psychology Bulletin*, *9*, 371-375.

Legendre, C., Allanore, Y., Ferrand, I., & Kahan, A. (2005). Evaluation of depression and anxiety in patients with systemic sclerosis. *Joint Bone Spine, 72,* 408-411.

Leite, C. C., & Maia, Â. C. (2013). Symptoms of disease and psychological adaptation in Brazilian scleroderma patients. *Revista Brasileira de Reumatologia*, *53*, 405-411.

Leroy, E. C., Black, C., Fleischmajer, R., Jablonska, S., Krieg, T., Medsger, T. A., ... Wollheim, F. (1988). Scleroderma (systemic sclerosis): Classification, subsets and pathogenesis. *Journal of Rheumatology, 15*, 202-205.

Levinson, C. A., & Rodebaugh, T. L. (2011). Validation of the Social Appearance Anxiety Scale: Factor, convergent, and divergent validity. *Assessment*, *18*, 350-356.

Levis, B., Hudson, M., Knafo, R., Baron, M., Nielson, W. R., Hill, M., & Thombs, B. D. (2012*a*). Rates and correlates of sexual activity and impairment among women with systemic sclerosis. *Arthritis Care & Research*, *64*, 340-350.

Levis, B., Burri, A., Hudson, M., Baron, M., Thombs, B. D., & Canadian Scleroderma Research Group. (2012*b*). Sexual activity and impairment in women with systemic sclerosis compared to women from a general population sample. *PloS one*, *7*, e52129.

Liu, H., Cella, D., Gershon, R., Shen, J., Morales, L. S., Riley, W., & Hays, R. D. (2010). Representativeness of the patient-reported outcomes measurement information system internet panel. *Journal of clinical epidemiology*, *63*(11), 1169-1178.

Lo, Y., Mendell, N., & Rubin, D. (2001). Testing the number of components in a normal mixture. *Biometrika*, *88*, 767-778.

Perrig-Chiello, P., Hutchison, S., & Morselli, D. (2015). Patterns of psychological adaptation to divorce after a long-term marriage. *Journal of Social and Personal Relationships*, *32*, 386-405.

Macgregor, F. (1979). *After plastic surgery: Adaptation and adjustment*. New York: Praeger.

Malcarne, V. L., Fox, R. S., Mills, S. D., & Gholizadeh, S. (2013). Psychosocial aspects of systemic sclerosis. *Current Opinion in Rheumatology, 25,* 707-713.

Malcarne, V. L., Handsdottir, I., Greenbergs, H.L., Clements, P. J., & Weisman, M. H. (1999). Appearance self-esteem in systemic sclerosis. *Cognitive Therapy and Research*, 23, 197-208.

Malcarne, V. L., Handsdottir, I., McKinney, A., Upchurch, R., Greenbergs, H. L., Henstorf, G. H., ... Weisman, M. H. (2007). Medical Signs and Symptoms Associated with Disability, Pain, and Psychosocial Adjustment in Systemic Sclerosis. *Journal of Rheumatology, 34*, 359-367.

Makol, A., Reilly, M. J., & Rosenman, K. D. (2011). Prevalence of connective tissue disease in silicosis (1985–2006)—a report from the state of Michigan surveillance system for silicosis. *American Journal of Industrial Medicine*, *54*, 55-262.

Mattick, R. P., & Clarke, J. C. (1998). Development and validation of measures of social phobia scrutiny fear and social interaction anxiety. *Behaviour Research and Therapy*, *36*, 455-470.

Mayes, M. D., Lacey, J. V., Beebe-Dimmer, J., Gillespie, B. W., Cooper, B., Laing, T. J., & Schottenfeld, D. (2003). Prevalence, incidence, survival, and disease characteristics of systemic sclerosis in a large US population. *Arthritis & Rheumatism*, *48*, 2246-2255.

McCormic, Z. D., Khuder, S. S., Aryal, B. K., Ames, A. L., & Khuder, S. A. (2010). Occupational silica exposure as a risk factor for scleroderma: a meta-analysis. *International archives of occupational and Environmental Health*, *83*, 763-769.

McLachlan, G., & Peel, D. (2000). *Finite Mixture Models.* New York: John Wiley & Sons.

Medsger, T. A., Bombardieri, S., Czirjak, L., Scorza, R., Rossa, A. D., & Bencivelli, W. (2003). Assessment of disease severity and prognosis. *Clinical and Experimental Rheumatology*, *21*(3; SUPP/29), S42-S46.

Merkel, P. A., Clements, P. J., Reveille, J. D., Suarez-Almazor, M. E., Valentini, G., & Furst, D. E. (2003). Current status of outcome measure development for clinical trials in systemic sclerosis. Report from OMERACT 6. *The Journal of Rheumatology*, *30*, 1630-1647.

Merz, E. L., & Roesch, S. C. (2011). A latent profile analysis of the Five Factor Model of personality: modeling trait interactions. *Personality and Individual Differences*, *51*, 915-919.

Milligan, G. W., & Cooper, M. C. (1987). Methodology review: Clustering methods. *Applied Psychological Measurement*, *11*, 329-354.

Mills, S. D., Gholizadeh, S., Fox, R. S., Jewett, L. R., Carrier, M., Kwakkenbos, L., Gottesman, K., Roesch, S. C., Thombs, B. D., Malcarne, V. L., & SPIN Investigators. *Psychometric Properties of the Social Appearance Anxiety Scale in Patients with Limited and Diffuse Systemic Sclerosis: Analysis from the Scleroderma Patient-centered Intervention Network Cohort.* 2015*a*; Poster presented at the annual meeting of the American College of Rheumatology/ Association of Rheumatology Health Professionals, San Francisco, CA.

Mills, S. D., Fox, R. S., Merz, E. L., Clements, P. J., Kafaja, S., Malcarne, V. L., ... & Khanna, D. (2015*b*). Evaluation of the Satisfaction with Appearance Scale and Its Short Form in Systemic Sclerosis: Analysis from the UCLA Scleroderma Quality of Life Study. *The Journal of Rheumatology*, *42*, 1624-1630.

Morin, A. J., Morizot, J., Boudrias, J. S., & Madore, I. (2010). A multifoci person-centered perspective on workplace affective commitment: A latent profile factor mixture analysis. *Organizational Research Methods, 14,* 58-90.

Moss, T. P. (2005). The relationships between objective and subjective ratings of disfigurement severity, and psychological adjustment. *Body Image*, *2*, 151-159.

Müller, H., Rehberger, P., Günther, C. & Schmitt, J. (2012). Determinants of disability, quality of life and depression in dermatological patients with systemic scleroderma. *British Journal of Dermatology, 166*, 343-353.

Muth, J. L., & Cash, T. F. (1997). Body-Image Attitudes: What Difference Does Gender Make? *Journal of Applied Social Psychology*, *27*, 1438-1452.

Muthén, B. (2006). Should substance use disorders be considered as categorical or dimensional? *Addiction, 101*, 6-16.

Muthén, L. K., & Muthén, B. O. (1998-2012). *Mplus User's Guide* (Seventh Edition ed.). Los Angeles, CA: Muthén & Muthén.

Newell, R., & Marks, I. (2000). Phobic nature of social difficulty in facially disfigured people. *The British Journal of Psychiatry*, *176*, 177-181.

Norton, S., Hughes, L. D., Chilcot, J., Sacker, A., van Os, S., Young, A., & Done, J. (2014). Negative and positive illness representations of rheumatoid arthritis: a latent profile analysis. *Journal of Behavioral Medicine*, *37*, 524-532.

Nusbaum, J. S., Gordon, J. K., & Steen, V. D. (2015). African American race associated with body image dissatisfaction among patients with systemic sclerosis. *Clinical and Experimental Rheumatology*, *34*, 70-73.

Nylund, K. L., Asparouhov, T., & Muthén, B. O. (2007). Deciding on the number of classes in latent class analysis and growth mixture modeling: A Monte Carlo simulation study. *Structural Equation Modeling*, *14*, 535-569.

Park, C. L. (2010). Making sense of the meaning literature: an integrative review of meaning making and its effects on adjustment to stressful life events. *Psychological Bulletin*, *136*, 257-301.

Park, C. L., & Folkman, S. (1997). Meaning in the context of stress and coping. *Review of General Psychology, 1*, 115–144.

Peters, L., Sunderland, M., Andrews, G., Rapee, R. M., & Mattick, R. P. (2012). Development of a short form Social Interaction Anxiety (SIAS) and Social Phobia Scale (SPS) using nonparametric item response theory: The SIAS-6 and the SPS-6. *Psychological Assessment*, 24, 66–76.

Poormoghim, H., Lucas, M., Fertig, N., & Medsger Jr, T. A. (2000). Systemic sclerosis sine scleroderma: demographic, clinical, and serologic features and survival in forty-eight patients. *Arthritis & Rheumatology*, *43*, 444-451.

Pope, J. (2011). Measures of systemic sclerosis (scleroderma): Health Assessment Questionnaire (HAQ) and Scleroderma HAQ (SHAQ), Physician-and Patient-Rated Global Assessments, Symptom Burden Index (SBI), University of California, Los Angeles, Scleroderma Clinical Trials Consortium Gastrointestinal Scale (UCLA SCTC GIT) 2.0, Baseline Dyspnea Index (BDI) and Transition Dyspnea Index (TDI)(Mahler's Index), Cambridge Pulmonary Hypertension Outcome Review (CAMPHOR), and Raynaud's Condition Score (RCS). *Arthritis Care & Research*, *63*, S98-S111

Pruzinsky, T. (1992). Social and psychological effects of major craniofacial deformity. *The Cleft palate-craniofacial journal*, *29*, 578-584.

Puckett, J. A., Levitt, H. M., Horne, S. G., & Hayes-Skelton, S. A. (2015). Internalized heterosexism and psychological distress: The mediating roles of self-criticism and community connectedness. *Psychology of Sexual Orientation and Gender Diversity*, *2*, 426-435.

Ramaswamy, V., Desarbo, W. S., Reibstein, D. J., & Robinson, W. T. (1993). An empirical pooling approach for estimating marketing mix elasticities with PIMS data. *Marketing Science*, *12*, 103-124.

Razykov, I., Hudson, M., Baron, M., & Thombs, B. D. (2013). Utility of the Patient Health Questionnaire-9 to Assess Suicide Risk in Patients with Systemic Sclerosis. *Arthritis Care & Research*, *65*, 753-758.

Richards, H. L., Herrick, A. L., Griffin, K., Gwilliam, P. D., Loukes, J., & Fortune, D. G. (2003). Systemic sclerosis: patients' perceptions of their condition. *Arthritis and Rheumatism, 49*, 689-696.

Richards, H. L., Herrick, A. L., Griffin, K., Gwilliam, P. D. H., & Fortune, D. G. (2004). Psychological adjustment to systemic sclerosis—exploring the association of disease factors, functional ability, body related attitudes and fear of negative evaluation. *Psychology, Health & Medicine*, *9*, 29-39.

Robinson E. (1997). Psychological research on visible differences in adults. In: R. Lansdown, N. Rumsey, E. Bradbury, T. Carr, J. Partridg, eds. *Visibly different: Coping with disfigurement* (pp. 102-111). London: Butterworth.

Roesch, S. C., Villodas, M., & Villodas, F. (2010). Latent class/profile analysis in maltreatment research: A commentary on Nooner et al., Pears et al., and looking beyond. *Child Abuse & Neglect*, *34*, 155-160.

Rosen, J. C., Srebnik, D., Saltzberg, E., & Wendt, S. (1991). Development of a body image avoidance questionnaire. *Psychological Assessment: A Journal of Consulting and Clinical Psychology*, *3*, 32-37.

Rumsey, N. (2002*a*). Body image & congenital conditions with visible differences. In T. F. Cash & T. Pruzinsky (Eds.), *Body image: A handbook of theory, research, and clinical practice* (pp. 226–233). New York: Guilford.

Rumsey, N. (2002*b*). Optimizing body image in disfiguring congenital conditions. In T. F. Cash & T. Pruzinsky (Eds.), *Body image: A handbook of theory, research, and clinical practice* (pp. 431–439). New York: Guilford.

Rumsey, N., Clarke, A., & Musa, M. (2002). Altered body image: the psychosocial needs of patients. *British Journal of Community Nursing*, *7*, 563-566.

Rumsey, N., & Harcourt, D. (2004). Body image and disfigurement: issues and interventions. *Body Image*, *1*, 83-97.

Sandusky, S. B., McGuire, L., Smith, M. T., Wigley, F. M., & Haythornthwaite, J. A. (2009). Fatigue: an overlooked determinant of physical function in scleroderma. *Rheumatology*, *48*, 165-169.

Schalet, B. D., Pilkonis, P. A., Yu, L., Dodds, N., Johnston, K. L., Yount, S., ... & Cella, D. (2016). Clinical validity of PROMIS Depression, Anxiety, and Anger across diverse clinical samples. *Journal of Clinical Epidemiology*, *73*, 119-127.

Schieir, O., Thombs, B. D., Hudson, M., Boivin, J. F., Steele, R., Bernatsky, S., ...Baron, M., & the Canadian Scleroderma Research Group. (2010). Prevalence, severity, and clinical correlates of pain in patients with systemic sclerosis. *Arthritis Care and Research, 62,* 409-417.

Schouffoer, A. A., Van Der Marel, J., Ter Kuile, M. M., Weijenborg, P. T. M., Voskuyl, A., Vliet Vlieland, C. S., ...Vliet Vlieland, T. P. M. (2009). Impaired sexual function in women with systemic sclerosis: A cross-sectional study. *Arthritis and Rheumatism, 61*, 1601-1608.

Schumacker, R. E., & Lomax, R. G. (2004). *A beginner's guide to structural equation modeling*. 2nd ed. Mahwah, NJ: Erlbaum.

Schwartz, M. B., & Brownell, K. D. (2004). Obesity and body image. *Body Image*, *1*, 43-56.

Schwarz, G. (1978). Estimating the dimensions of a model. *Annals of Statistics*, *6*, 461-464.

Seibold, J. R. (2005). Scleroderma. *Kelley's textbook of rheumatology. 7th ed. Philadelphia: Elsevier*, 1279-308.

Sharif, R., Mayes, M. D., Nicassio, P. M., Gonzalez, E. B., Draeger, H., McNearney, T. A. ... Assassi, S. for the GENISOS study group (2011). Determinants of work disability in patients with systemic sclerosis: A longitudinal study of the GENISOS Cohort. Seminars in Arthritis and *Rheumatism, 41*, 38-47.

Sirois, F. M., & Gick, M. L. (2016). An appraisal-based coping model of attachment and adjustment to arthritis. *Journal of Health Psychology*, *21*, 821-831.

Speece, D. L. (1994). Cluster analysis in perspective. *Exceptionality*, *5*, 31-44.

Stanton, A. L., & Snider, P. R. (1993). Coping with a breast cancer diagnosis: A prospective study. *Health Psychology*, *12*, 16-23.

Steen, V. D., & Medsger, T. A. (1997). The value of the health assessment questionnaire and special patient-generated scales to demonstrate change in systemic sclerosis patients over time. *Arthritis & Rheumatism*, *40*, 1984-1991.

Steen, V. D., Powell, D. L., & Medsger, T. A. (1988). Clinical correlations and prognosis based on serum autoantibodies in patients with systemic sclerosis. *Arthritis & Rheumatism*, *31*, 196-203.

Sullivan, P. F., Smith, W., & Buchwald, D. (2002). Latent class analysis of symptoms associated with chronic fatigue syndrome and fibromyalgia. *Psychological Medicine*, *32*, 881-888.

Szymanski, M. L., & Cash, T. F. (1995). Body-image disturbances and self-discrepancy theory: Expansion of the Body-Image Ideals Questionnaire. *Journal of Social and Clinical Psychology*, *14*, 134-146.

Taleporos, G., & McCabe, M. P. (2001). The impact of physical disability on body esteem. *Sexuality and Disability*, *19*, 293-308.

Tein, J. Y., Coxe, S., & Cham, H. (2013). Statistical power to detect the correct number of classes in latent profile analysis. *Structural Equation Modeling: A Multidisciplinary Journal*, *20*, 640-657.

Thombs, B. D., Bassel, M., McGuire, L., Smith, M. T., Hudson, M., & Haythornthwaite, J. A. (2008*a*). A systematic comparison of fatigue levels in systemic sclerosis with general population, cancer and rheumatic disease samples. *Rheumatology, 47*, 1559-1563.

Thombs, B. D., Hudson, M., Taillefer, S. S., & Baron, M. (2008). Prevalence and clinical correlates of symptoms of depression in patients with systemic sclerosis. *Arthritis Care & Research*, *59*, 504-509.

Thombs B.D., Jewett L.R., Kwakkenbos L., Hudson M., Baron M., & the Canadian Scleroderma Research Group (In Press). Major depression diagnoses are often transient among patients with systemic sclerosis: Baseline and 1-month follow-up. *Arthritis Care and Research.*

Thombs, B. D., van Lankveld, W., Bassel, M., Baron, M., Buzza, R., Haslam, S., ... Worron-Sauve, M., (2010). Psychological health and well-being in systemic sclerosis: State of the science and consensus research agenda. *Arthritis Care and Research, 62,* 1181-1189.

Thompson, J. K. (2004). The (mis) measurement of body image: ten strategies to improve assessment for applied and research purposes. *Body Image*, *1*, 7-14.

Thompson, J.K. (2011). Body Image Issues in Dermatology In Cash, T. F., & Smolak, L. (Eds.). (2011). *Body image: A handbook of science, practice, and prevention* (2nd ed., pp. 323-342). New York, NY: Guilford Press.

Thompson, J. K., & Heinberg, L. J. (1999). The media's influence on body image disturbance and eating disorders: We've reviled them, now can we rehabilitate them?. *Journal of Social Issues*, *55*, 339-353.

Thompson, J. K., Heinberg, L. J., Altabe, M., & Tantleff-Dunn, S. (1999). Exacting beauty: Theory, assessment, and treatment of body image disturbance. Washington, DC: American Psychological Association.

Thompson, J. K., Roehrig, M., Cafri, G., & Heinberg, L. J. (2005). *Assessment of body image disturbance*. In J.E. Mitchell & C.B. Peterson (Eds.), Assessment of eating disorders, (pp. 175-202). New York: Guilford Press.

Tiggemann, M. (2011). Sociocultural perspectives on human appearance and body image. In T. F. Cash & L. Smolak (Eds.), *Body image: A handbook of science, practice, and prevention* (2nd ed., pp. 12–19). New York: Guilford Press.

Titchener, K., & Wong, Q. J. (2015). A weighty issue: Explaining the association between body mass index and appearance-based social anxiety. *Eating Behaviors*, *16*, 13-16.

Van den Hoogen, F., Khanna, D., Fransen, J., Johnson, S. R., Baron, M., Tyndall, A., ... & Riemekasten, G. (2013). 2013 classification criteria for systemic sclerosis: an American College of Rheumatology/European League against Rheumatism collaborative initiative. *Arthritis & Rheumatism*, *65*, 2737-2747.

Van Lankveld, W. G. J. M., Vonk, M. C., Teunissen, H., & van den Hoogen, F. H. J. (2007). Appearance self-esteem in systemic sclerosis—subjective experience of skin deformity and its relationship with physician-assessed skin involvement, disease status and psychological variables. *Rheumatology*, *46*, 872-876.

Vermunt, J. K. (2010). Latent class modeling with covariates: Two improved three-step approaches. *Political Analysis*, 18, 450–469.

Wigley, F. M., & Hummers, L. K. (2003). Clinical features of systemic sclerosis. *Rheumatology*, *3*, 1463-80.

White, A. (1982). Psychiatric study of patients and severe burn injuries. *British Medical Journal*, 284, 465–467.

Wilson, A., Hickie, I., Wilson, A., Hickie, I., Hadzi-Pavlovic, D., Wakefield, D., ... & Hinds, G. (2001). What is chronic fatigue syndrome? Heterogeneity within an international multicentre study. *Australian and New Zealand Journal of Psychiatry*, *35*, 520-527.

Yacoub, Y. I., Amine, B., Bensabbah, R., & Hajjaj-Hassouni, N. (2012). Assessment of fatigue and its relationship with disease-related parameters in patients with systemic sclerosis. *Clinical Rheumatology, 31*, 655-660.
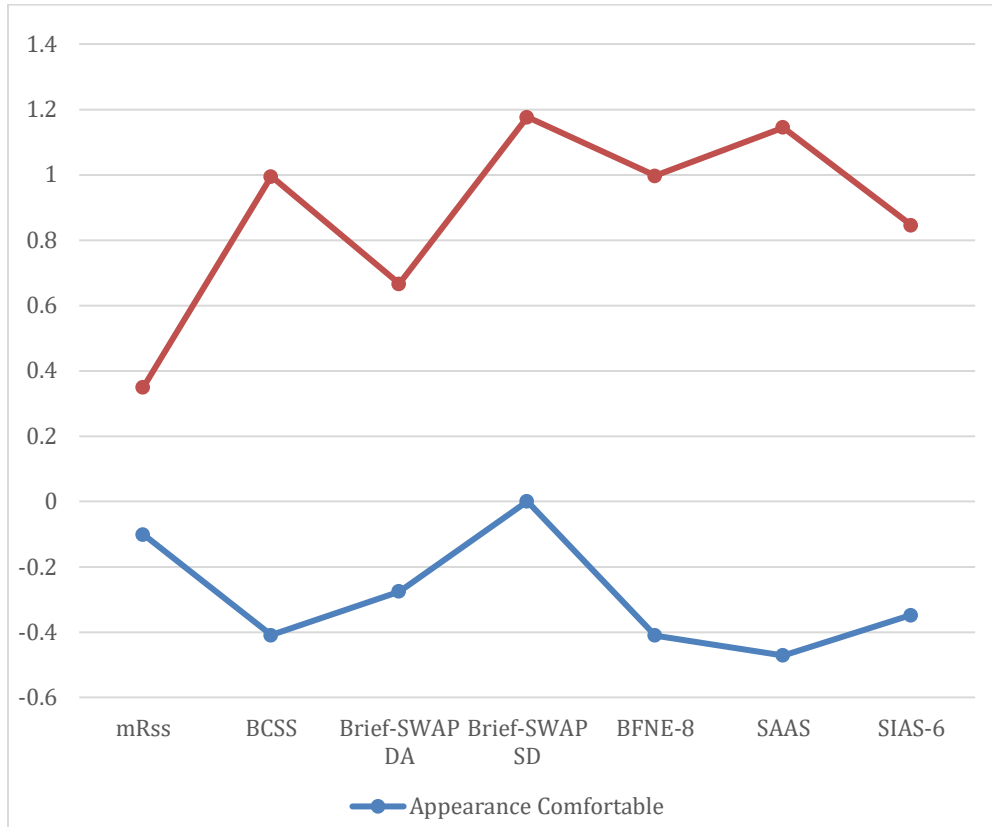
Figure 1. Standardized conditional response means for objective body image, subjective body image, and subjective appearance-related social anxiety variables in Sample 1
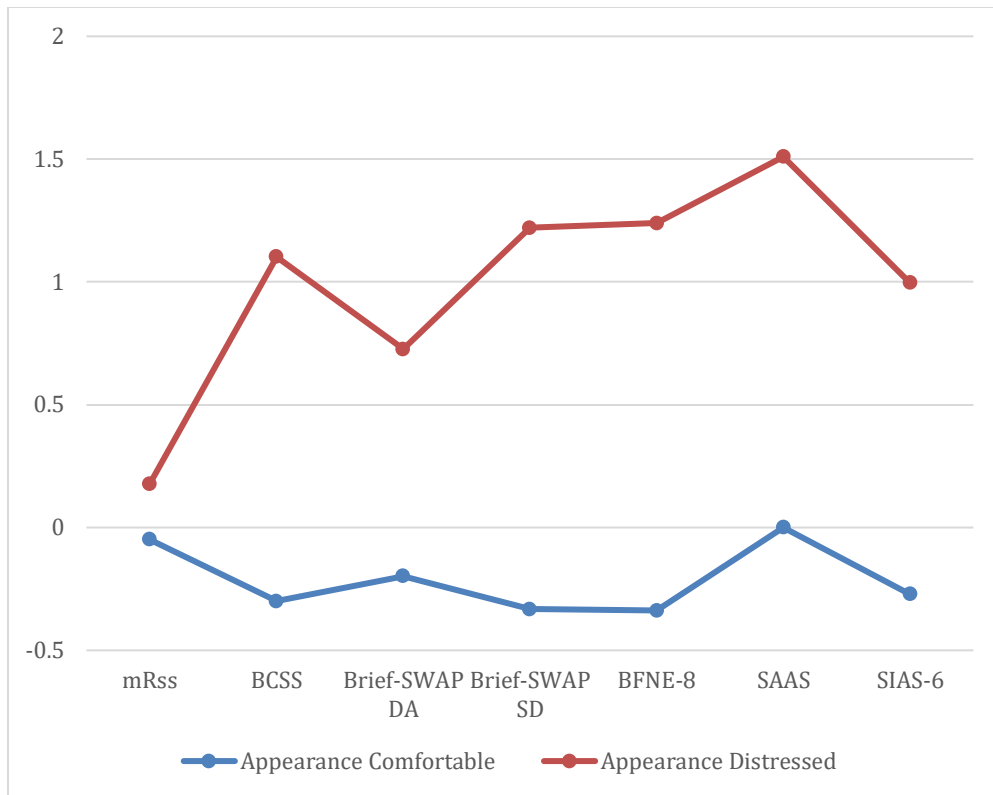
Figure 2. Standardized conditional response means for objective body image, subjective body image, and subjective appearance-related social anxiety variables in Sample 2

Table 1. Study characteristics

Variable	Total	Sample 1	Sample 2
	(N = 942)	(N = 469)	(N = 473)
Sociodemographic variables			
Gender[a]			
Female	823 (87.4)	415 (88.5)	405 (86.3)
Male	119 (12.6)	54 (11.5)	65 (13.7)
Ethnicity[a]			
White	751 (79.8)	380 (81.0)	371 (78.4)
Black	55 (5.8)	28 (6.0)	27 (5.7)
Other	135 (14.3)	60 (12.8)	75 (15.9)
Missing	1 (0.1)	1 (0.2)	0 (0.0)
Education[b,c]	15.14 (3.34)	15.17 (3.6)	15.11 (3.4)
Marital status[a]			
Married/Common Law	687 (72.9)	339 (72.3)	348 (73.6)
Not married	255 (27.1)	130 (27.7)	125 (26.4)
Single	109 (11.6)	60 (12.8)	49 (10.4)
Separated/Divorced	109 (11.6)	50 (10.7)	59 (12.5)
Widowed	37 (3.9)	20 (4.3)	17 (3.6)
Age[b]	55.30 (12.4)	55.81 (12.9)	54.79 (11.8)

Note. [a]n (%); [b]M (SD);[c]Years of education completed starting with elementary/primary school; [d]mRss: modified Rodnan skin score

Table 1: Continued

Variable	Total Sample	Group 1	Group 2
	$(N = 942)$	$(N = 469)$	$(N = 473)$
Medical variables			
Years since first non-Raynaud's symptom[b]	11.15 (8.5)	11.39 (8.5)	10.92 (8.4)
Disease Subtype			
Limited	558 (59.2)	276 (58.8)	282 (59.6)
Diffuse	384 (40.8)	193 (41.2)	191 (40.4)
mRss[d]	7.97 (8.4)	7.86 (8.1)	8.07 (8.7)
Telangiectasia			
No	269 (28.6)	138 (29.4)	131 (27.7)
Yes	661 (70.2)	327 (69.7)	334 (70.6)
Missing	12 (1.3)	4 (0.9)	8 (1.7)
Pigmentation			
No	613 (65.1)	312 (66.5)	301 (63.6)
Yes	277 (29.4)	134 (28.6)	143 (30.2)
Missing	52 (5.5)	23 (4.9)	29 (6.1)
Scleroderma			
No	430 (45.6)	220 (46.9)	210 (44.4)
Yes	510 (54.1)	248 (52.9)	262 (55.4)
Missing	2 (0.2)	1 (0.2)	1 (0.2)

Table 1: Continued

Variable	Total Sample	Group 1	Group 2
	($N = 942$)	($N = 469$)	($N = 473$)
Sclerodactly			
No	141 (15.0)		
Yes	798 (84.7)		
Missing	3 (0.3)		
Psychosocial variables			
BCSS[b]	7.68 (8.5)	8.10 (8.8)	7.25 (8.1)
BFNE-8[b]	16.04 (8.1)	16.14 (8.4)	15.9 (7.8)
Brief-SWAP-DA[b]	9.09 (5.2)	9.16 (5.2)	9.02 (5.07)
Brief-SWAP-SD[b]	5.18 (5.1)	5.40 (5.3)	4.97 (5.0)
SIAS-6[b]	2.43 (3.8)	2.30 (3.7)	2.55 (3.9)
SAAS[b]	28.20 (13.2)	28.08 (13.3)	28.22 (13.1)
PHQ-8[b]	5.89 (5.2)	5.71 (5.1)	6.07 (5.2)
SHAQ-DI[b]	0.74 (0.7)	0.74 (0.7)	0.72 (0.7)

Note. [a]*n* (%); [b]*M* (*SD*); BCSS: Body Concealment Score; BFNE-8: Brief Fear of Negative Evaluation Scale-8; Brief SWAP-DA: Brief Satisfaction with Appearance Scale- Social Discomfort Subscale; Brief-SWAP –SD: Brief Satisfaction with Appearance Scale-Social Discomfort Subscale; SIAS-6: Social Interaction Anxiety Scale-6: SAAS: Social Appearance Anxiety Scale; PHQ-8: Patient Health Questionnaire-8; SHAQ-DI: Scleroderma Health Assessment Questionnaire-Disability Index.

Table 2a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 1

Solution	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	22099.844	22113.520	---	----	---
2 profile	21095.964	21117.453	0.0002	$< .001$	0.904
3 profile	20771.658	20800.962	0.1443	$< .001$	0.922
4 profile	20631.304	20668.422	0.5930	$< .001$	0.920
5 profile	20487.035	20531.968	0.0826	$< .001$	0.929

Note. AIC: Akaike Information Criterion; sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Table 2b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2

Solution	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	22172.886	22186.680	---	----	---
2 profile	21187.959	21209.635	$< .001$	$< .001$	0.936
3 profile	20873.273	20902.831	0.0027	$< .001$	0.883
4 profile	20769.414	20806.854	0.1946	$< .001$	0.908
5 profile	20676.350	20721.672	0.4276	$< .001$	0.905

Note. AIC: Akaike Information Criterion; sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Table 3a. Sample means and group conditional response means for Sample 1

	M (SE)		
	Full sample (N = 469)	Appearance Comfortable (n = 334)	Appearance Distressed (n =135)
Subjective Body Image Measures			
BCSS	8.104 (0.404)	4.518 (0.462)	16.806 (1.178)
Brief SWAP-DA	9.159 (0.241)	7.721 (0.374)	12.636 (0.356)
Brief SWAP-SD	5.397 (0.245)	2.823 (0.407)	11.640 (0.433)
Social Anxiety			
BFNE-8	16.139 (0.385)	12.712 (0.410)	24.452 (1.275)
SAAS	28.079 (0.612)	21.825 (0.545)	43.250 (2.222)
SIAS-6	2.305 (0.170)	1.021 (0.110)	5.419 (0.691)
Objective Skin Involvement			
mRss	7.861 (0.376)	7.032 (0.524)	9.872 (0.909)

Note. SE: standard error.

Table 3b. Sample means and group conditional response means for Sample 2

	M (SE)		
	Full sample (N = 473)	Appearance Comfortable (n = 375)	Appearance Distressed (n =98)
Subjective Body Image Measures			
BCSS	7.254 (0.373)	4.819 (0.357)	16.192 (1.309)
Brief SWAP-DA	9.019 (0.233)	8.016 (0.295)	12.701 (0.447)
Brief SWAP-SD	4.966 (0.228)	3.319 (0.293)	11.015 (0.436)
Social Anxiety			
BFNE-8	15.945 (0.359)	13.309 (0.339)	25.624 (1.161)
SAAS	28.328 (0.604)	22.703 (0.483)	48.981 (2.066)
SIAS-6	2.545 (0.181)	1.477 (0.151)	6.468 (0.753)
Objective Skin Involvement			
mRss	8.072 (0.398)	7.651 (0.454)	9.617 (1.041)

Note. SE: standard error.

Table 4a. Differences in sociodemographic characteristics among groups for Sample 1

Variable	Appearance Comfortable (*n* = 334)	Appearance Distressed (*n* =135)	Statistics	
			Overall Model $\chi^2(1)$	*p*
Age[a]	58.01 (12.08)	50.36 (13.36)	33.49	**< 0.001**
Gender[b]			16.875	**< 0.001**
Female	285 (85.2)	130(96.3)		
Male	49 (14.8)	5 (3.7)		
Marital Status[b]	256 (76.8)	83 (61.5)	8.660	**0.003**
Race[b]				
White	269 (80.6)	112 (82.7)	0.274	0.872
Black	20 (6.0)	8 (5.9)		
Other	45 (13.4)	15 (11.4)		
Education[a,c]	14.98 (5.15)	15.74 (5.25)	0.204	0.652

Note. [a]*M* (*SD*); [b]*n* (%);[c]Years of education completed starting with elementary/primary school.

Table 4b. Differences in medical characteristics among groups for Sample 1

Variable	Appearance Comfortable ($n = 334$)	Appearance Distressed ($n = 135$)	Statistics Overall Model $\chi^2(1)$	p
Disease Subtype[b]			19.500	**<0.001**
Limited	221 (66.3)	56 (41.3)		
Diffuse	113 (33.7)	79 (58.7)		
Years Since 1st Non-Raynaud's[a]	10.79 (8.13)	12.74 (9.26)	4.553	**0.033**
Years Since Diagnosis[a]	9.35 (7.68)	11.01 (9.04)	3.519	0.061
Lower Hand Thickening[b]	274 (82.4)	122 (90.7)	5.074	**0.017**
Scleroderma[b]	153 (46.0)	94 (69.7)	21.30	**<0.001**
Fingertip Scars[b]	117 (35.2)	72 (53.7)	11.96	**0.001**
Telangiectasias[b]	235 (70.4)	94.5 (70.0)	0.007	0.931
Pigmentation[b]	86 (25.8)	45 (35.2)	7.079	**0.029**
BMI[a]	25.90 (6.31)	25.66 (6.07)	0.077	0.781

Note. [a]*M* (*SD*); [b]*n* (%)

Table 4c. Differences in psychosocial characteristics among groups for Sample 1

Variable	Appearance Comfortable ($n = 334$)	Appearance Distressed ($n = 135$)	Statistics	
			Overall Model $\chi^2(1)$	p
PHQ-8[a]	3.89 (3.45)	10.17 (5.69)	143.18	**<0.001**
SHAQ-DI[a]	0.60 (0.60)	1.08 (0.71)	47.64	**<0.001**
PROMIS-Anxiety[a]	47.76 (8.30)	59.15 (8.21)	183.96	**<0.001**

Note. [a]*M* (*SD*); [b]*n* (%)

Table 5a. Differences in sociodemographic characteristics among groups for Sample 2

Variable	Appearance Comfortable (*n* = 375)	Appearance Distressed (*n* =98)	Statistics	
			Overall Model $\chi^2(1)$	*p*
Age[a]	55.90 (11.4)	50.69 (12.4)	14.167	**<0.001**
Gender[b]			1.848	0.174
Female	319 (85.2)	88 (90.2)		
Male	55 (14.8)	10 (9.80)		
Marital Status[b]	283 (75.4)	66 (67.0)	2.255	0.133
Race[b]			3.776	0.151
White	303 (80.5)	69.48 (70.9)		
Black	21 (5.6)	6 (5.9)		
Other	51 (13.9)	23 (23.2)		
Education[a,c]	15.06 (5.15)	15.43 (5.25)	0.244	0.652

Note. [a]*M* (*SD*); [b]*n* (%);[c]Years of education completed starting with elementary/primary school

Table 5b. Differences in medical characteristics among groups for Sample 2

Variable	Appearance Comfortable (*n* = 375)	Appearance Distressed (*n* =98)	Statistics	
			Overall Model $\chi^2(1)$	*p*
Disease Subtype[b]			5.734	**0.020**
Limited	237 (63.1)	46 (46.9)		
Diffuse	138 (36.9)	52 (53.1)		
Years Since 1st Non-Raynaud's[a]	10.97 (8.8)	10.75 (8.4)	0.054	0.817
Years Since Diagnosis[a]	9.26 (8.0)	9.33 (7.9)	0.005	0.945
Sclerodactyly[b]	322 (86.1)	79 (81.7)	0.949	0.330
Lower hand thickening[b]	202 (53.8)	60 (61.7)	1.713	0.191
Fingertip Scars[b]	152 (40.3)	47 (46.5)	1.029	0.310
Telangiectasias[b]	27 (73.3)	65 (66.6)	1.542	0.214
Pigmentation[b]	107 (28.6)	44 (45.0)	7.534	**0.006**
BMI[a]	25.72 (5.8)	26.18 (6.0)	0.484	0.487

Note. [a]*M* (*SD*); [b]*n* (%)

Table 5c. Differences in psychosocial characteristics among groups for Sample 2

Variable	Appearance Comfortable ($n = 375$)	Appearance Distressed ($n = 98$)	Statistics	
			Overall Model $\chi^2(1)$	p
PHQ	4.47 (4.0)	10.82 (5.4)	119.24	**<.001**
SHAQ-DI	0.64 (0.6)	1.04 (0.6)	28.12	**<.001**
PROMIS-Anxiety	48.15 (8.4)	60.36 (7.4)	201.558	**<.001**

Note. [a]*M* (*SD*); [b]*n* (%)

Appendix A. SPIN cohort description

SPIN utilizes a novel research design called the cohort multiple RCT (cmRCT) design (Kwakkenbos et al., 2013). The design, a type of pragmatic trial design, was first described by Relton and colleagues (2010) as a response to some of the criticisms (e.g., limited external validity) of "gold standard" randomized controlled trials. The key characteristics of the cmRCT are described below:

    I.   Recruitment of participants with condition of interest by way of a large, observational cohort

    II.   The whole cohort participates in regular outcome measurement

    III.   The design allows for multiple randomized controls trials for the duration nof the study period

        a.   All eligible patients are identified from the entire cohort (e.g., patients who screen positive for depression)

        b.   A random selection of the eligible patients is selected for intervention and their scores are compared to those eligible but not selected (i.e., receiving usual care)

Kwakkenbos and colleagues (2013) noted that cmRCT is especially well-suited for rare diseases, such as scleroderma, in that it allows researchers to utilize participants from the same patient cohort for multiple trials (each with its own inclusion and exclusion criteria) and the consent process more closely mirrors actual clinical rather than research settings. Specifically, patients are informed in in the initial consenting that there are a number of

trials for which they may be eligible as part of the cohort, and they they will only be notified for trials where they have been offered the intervention, although their data may be used for comparative purposes. Kwakkenbos et al. (2013) described that, "In real-life healthcare situations, patients are only told of treatments that their healthcare provider can provide with certainty, and they are not told that the treatment they receive will be decided by chance." (p. 4).

Appendix B

<u>Exploratory Analysis 1 Including Only Subjective Indicators</u>

Table 6a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 1[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted *p*-value	Parametric bootstrapped LRT *p*-value	Entropy
1 profile	18798.118	18809.839	---	----	---
2 profile	17801.696	17820.255	0.0005	< .001	0.904
3 profile	17483.636	17509.033	0.1612	< .001	0.920
4 profile	17392.722	17424.956	0.2724	< .001	0.898
5 profile	17207.903	17246.976	0.0897	< .001	0.923

Note. [1] Variables included: BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test

Table 6b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	18783.991	18795.814	---	----	---
2 profile	17800.747	17819.467	< .001	< .001	0.938
3 profile	17487.092	17512.709	0.0035	< .001	0.884
4 profile	17385.241	17417.755	0.4700	< .001	0.903
5 profile	17295.027	7334.438	0.5786	< .001	0.895

Note. [1] Variables included: BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Exploratory Analysis 2 Including Only Subjective Indicators and Only Females

Table 7a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 1[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted *p*-value	Parametric bootstrapped LRT *p*-value	Entropy
1 profile	16697.908	16708.168	---	----	---
2 profile	15843.601	15859.846	0.0018	< .001	0.896
3 profile	15558.488	15580.719	0.1820	< .001	0.922
4 profile	15445.500	15473.716	0.0599	< .001	0.925
5 profile	15309.643	15343.844	0.1155	< .001	0.924

Note. [1] Variables included: BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Table 7b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted *p*-value	Parametric bootstrapped LRT *p*-value	Entropy
1 profile	16228.601	16238.658	---	----	---
2 profile	15402.092	15418.016	< .001	< .001	0.935
3 profile	15115.449	15137.240	0.0119	< .001	0.884
4 profile	15002.439	15030.096	0.3589	< .001	0.905
5 profile	14933.527	4967.051	0.4107	< .001	0.882

Note. [1] Variables included: BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Exploratory Analysis 3 Including All Indicators and Only Females

Table 8a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile solutions for Sample 1[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	19632.323	19644.293	---	----	---
2 profile	18768.654	18787.464	0.0002	< .001	0.899
3 profile	18476.396	18502.047	0.0851	< .001	0.924
4 profile	18350.699	18383.190	0.0164	< .001	0.935
5 profile	18216.732	18256.063	0.1592	< .001	0.929

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Table 8b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT ' p-value	Entropy
1 profile	19122.777	19134.511	---	----	---
2 profile	18294.388	18312.826	< .001	< .001	0.932
3 profile	18003.323	18028.467	0.0129	< .001	0.884
4 profile	17891.867	17923.715	0.3631	< .001	0.904
5 profile	17824.281	17862.833	0.4658	< .001	0.903

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6 AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Exploratory Analysis 4 Including All Indicators + Psychosocial Indicators

Table 9a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	25928.244	25945.826	---	----	---
2 profile	24721.019	24748.369	< .001	< .001	0.914
3 profile	24355.597	24392.716	0.0341	< .001	0.928
4 profile	24175.470	24222.356	0.0425	< .001	0.943
5 profile	24073.941	24130.595	0.2134	< .001	0.942

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6, PHQ-8, PROMIS-Anxiety, SHAQ-Disability; [2] AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Table 9b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted *p*-value	Parametric bootstrapped LRT *p*-value	Entropy
1 profile	26031.621	26049.356	---	----	---
2 profile	24879.907	24907.494	< .001	< .001	0.920
3 profile	24487.382	24524.822	0.0167	< .001	0.888
4 profile	24366.275	24413.568	0.1027	< .001	0.902
5 profile	24230.532	24287.677	0.1605	< .001	0.942

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6, PHQ-8, PROMIS-Anxiety, SHAQ-Disability; [2] AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.

Exploratory Analysis 5 Including All Indicators + Psychosocial Indicators + Only
Females

Table 10a. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for
Sample 1[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted p-value	Parametric bootstrapped LRT p-value	Entropy
1 profile	23050.387	23065.778	---	----	---
2 profile	22012.892	22036.833	0.0013	< .001	0.906
3 profile	21682.204	21714.695	0.0924	< .001	0.931
4 profile	21510.028	21551.069	0.5601	< .001	0.935
5 profile	21412.086	21461.678	0.0901	< .001	0.943

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8,
SAAS, SIAS-6, PHQ-8, PROMIS-Anxiety, SHAQ-Disability; [2] AIC: Akaike Information
Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT:
Bootstrapped Likelihood Ratio Test.

Table 10b. Model fit indices for the 1-, 2-, 3-, 4- & 5-profile -profile solutions for Sample 2[1]

Solution[2]	AIC	sBIC	Lo-Mendell Rubin Adjusted *p*-value	Parametric bootstrapped LRT *p*-value	Entropy
1 profile	22454.725	22469.810	---	----	---
2 profile	21461.068	21484.535	< .001	< .001	0.912
3 profile	21118.898	21150.745	0.0786	< .001	0.886
4 profile	20978.835	21019.064	0.2380	< .001	0.910
5 profile	20887.473	20936.083	0.2422	< .001	0.911

Note. [1] Variables included: mRss, BCSS, Brief-SWAP-DA, Brief-SWAP-SD, BFNE-8, SAAS, SIAS-6, PHQ-8, PROMIS-Anxiety, SHAQ-Disability; [2] AIC: Akaike Information Criterion; [2]sBIC: Sample-size adjusted Bayesian Information Criterion; BLRT: Bootstrapped Likelihood Ratio Test.