Living with a Visible Difference:
Assessing Body Image and Appearance Concerns in Scleroderma

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Abstract

People living with visible appearance differences, whether present from birth or acquired through injury or illness, often experience psychosocial and psychological challenges. These include negative self-perceptions such as low self-esteem, shame, body image dissatisfaction, and depression; as well as challenges related to social interactions such as fear of negative evaluation, social avoidance, and social anxiety. Systemic sclerosis, or scleroderma (SSc), is a rare, chronic, autoimmune connective tissue disease that leads to significant physical appearance changes, which have been linked to negative outcomes including body image dissatisfaction, social discomfort and anxiety, as well as general psychological distress. In order to advance research in this area, however, and support the design of adequate psychosocial interventions, more disease-specific patient-reported outcome measures related to body image concerns are needed. In addition, an increased understanding of how disease and sociodemographic factors are associated with these outcomes is required to inform intervention content. In order to address these gaps, four independent studies were conducted and included in this thesis. Study One involved the development and validation of a new patient-reported outcome measure of body concealment and avoidance behaviours specific to SSc, the Body Concealment Scale for Scleroderma (BCSS), including examination of its basic psychometric properties. Study Two constituted a replication of Study One, with the extension of determining the measurement equivalency of BCSS items across sex and age groups. The findings of this study replicated those of Study One and further demonstrated that BCSS scores can be compared and combined across females and males with SSc and those of different ages without concern that measurement differences influence results. Study Three examined the measurement equivalency of a commonly used measure of social discomfort and body image dissatisfaction relative to acquired disfigurements in SSc, the Satisfaction with Appearance Scale (SWAP), across language groups. Findings from this study enhanced existing psychometric evidence by showing that SWAP items do not function differently across English-and French-speaking Canadian SSc patients and that scores can be pooled across these language groups. Study Four explored the association of key sociodemographic variables with the presence of appearance concerns among patients living with SSc. Main findings from this study revealed that female patients had significantly higher odds of reporting appearance concerns than male patients and that older age was significantly associated with a reduction in the odds of reporting appearance concerns. There was no significant difference in the odds of reporting appearance concerns between Black and White patients, after accounting for the influence of sociodemographic and disease characteristics. Finally, women and men with SSc were more likely to report appearance concerns than members of the general population. In sum, the body of the presented research involved the creation of a new disease-specific patient-reported outcome measure and potentially useful clinical assessment tool of body concealment behaviours in SSc, including establishing its psychometric properties. This thesis also highlighted the importance of evaluating the measurement equivalency of body image questionnaires across diverse groups of patients with SSc and demonstrated differences in key sociodemographic and disease characteristics, such as sex, age, and presence of hand contractures, in the experiencing of body image concerns. Implications and future directions for research are discussed.

Résumé

Les personnes vivant avec des différences visibles au niveau de l'apparence, qu'elles soient présentes dès la naissance ou acquises suite à une blessure ou une maladie, éprouvent souvent des difficultés psychosociales et psychologiques. Ces difficultés regroupent des perceptions de soi négatives telles qu'une faible estime de soi, un sentiment de honte, une insatisfaction liée à l'image corporelle et la dépression. Certains défis rencontrés sont liés aux interactions sociales telles que la peur d'être jugé négativement, l'évitement social et l'anxiété sociale. La sclérodermie systémique, ou sclérodermie (SSc), est une maladie du tissu conjonctif auto-immune, rare et chronique qui entraîne des changements importants au niveau de l'apparence physique. Ces changements ont des conséquences négatives comme une insatisfaction liée à l'image corporelle, un inconfort social, de l'anxiété sociale ainsi qu'une détresse psychologique générale. Cependant, afin de faire avancer la recherche dans ce domaine et de soutenir l'élaboration d'interventions psychosociales adéquates, il faut davantage de mesures de résultats déclarés par les patients, spécifiques aux maladies et liées aux problèmes d'image corporelle. De plus, une meilleure compréhension de la façon dont la maladie et les facteurs sociodémographiques sont associés à ces résultats est requise pour développer le contenu de ces interventions. Afin de combler ces lacunes, quatre études indépendantes ont été menées et intégrées dans cette thèse. La première étude constituait le développement et la validation d'une nouvelle mesure de résultats déclarés par les patients sur la dissimulation corporelle et de comportements d'évitement spécifiques à la SSc, la Body Concealment Scale for Scleroderma (BCSS), incluant l'évaluation de ses composantes psychométriques de base. La deuxième étude comprenait une réplication de la première étude, à laquelle s'ajoutait la détermination de l'équivalence de mesures des items de l'échelle BCSS entre les sexes et les groupes d'âge. Les résultats de cette étude ont reproduit ceux de la première étude et ont démontré que les scores

BCSS peuvent être comparés et combinés entre les femmes et les hommes atteints de SSc issus de différents groupes d’âge sans que ces différences de mesures influencent les résultats. La troisième étude évaluait l’équivalence de mesure de l’échelle de satisfaction de l’apparence physique (SWAP) qui est couramment utilisée pour mesurer l’inconfort social et l’insatisfaction de l’image corporelle par rapport aux défigurations reliées à la SSc, à travers les groupes linguistiques. Les résultats de cette étude ont enrichi les données psychométriques existantes en montrant que les items du SWAP ne fonctionnent pas différemment chez les patients anglophones que chez les patients francophones du Canada. Ainsi, les scores peuvent être mis en commun entre ces groupes linguistiques. La quatrième étude portait sur l’association des variables sociodémographiques principales à la présence de préoccupations en lien avec l’apparence chez les patients atteints de SSc. Les principaux résultats de cette étude ont révélé que les femmes atteintes de SSc avaient des probabilités significativement plus élevées de déclarer des problèmes d’apparence que les hommes atteints de SSc et qu’un âge plus élevé était significativement associé à une probabilité plus faible de déclarer des problèmes d’apparence. Il n’y avait pas de différence significative dans les probabilités de déclarée les problèmes d’apparence entre les patients d’ethnicité noire ou blanche, après avoir tenu compte de l’influence des caractéristiques sociodémographiques et de la maladie. Enfin, les femmes et les hommes atteints de SSc étaient plus susceptibles de déclarer des problèmes d’apparence que les membres de la population générale. En somme, la présente recherche a impliqué la création d’une nouvelle mesure de résultats déclarés par les patients, spécifiques aux maladies et d’un outil d’évaluation clinique potentiellement utile pour les comportements de dissimulation du corps avec la SSc, y compris l’élaboration de ses propriétés psychométriques.

Cette thèse a également permis de souligner l'importance de l'évaluation de l'équivalence des questionnaires d'image corporelle entre divers groupes de patients atteints de SSc. Elle a également démontré les différences entre les principales caractéristiques sociodémographiques et caractéristiques en lien avec la maladie telles que le sexe, l'âge et la présence de contractures aux mains, à travers l'expérience de préoccupations en lien avec l'image corporelle. Les implications et les pistes de recherches futures sont discutées.

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Contribution of Authors

The four studies included in the present thesis constitute original research projects. I am the first author on all four study manuscripts, as I contributed to the conception and design of each project; conducted the acquisition, analysis, and interpretation of data; completed the original draft of each manuscript; incorporated critical feedback from co-authors and reviewers; and prepared and submitted the final draft of each manuscript.

The first study manuscript is co-authored by Dr. Vanessa Malcarne, Dr. Linda Kwakkenbos, Dr. Diana Harcourt, Dr. Nichola Rumsey, Dr. Annett Körner, Dr. Russell Steele, Dr. Marie Hudson, Dr. Murray Baron, Dr. Jennifer Haythornthwaite, Dr. Leslie Heinberg, Dr. Fredrick Wigley, Dr. Brett Thombs, and the Canadian Scleroderma Research Group (CSRG). Dr. Thombs contributed to the conception and design of the manuscript; the analysis and interpretation of data; editing and revising the manuscript; and approving of the final version. Drs. Malcarne, Kwakkenbos, and Steele contributed to the analysis and interpretation of data; editing and revising the manuscript; and approving of the final version. Drs. Harcourt, Rumsey, and Körner contributed to editing and revising the manuscript and approving of the final version. Drs. Hudson, Baron, Haythornthwaite, Heinberg, and Wigley contributed to the acquisition of data; editing and revising the manuscript; and approving of the final version. The CSRG contributed to the acquisition of data.

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Chapter 1

Introduction

Literature Review

Visible Differences and Body Image Concerns

It is estimated that approximately one person in 100 has a visible difference (Changing Faces, 2016). Visible differences refer to any condition that alters an individual's physical appearance in such a way that it is distinct from the perceived norm, whatever that may be (Rumsey & Harcourt, 2004). Visible differences result from a variety of sources but are generally divided into categories of congenital anomalies and acquired disfigurements (Bessell, Dures, Semple, & Jackson, 2012; Rumsey & Harcourt, 2004). Congenital anomalies denote conditions that are fully present from birth (e.g., cleft lip and palate, birthmarks) as well as those that become more apparent over time (e.g., neurofibromatosis). Acquired disfigurements include those caused by injuries (e.g., burns, accidents), medical and genetic conditions (e.g., acne, vitiligo), and surgical interventions (e.g., treatment for head and neck cancer; Rumsey & Harcourt, 2004). Visible differences generally exclude individuals whose appearance concerns directly relate to a psychiatric condition (e.g., body dysmorphic disorder) and those resulting from a physical disability (e.g., missing limb; Rumsey & Harcourt, 2004).

Whether present from birth or acquired later in life, visible differences are associated with substantial psychological and psychosocial impact (Clarke, Thompson, Jenkinson, Rumsey, & Newell, 2014; Rumsey & Harcourt, 2004; 2012). The most common difficulties experienced by individuals with visible differences relate to unfavourable self-perceptions and challenges with social interactions (Rumsey & Harcourt, 2004). These involve negative emotional states such as low self-esteem, embarrassment, shame, body image dissatisfaction, depression, and social anxiety; thought processes including fear of negative social evaluation; as well as adverse behavioural responses such as concealment of affected body parts, unhelpful safety behaviours, and social withdrawal and avoidance (Bessell et al., 2012; Cash & Pruzinsky, 2002; Newell, 1999; Rosser, Moss, & Rumsey, 2010; Rumsey & Harcourt, 2004).

Individuals with visible differences are also frequently subject to negative reactions from others, such as staring, unwanted attention and questions, hostility, and avoidance (Jewett et al., 2018; Kent, 2000; Macgregor, 1990). These experiences can trigger appearance-related distress and concerns about not meeting cultural norms for attractiveness (Bessell et al., 2012). Social anxiety may develop when actual experiences of exclusion or rejection by others lead to an ongoing fear of being evaluated negatively based on one's appearance (Baumeister & Leary, 1995; Kent, 2000). Thus, visible differences have been described as a form of social disability, due to their impact on the thoughts, feelings, and behaviours of those affected but also because of their noticeability and potential to lead to negative reactions from others (Macgregor, 1990). The social impairment from disfigurements can add substantially to already high levels of physical discomfort and disability experienced by those with acquired visible differences from an injury or medical illness (Green, 2010; Woolf & Pfleger, 2003).

Concerns about appearance may be particularly salient among people with disfigurements from an injury or medical condition (Rumsey & Harcourt, 2004; 2012). The acquisition of a visible difference can sometimes worsen pre-existing body image concerns and feelings of separation from appearance ideals (Bessell et al., 2012); if appearance is altered as the result of a traumatic injury, additional rehabilitation and psychological considerations may be important to promote reintegration into the community after hospitalization and adjustment post-injury (Rumsey & Harcourt, 2012). Furthermore, acquired appearance changes from medical causes such as dermatological conditions, rheumatic and connective tissue diseases, and cancers often carry with them indicators of personal health status, which can involve additional issues of disclosure and related discomfort (Rumsey & Harcourt, 2012). It has been documented that individuals with acquired disfigurements have difficulty achieving healthy social interactions, experience body image dissatisfaction and self-consciousness, as well as fear of stigmatization and rejection (Basra & Shahrukh, 2009; Bessell et al., 2012; Fobair et al., 2006; Gupta & Gupta, 2003; Hong, Koo, & Koo, 2008; Jorge et al., 2010; Levine, Degutis, & Pruzinsky, 2005; Pruzinsky, 1992; Rumsey & Harcourt, 2012). These feelings often lead to social anxiety, withdrawal, and isolation (Basra & Shahrukh, 2009), all of which can be highly debilitating and may lead to generalized helplessness and depression (Gupta & Gupta, 2003).

Scleroderma and Body Image Concerns

Systemic sclerosis, or scleroderma (SSc), is an example of a rare, chronic, multisystem autoimmune connective tissue disease that leads to substantial physical disability and significant appearance changes (Gholizadeh et al., 2017; Kwakkenbos et al., 2015; Mayes, 2008). SSc is characterized by three main pathological processes: (1) vascular injury, (2) immune dysfunction (autoimmunity), and (3) abnormal fibrosis (Boin & Wigley, 2012); and it commonly affects organ systems including the skin, lungs, gastrointestinal tract, and heart (Boin & Wigley, 2012; Wigley & Hummers, 2003; Mayes, 2008). As such, SSc often leads to a complex set of serious medical problems, such as Raynaud’s phenomenon (Merkel et al., 2002), arthritis and joint contractures (Seibold et al., 2005), esophageal disease and chronic gastrointestinal symptoms (Ntoumazios et al., 2006; Younho et al., 2015), pulmonary hypertension and disease (Wigley & Hummers, 2003), interstitial lung disease (Seibold et al., 2005), and renal failure (Wigley & Hummers, 2003); and it can sometimes be fatal.

Disease onset typically occurs between the ages of 30 and 50 years, and approximately 80% of people with SSc are women (Mayes, 2008). In Canada, SSc affects around 74 persons per 100,000 for women and 13 per 100,000 for men. Overall, it is estimated that 16,000 Canadians may have the condition (Bernatsky, Joseph, Pineau, Belisle, Hudson, & Clarke, 2009). There is no cure for SSc, and the median survival time from diagnosis is approximately 11 years, with patients 3.7 times more likely to die within 10 years of diagnosis (44.9% mortality) than age, race, and sex-matched persons without the disease (12.0% mortality; Mayes et al., 2003).

The term *scleroderma* is derived from the Greek words *skleros* (hard) and *derma* (skin), which reflect the hallmark feature of skin disfigurement experienced by many SSc patients, stemming from abnormal fibrotic processes and excessive production of collagen (Mayes, 2008). However, appearance changes in SSc due to disease processes extend beyond thickening and hardening of the skin and can include hypo- and/or hyper-pigmentation of the skin; telangiectasias (dilated blood vessels visible beneath the skin); hand contractures; sclerodactyly (skin tightening on the digits that often leads to ulcerations); deformity of the digits; calcinosis (calcium deposits under or protruding through the skin); and altered facial features, including a pinched appearance to the nose and eyes, loss of lip flexibility, and a decreased ability to fully open the mouth (Gholizadeh et al., 2017). Appearance changes from SSc can be drastic and extensive and commonly affect body parts that are highly visible and play a central role in social interactions, such as the face, mouth, and hands (Gholizadeh et al., 2017; Kwakkenbos et al., 2015). Furthermore, they are typically not alleviated by treatments, and in some cases, treatments may lead to changes in appearance. For instance, certain immunosuppressant drugs cause hair loss, and long-term steroid use may contribute to a rounded appearance of the face (Gholizadeh et al., 2017).

The psychological and psychosocial impact of the visible differences experienced by SSc patients can be significant. For instance, appearance changes in SSc are associated with poor appearance self-esteem, social discomfort, and general psychological distress (Benrud-Larson et al., 2003; Jewett et al., 2012; van Lankveld, Vonk, Teunissen, & van den Hoogen, 2007). Disfigurements to the face and hands are related to dissatisfaction with appearance and symptoms of depression (Benrud-Larson et al., 2003; Gholizadeh et al., 2017; Malcarne, Hansdottir, Greenberg, Clements, & Weisman, 1999; van Lankveld, et al., 2007). Beyond this, the presence, subjective severity, and perceived noticeability of appearance changes are also linked to greater body image dissatisfaction and poorer psychological functioning, as well as social discomfort and psychosocial difficulties among SSc patients (Amin et al., 2011; Kwakkenbos et al., 2015; Sivakumar et al., 2010). Frustration, worry, and fear of negative evaluation are also reported by SSc patients, as feelings associated with the physical changes and unpredictability of the disease, in addition to the unwanted attention received from others (Joachim & Acorn, 2003; Richards, Herrick, Griffin, Gwilliam, & Fortune, 2004). It has been suggested that people with SSc may be susceptible to social avoidance and anxiety, in a similar manner to other groups of individuals with acquired visible differences from medical conditions, given the nature and extent of the disfiguring appearance changes that are often sustained (Haythornthwaite, Heinberg, & McGuire, 2003).

Cognitive-behavioural Theoretical Approach to Body Image

A cognitive-behavioural theoretical approach to body image (Cash & Pruzinsky, 2002) can explain how such negative outcomes may occur among SSc patients with visible differences and provides an orienting model for the manuscripts of this thesis. According to this perspective, two basic schemas, investment in appearance and evaluation of appearance, serve as organizing constructs for the cognitive, emotional, and behavioural processes related to body image (Cash & Pruzinsky, 2002). Investment refers to the importance people place on their appearance (e.g., cognitive, emotional, and behavioural significance of body for self-perception), and evaluation pertains to positive versus negative appraisals of and beliefs about one's appearance (e.g., body image satisfaction versus dissatisfaction; Cash & Pruzinsky, 2002). The present thesis focuses on manifestations of the evaluation schema. According to cognitive-behavioural theory, body image evaluations stem from the degree of discrepancy between self-perceived physical characteristics and personally valued appearance ideals (Cash & Pruzinsky, 2002). Historical aspects such as culture, socialization, past interpersonal experiences, as well as current situational events, can activate schema-driven processing of information about one's physical appearance (Cash & Pruzinsky, 2002). This leads to automatic thoughts and interpretations about one's looks (e.g., dissatisfaction with the appearance of certain body parts), as well as emotions that are often distressing, whether anticipated (e.g., fear of negative social evaluation) or real (e.g., low self-esteem, depressive mood). In order to manage these thoughts and emotions as well as adapt to triggering situations, individuals may engage in a range of actions, such as avoidance and body concealment, among others (e.g., appearance-correcting rituals, seeking social reassurance; Cash & Pruzinsky, 2002). These behavioural responses may serve to complete a negative feedback loop, whereby schemas are reinforced through escape or other self-regulatory behaviours and short-term reduction in body image distress is achieved.

Research on Body Image Issues in Scleroderma

Due to the drastic nature of many visible differences common to SSc, as well as the potential for significant psychosocial and psychological effects, there is emerging recognition of the importance of investigating body image and appearance-related concerns among patients (Gholizadeh et al., 2017; Kwakkenbos et al., 2015; Thombs et al., 2010). Calls have also been made for the development and testing of interventions to meet the rehabilitation and psychosocial needs of SSc patients, including those related to living with a unique and visibly different appearance (Kwakkenbos et al., 2015; Thombs et al., 2012). Recently, an international consortium, the Scleroderma Patient-centered Intervention Network (SPIN), was established for this purpose and is in the process of creating a large patient cohort from treatment centres in Canada, the United States, and Europe, to aid in the design, development, and testing of programs for SSc patients, including a body image intervention (Kwakkenbos et al., 2013). However, in order to further the research on appearance-related concerns in SSc and to support the design of interventions that target associated distress, the development of assessment tools that measure relevant body image constructs for patients is much needed, as well as an increased understanding of how disease and sociodemographic factors are associated with these outcomes. While some research has been conducted in these areas, there remains a lack of measures that assess disease-specific body image issues in SSc, including the absence of validity evidence for existing tools across different subgroups of patients; as well as a dearth of information on the associations of important factors such as age, sex, race/ethnicity, and disfiguring disease characteristics, with appearance concerns.

One body image measure, the Satisfaction with Appearance Scale (SWAP; Lawrence et al., 1998) has been studied fairly extensively in SSc, including adaptation and validation in different samples of patients (Heinberg et al., 2007; Mills et al., 2015). A shortened version of the questionnaire, the Brief-SWAP, has also been developed and found to be a valid measure of dissatisfaction with appearance and social discomfort related to appearance changes from the disease (Jewett et al., 2010; 2012; Mills et al., 2015) that is also quicker to administer in practice than the full-length SWAP scale. While the psychometric properties of both the SWAP and Brief-SWAP have been evaluated and established, the validity of the measure for use across different characteristics of patients that may potentially influence scores (e.g., sex, age, and language) has yet to be examined. Furthermore, this measure only assesses two dimensions of the body image construct, namely social discomfort based on disfiguring appearance changes and dissatisfaction with the appearance of particular body parts. The SWAP does not capture other potentially relevant body image domains for SSc patients, such as body concealment and social and behavioural avoidance due to appearance-related distress, which are recognized as common coping strategies among people with visible differences (Clarke et al., 2014). Furthermore, as Thompson (2004) highlights, proper and comprehensive measurement of body image constructs occurs when outcome measures assess specific and relevant dimensions of body image (e.g., subjective satisfaction, behavioural avoidance and concealment), are appropriately adapted for particular target groups, and are properly validated in these groups.

Preliminary investigations have also explored the associations of sociodemographic and disease factors with body image outcomes in SSc. One study of 489 Canadian SSc patients used the Brief-SWAP items to model relationships of dissatisfaction with appearance and social discomfort, separately, with age, sex, education, marital status, race/ethnicity, disease duration, skin involvement, telangiectasias, skin pigmentation changes, and hand contractures (Jewett et al., 2012). This study found that extent of skin involvement was significantly associated with both dissatisfaction with appearance and social discomfort, as was skin involvement specifically on the face. Greater social discomfort was linked to younger age and upper-body telangiectasias, and dissatisfaction with appearance was associated with the presence of hand contractures

(Jewett et al., 2012). In another study of 98 American SSc patients, Black patients had greater body image dissatisfaction related to digital ulcers, pruritus (itching), and skin pigmentation changes, than White patients, based on a non-validated, three-point scale (Nusbaum, et al., 2016).

Research Objectives

The research conducted to date on body image in SSc has identified significant appearance-related concerns among patients and identified the SWAP as a valid measure of two important body image outcomes relative to the acquired disfigurements in SSc, namely social discomfort and dissatisfaction with appearance. This measure lacks validity data, however, for distinct subgroups of patients, and it does not address other potentially relevant appearance concerns for people with the disease, such as those related to body concealment and avoidance. While previous studies have also provided some initial evidence of disease (e.g., skin involvement) and sociodemographic (e.g., age and sex) characteristics that may impact appearance-related distress among patients, replication of these findings is needed, as well as further investigations into relationships among these and other factors that may affect body image concerns, such as race/ethnicity.

Therefore, the objectives of the present research are to enhance the scope and validity of available disease-specific measures of central body image concerns in SSc and to better understand the associations of both sociodemographic and disease factors with appearance-related distress among patients. As such, four independent studies were conducted and included in this thesis. First, a study was undertaken to validate a new patient-reported outcome measure of body concealment and avoidance behaviours specific to SSc, the Body Concealment Scale for Scleroderma (BCSS), including evaluation of its basic psychometric properties.

Next, a study was conducted to examine the psychometric properties of the BCSS in a separate sample of SSc patients and to assess the tool's measurement equivalency across sex and age groups, which are characteristics that may be expected to influence item responses. It is possible that race/ethnicity and language background may also influence item responses; however, it was not possible to examine the measurement equivalency of BCSS items across racial/ethnic or language groups in this study, given the limited number of non-English speakers and non-White patients in the sample.

The third study similarly evaluated measurement equivalency, but, in this case, of the SWAP, the most commonly used assessment tool of social discomfort and body image dissatisfaction in SSc. The SWAP is currently administered as part of an international cohort of patients with SSc (Kwakkenbos et al., 2013); however, the degree to which scores are equivalent across linguistic groups has yet to be determined. This is an important consideration because patients who complete the SWAP in countries with more than one common language, such as Canada, or as part of an international cohort, may do so in different languages. As such, the objective of the third study was to assess the equivalence of SWAP item scores across English- and French-speaking SSc patients in a large Canadian cohort. We did not examine the measurement equivalency of SWAP items across racial and ethnic groups due to limited numbers of non-White patients in the study sample, nor did we examine measurement equivalency across age and sex, given the aim to focus on potential linguistic differences across SWAP item responses.

Finally, the fourth study examined the association of key sociodemographic factors (e.g., sex, age, race/ethnicity) and disease variables (e.g., skin pigmentation changes, hand contractures) with the presence of appearance concerns, controlling for factors that are known to influence appearance concerns, in a large, international cohort of SSc patients. This fourth study also compared the percentage of patients with SSc who reported the presence of appearance concerns to rates previously published from a UK general population sample, stratified by sex and age groups.

Chapter 2

Study One

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Development and Validation of the Body Concealment Scale for Scleroderma (BCSS)

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Abstract

Body concealment is a component of social avoidance among people with visible differences from disfiguring conditions, including systemic sclerosis (SSc). The objective was to develop a measure of body concealment related to avoidance behaviours in SSc. Initial items for the Body Concealment Scale for Scleroderma (BCSS) were selected using item analysis in a development sample of 93 American SSc patients. The factor structure of the BCSS was evaluated in 742 Canadian patients with one-factor, two-factor, and bifactor confirmatory factor analysis models. Convergent and divergent validity were assessed by comparing the BCSS total score with a measure of body image distress and measures of depressive symptoms and pain. A two-factor model fit substantially better than a one-factor model for the 9-item BCSS, but the Concealment with Clothing and Concealment of Hands factors were highly correlated. The bifactor model also fit well. In the bifactor model, the omega coefficient was high for the general factor but low for the Concealment with Clothing and Concealment of Hands factors. The BCSS demonstrated strong internal consistency reliability and construct validity. The BCSS sum score is a valid indicator of body concealment in SSc that extends the concepts of body concealment and avoidance beyond the realms of body shape and weight to concerns of individuals with visible differences from SSc.

Keywords: systemic sclerosis; scleroderma; disfigurement; body image avoidance; body concealment; measurement

Development and Validation of the Body Concealment Scale for Scleroderma (BCSS)

Systemic sclerosis (SSc) or scleroderma, is a rare chronic autoimmune connective tissue disease that affects organs including the skin, lungs, gastrointestinal tract, and kidneys. SSc often alters physical appearance in visible and socially relevant body parts, including the face, mouth, neck, and hands (Boin & Wigley, 2012; Heinberg et al., 2007; Mayes, 2008). Treatments can mitigate the impact of some symptoms but do not alleviate manifestations of irreversible tissue damage that affect appearance.

Concerns about appearance are particularly salient among people with visible differences from injury or illness (Krawczyk, Menzel, & Thompson, 2012; Rumsey & Harcourt, 2004). Many people with acquired disfigurements from injury or illness have difficulty achieving healthy social interactions, and some experience negative interpersonal relations due to their different appearance (Fobair et al., 2006; Levine, Degutis, & Pruzinsky, 2005; Pruzinsky, 1992; Rumsey & Harcourt, 2012). Many appearance changes common in SSc, such as changes in skin pigmentation, telangiectasias (visible dilation of blood vessels beneath skin), hand contractures, and altered facial features, are distinct from appearance changes in other medical conditions. Visual manifestations of SSc are associated with greater body image dissatisfaction and social discomfort, poorer appearance self-esteem, more depressive symptoms, and reduced overall psychosocial functioning (Amin et al., 2011; Benrud-Larson et al., 2003; Malcarne et al., 1999; van Lankveld et al., 2007; Jewett et al., 2012; Sivakumar et al., 2010).

Appearance concerns among individuals with visible differences can lead to fear of negative evaluation and social anxiety (Rumsey & Harcourt, 2004), which, in turn, can lead to behaviours aimed at concealing visible differences or avoiding social situations where attention may be given to physical appearance (Cash & Pruzinsky, 2002; Lethem, Slade, Troup, &

Bentley, 1983; Newell, 1999; Newell, & Marks, 2000; Rosen, Srebnik, Satlzberg, & Wendt, 1999; Rumsey & Harcourt, 2004). Body image avoidance generally refers to avoidance behaviours associated with negative body image, such as avoiding looking at one's own body, avoiding being looked at by others, avoiding body image-related media, or concealing the body (Rosen et al., 1999). Concealment behaviours may take a variety of forms, including the use of clothing to hide certain body parts (Clarke et al., 2014); however, in SSc, appearance changes to the hands or face are difficult to conceal via clothing. This may have significant implications for people with SSc, since joint contractures, digital ulcers, loss of digits, changes in pigmentation, calcinosis, and Raynaud's Phenomenon are common manifestations affecting these body parts (Mayes, 2008).

Body image-related concealment has not been studied in SSc, and there are no measures assessing concealment behaviours that reflect disease-specific appearance changes. The Body Image Avoidance Questionnaire (BIAQ; Rosen et al., 1999) includes several items related to concealment with clothing, but it focuses on concealment of weight, shape, or size and has no subscale that assesses concealment specifically. The five-item Body Concealment subscale of the Body Image Behaviour Scale (Davidson & McCabe, 2005) is based on items from the BIAQ (Rosen et al., 1999) and the Attention to Body Shape Scale (Beebe, 1995) but it is not validated, and its items similarly focus on general concealment, but not strategies associated with disfiguring conditions. More general measures of distress related to physical appearance, such as the Derriford Appearance Scale (Carr, Harris, & James, 2000; Carr, Moss, & Harris, 2005) include items that reflect attempts at concealment, but these items do not generate a single score that reflects concealment behaviours.

Given the significance of concealment as a coping strategy among people with visible differences, including the popularity of clothing, make-up, and accessories as concealment methods (Clarke et al., 2014), it is important to be able to accurately measure appearance-related concealment. Some concealment behaviours in SSc may be similar to behaviours common to other visible differences. The distinctive physical appearance changes (e.g., telangiectasias, skin hardening and tightening, unique facial changes, hand involvement) in SSc, however, make it essential to assess concealment behaviours in a manner that reflects the experience of individuals with the disease. Concealment of the hands, for instance, may be a behaviour particularly common in SSc, but less so in other conditions.

The objective of this study was to develop and validate the Body Concealment Scale for Scleroderma (BCSS). First, in a development sample, we conducted preliminary testing of items from the BIAQ potentially relevant to body image concerns in SSc, as well as original items developed to reflect concealment behaviours in SSc. Second, in a larger validation sample, we assessed the BCSS factor structure and convergent and discriminant validity. We hypothesized that the BCSS would correlate strongly with other measures of body image distress, moderately with measures of depressive symptoms, and to a lesser degree with a measure of pain.

Method

Participants and Procedures

Development Sample. The development sample consisted of patients diagnosed with SSc based on American College of Rheumatology criteria (LeRoy, Black, Fleishmajer, Jablonksa, Krieg, & Medsger, 1988) who were undergoing treatment at the Johns Hopkins Scleroderma Center, enrolled in a longitudinal study examining psychosocial adjustment to SSc, and had completed all items of a preliminary version of the BCSS. Items under consideration for inclusion in the BCSS were initially generated by two psychologists and were then reviewed by three experts in body image research. Subsequently, the revised items were reviewed by two rheumatologists and a nurse with expertise in SSc. Items were administered to SSc patients between August 2004 and January 2005. The study was approved by the Johns Hopkins University School of Medicine Internal Review Board, and informed consent was obtained from all participants.

Validation Sample. The validation sample consisted of SSc patients enrolled in the Canadian Scleroderma Research Group's (CSRG's) multi-center Registry who completed the BCSS between July 2008 and May 2013 with fewer than three items missing. Registry patients must have a diagnosis of SSc from a Registry rheumatologist, be $\geq$18 years of age, and be fluent in English or French. Only patients who completed the BCSS in English were included in the present study. Over 98% of patients in the Registry meet the 2013 ACR/EULAR classification criteria for SSc (Alhajeri et al., 2013; van den Hoogen et al., 2013). The CSRG Registry was approved by the Research Ethics Office of McGill University. All patients provided informed consent.

In both the development and validation samples, some patients completed study measures more than once, but only data from the first administration were analyzed.

Measures

Disease-related Characteristics. Disease duration was measured in years since onset of the first non-Raynaud's Phenomenon disease symptoms. Limited SSc was defined as skin involvement distal to the elbows and knees only, whereas diffuse SSc was defined as skin involvement proximal to the elbows and knees, and/or the trunk (LeRoy et al., 1988).

The Body Concealment Scale for Scleroderma (BCSS). The preliminary version of the

BCSS administered to the development sample was comprised of 13 items, including three items from the BIAQ (Rosen et al., 1991) and 10 newly developed items that reflected SSc-specific body concealment behaviours. Items retained from the BIAQ included three of nine items on the BIAQ Clothing factor: *I wear clothes I do not like*; *I avoid wearing revealing clothes (e.g., bathing suits, tank tops, shorts);* and *I wear clothes that will divert attention from my appearance* ("appearance" changed from "weight" in the original BIAQ). All other BIAQ items were unrelated to concealment. Ten newly developed SSc-specific concealment-related items included: *I wear long sleeves to hide skin changes*; *I wear clothes that hide the changes to my skin; I wear gloves to hide my hands; I wear make-up to hide skin discoloration; I avoid shaking hands with people; I get regular manicures; I wear high-necked shirts; I hide my hands so that people don't see them; I consider having plastic or reconstructive surgery;* and *I avoid directly giving change or other items to people*. Analogous to the BIAQ, BCSS item response categories are scored on a 0-5 scale and reflect frequency of current behaviours (*never, rarely, sometimes, often, usually, always).*

The Brief-Satisfaction with Appearance Scale (Brief-SWAP). The six-item Brief-SWAP (Jewett et al., 2010), which was adapted from the original 14-item SWAP (Lawrence et al., 1998), assesses dissatisfaction with appearance and social discomfort related to visible differences from SSc. Items are scored on a seven-point scale ranging from 0 (*strongly disagree)* to 6 (*strongly agree*) and are summed to create subscale scores (Dissatisfaction with Appearance and Social Discomfort). Higher scores indicate greater dissatisfaction or social discomfort. In a study of 654 CSRG Registry patients (Jewett et al., 2010), internal consistency reliability was good for both subscales (α = .81 for both). In the present study, α = .80 in the validation sample for both subscales. The Brief-SWAP subscales were used to assess convergent validity, with each subscale hypothesized to correlate strongly with the BCSS total score (i.e., $r \geq 0.50$), and with the Social Discomfort subscale hypothesized to correlate more strongly than the Dissatisfaction with Appearance subscale.

Center for Epidemiologic Studies Depression Scale (CES-D). The 20-item CES-D assesses frequency of depressive symptoms during the past week on a 0-3 scale (*rarely, none of the time, most of the time, all of the time*). The CES-D had good internal consistency reliability ($\alpha = .87$) and convergent validity with related self-report measures among 470 CSRG Registry patients (Thombs, Hudson, Schierir, Taillefer, & Baron, 2008) and good internal consistency reliability ($\alpha = .79$) in the present validation sample. The CES-D was included as a measure of discriminant validity and to assess the degree to which the BCSS specifically measures body image-related distress versus general psychological distress as reflected by CES-D scores. It was hypothesized to have a moderate correlation (i.e., $.30 \leq r < .50$) with the BCSS total score.

Patient Health Questionnaire-9 (PHQ-9). The nine-item PHQ-9 assesses frequency of depressive symptoms over the past two weeks on a 0-3 scale (*not at all* to *nearly every day).* The PHQ-9 had good internal consistency reliability ($\alpha = .87$) and convergent validity in a study of 566 CSRG Registry patients (Milette, Hudson, Baron, & Thombs, 2010) and good internal consistency reliability ($\alpha = .88$) in the present validation sample. As with the CES-D, the PHQ-9 was used to assess the degree to which the BCSS specifically measures body image distress versus more general psychological distress, with a moderate correlation (i.e., $.30 \leq r < 0.50$) hypothesized.

McGill Pain Questionnaire (MPQ). The MPQ (Melzack, 1987) includes 15 descriptors that reflect sensory (11 items) and affective (four items) pain, with higher scores reflecting greater pain. Each descriptor is ranked on a four-point intensity scale (0-3; *none* to *severe*), and total scores range from 0-45. The MPQ has been used extensively in various populations and has excellent psychometric properties (Melzack, 1987), including good internal consistency reliability ($\alpha = .92$) in the present validation sample. The MPQ was used to examine discriminant validity and was hypothesized to have a relatively low correlation (i.e., $r < .30$) with the BCSS total score because patients with worse disease may experience greater pain and body image distress; however, it is likely that this relationship will be less robust compared to self-report measures of body image and depressive symptoms.

Data Analysis

Preliminary Assessment of BCSS Items in the Development Sample. Preliminary item analysis was conducted with the initial 13-item version of the BCSS to determine items to be administered in the CSRG Registry validation sample. Items from the preliminary version were administered in the validation sample if the corrected item-total correlations (i.e., correlations of each item with the total from all other items) were ≥ .40 and if removal did not substantively reduce internal consistency reliability (DeVellis, 2012). Items were examined and removed iteratively, one at a time, beginning with the items with the lowest item-total correlations.

Assessment of the Factor Structure of the BCSS in the Validation Sample. Confirmatory factor analysis (CFA) was conducted with Mplus, Version 7 in the validation sample, and three alternative models of the factor structure were tested: a single-factor model, a two-factor model (consistent with expected concealment strategies related to Concealment of Hands and Concealment with Clothing), and a bifactor model.

Bifactor models evaluate the degree to which covariance among a set of item responses can be accounted for by a single general factor that reflects common variance among all scale items, as well as specific factors reflecting additional common variance among clusters of items with similar content (Cook, Kallen, & Amtmann, 2009; Reise, 2012; Reise, Moore, & Haviland, 2010). The bifactor model can provide a useful alternative to standard correlated factor models for assessing aspects of multidimensionality and can be useful for providing possible explanations when there is not clarity about dimensionality. The bifactor model can also be useful for evaluating whether a unit-weighted composite score for a single latent trait can be reasonably interpreted, versus creating subscales, in the context of identifiable multidimensionality (Cook et al., 2009; Reise, 2012; Reise et al., 2010).

In the bifactor model, the general factor represents the broad overarching construct that is being measured, and the group factors represent more narrowly defined subdomains (Cook et al., 2009). In the case of the BCSS, the general factor represented the use of strategies to conceal disfiguring aspects of appearance. The subdomains reflected the more specific subscales, represented by the two proposed factors, Concealment of Hands and Concealment with Clothing (Cook et al., 2009). In the bifactor model, all items were specified to load on the general factor plus their designated specific factor (Concealment of Hands or Concealment with Clothing), and the general and specific factors were specified to be orthogonal. In order to assess the contribution of the general factor and the specific factors to explaining item covariance, we calculated explained common variance (ECV), which is the ratio of variance explained by the general factor divided by variance explained by the general plus the specific factors (Cook et al., 2009). In addition, coefficient omega was generated for the full scale BCSS, which is a model-based reliability estimate analogous to coefficient alpha. Coefficient omega hierarchical was also calculated for the general factor and the two specific factors, in order to evaluate the degree to which the subscales reflected reliable variance beyond variance captured by the general factor (Cook et al., 2009).

For all models, item responses for the BCSS were modeled as ordinal Likert data in Mplus, using the weighted least squares estimator with a diagonal weight matrix and robust standard errors and a mean- and variance-adjusted chi-square statistic with delta parameterization (Muthén & Muthén, 2012). A full information maximum likelihood (FIML) procedure was used to estimate models that included patients with missing data. Modification indices were used in an attempt to identify pairs of items within scales for which model fit would improve if error estimates were freed to covary and for which there appeared to be theoretically justifiable shared method effects (e.g., similar wording; McDonald & Ringo Ho, 2002). A chi-square goodness-of-fit test and three fit indices were used to assess model fit, including the Tucker-Lewis Index (TLI; Tucker & Lewis, 1973), the Comparative Fit Index (CFI; Bentler, 1990) and the Root Mean Square Error of Approximation (RMSEA; Steiger, 1990). Since the chi-square test is highly sensitive to sample size and can lead to the rejection of well-fitting models, practical fit indices were emphasized (Reise, Widaman, & Pugh, 1993). Models with a TLI and CFI close to .95 or higher, and RMSEA close to .06 or lower are representative of good fitting models (Hu & Bentler, 1999). A CFI of .90 or above (Kline, 2005) and a RMSEA of .08 or less (Browne & Cudeck, 1993) may also be considered to represent reasonably acceptable model fit.

Assessment of the Reliability and Validity of the BCSS in the Validation Sample. Cronbach's alpha was computed for the BCSS total score to measure internal consistency reliability. Convergent and discriminant validity were assessed by calculating Pearson's bivariate correlations with 95% confidence intervals of the BCSS total summed score and the summed scores of the items on the two proposed factors with the Brief-SWAP subscale scores, and CES-D, PHQ-9, and MPQ total scores. These analyses were conducted using SPSS, Version 20.

Results

Sample Characteristics

As shown in Table 1, in the development sample, 93 patients had complete data on all initial BCSS items and were included in analyses. Eleven patients who were missing one to three items were not included in analyses. The majority of patients were women (90%), White (85%), and had limited SSc (76%). Mean age was 54.8 years (*SD* = 11.6).

There were 742 English-speaking patients in the CSRG validation sample included in analyses (730 with no missing items on the BCSS and 12 with one missing item). Three patients were excluded because they were missing more than three items. The majority of patients were women (88%), White (88%), and had limited SSc (71%). Mean age was 56.9 years (*SD* = 11.9). See Table 1.

Preliminary Item Analysis

Four of the original 13 BCSS items had corrected item-total correlations < .40 and were not administered in the CSRG validation sample as their removal did not negatively affect internal consistency reliability. These excluded items were: (1) *I get regular manicures,* (r = -.03), (2) *I consider having plastic or reconstructive surgery* (r = .12), (3) *I wear high-necked shirts* (r = .24), and (4) *I wear make-up to hide skin discoloration* (r = .33). This left nine items with corrected item-total correlations ≥ .40 for possible inclusion in the final version of the BCSS (α = .79).

Assessment of the Factor Structure of the BCSS

Model fit for the single-factor CFA was good based on the CFI and TLI, but less than ideal based on the RMSEA ($\chi^2(27)$ = 503.96, CFI = .95, TLI = .94, RMSEA = .15). The two-factor model, consisting of items assigned based on content to either a Concealment of Hands or

Concealment with Clothing subscale fit well ($\chi^2(26) = 156.63$, CFI = .99, TLI = .98, RMSEA = .08). The correlation between the Concealment with Clothing and Concealment of Hands latent factors was .79.

Model fit was also strong for the bifactor model, ($\chi^2(19) = 106.99$, CFI = .99, TLI = .99, RMSEA = .08). Coefficient omega was .88, and coefficient omega hierarchical for the general factor was .80. The coefficient omega was lower for the Concealment of Hands ($\omega = .33$) and negligible for the Concealment of Clothing ($\omega = .01$) factors. No item error estimates were freed to covary in any models. Factor loadings, parameter estimates, and item statistics for all models are shown in Table 2.

Assessment of the Reliability and Validity of the BCSS Total Score

Cronbach's alpha for the BCSS total score was .88. All convergent and discriminant validity correlations were consistent with hypotheses. Specifically, the BCSS total score and the Concealment with Clothing and Concealment of Hands subscale scores correlated strongly with the Social Discomfort subscale of the Brief-SWAP, moderately to strongly with the Brief-SWAP Dissatisfaction with Appearance subscale, moderately with the PHQ-9 and CES-D, and somewhat less with the MPQ. See Table 3.

Discussion

In the present study, we found that a single-factor model of the 9-item BCSS fit well based on the CFI and TLI fit indices, but not based on the RMSEA. A two-factor model, which specified Concealment with Clothing and Concealment of Hands factors, provided a better fit to the data. However, the correlation between the two factors was high (.79). Consistent with this, convergent and discriminant validity correlations with other measures were similar for the BCSS total score and the two subscale scores.

The bifactor model can be useful in contexts where multidimensionality is identified, but when it is not clear whether a multidimensional framework adds meaningfully to a single score approach. When the omega coefficient for the general factor is high, a single summed score can be interpreted as reflecting a single common source, even when data are multidimensional (Reise, 2012). Analysis with the bifactor model in our context identified a strong general factor that explained a large proportion of item covariance. On the other hand, reliability indices calculated for the specific Concealment with Clothing and Concealment of Hands factors were very low, suggesting that their interpretation as reliable indicators of unique constructs would be extremely limited, and that little reliable variance exists beyond that explained by the general factor (Reise, 2012). The presence of identifiable multidimensionality alone does not necessitate the creation and scoring of subscales or substantively hinder the interpretability of a single unit-weighted composite score when the general factor accounts for a large proportion of item covariance, but the specific factors do not. Thus, the totality of the evidence from the present study supports the use and interpretation of a single summed score for the BCSS.

Results from this study build upon pre-existing measures that assess behaviours related to body image distress and self-consciousness (Carr et al., 2000; 2005) body image avoidance among individuals with weight concerns (Rosen et al., 1999) and more general body image concealment issues (Davidson & McCabe, 2005), to provide a mechanism to assess body concealment related to avoidance among individuals with visible difference due to SSc. This addresses an important gap in the study of body image concerns related to acquired disfigurement from medical illnesses like SSc and will be useful in assessing programs designed to address body image concerns in this patient group. Social interaction skills training programs (Kapp-Simon, Simon, & Kristovich, 1992; Robinson, Rumsey, & Partridge, 1996) and cognitive behavioural therapy (CBT; e.g., Newell, 1999) have been recommended as strategies to reduce avoidance behaviours and body image distress among individuals with visible differences. These interventions focus on teaching techniques that allow people with a visibly different appearance to effectively anticipate and manage the reactions of others and to increase their own confidence and self-esteem in social settings, as well on helping these individuals re-evaluate the importance of appearance for their self-worth (Rumsey & Harcourt, 2012). A review of interventions for individuals with visible differences (Bessell & Moss, 2007) found some evidence in support of social interaction skills training and CBT, although there remains a lack of strong evidence for any one particular therapeutic intervention (Rumsey & Harcourt, 2012). Furthermore, none of these types of interventions have been tested in patients with visible differences resulting from SSc and none address the particular concealment behaviours relevant to the disease, such as those related to hiding of the hands or covering up the skin. The development of the BCSS is a first step toward better assessment of body image avoidance behaviours through the mechanism of concealment in SSc. Future research is needed regarding interventions that address such avoidance behaviours as a result of anxiety related to appearance changes in SSc, and currently, the Scleroderma Patient-centered Intervention Network (SPIN) is developing an online intervention for this purpose (Thombs et al., 2012).

There are limitations that should be considered when interpreting results from this study. First, both the development and validation samples were convenience samples of SSc patients receiving treatment at either the Johns Hopkins Scleroderma Center or from centers involved in the CSRG Registry, and results may depart to some degree from what might be found in a community setting and for different SSc patient groups, given the fact that our study was conducted with cases from such specialty centers. Second, items for the BCSS were generated by a group of experts in body image research, but did not include patients with SSc; therefore, no direct feedback was given on the items from people living with the disease. While it would have been preferable to involve SSc patients in the development of BCSS items, as a first step, the approach taken was able to demonstrate that the items generated worked well. Third, female and male participants were combined in the current study, and it is possible that there are differences between sexes in terms of the manifestations of body concealment behaviours. However, there were too few men in the study to test separate factor structures by sex. Finally, the majority of our sample (88%) was White, and as such, our results may not be representative of the experience of body concealment among individuals from different racial backgrounds.

In sum, body concealment related to avoidance is a significant issue for individuals with visible differences. Prior to this study, there were no validated measures of body concealment behaviours and no measures that address the unique appearance changes central to SSc, such as disfigurement of the hands and skin. The present study involved the development and validation of a brief, disease-specific measure to assess specific concealment behaviours relevant to SSc, the BCSS. Results from this study highlight body concealment as an important mechanism of avoidance for people with SSc and provide evidence for the validity of the BCSS.

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Table 1.

Sociodemographic and Disease Variables for Scleroderma Patients in Development and Validation Sample

Sociodemographic Variables	Johns Hopkins (Development Sample)	Canadian Scleroderma Research Group (Validation Sample)
Age (*M, SD)*	54.8 (12.1)[a]	56.9 (11.9)
Female *(n, %)*	82 (90)[b]	650 (88)
White *(n, %)*	77 (85)[b]	649 (88)
Greater than High School Education (*n, %)*	63 (68)	380 (51)[c]
Employed *(n, %)*	42 (46)[d]	295 (40)[e]
Married or Living as Married *(n, %)*	64 (69)	443 (60)
Disease Variables		
Diffuse Scleroderma *(n, %)*	22 (24)	195 (28)[f]
Disease Duration in Years (*M, SD)*	12.5 (8.1)[g]	12.9 (9.4)[h]

Note. Development Sample N = 93. *Validation Sample N* = 742. Due to missing values: [a]*N* = 83; [b]*N* = 91; [c]*N* = 741; [d]*N* = 92; [e]*N* = 738; [f]*N* = 693; [g]*N* = 73; [h]*N* = 732.

Table 2.

Confirmatory Factor Analysis Models and Parameter Estimates for BCSS[a]

		Single-Factor Model	Two-Factor Model		Bifactor Model			
Item	*M* (*SD*)	Factor Loading[b]	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	Factor Loading:[b] General	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	θ[e]
1. I wear clothes I do not like	.45 (.90)	.67	.70		.77	-.34		.53
2. I wear long sleeves to hide skin changes	.73 (1.26)	.84	.84		.83	.14		.54
3. I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts)	1.60 (1.86)	.82	.82		.79	.23		.57
4. I wear clothes that hide the changes to my skin	1.01 (1.48)	.95	.95		.92	.36		.38
5. I wear clothes that will divert attention from my appearance	.67 (1.24)	.83	.86		.87	.05		.49
6. I wear gloves to hide my hands	.50 (1.08)	.73		.75	.50		.57	.65
7. I avoid shaking hands with people	.80 (1.40)	.79		.85	.69		.47	.55
8. I hide my hands so that people don't see them	.90 (1.40)	.82		.82	.65		.62	.43
9. I avoid directly giving change or other items to people	.52 (1.15)	.83		.89	.73		.48	.49

Note. *N* = 742. [a]BCSS responses are scored on a six-point scale ranging from 0-5 (0 = never, 1 = rarely, 2 = sometimes, 3 = often, 4 = usually, 5 = always). [b]Factor loadings are unstandardized, raw factor loadings. [c]Clothing = Concealment with Clothing factor. [d]Hands = Concealment of Hands factor. [e]θ = Square root of the error variance. Sum of error variances = 6.55. ECV (explained common variance) = .79. Coefficient ω (omega) = .88. Coefficient $\omega_{H\text{-}GEN}$ (omega hierarchical – general = .80. $\omega_{H\text{-}Clothing}$ = .01. $\omega_{H\text{-}Hands}$ = .33. PUC (percent of contaminated correlations) = 56%.

Table 3.

Correlations and 95% Confidence Intervals of Summed BCSS Score and Summed Subscale Scores with Other Patient-reported Outcome Measures

Measure	BCSS Total	Concealment with Clothing Subscale	Concealment of Hands Subscale
Brief-Satisfaction with Appearance: Social Discomfort Subscale	.59 (.54 - .63)	.50 (.44 - 0.55)	.57 (.52 - .61)
Brief-Satisfaction with Appearance: Dissatisfaction with Appearance Subscale	.53 (.48 - .58)	.49 (.44 - 0.55)	.47 (.41 - .53)
Patient Health Questionnaire-9	.39 (.32 - .45)	.36 (.29 - .42)	.35 (.28 - .41)
Center for Epidemiologic Studies Depression Scale	.39 (.33 – .45)	.36 (.30 - .43)	.34 (.27 - .40)
McGill Pain Questionnaire	.30 (.22 - .38)	.26 (.17 - .34)	.29 (.21 - .37)

Linking Studies One and Two

Body concealment has been identified as a common coping mechanism among people with visible differences who experience body image distress and concerns about negative social evaluation from others (Clarke et al., 2014; Rumsey & Harcourt, 2012). The first study in the present thesis highlighted how body concealment is also a significant means for body image avoidance among individuals with SSc, particularly in relation to disfiguring appearance changes to the hands and skin. As a result, the Body Concealment Scale for Scleroderma (BCSS) was developed as a new disease-specific patient-reported outcome measure to address body concealment behaviours relevant to people with SSc, and its basic psychometric properties were established. In particular, it was found that the BCSS demonstrated strong internal consistency reliability and construct validity. Despite identifiable multidimensionality, with BCSS items pertaining to separate Concealment of Hands and Concealment with Clothing factors, a bifactor model fit the data best, including a strong general factor that explained a large proportion of item covariance and low reliability indices for the separate group factors. As such, evidence from Study One of this thesis suggested that the BCSS be scored as a single, summed total.

All research findings, including those related to measurement, are subject to chance and should be replicated (Thompson, 2004). Therefore, findings from the BCSS development study regarding the psychometric properties of this newly developed tool should be re-examined in a separate sample of patients with SSc. As such, this constituted the first objective of the second study of this thesis.

Part of establishing evidence of the validity of any patient-reported outcome measure involves determining its measurement equivalency across diverse subgroups of potential respondents (Mokkink et al., 2010). Measurement equivalency occurs when individuals from different groups with similar levels of a construct being measured obtain similar scores on the measure and respond similarly to individual items of the measure (Mokkink et al., 2010). It is only when measurement equivalency is established for diverse groups on a given measure that results from these individuals can be combined within the same research study (Mokkink et al., 2010). Establishing measurement equivalency becomes of central importance in the context of rare diseases like SSc, where researchers increasingly combine results across groups with distinct characteristics in order to attain large enough sample sizes. The development study of the BCSS did not examine the degree to which items on the measure function equally across characteristics of SSc patients that might be expected to influence scores, such as age and sex, as it is well documented in the visible difference literature that younger individuals and those of the female sex tend to report more body image distress and appearance-related concerns (Clarke et al., 2014; Rumsey, Clarke, White, Wyn-Williams, & Garlick, 2004; Tiggeman, 2004; Thompson & Kent, 2001). As such, a second objective of Study Two was to evaluate the measurement equivalency of the BCSS items across age groups and sex in a large, international cohort of SSc patients.

Chapter 3

Study Two

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Validation of the Body Concealment Scale for Scleroderma (BCSS): Replication in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort

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Abstract

Body concealment is an important component of appearance distress for individuals with disfiguring conditions, including scleroderma (SSc). The objective was to replicate the validation study of the Body Concealment Scale for Scleroderma (BCSS) among 897 SSc patients. The factor structure of the BCSS was evaluated using confirmatory factor analysis, and the Multiple-Indicator Multiple-Cause (MIMIC) model examined differential item functioning of BCSS items for sex and age. Internal consistency reliability was assessed via Cronbach's alpha. Construct validity was assessed by comparing the BCSS with a measure of body image distress and measures of mental health and pain intensity. Results replicated the original validation study, where a bifactor model provided the best fit. The BCSS demonstrated strong internal consistency reliability and construct validity. Findings further support the BCSS as a valid measure of body concealment in scleroderma and provide new evidence that scores can be compared and combined across sexes and ages.

Keywords: systemic sclerosis; scleroderma; disfigurement; body image avoidance; body concealment; measurement

Validation of the Body Concealment Scale for Scleroderma (BCSS): Replication in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort

Systemic sclerosis (SSc), or scleroderma, is a rare, chronic, autoimmune connective tissue disease with no known cure. Onset of SSc is highest between the ages of 30-50 years, and women are four to five times more likely to be diagnosed with SSc than men (Mayes, 2003). Some research suggests that individuals from non-White racial/ethnic backgrounds have higher prevalence rates of SSc than White individuals; however, this evidence is limited (Mayes, 2005). Disfiguring appearance changes, including hand contractures, digital ulcers, telangiectasias, hyper- and hypo-pigmentation of the skin, and altered facial features, are common among many SSc patients (Boin & Wigley, 2012). These appearance changes often affect highly visible body parts that are important for social interactions, such as the face, mouth, and hands. The psychological and psychosocial impact of visible differences experienced by people with SSc can be significant, and no treatments alleviate disfiguring disease manifestations. Appearance changes in SSc are associated with body image dissatisfaction, poor appearance self-esteem, social discomfort, and depressive symptoms (Benrud-Larson et al., 2003; Jewett et al., 2012; van Lankveld et al., 2007).

Among people with visible differences, appearance concerns can generate fear of negative evaluation and social anxiety, which, in turn, can lead to avoidance behaviours, such as social withdrawal or concealment of body parts (Newell, 1999; Rumsey & Harcourt, 2004). Body concealment is used as a coping strategy among many individuals with visible differences. People with SSc experience unique appearance changes, particularly skin- and hand-related changes; thus, it is important to assess concealment behaviours specific to SSc in order to increase understanding about the etiology and consequences of these concerns and to improve the scope of clinical assessment of such concerns. The brief self-report measure, the Body Concealment Scale for Scleroderma (BCSS; Jewett et al., 2016), was developed to assess disease-specific body concealment behaviours related to appearance changes in SSc. The initial validation of the BCSS (N = 742) found that a bifactor structure fit best. The BCSS had two identifiable factors (Concealment with Clothing and Concealment of Hands), but a general factor explained the majority of item covariance, supporting the use of a single total score (Jewett et al., 2016). The BCSS total score demonstrated strong internal consistency reliability (α = .88) and good construct validity (Jewett et al., 2016).

All research findings, including those related to measurement, are subject to error and chance, and should therefore be replicated (Thompson, 2004). As such, the first objective of the present study was to replicate findings from the initial BCSS validation study in a large, international SSc cohort. The second objective was to examine the measurement equivalency of the BCSS across sex and age. Measurement equivalency occurs when individuals from different groups (e.g., females and males) with similar levels of a construct being measured obtain similar scores on the measure and respond similarly to individual items of the measure (Mokkink et al., 2010). Differential item functioning (DIF), on the other hand, occurs when an item has different measurement properties for one group compared to another, apart from any true differences in the construct being measured (Mokkink et al., 2010). DIF is generally evaluated by identifying differences in individual item scores across groups that remain present after controlling for levels of the overall construct being measured (e.g., body concealment behaviours; Mokkink et al., 2010). When DIF is present, it is assumed that scores on the item are affected by group characteristics that are not directly related to the construct being measured (Mokkink et al., 2010). Measurement equivalency is a central component of assessing the validity of any measure, and it becomes especially important in the context of rare diseases like SSc, where researchers increasingly combine results across groups with distinct characteristics in order to attain large enough sample sizes. However, results should only be combined if measurement equivalency is established (Mokkink et al., 2010). To date, the measurement equivalency of the BCSS has not been established for characteristics that might be expected to influence scores, such as age and sex, as it is well documented in the visible difference literature that younger individuals and those of the female sex tend to report more body image distress and appearance-related concerns (Clarke et al., 2014; Rumsey et al., 2004; Tiggeman, 2004; Thompson & Kent, 2001).

Consistent with Jewett et al. (2016), we hypothesized that a bifactor model would provide the best fit for the BCSS. Further, we hypothesized that the BCSS total score would correlate strongly (i.e., $r \geq .50$) with the Brief-Satisfaction with Appearance (Brief-SWAP) Social Discomfort subscale score; to a moderate or strong degree (i.e., $r \geq .30$ to $\leq .50$) with the Brief-SWAP Dissatisfaction with Appearance subscale score, Brief Fear of Negative Evaluation (BFNE-II) total score, Social Interaction Anxiety Scale (SIAS-6) total score, and the Patient-Reported Outcomes Measurement Information System (PROMIS-29v2) Health Profile depression domain score; and to a small to moderate degree (i.e., $r < .30$) with the PROMIS-29v2 pain intensity domain score.

Method

Patients and Procedure

The sample consisted of patients enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort (Kwakkenbos et al., 2013) who completed baseline study questionnaires between April 2014 and February 2016. Patients were enrolled at 27 SPIN centers in Canada, the USA, and the UK. To be eligible for the SPIN Cohort, patients must be classified as having SSc according to the 2013 American College of Rheumatology/European League Against Rheumatism classification criteria (van den Hoogen et al., 2013), be ≥ 18 years of age, have the ability to give informed consent, be fluent in English or French, and be able to respond to questionnaires via the Internet. The SPIN sample is a convenience sample. Eligible patients are invited by physicians or supervised nurse coordinators from SPIN centers to participate in the SPIN Cohort, and written informed consent is obtained. Local SPIN physicians or nurse coordinators complete an online medical data form to initiate registration. An automated welcoming email is sent to participants with instructions for activating their SPIN account and completing SPIN Cohort measures. SPIN Cohort patients complete outcome measures via the Internet upon enrollment and subsequently every three months (Kwakkenbos et al., 2013). SSc patients in the SPIN Cohort who had complete data at their baseline assessment for all BCSS items were included in the present analyses. The SPIN Cohort study was approved by the Research Ethics Committee of the Jewish General Hospital, Montreal, Canada and by the research ethics committees of each participating center.

Measures

Sociodemographic information was collected via patient self-report, and disease characteristics were obtained from physicians’ reports. Disease duration was defined as years since onset of the first non-Raynaud’s Phenomenon symptoms. Limited SSc was defined as skin involvement distal to the elbows and knees and diffuse SSc as skin involvement proximal to the elbows and knees or the trunk (van den Hoogen et al., 2013).

Body Concealment Scale for Scleroderma (BCSS). The 9-item BCSS assesses body concealment behaviours in SSc (Jewett et al., 2016). BCSS item responses reflect frequency of current behaviours (0 = *never* to 5 = *always*). Higher total scores reflect more concealment behaviours.

Brief-Satisfaction with Appearance Scale (Brief-SWAP). The 6-item Brief-SWAP (Jewett et al., 2010) assesses body image-related distress in SSc and includes two subscales, Dissatisfaction with Appearance and Social Discomfort. Items are scored on a 7-point scale (0 = *strongly disagree* to 6 = *strongly agree*). Higher scores indicate greater dissatisfaction or social discomfort. The Brief-SWAP has strong internal consistency reliability ($\alpha = .81$ for both subscales) and good construct validity (Jewett et al., 2010). In the present study, Cronbach's alpha for the Brief-SWAP Social Discomfort subscale was .88 and was .80 for the Brief-SWAP Dissatisfaction with Appearance subscale.

Brief Fear of Negative Evaluation Scale (BFNE-II). The 12-item BFNE-II assesses worry about how one is perceived and evaluated by others (Carleton, Collimore, & Asmundson, 2007). Items are rated on a 5-point scale (0 = *not characteristic of me at all* to 4 = *extremely characteristic of me*). Higher summed total scores indicate greater fear of negative evaluation. The BNFE-II has good internal consistency reliability and validity (Carleton et al., 2007). In the present study, Cronbach's alpha for the BFNE-II was .97.

Social Interaction Anxiety Scale (SIAS-6). The 6-item SIAS-6 assesses anxiety from social interactions (Peters, Sunderland, Andrews, Rapee, & Mattlick, 2012). Items are scored on a 5-point scale (0 = *not at all characteristic of me* to 4 = *extremely characteristic of me*). Higher summed total scores indicate greater interactional anxiety. The SIAS-6 has strong psychometric properties including internal consistency reliability and convergent validity (Peters et al., 2012). In the present study, Cronbach's alpha for the SIAS-6 was .89.

Patient-Reported Outcomes Measurement Information System Health Profile Depression and Pain Domain Scores (PROMIS-29v2). The PROMIS-29v2 assesses patient-reported health status on seven domains, including depression (four items scored on a 5-point scale ranging from 1 = *never* to 5 = *always*), plus a single item for pain intensity (scored on an 11-point rating scale; Hinchcliff et al., 2011). Higher scores represent more of the measured domain, and raw scores are converted into T-scores standardized based on a general US population sample (*M* = 50, *SD* = 10; Hinchcliff et al., 2011). The PROMIS-29v2 has strong psychometric properties, including construct validity (Hinchcliff et al., 2011). In the present study, Cronbach's alpha for the PROMIS-29v2 depression domain score was .93 and was .97 for the pain domain score.

Data Analysis

The factor structure of the BCSS was evaluated first in the total sample using confirmatory factor analysis (CFA). Given that in the original BCSS development study, single-factor, two-factor (Concealment with Clothing and Concealment of Hands), and bifactor models were tested, the same three models were replicated in the present study. Bifactor models evaluate the degree to which covariance among a set of item responses can be accounted for by a single general factor that reflects common variance among all items, plus specific factors reflecting additional common variance among clusters of items with similar content (Cook et al., 2009; Reise et al., 2010). Bifactor models can also help assess multidimensionality and evaluate whether a unit-weighted composite score for a single latent trait can be reasonably interpreted, versus creating subscales, in the context of identifiable multidimensionality (Cook et al., 2009; Reise et al., 2010). In the bifactor model, all items were specified to load on the general factor in addition to their designated specific factor (Concealment with Clothing and Concealment of

Hands), and the general and specific factors were specified as orthogonal. To assess the contribution of the general and specific factors to explaining item covariance, we calculated explained common variance (ECV), the ratio of variance explained by the general factor divided by variance explained by the general plus specific factors (Reise, 2012). In addition, coefficient omega, a model-based reliability estimate analogous to coefficient alpha, was calculated for the general and specific factors to evaluate the degree to which the subscales reflected reliable variance beyond variance captured by the general factor (Reise, 2012).

Item responses for the BCSS were modeled as ordinal Likert data, using the weighted least squares estimator with a diagonal weight matrix and robust standard errors and a mean- and variance-adjusted chi-square statistic with delta parameterization (Muthén & Muthén, 2012). A chi-square goodness-of-fit test and three fit indices were used, including the Tucker-Lewis Index (TLI; Tucker & Lewis, 1973), Comparative Fit Index (CFI; Bentler, 1990), and Root Mean Square Error of Approximation (RMSEA; Steiger, 1990). Since the chi-square test is highly sensitive to sample size and can lead to the rejection of well-fitting models, practical fit indices were emphasized (Reise et al., 1993). Models with a TLI and CFI close to .95 or higher, and a RMSEA close to .06 or lower, are generally representative of good fitting models (Hu & Bentler, 1999).

In order to determine if the BCSS exhibited DIF for female versus male SSc patients and for younger versus older patients, the Multiple-Indicator Multiple-Cause (MIMIC) model was utilized. MIMIC models for DIF assessment are based on structural equation models, in which the group variable (e.g., sex) is added to the basic CFA model as an observed variable. Therefore, the base MIMIC model consists of the CFA factor model with the additional direct effect of group on the latent factors, which serves to control for group differences on the level of the latent factors (Mokkink et al., 2010). Ideally for DIF assessment, the simplest factor structure with reasonable fit is used.

To assess for potential DIF, the direct effect of group on BCSS items was assessed for each item separately, by regressing the items, one at a time, on group (see Figure 1). Each item was tested separately to determine if there was statistically significant DIF, which is represented by a statistically significant association in the model from group (e.g., sex) to the item, after controlling for any differences in the overall level of the latent factor between groups. If statistically significant DIF was present for one or more items, the item with the largest magnitude of DIF was considered to have DIF, and the link between the group variable (e.g., sex) and that item was included in the model. This procedure was then repeated until none of the remaining items showed statistically significant DIF. Hommel's correction for multiple testing (Hommel, 1988) was applied to determine items with statistically significant DIF.

Once all items with significant DIF were identified, the potential magnitude of all DIF items was evaluated collectively, by conducting comparisons of the difference on the latent factor between groups in the baseline model (not correcting for DIF) and after controlling for DIF. The magnitude of this difference was interpreted using Cohen's effect size suggestions, with ≤ .20 *SD* indicating small, .50 *SD* indicating moderate, and .80 *SD* indicating large differences (Cohen, 1988). CFA and DIF analyses were conducted using Mplus, Version 7.

Cronbach's alpha was computed for the BCSS to assess internal consistency reliability. Construct validity was assessed by calculating Pearson's bivariate correlations of the BCSS total score with the Brief-SWAP subscale scores, PROMIS-29v2 depression and pain domain scores, and BFNE-II and SIAS-6 total scores. These analyses were conducted using SPSS, Version 20. Because some SSc patients from the SPIN Cohort could have been included in the Canadian

Scleroderma Research Group (CSRG) Registry sample used in the original BCSS validation study, we also calculated the maximum possible patient overlap between the two samples based on sites that contribute data to both cohorts. As a sensitivity analysis, we replicated analyses after removing patients from CSRG sites who could have possibly been in the SPIN sample, as it was not possible to determine patient-by-patient overlap.

Results

Sample Characteristics

Of 943 patients, 897 had complete BCSS item data and were included in present analyses. The majority of the sample was female (*N* = 782, 87%) and White (*N* = 774, 86%). Mean age was 55.4 years (*SD* = 12.1), and 41% of patients had diffuse SSc, which is a rate similar to that seen among SSc patients more generally (Mayes, 2005). See Table 1 for a breakdown of all sociodemographic and disease characteristics. There were 214 patients (24%) from CSRG sites whose diagnosis was made before the date of the initial BCSS validation study using CSRG data and who could have possibly been in both cohorts.

Assessment of the Factor Structure (CFA) of the BCSS

Model fit for the single-factor model was suboptimal ($\chi^2(27) = 753.32$, CFI = .94, TLI = .92, RMSEA = .17). Model fit for the two-factor model was substantially better, ($\chi^2(26) = 165.27$, CFI = .99, TLI = .98, RMSEA = .08). The correlation between the Concealment with Clothing and Concealment of Hands latent factors was .75. Model fit was strongest for the bifactor model ($\chi^2(18) = 81.84$, CFI = .99, TLI = .99, RMSEA = .06). The ECV was .79. Coefficient omega for the Concealment with Clothing subscale was .01 and was .34 for the Concealment of Hands subscale. See Table 2 for all model fit parameters.

Assessment of the Differential Item Functioning (DIF) of the BCSS

MIMIC Base Model. Given that for DIF assessment, the simplest factor structure with reasonable fit is used, the base model consisted of the two-factor model of Concealment with Clothing and Concealment of Hands. This two-factor model was extended to include direct effects of the Concealment with Clothing and Concealment of Hands latent factors on sex (female versus male) and age (dichotomized into age bands of less than 60 years, and 61 years and older), separately. These age bands were determined based on an assessment of the age distribution of the present sample.

The MIMIC base models for sex and age both demonstrated strong fit, $\chi^2(33) = 183.60$, $p < .001$, CFI = .99, TLI = .98, RMSEA = .07, and $\chi^2(33) = 183.15$, $p < .001$, CFI = .99, TLI = .98, RMSEA = .07, respectively. Prior to accounting for DIF, male patients had .57 *SD* lower latent scores than female patients for the Concealment with Clothing factor (95% Confidence Interval [CI] -.82 to -.33) and .16 *SD* lower latent scores than female patients for the Concealment of Hands factor (95% CI -.39 to .07). Prior to accounting for DIF, older patients had .22 *SD* lower latent scores than younger patients for the Concealment with Clothing factor (95% CI -.38 to -.06) and .41 *SD* lower latent scores than younger patients for the Concealment of Hands factor (95% CI -.57 to -.25).

DIF Assessment. There were no items that demonstrated statistically significant DIF based on sex. Three items were identified with statistically significant DIF based on age: items 2, (*I wear long sleeves to hide skin changes*), 3 (*I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts)*), and 8 (*I hide my hands so that people don't see them*). More specifically, compared to younger patients, older patients had higher scores (more concealment-related behaviours) than would be expected on items 2 ($z = 2.80$, $p = .005$) and 3 ($z = 3.65$, $p < .001$), based on their latent levels of the Concealment with Clothing factor. Compared to younger patients, older patients had lower scores than would be expected on item 8 (*z* = -2.87, *p* = .004), based on their latent levels of the Concealment of Hands factor.

After correcting for DIF, the difference on the Concealment with Clothing latent factor between younger versus older patients increased from -.22 *SD* (95% CI -.38 to -.06) to -.32 *SD* (95% -.49 to -.15). The difference on the Concealment of Hands latent factor between younger versus older patients decreased slightly from -.41 *SD* (95% CI -.57 to -.25) to -.35 *SD* (95% CI -.52 to -.18). Despite the statistically significant DIF found for three BCSS items, the magnitude was small (all ≤ .20 *SD*s) and did not influence scores substantively. Table 3 shows the baseline CFA parameters, before assessing for DIF, as well as the DIF-corrected model parameters for age.

Assessment of the Reliability and Validity of the BCSS Total Score

Cronbach's alpha for the BCSS total score was .89. Consistent with hypotheses, the BCSS total score was strongly correlated with the Brief-SWAP Social Discomfort subscale (*r* = .63) and moderately correlated with the Brief-SWAP Dissatisfaction with Appearance subscale (*r* = 0.44), BFNE-II total score (*r* = .43), SIAS-6 total score (*r* = .40), and PROMIS-29v2 depression domain score (*r* = .48). The correlation with the PROMIS-29v2 pain intensity score was slightly stronger than hypothesized (*r* = .46).

When analyses were replicated removing possibly overlapping patients from CSRG sites, sociodemographic data, factor analysis results, and reliability and validity indices were virtually unchanged (see Appendix A). Given this, DIF analyses were only run with the main model, including the full sample, as it is reasonable to assume that no differences in terms of the measurement equivalency of BCSS items would emerge if overlapping patients were removed.

Discussion

Results replicated Study One of this thesis, the initial BCSS validation study (Jewett et al., 2016). The bifactor model fit best, with the general factor explaining 79% of item covariance. Reliability indices for the specific factors were very low, suggesting that they do not reliably measure substantive variance beyond that explained by the general factor (Reise, 2012). Thus, consistent with the initial BCSS validation study, evidence from the present study supports the use and interpretation of a single summed score for the BCSS. Beyond this, BCSS items demonstrated measurement equivalency across female and male patients, and while three items had statistically significant DIF for younger versus older patients, the magnitude of the differences was small, and the influence on scores was negligible.

The present study also found evidence of good internal consistency reliability and construct validity for the BCSS, similar to the initial validation study. As expected, scores on the BCSS correlated most strongly with scores on the Brief-SWAP Social Discomfort subscale, and this relationship highlights the social implications of the disfiguring appearance changes experienced by many SSc patients. The moderate correlation found between the BCSS and Brief-SWAP Dissatisfaction with Appearance subscale scores is not surprising, because BCSS items pertain less to body image dissatisfaction, and more to social and behavioural avoidance. Similarly, the moderate correlations found between scores on the BCSS and scores on the BFNE-II, SIAS-6, and PROMIS-29v2 depression domain score likely reflect that these latter measures tap into mental health constructs rather than body image-related avoidance mechanisms.

Evidence provided in the current study points to the utility of the BCSS as a patient-reported outcome in programs designed to address body image concerns among people with SSc, with essentially equivalent measurement properties for both female and male patients and those of different ages. Currently, there are programs being developed (Kwakkenbos et al., 2013) that target disfigurement-related distress and concealment behaviours specific to SSc, building on strategies from social interaction skills training and CBT (Bessell et al., 2012; Clarke et al., 2014) that have been used more generally among individuals with visible differences. The BCSS could be used as an outcome measure to assess body concealment behaviours in such programs.

There are limitations to consider. First, the SPIN Cohort constitutes a convenience sample of SSc patients receiving treatment at SPIN centers, and patients at these centers may differ from those in other settings. SPIN Cohort patients complete questionnaires online, which may further limit the generalizability of findings. Next, the present sample combined both female and male patients, and body concealment behaviours may differ by sex. However, there were not enough men to examine measurement properties separately by sex. Additionally, the majority of our sample (86%) was White; therefore, results may not be generalizable to individuals from non-White racial/ethnic backgrounds. Finally, there was overlap between sites that recruited SSc patients for the SPIN Cohort and the CSRG Registry used in the original BCSS validation study, and it is possible that some patients were included in both. However, even if all patients from those sites were enrolled in both studies, the overlap would have been < 25%. The actual amount of overlap is likely much smaller because the studies were conducted several years apart. Furthermore, results were virtually identical when possibly overlapping patients were excluded.

In summary, the present study replicated previous findings and provided further evidence for the validity of the BCSS by showing that scores can be compared and combined across females and males with SSc, and those of different ages without concern that measurement differences may substantially influence results. The BCSS assesses disease-specific concealment behaviours related to the unique skin and hand appearance changes experienced by SSc patients and highlights body concealment as a pertinent issue for body image and social interactions for individuals with the disease. The BCSS can be used as a clinical assessment tool and as a patient-reported outcome measure in research to assess body concealment behaviours among people with SSc.

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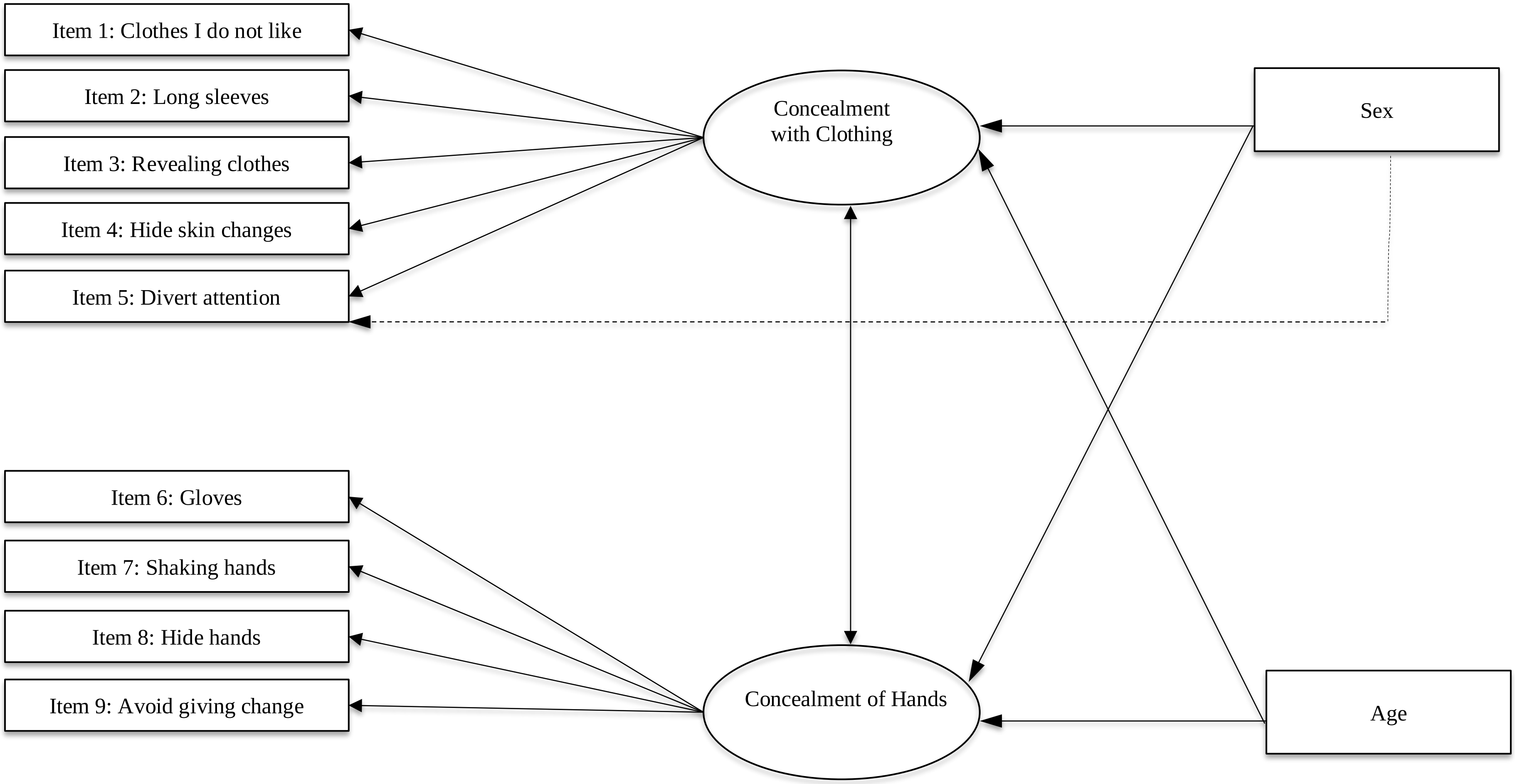
Figure 1. The MIMIC Model for the Body Concealment Scale for Scleroderma (BCSS)



Note. Dotted line represents the potential DIF effect of grouping variable on BCSS items.

Table 1.
Sociodemographic and Disease Variables for Scleroderma Patients

Sociodemographic Variables	
Age (*M, SD)*	55.4 (12.1)[a]
Female *(n, %)*	782 (87.2)
White *(n, %)*	774 (86.3)
Years of Formal Education (*M, SD)*	15.3 (3.2)
Employed Full or Part-time *(n, %)*	357 (64.4)[b]
Married or Living as Married *(n, %)*	654 (72.9)
Disease Variables	
Diffuse Scleroderma *(n, %)*	366 (40.8)
Disease Duration in Years (*M, SD)*	11.6 (8.8)[c]

Note. *N* = 897. Due to missing values: [a]*N* = 894; [b]*N* = 554; [c]*N* = 824.

Table 2.

Confirmatory Factor Analysis Models and Parameter Estimates for BCSS[a]

		Single-Factor Model	Two-Factor Model		Bifactor Model			
Item	*M* (*SD*)	Factor Loading[b]	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	Factor Loading:[b] General	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	θ[e]
1. I wear clothes I do not like	.72 (1.11)	.69	.72		.75	-.21		.62
2. I wear long sleeves to hide skin changes	.80 (1.33)	.86	.88		.84	.23		.50
3. I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts)	1.63 (1.75)	.79	.82		.80	.16		.58
4. I wear clothes that hide the changes to my skin	1.06 (1.46)	.92	.95		.92	.38		.40
5. I wear clothes that will divert attention from my appearance	.80 (1.25)	.82	.86		.89	-.12		.46
6. I wear gloves to hide my hands	.81 (1.28)	.72		.77	.59		.50	.64
7. I avoid shaking hands with people	.92 (1.37)	.78		.83	.60		.61	.52
8. I hide my hands so that people don’t see them	.98 (1.32)	.83		.89	.68		.56	.47
9. I avoid directly giving change or other items to people	.69 (1.22)	.80		.87	.67		.52	.53

Note. $N = 897$. [a]BCSS responses are scored on a 6-point scale ranging from 0-5 (0 = never, 1 = rarely, 2 = sometimes, 3 = often, 4 = usually, 5 = always). [b]Factor loadings are unstandardized, raw factor loadings. [c]Concealment with Clothing factor. [d]Concealment of Hands factor. [e]θ = Square root of the error variance. Sum of error variances = 6.50. ECV (explained common variance) = .79. Coefficient ω (omega) = .89. Coefficient $\omega_{\text{H-GEN}}$ (omega hierarchical – general = .80. $\omega_{\text{H-Clothing}}$ = .01. $\omega_{\text{H-Hands}}$ = .34. PUC (percent of contaminated correlations) = 56%.

Table 3.

Factor Loadings of the BCSS Concealment with Clothing and Concealment of Hands Latent Factors and Influence on the Overall Estimates of Concealment with Clothing and Concealment of Hands Latent Factor Scores for Age

Item	Base Model[a]	DIF Corrected Model[b]
	Factor Loading (95% CI)	Factor Loading (95% CI)
Concealment with Clothing Latent Factor		
1. I wear clothes I do not like.	.72 (.68, .76)	.72 (.68, .76)
2. I wear long sleeves to hide skin changes.	.88 (.86, .90)	.88 (.86, .90)
3. I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts).	.82 (.79, .85)	.83 (.80, .86)
4. I wear clothes that hide the changes to my skin.	.95 (.93, .97)	.94 (.92, .96)
5. I wear clothes that will divert attention from my appearance.	.85 (.82, .88)	.85 (.82, .88)
Concealment of Hands Latent Factor		
6. I wear gloves to hide my hands.	.77 (.73, .81)	.77 (.73, .81)
7. I avoid shaking hands with people.	.82 (.79, .85)	.82 (.79, .85)
8. I hide my hands so that people don't see them.	.89 (.87, .91)	.89 (.87, .91)
9. I avoid directly giving change or other items to people.	.86 (.83, .89)	.86 (.83, .89)
Correlation of Concealment with Clothing and Concealment of Hands Latent Factors	.75 (.71, .79)	.75 (.71, .79)
Direct Effects on Items Attributable to Age		
Item 2. Long sleeves to hide skin changes	---	.17 (.05, .29)
Item 3. Avoid wearing revealing clothes	---	.21 (.10, .32)
Item 8. Hide my hands	---	-.17 (-.29, -.05)
Structural Effect of Age on Latent Factors		
Age on Concealment with Clothing factor	-.22 (-.38, -.06)	-.32 (-.49, -.15)
Age on Concealment of Hands factor	-.41 (-.57, -.25)	-.35 (-.52, -.18)

Note. CI = Confidence Interval. [a]Not corrected for Differential Item Functioning (DIF); [b]Corrected for DIF on items 2, 3, 8.

Linking Studies Two and Three

Study Two of this thesis replicated Study One’s findings related to the psychometric properties of a new patient-reported outcome measure of body concealment behaviours in patients with SSc, the BCSS. It also extended results from the validation study by establishing the measurement equivalency of BCSS items across females and males with SSc and those of different ages. As such, it was determined that, not only can the BCSS be used as a possible clinical assessment tool to address body concealment behaviours among patients with SSc, but item responses across sex and age groups can also be combined in research studies without concern that measurement differences will substantially influence results.

Measurement equivalency is an important component of assessing the validity of any patient-reported outcome measure, and it may be affected by characteristics beyond respondents’ sex and age. For instance, when measures are translated into other languages, it is possible that translations may alter the meaning, interpretation, or severity of individual items (Zumbo, 1999; 2007), thus leading responses to be influenced by such changes, as opposed to any true differences in the construct being measured. Therefore, it is also important to investigate measurement equivalency across diverse linguistic or cultural groups who may complete a given questionnaire as part of an international cohort, for instance, or whose responses may be pooled in research studies, both of which are increasingly common in the context of rare diseases like SSc. For instance, SPIN’s recently established largescale cohort of SSc patients currently completes many measures related to body image and appearance concerns as part of regular assessments and as outcomes for planned trials of psychosocial needs among patients (Kwakkenbos et al., 2013).

One such measure that is presently administered in the SPIN Cohort, as well as within the largescale national CSRG Registry, is the Satisfaction with Appearance Scale (SWAP; Lawrence et al., 1998). The basic psychometric properties of the SWAP have been established in several SSc samples to date (e.g., Heinberg et al., 2007; Jewett et al., 2010, 2012; Mills et al., 2015); however, the measurement equivalency of the tool for different linguistic or cultural groups has yet to be examined. Therefore, the objective of the third study of the present thesis was to extend validity evidence of the SWAP by evaluating the equivalence of item scores across English- and French-speaking SSc patients in a large Canadian cohort, the CSRG.

Chapter 4

Study Three

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Assessment of English-French Differential Item Functioning of the Satisfaction with Appearance Scale (SWAP) in Systemic Sclerosis

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Abstract

The Satisfaction with Appearance Scale (SWAP) has been used to assess body image distress among people with the rare and disfiguring disease systemic sclerosis, or scleroderma (SSc); however, it has not been validated across different language groups. The objective was to examine differential item functioning of the SWAP among 856 Canadian English- or French-speaking SSc patients. Confirmatory factor analysis was used to evaluate the SWAP two-factor structure (Dissatisfaction with Appearance and Social Discomfort). The Multiple-Indicator Multiple-Cause (MIMIC) model was utilized to assess differential item functioning. Results revealed that the established two-factor model of the SWAP demonstrated relatively good fit. Statistically significant, but small-magnitude differential item functioning was found for three SWAP items based on language; however, the cumulative effect on SWAP scores was negligible. Findings provided empirical evidence that SWAP scores from Canadian English- and French-speaking patients can be compared and pooled without concern that measurement differences may substantially influence results.

Keywords: Satisfaction with Appearance Scale; systemic sclerosis; body image; measurement; differential item functioning

Assessment of English-French Differential Item Functioning of the Satisfaction with Appearance Scale (SWAP) in Systemic Sclerosis

Systemic sclerosis (SSc), or scleroderma, is a rare, chronic autoimmune connective tissue disease. SSc is characterized by abnormal fibrotic processes and excessive production of collagen, which commonly manifest in thickening and hardening of the skin (Boin & Wigley, 2012; Mayes, 2008). As a result, people with SSc often develop visible differences in the texture of the skin, in addition to other disfiguring appearance changes including telangiectasias, hand contractures, skin pigmentation changes, and altered facial features (Mayes, 2008). These appearance changes commonly affect body parts that are highly visible and play a central role in social interactions, such as the face, mouth, and hands (Boin & Wigley, 2012; Rumsey & Harcourt, 2005) and are not alleviated by treatments.

Disfiguring aspects of SSc have been associated with concerns about appearance, attempts to conceal affected body parts, social discomfort, and poor psychological functioning (Jewett et al., 2010, 2016; Malcarne et al., 1999; van Lankveld et al., 2007). Increasingly, research examining body image in SSc has focused on the development and validation of measures that accurately reflect disease-specific body image concerns (Jewett et al., 2010, 2016; Mills et al., 2015). One such measure, the Satisfaction with Appearance Scale (SWAP), which was originally developed with burn patients (Lawrence et al., 1998), but has been adapted and validated in SSc, is commonly used as an outcome to assess dissatisfaction with appearance and social discomfort relative to acquired disfigurements from the disease (Jewett et al., 2010, 2012; Mills et al., 2015).

The measurement properties of the SWAP have been examined for SSc patients in several studies (e.g., Heinberg et al., 2007; Jewett et al., 2010, 2012; Mills et al., 2015), including those using the same cohort of patients as the present study (Jewett et al., 2010; 2012), which have collectively established strong evidence of internal consistency reliability, construct validity, and a two-factor structure with Social Discomfort and Dissatisfaction with Appearance factors. However, the degree to which the SWAP generates scores that are equivalent across linguistic or cultural groups has yet to be explored. This is an important consideration because patients who complete the SWAP in countries with more than one common language, such as Canada, or as part of an international cohort (Kwakkenbos et al., 2013), which is often the case in rare diseases like SSc, may do so in different languages (e.g., French, English, Spanish).

Importantly, results pooled across language groups are only valid to the extent that the measurement metric is equivalent, meaning that scores are not affected by linguistic or cultural differences, beyond actual differences in the construct being measured (Mokkink et al., 2010). When the measurement metric is equivalent, individuals from different linguistic groups with similar levels of the construct being measured (e.g., social discomfort) should obtain similar scores on the measure (e.g., SWAP) and respond similarly to individual items of the measure. Differential item functioning (DIF), on the other hand, is said to occur when an item has different measurement properties for one group compared to another, apart from any true differences in the construct being measured (Mokkink et al., 2010). When DIF is present, the scores on the item are likely influenced by group characteristics (e.g., language) that are not directly related to the construct being measured (Mokkink et al., 2010). DIF in cross-linguistic comparisons may occur, for instance, because translations shift meanings, formats, or severity of items used in measures (Zumbo, 1999). As such, it is possible that individuals' endorsement of items reflects these changes, or some other characteristic of the test item or testing situation (Zumbo, 2007), beyond actual levels of the construct being measured (e.g., social discomfort due to appearance changes).

Evaluation of DIF is essential to disambiguate group comparisons and to determine if scoring differences are actual functions of the outcome being measured, versus an artifact of the measurement process, such as interpretation of item meaning (Teresi & Fleishman, 2007). To date, no studies have assessed DIF for the SWAP in SSc for patients from different linguistic groups. As such, the objective of the present study was to assess the equivalence of SWAP item scores across English- and French-speaking SSc patients in a large Canadian cohort.

Method

Patients and Procedure

The study sample consisted of SSc patients recruited from 15 Canadian Scleroderma Research Group (CSRG) Registry sites. To be eligible for the Registry, patients must be at least 18 years of age, classified as having SSc by a CSRG rheumatologist, and fluent in English or French. Patients completed the SWAP in their preferred language. Over 98% of patients in the Registry meet the 2013 ACR/EULAR classification criteria for SSc (Alhajeri et al., 2015). Each year at their annual Registry visit, patients complete clinical evaluations and fill out a series of self-report questionnaires, including the SWAP. Only patients with complete data for all SWAP items were included in this study. The first available assessment with complete SWAP data was included for patients who had filled out the SWAP on more than one occasion. All patients provided informed consent, and the research ethics board of each participating center approved the data collection protocol.

Measures

Sociodemographic and Disease Characteristics. Patients enrolled in the CSRG Registry provided sociodemographic data, including age, sex, race/ethnicity, education level (greater than a high school level of education versus high school or less), marital status (married or living as married versus unmarried). CSRG rheumatologists provided medical information, including disease duration based on time since onset of a patient’s first symptom (calculated for both first Raynaud’s or non-Raynaud’s symptoms) and disease subtype (limited or diffuse cutaneous SSc). Limited cutaneous SSc was defined as skin involvement distal to the elbows and knees only (LeRoy et al., 1988). Patients with sine SSc, which is SSc without skin involvement, were included in the same group as patients with limited cutaneous SSc for the purposes of analysis (Hachulla & Launay, 2011). Diffuse cutaneous SSc was defined as skin involvement proximal to the elbows and knees or the trunk (LeRoy et al., 1988).

The Satisfaction with Appearance Scale (SWAP). The 14-item SWAP was originally designed to measure non-weight related body image concerns among individuals with burn injuries (Lawrence et al., 1998). Items pertain to social discomfort relative to disfigurement (Social Discomfort subscale) and satisfaction with the appearance of particular body parts (Dissatisfaction with Appearance subscale). Respondents rate the degree to which they feel each item reflects their thoughts and feelings about their appearance on a 7-point scale ranging from 0 (*strongly disagree)* to 6 (*strongly agree*). Item scores are summed to calculate total subscale scores, reverse-scoring items pertaining to satisfaction with appearance. Higher scores indicate greater social discomfort and dissatisfaction with appearance (Lawrence et al., 1998). The original SWAP was adapted for SSc by replacing the word “burn” with the word “scleroderma” as indicated (Heinberg et al., 2007). The SWAP has consistently demonstrated strong psychometric properties, including internal consistency reliability estimates (i.e., Cronbach’s alphas ≥ .88 for both subscales) and construct validity (i.e., positive moderate correlations with measures of depressive symptoms, quality of life, physical functioning, and disease severity; and low negative correlations with measures of various dimensions of pain; Jewett et al., 2010, 2012;

Mills et al., 2015). Previous studies of the SWAP in SSc samples have consistently found that a two-factor structure (Social Discomfort and Dissatisfaction with Appearance), but not a one-factor structure, fit the data well (Heinberg et al., 2007; Jewett et al., 2010, 2012).

Data Analysis

Descriptive statistics were calculated for all sociodemographic and disease variables, including means and standard deviations (*SD*s) for continuous variables. Sociodemographic and disease-related variables were compared between English- and French-speaking patients using chi-square tests for categorical variables and *t*-tests for continuous variables.

The factor structure of the SWAP was evaluated first in the total sample using confirmatory factor analysis (CFA). Ideally for DIF assessment, the simplest structure with reasonable fit is used. Given the previously established two-factor structure of Social Discomfort and Dissatisfaction with Appearance for the SWAP (Heinberg et al., 2007; Jewett et al., 2010, 2012), it was expected that a two-factor structure would fit the data reasonably well in the present study.

Item responses for the SWAP are ordinal Likert data, so the weighted least squares estimator with a diagonal weight matrix, robust standard errors, and a mean- and variance-adjusted chi-square statistic was used with delta parameterization (Muthén & Muthén, 2012). Modification indices were used to identify pairs of items within scales for which model fit would improve if error estimates were freed to covary and for which there appeared to be theoretically justifiable shared method effects (e.g., similar wording; McDonald & Ringo Ho, 2002). To assess model fit, the chi-square test, the Tucker-Lewis Index (TLI; Tucker & Lewis, 1973), the Comparative Fit Index (CFI; Bentler, 1990), and the Root Mean Square Error of Approximation (RMSEA; Steiger, 1990) were used. Since the chi-square test is highly sensitive to sample size, it can lead to the rejection of well-fitting models (Reise et al., 1993); therefore, the TLI, CFI, and RMSEA fit indices were emphasized. Good fitting models are indicated by a TLI and CFI ≥ .95 and RMSEA ≤ .06 (Hu & Bentler, 1999).

A general method for assessing DIF involves identifying differences in individual item scores across groups that remain present after controlling for levels of the overall construct being measured (Mokkink et al., 2010). In order to assess if SWAP items exhibited DIF for English- versus French-speaking SSc patients, the Multiple-Indicator Multiple-Cause (MIMIC) model was utilized. MIMIC models for DIF assessment are based on structural equation models, in which the group variable (i.e., language) is added to the basic CFA model as an observed variable. As such, the base MIMIC model consists of the CFA factor model with the additional direct effect of group on the latent factors, which serves to control for group differences on the level of the latent factors (Mokkink et al., 2010).

To assess for potential DIF, the direct effect of group on SWAP items was assessed for each item separately, by regressing the items, one at a time, on language (see Figure 1). Each item was tested separately to determine if there was statistically significant DIF, which is represented by a statistically significant association in the model between language and the item, after controlling for any differences in the overall level of the latent factor between language groups. If statistically significant DIF was present for one or more items, the item with the largest magnitude of DIF was considered to have DIF, and the link between language and that item was included in the model. This procedure was then repeated until none of the remaining items showed statistically significant DIF. Hommel’s correction for multiple testing (Hommel, 1988) was applied.

Once all items with significant DIF were identified, the potential magnitude of all DIF items was evaluated collectively, by conducting comparisons of the difference on the latent factor between groups in the baseline CFA model and after controlling for DIF. The magnitude of this difference was interpreted using Cohen's effect size suggestions, with ≤ .20 *SD* indicating small, .50 *SD* indicating moderate, and .80 *SD* indicating large differences (Cohen, 1988). Both CFA and DIF analyses were conducted using Mplus Version 7 (Muthén & Muthén, 2012), and all other analyses were conducted using SPSS Statistics, Version 22.

Results

Sample Characteristics

In total, 856 SSc patients had complete data for all SWAP items and the language variable. Mean age in the total sample was 56.8 (*SD* = 11.5) years. The majority of patients were female (88%), English-speaking (76%), White (90%), married or living as married (70%), and had limited cutaneous SSc (*N* = 69%). The mean time since onset of the first disease symptom was 16.3 (*SD* = 12.2) years, and the mean time since onset of the first non-Raynaud's disease symptom was 12.2 (*SD* = 9.4) years. The mean SWAP Social Discomfort subscale score was 8.6 (*SD* = 8.8), and the mean Dissatisfaction with Appearance subscale score was 19.1 (*SD* = 12.6). Cronbach's alpha for the SWAP Social Discomfort subscale was .89 and was .92 for the Dissatisfaction with Appearance subscale. There were no statistically significant differences between English- and French-speaking patients on sociodemographic and disease characteristics, or on SWAP subscale scores (see Table 1).

Confirmatory Factor Analysis (CFA)

For the previously established two-factor structure (Social Discomfort and Dissatisfaction with Appearance), model fit was acceptable based on the CFI and TLI indices, but less than acceptable based on the RMSEA, $\chi^2(76) = 1169.68$, $p < .001$, CFI = .96, TLI = .96, RMSEA = .13. Inspection of the modification indices indicated that freeing error terms to covary for SWAP items 1 and 2 ("*Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my family,"* and "*Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my friends"*) would improve model fit, and there was clear overlap in these items' content. Modification indices also revealed that freeing error terms to covary for SWAP items 5 and 6 ("*I feel that my scleroderma is unattractive to others,"* and "*I don't think people would want to touch me"*) would improve model fit as well. Given these modification indices, and the shared content and meaning of the items (McDonald & Ringo Ho, 2002), the model was refitted to the data, allowing the error terms for these items to be correlated. This change resulted in a model with improved fit, $\chi^2(74) = 846.76$, $p < .001$, CFI = .97, TLI = .97, RMSEA = .11; however, it was still slightly less than acceptable based on the RMSEA, although sufficiently good-fitting to establish dimensionality for evaluation of DIF (Mokkink et al., 2010). There were no additional modification indices that would have improved model fit substantively. The correlation between the Social Discomfort and Dissatisfaction with Appearance latent factors was .58.

Differential Item Functioning (DIF)

MIMIC Base Model. To assess possible DIF, the two-factor model was extended to include direct effects of the Social Discomfort and Dissatisfaction with Appearance latent factors on language (English versus French), which demonstrated acceptable fit, $\chi^2(86) = 952.83$, $p < .001$, CFI = .97, TLI = .97, RMSEA = .11. Table 2 shows the baseline CFA model parameters, before assessing for DIF. Prior to accounting for DIF, there were no statistically significant differences between English- and French-speaking patients for either the Social Discomfort factor (95% Confidence Interval [CI] -.10 to .27) or the Dissatisfaction with Appearance factor (95% CI -.12 to .20).

DIF Assessment. Three items were identified with statistically significant DIF based on language: items 1 ("*Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my family"*), 2 ("*Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my friends"*), and 5 ("*I feel that my scleroderma is unattractive to others"*). Compared to English-speaking patients, French-speaking patients had higher scores than would be expected on item 1 ($z = 5.60$, $p < .001$) and item 2 ($z = 2.92$, $p = .003$), and lower scores on item 5 ($z = -6.94$, $p < .001$), based on their latent levels of the Social Discomfort factor.

As shown in Table 2, after correcting for DIF, the difference on the Social Discomfort latent factor between English- and French-speaking patients was non-significant (.09 *SD*, 95% CI -.10 to .27 to .06 *SD*, 95% CI -.13 to .24). As all three DIF items load onto the Social Discomfort factor, there was no difference on the Dissatisfaction with Appearance latent factor after correcting for DIF. Thus, despite the fact that there was statistically significant DIF for three SWAP items, the cumulative DIF effects across these items did not result in substantive differences in estimates of latent factor levels between English- and French-speaking patients.

Discussion

The main finding of this study was that, although three items demonstrated statistically significant DIF for SSc patients who completed the SWAP in English versus French, the cumulative amount of DIF across these items was small, and the effect on overall SWAP Social Discomfort scores was negligible. Given the large number of tests conducted, interpretations for the differences found at the item-level cannot be made with confidence, and replication of item-specific DIF results is necessary. Taken altogether, however, current findings suggest that SWAP scores can be validly compared and summed across Canadian English-and French-speaking patients without concern that outcomes will be influenced substantively by differences in scoring metrics.

In DIF assessments, many analyses are conducted, and, particularly when sample size is large, as in the present study, statistically significant findings are not uncommon. In the present study, because the magnitude of the DIF identified was small and the impact on measurement of the SWAP latent constructs was minimal, we did not seek to identify linguistic differences in items identified with DIF.

As largescale, national and international collaborations in rare chronic diseases like SSc become increasingly common, researchers are often required to integrate data across multiple language groups in order to attain adequate sample sizes. The SWAP is currently used as a key patient-reported outcome measure, for example, in SPIN, which collects data from SSc patients in Canada, the USA, and Europe (Kwakkenbos et al., 2013). The SWAP will serve as a primary outcome in a planned SPIN trial to evaluate an online intervention to help people with SSc cope with appearance concerns and body image distress (Kwakkenbos et al., 2013). Results from the present study provide evidence that SWAP scores in English and French can be reasonably combined. Furthermore, it is reasonable to assume, based on the present findings, that items on the 6-item Brief-SWAP (Jewett et al., 2010) would function similarly in terms of DIF, therefore providing evidence that scores in English and French can be similarly combined for this shorter version of the measure as well.

The present study has limitations that should be considered when interpreting results. First, the CSRG cohort constitutes a convenience sample of SSc patients receiving treatment at one of the sites associated with the Registry. Thus, it was not possible to compare data for patients with complete SWAP results to all eligible patients. Also, results may depart to some degree from what might be found in a community setting and for different SSc patient groups. However, it has been documented that SSc patients included in the CSRG Registry are similar to patients in other large cohorts in terms of features including age and sex distribution (Hudson et al., 2009). Furthermore, the Registry includes SSc patients across the spectrum of disease subtypes and severity levels, thus, there is evidence supporting that patients sampled in the current study are representative of the general population of SSc patients and those seen in rheumatology clinics more broadly (Hudson et al., 2009). An additional limitation of the present study is that the SSc sample was from Canada only, and, as such, the present findings require replication in other samples of French-speaking patients. Relatedly, DIF was only assessed for English- versus French-speaking patients, and not any for other language groups. We used a MIMIC model to assess DIF. This model implicitly assumes configural invariance, which is a strong assumption (Bauer, 2017). In the case of the SWAP, which was designed to reflect two underlying factors and scored as two subscales, this is likely to be a reasonable assumption, particularly since all previous studies have replicated the two-factor structure. Compared to a multi-group CFA model, which can be used to fully test measurement invariance, including configural invariance, the MIMIC model can provide greater power to detect DIF, if present (Bauer, 2017). This was an important consideration in the present study, since there were only 208 French-speaking SSc patients, and coverage of some parts of the item response spectrum would have been sparse if modelled separately.

In sum, the present study provided evidence that the SWAP is a valid measure of Social Discomfort and Dissatisfaction with Appearance and that items do not function differently across

English- and French-speaking SSc patients. There were some SWAP items that exhibited DIF based on language; however, none of the resulting differences influenced overall scores substantively. Thus, scores on the Social Discomfort and Dissatisfaction with Appearance subscales of the SWAP can be compared and pooled across these languages groups. Replications of the current study with other samples of French-speaking patients are needed.

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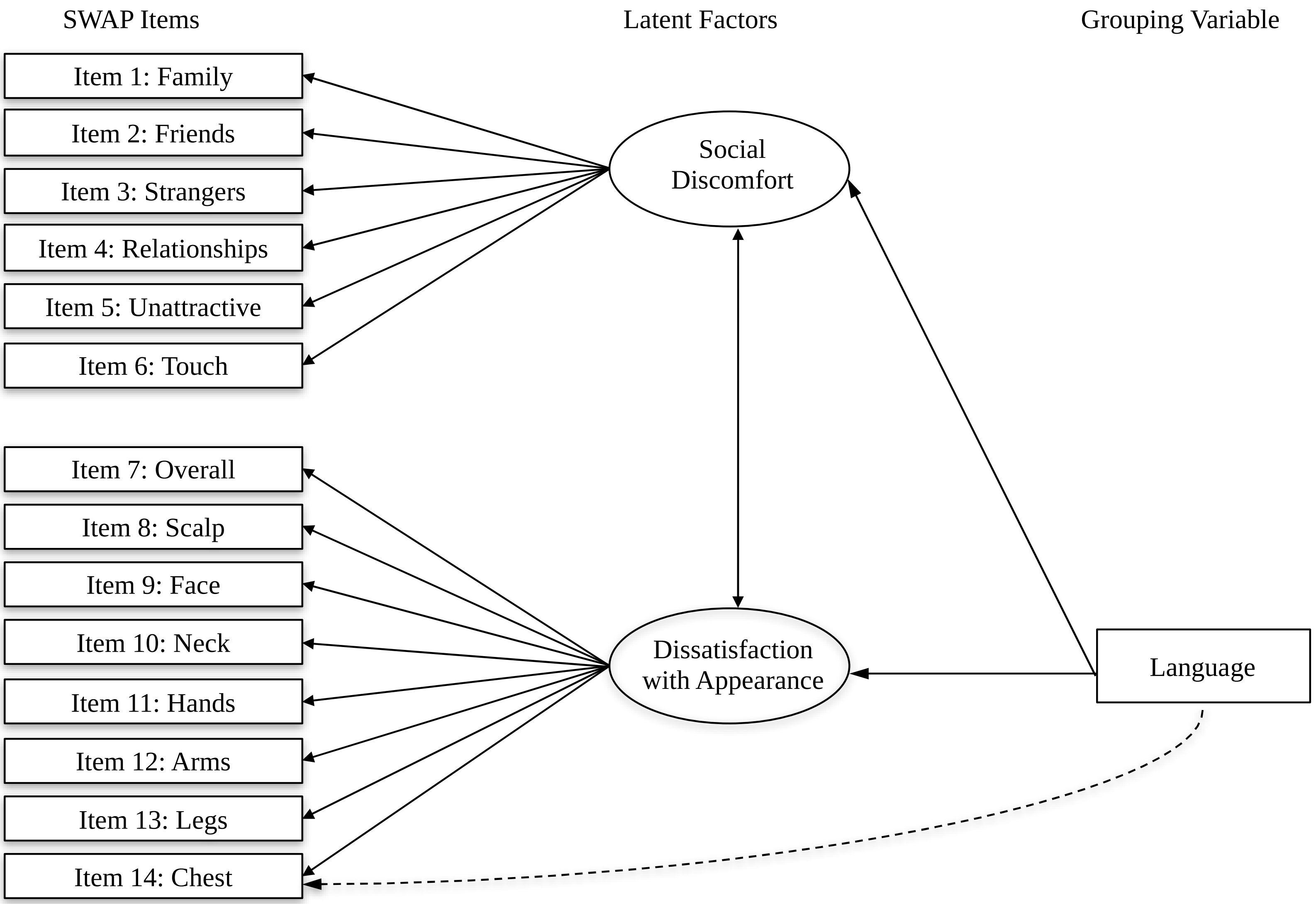
Figure 1. The MIMIC Model for the Satisfaction with Appearance Scale (SWAP)



Note. Dotted line represents the potential DIF effect of language on SWAP items.

Table 1.

Sociodemographic and Disease Characteristics by Total and Language Group Samples

Variable	Total *N* = 856	English *N* = 648	French *N* = 208	*p* value
Female *n, (%)*	751 (87.7)	566 (87.3)	185 (88.9)	.541
White *n, (%)*	762 (90.1)[a]	570 (89.2)[d]	192 (98.2)[g]	.138
Limited cutaneous SSc *n, (%)*	594 (69.4)	457 (70.5)	137 (65.9)	.205
Age (years) *mean* ± *SD*	56.8 ± 11.5	56.4 ± 11.9	57.9 ± 10.2	.105
Education (> high school), *n, (%)*	406 (47.7)[b]	313 (48.6)[e]	93 (44.7)	.329
Married/living as married *n, (%)*	594 (69.5)[c]	454 (70.2)[f]	140 (67.3)	.436
Disease duration (years), *mean* ± *SD*	16.3 ± 12.2[a]	16.7 ± 12.4[d]	15.0 ± 11.3[g]	.080
SWAP Social *mean* ± *SD*	8.6 ± 8.8	8.4 ± 8.8	8.9 ± 8.9	.478
SWAP Dissat *mean* ± *SD*	19.1 ± 12.6	19.0 ± 12.9	19.3 ± 11.5	.756

Note. SSc = systemic sclerosis; *SD* = Standard Deviation; Social = SWAP Social Discomfort subscale; Dissat = SWAP Dissatisfaction with Appearance subscale. Disease duration = time since onset of first symptoms (either Raynaud's nor non-Raynaud's). Due to missing values: [a]*N* = 846; [b]*N* = 852; [c]*N* = 855; [d]*N* = 639; [e]*N* = 644; [f]*N* = 647; [g]*N* = 207.

Table 2.

Factor Loadings of the SWAP Social Discomfort and Dissatisfaction with Appearance Latent Factors and Influence on the Overall Estimates of Social Discomfort and Dissatisfaction with Appearance Latent Factor Scores

Item	Base Model[a]	DIF Corrected Model[b]
	Factor Loading (95% CI)	Factor Loading (95% CI)
Social Discomfort Latent Factor		
1. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my family.	.84 (.81, .87)	.84 (.81, .87)
2. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my friends.	.93 (.91, .94)	.93 (.91, .94)
3. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of strangers.	.95 (.93, .97)	.95 (.93, .97)
4. Changes in my appearance have interfered with my relationships.	.76 (.73, .80)	.76 (.73, .80)
5. I feel that my scleroderma is unattractive to others.	.78 (.75, .81)	.78 (.75, .81)
6. I don’t think people would want to touch me.	.70 (.66, .74)	.70 (.66, .74)
Dissatisfaction with Appearance Latent Factor		
7. I am satisfied with my overall appearance.	.86 (.84, .88)	.86 (.84, .88)
8. I am satisfied with the appearance of my scalp.	.67 (.64, .71)	.67 (.64, .71)
9. I am satisfied with the appearance of my face.	.89 (.88, .91)	.89 (.88, .91)
10. I am satisfied with the appearance of my neck.	.85 (.83, .87)	.85 (.83, .87)
11. I am satisfied with the appearance of my hands.	.72 (.68, .75)	.72 (.68, .75)
12. I am satisfied with the appearance of my arms.	.86 (.85, .88)	.86 (.85, .88)

13. I am satisfied with the appearance of my legs.	.79 (.76, .81)	.79 (.76, .81)
14. I am satisfied with the appearance of my chest.	.87 (.86, .89)	.87 (.86, .89)
Correlation of Social Discomfort and Dissatisfaction with Appearance Latent Factors	.58 (.53, .62)	.58 (.53, .62)
Direct Effects on Items Attributable to French Language		
Item 1. Family	---	.36 (.23, .49)
Item 2. Friends	---	.17 (.06, .28)
Item 5. Unattractive	---	-.40 (-.52, -.29)
Structural Effect of French Language on Latent Factors		
Language on Social Discomfort Factor	.09 (-.10, .27)	.06 (-.13, .24)
Language on Dissatisfaction with Appearance Factor	.04 (-.12, .20)	.04 (-.12, .20)

Note. Factor loadings represent raw model results. CI = Confidence Interval. [a]Not corrected for Differential Item Functioning (DIF); [b]Corrected for DIF on items 1, 2, 5.

Linking Studies Three and Four

Elements of the first three studies of this thesis explored the impact of sociodemographic factors such as sex, age, and language background on item responses of two disease-specific measures of central body image concerns among SSc patients, the BCSS and the SWAP. A potentially significant factor that was not examined in these studies, but that may relate meaningfully to appearance outcomes in patients with SSc, is race/ethnicity. For instance, some evidence from the visible difference literature suggests that people from non-White racial/ethnic backgrounds experience greater worry and concern about their appearance than White individuals (Rumsey et al., 2004). The link between race/ethnicity and body image distress across groups with visible differences, however, remains unclear. It is even less well understood in SSc, as only one study to date (Nusbaum, Gordon, & Steen, 2016) has investigated the degree to which race/ethnicity may be associated with appearance concerns among patients. We may expect there to be racial/ethnic differences, though, given the heterogeneity of disfiguring manifestations of the disease across racial/ethnic groups. For instance, skin involvement and pigmentation changes are more common among Black patients than White patients, whereas White patients are more likely to have other appearance-altering features such as telangiectasias (Reveille et al., 2001).

Given this, the primary objective of the fourth study of this thesis was to examine the association of race/ethnicity with the presence of appearance concerns, controlling for factors that are known to influence appearance concerns, in a large, international cohort of SSc patients. While Study Two highlighted negligible influences of sex and age in terms of the measurement equivalency of BCSS items in SSc, previous research has identified these both as factors associated with body image distress among patients (e.g., Jewett et al., 2012); therefore, the associations of sex and age with the presence of appearance concerns were also assessed. We also explored potential associations of disfiguring disease characteristics with the presence of appearance concerns, as research has documented that some features of the disease are linked to negative body image (e.g., Jewett et al., 2012; Malcarne et al., 1999; van Lankveld et al., 2007). A secondary objective of this fourth study was to compare the percentage of patients with SSc who reported the presence of appearance concerns to rates from a UK general population sample (Harris & Carr, 2001).

Chapter 5

Study Four

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

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Abstract

Appearance concerns are common in scleroderma (SSc) and have been linked to younger age and more severe disease. No study has examined their association with sex or race/ethnicity. SSc patients were sampled from the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. Presence of appearance concerns was assessed with a single item, and medical and sociodemographic information were collected. Of 644 patients, appearance concerns were present in 72%, including 421 of 565 women (75%), 42 of 79 men (53%), 392 of 550 patients who identified as White (71%), 35 of 41 who identified as Black (85%), and 36 of 53 who identified as another race/ethnicity (68%). In multivariate analysis, women had significantly greater odds of reporting appearance concerns than men (odds ratio (OR) = 2.97, 95% confidence interval (CI) = 1.78-4.95, $p < .001$). Black patients had significantly greater odds of appearance concerns than White patients in unadjusted (OR = 2.64, 95% CI = 1.01-6.34, $p = .030$), but not multivariate analysis (OR = 1.76, 95% CI = 0.67-4.60, $p = .250$). Compared to a general population sample, appearance concerns were substantially more common in SSc, particularly for men across all age groups and for younger women. The most commonly reported features of concern were related to the face and head, followed by the hands and fingers; this did not differ by sex or race/ethnicity. Appearance concerns were common in SSc. Women were substantially more likely than men to have appearance concerns. Although non-significant in multivariate analysis, Black patients were more likely to have concerns than White patients, likely due to more severe changes in appearance.

Keywords: scleroderma; appearance concerns; sex; race/ethnicity

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

Disfiguring appearance changes, including telangiectasias, hand contractures, skin pigmentation changes, digital ulcers, and altered facial features, are common in systemic sclerosis (SSc). These appearance changes often affect body parts that are highly visible and that play a central role in social interactions, such as the face, mouth, and hands (Boin & Wigley, 2012; Mayes, 2008; Rumsey & Harcourt, 2005). Treatments can lessen the impact of some SSc symptoms but do not alleviate manifestations of irreversible tissue damage that affect appearance.

Among individuals with visible differences due to medical illness or injury, there is a well-established relationship between the extent and severity of appearance changes and psychological outcomes (Clarke et al., 2014; Ong et al., 2007). Consistent with this, in SSc, the presence, severity, and perceived noticeability of appearance changes are associated with greater body image dissatisfaction and social discomfort, as well as poorer overall psychosocial and psychological functioning (Amin et al., 2011; Benrud-Larson et al., 2003; Jewett et al., 2012; Kwakkenbos et al., 2015; Malcarne et al., 1999; Sivakumar et al., 2010; van Lankveld et al., 2007). The association of younger age and greater appearance concerns is also well-established, both among people with visible differences and in the general population (Clarke et al., 2014; Harris & Carr, 2001; Tiggemann, 2004). Among 2,100 randomly sampled members of the UK general population, 70% of women and 50% of men 30 years or younger reported at least one appearance concern compared to 33% of women and 21% of men 61 years or older (Harris & Carr, 2001). Younger patients with SSc have also been found to experience greater social discomfort related to appearance than older patients (Jewett et al., 2012).

No published studies have investigated the degree to which sex and race/ethnicity may be associated with appearance concerns in SSc, likely due to the small numbers of men and non-

White patients in most study samples. Among other groups of people with visible differences (e.g., skin conditions, burn scarring, limb disfigurement), however, women experience more worry about their appearance and greater general distress, social anxiety, and social avoidance than men (Hassan, Grogan, Clark-Carter, Richards, & Yates, 2009; Richards et al., 2014; Rumsey, 2014; Rumsey & Harcourt, 2004; Rumsey, et al., 2004; Thompson & Kent, 2001). Less is known about the association of race/ethnicity and appearance concerns due to visible differences. One survey of 458 adults with a variety of visible disfigurements found that people from non-White racial/ethnic backgrounds experienced significantly greater worry about their appearance and heightened concern that their condition was noticeable to others than White respondents (Rumsey et al., 2004). This may have been because people with darker skin tones are more vulnerable to visible changes in skin pigmentation and report greater psychosocial impact compared to individuals with lighter skin tones (van der Veen, 2007). In SSc, skin involvement and pigmentation changes are more common among Black patients than White patients, whereas White patients are more likely to have telangiectasias (Reveille et al., 2001).

The objective of the present study was to examine the association of sex and race/ethnicity with the presence of appearance concerns, controlling for factors that are known to influence appearance concerns (e.g., age, disease severity) in a large, international cohort of SSc patients. We also compared the percentage of patients with SSc who reported the presence of appearance concerns to rates previously published from a UK general population sample (Harris & Carr, 2001), stratified by sex and age groups.

Method

Participants and Procedure

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

Sociodemographic Characteristics. Patients enrolled in the SPIN Cohort provided sociodemographic data, including sex, race/ethnicity, age, education level, marital status, and employment status. Response options for race/ethnicity differed slightly for patients from Canada, the USA, and the UK, consistent with how racial/ethnic status is typically characterized in each country. In the Canadian sample, patients could identify as White, Black, Aboriginal, Asian, Latin American, or Arab. In the US sample, patients could identify as White (non-Hispanic), African American or Black, Hispanic or Latino, Asian, American Indian/Alaska

Native, Native Hawaiian/Other Pacific Islander, or Mixed-race. In the UK sample, patients could identify as White, African, or Asian. In the present study, racial/ethnic status was collapsed into three categories across Canadian, American, and UK samples, consisting of White, Black, or Other. Across countries, responses indicating White racial/ethnic status were combined to create one White race/ethnicity category; responses indicating Black, African American or Black, or African were combined to create one Black race/ethnicity category; and other responses were combined to create one Other race/ethnicity category.

Disease-related and Medical Characteristics. SPIN physicians or nurse coordinators provided disease and medical information, including time since onset of the first non-Raynaud's symptom (disease duration); disease subtype (limited or diffuse); and presence of telangiectasias, skin pigmentation changes, hand contractures, skin thickening on the fingers of both hands, and body mass index (BMI). Limited SSc was defined as skin involvement distal to the elbows and knees only, whereas diffuse SSc was defined as skin involvement proximal to the elbows and knees, and/or the trunk (Medsger et al., 2003). Telangiectasias were defined as the visible dilation of superficial cutaneous blood vessels that collapse upon pressure and fill slowly when pressure is released (van den Hoogen et al., 2013). Skin pigmentation changes included either hyper- or hypo-pigmentation of the skin. In the present study, telangiectasias and skin pigmentation changes were coded as present on the body and face, present on the body only, or none. Hand contractures, which entail limitations in the range of motion of a joint, secondary to tightening around the joint, were measured for small joints on the hands (i.e., proximal interphalangeal joints, metacarpals, and/or wrists) and categorized as None/Mild (0-25% limitation in range of motion), Moderate (25-50%), or Severe (>50%). Skin thickening on the fingers of both hands was defined as skin thickening or hardening extending proximal to the metacarpophalangeal joints (Reveille et al., 2001).

Presence of Appearance Concerns. The Derriford Appearance Scale (DAS-24; Carr, Moss, & Harris, 2005; Moss, Lawson, & White, 2015) is a self-report measure of distress related to problems with appearance. It assesses social anxiety and avoidance related to self-consciousness due to appearance. The DAS-24 has an introductory item, not included in the scoring of the measure, that asks, "*Is there any aspect of your appearance (however small) that concerns you at all?"* (yes/no). This item was used as the primary outcome in the present study. Published prevalence of appearance concerns using this question is available for the UK general population (Harris & Carr, 2001), which we used for comparison. In addition, for patients who answer yes to the appearance concern question, the DAS-24 includes a further inquiry, "*The aspect of my appearance about which I am most sensitive or self-conscious is …."* with space for open response. If patients reported more than one aspect of appearance concern, the first listed was used.

Data Analysis

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

The associations of sociodemographic variables (sex, race/ethnicity, age, marital status, education level), and disease/medical variables (telangiectasias, skin pigmentation changes, hand contractures, skin thickening on fingers, disease subtype, BMI) with the presence of appearance concerns were assessed using binary logistic regression. All variables included in the regression analysis were selected a priori based on previous research indicating variables likely to relate to appearance concerns in SSc. Discrimination and calibration of the multivariate model were assessed with the c-index and Hosmer-Lemeshow goodness-of-fit test statistic, respectively (Hosmer & Lemeshow, 2000). The c-index is the percentage of comparisons where patients with appearance concerns had a higher predicted probability of having appearance concerns than patients without appearance concerns for all possible pairs where patients were discrepant on outcome variable status. The Hosmer-Lemeshow goodness-of-fit statistic is a measure of the accuracy of the predicted number of cases of appearance concerns compared to the number of patients who actually reported appearance concerns across the spectrum of probabilities. A relatively large p value indicates a reasonably good model fit (Hosmer & Lemeshow, 2000). All analyses were conducted using SPSS, Version 22, and statistical tests were two-sided with $\alpha < .05$.

The percentage of women and men with SSc who reported appearance concerns was compared to levels of appearance concerns reported in a sample from the general UK population (Harris & Carr, 2001). The relative risk of reporting appearance concerns was calculated for women and men, separately, stratified by age groups. Race/ethnicity data were not provided for the UK population sample, thus comparisons for these groups separately could not be conducted.

Results

Sample Characteristics

In total, 757 SSc patients completed baseline assessments. There were 717 who answered the binary appearance concern item, of whom 644 had data for all variables in the multivariate model and were thus included in analyses. Of these, 463 (72%) indicated that there was an aspect of their appearance that caused them concern. Rates were similar among the 73 patients excluded for missing data, with 57 (78%) indicating that there was an aspect of their appearance that caused them concern.

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

There were 565 (88%) women and 79 (12%) men included. There were no statistically significant differences between women and men for any sociodemographic or disease/medical variables (see Table 1). There were 41 (6%) Black patients and 53 (8%) patients who identified as a member of another racial/ethnic group. As shown in Table 1, White patients were older and more likely to be married than Black patients, but less likely to have skin thickening on the fingers and to have diffuse SSc (statistically significant, $p < .0167$). There were also statistically significant differences between White and Black patients in presence of telangiectasias and skin pigmentation changes. White patients were older and less likely to have diffuse SSc than patients from other racial/ethnic groups (statistically significant, $p < .0167$). There were also statistically significant differences in skin pigmentation changes between White patients and those from other racial/ethnic groups (see Table 1).

Among women, 75% (421 of 565) reported appearance concerns, compared to 53% (42 of 79) of men ($p < .001$). Across racial/ethnic groups, 85% (35 of 41) of Black patients reported appearance concerns, compared to 71% (392 of 550) of White patients and 68% (36 of 53) from other racial/ethnic groups ($p = .122$).

Of the 463 patients who indicated that there was an aspect of their appearance that caused them concern, 444 listed the specific feature of greatest concern. As shown in Table 2, the most common category was the face/neck/head/mouth (41%), followed by the hands/fingers (27%). Results were similar for female and male patients and patients classified as White, Black, or Other racial/ethnic status.

As shown in Table 3, on an unadjusted basis, the odds of appearance concerns were greater for female patients compared to male patients (OR = 2.70, 95% Confidence Interval (CI) = 1.73 - 4.22, $p < .001$), and for Black patients versus the reference group, White patients (OR = 2.64, 95% CI = 1.01 - 6.34, $p = .030$). Patients with another racial/ethnic status did not have significantly different odds compared to White patients (OR = .92, 95% CI = .52 - 1.65, $p = .788$). Other variables that were significantly associated with the presence of appearance concerns on an unadjusted basis included younger age, diffuse disease subtype, presence of moderate hand contractures, and presence of skin thickening on the fingers.

In the multivariate analysis, only sex, age, and presence of moderate hand contractures were significantly associated with appearance concerns, after controlling for covariates of race/ethnicity, BMI, education level, marital status, disease subtype, telangiectasias, pigmentation changes, and skin thickening on the fingers. The odds of appearance concerns for female patients were significantly greater than for male patients (OR = 2.97, 95% CI = 1.78 - 4.95, $p < .001$). Older patients were also less likely to report appearance concerns than younger patients (OR = .98 per year, 95% CI = .96 - .99, $p = .007$), equivalent to a reduction of 18% in the OR for every 10-year increase in age. The odds of reporting appearance concerns did not differ significantly for Black versus White patients (OR = 1.76, 95% CI = .67 - 4.60, $p = .250$). The odds of reporting appearance concerns for patients with moderate hand contractures were almost twice those of patients with no or only mild hand contractures (OR = 2.05, 95% CI = 1.17 - 3.60, $p = .012$; see Table 3). Model fit for the 11 predictors included was less than ideal based on the Hosmer-Lemeshow test ($\chi^2(8, N = 644) = 10.65$, $p = .022$), and the c-index statistic was .69.

As shown in Table 4, rates of appearance concerns were higher, across all age groups, for women and men with SSc, compared to women and men from the general UK population sample (Harris & Carr, 2001). The relative risk of reporting appearance concerns among SSc patients compared to the general population was greater for men than for women across age groups. For both women and men, the relative risk was substantially higher among older patients.

Discussion

The main findings of this study were that women with SSc were substantially more likely than men to report appearance concerns, controlling for sociodemographic and disease variables; black racial/ethnic group membership was significantly associated with appearance concerns at the bivariate level; however, the association was not statistically significant after accounting for the influences of other sociodemographic and disease variables. Consistent with findings from previous studies, older age was significantly associated with reduced odds of appearance concerns. Among disease variables, moderate hand contractures were significantly associated with greater odds of appearance concerns. Compared to data from a general population sample, both women and men with SSc were more likely to report appearance concerns, although the relative risk was higher for men with SSc compared to women. In addition, for both women and men, the relative risk was substantially higher, compared to the general population, among older patients than among younger patients. The most commonly reported features of concern were related to the face and head, followed by the hands and fingers; this did not differ by sex or race/ethnicity.

The finding that female SSc patients had greater odds of experiencing appearance concerns is consistent with previous research on the relationship between sex and visible differences, which has highlighted that women tend to experience greater distress, social anxiety, and difficulty adjusting to disfiguring appearance changes than men (Hassan et al., 2009; Richards et al., 2014; Rumsey, 2014). Although 85% of Black patients reported appearance concerns compared to 71% of White patients, race/ethnicity was not independently associated with appearance concerns in the multivariate analysis. It is possible that the small number of Black people in the study could explain the non-significant result. This finding may also have occurred because Black patients tended to be younger, have diffuse disease, and have greater pigmentation changes and skin thickening on the fingers compared to White patients, as these are all factors associated with appearance concerns.

Although men with SSc were less likely to report appearance concerns than women with the disease, men with SSc had a greater relative risk of appearance concerns compared to the general population sample than women. In both SSc and the general population, women are more likely than men to have appearance concerns, but the increase in appearance concerns is much greater for men with SSc. A similar phenomenon can be seen with respect to age. For both women and men in the general population, the proportion of people with appearance concerns drops substantially in older groups, particularly for people aged 61 and older. The proportion also diminishes across age groups for people with SSc but much less dramatically, and in SSc, older people are more likely to report appearance concerns compared to the general population, where worries about appearance have mostly subsided among older adults.

The finding that the majority of patients across sexes and race/ethnicities listed the face and head body regions as the primary feature that caused them concern, followed by the hands and fingers, is consistent with previous research in SSc, which has reported that appearance changes to visible and socially relevant body regions are common and of the greatest distress to patients (Amin et al., 2011; Jewett et al., 2012; Malcarne et al., 1999; Mayes, 2008; van Lankveld et al., 2007). While the categories of appearance concern in the present study were broad, results showed that the general types of appearance concerns reported by patients were fairly consistent across sex and racial/ethnic groups.

The present study has limitations that should be considered in interpreting results. First, the SPIN Cohort constitutes a convenience sample of SSc patients receiving treatment at SPIN recruiting centers, and patients at these centers may differ from those in other settings. SSc patients in the SPIN Cohort also complete questionnaires online, which may further limit the generalizability of findings. An additional limitation relates to the nature of disease characteristics included in the analyses. Specifically, the majority of the variables consisted of fairly crude indicators of either the presence or absence of a particular disease factor and did not provide a measure of severity. This may have reduced the ability to identify any associations, if present, between the severity of visible differences from the disease and the presence of appearance concerns. An additional limitation is that the sample sizes of Black patients and those who self-identified as members of another racial/ethnic group were small. These limitations may have been reflected in the model fit statistics, which were less than ideal (Hosmer & Lemeshow, 2000). It is also possible that the relatively limited information obtained from the predictive model was due to the dichotomous outcome variable. Most patients reported the presence of appearance concerns; however, measuring appearance concerns may not be something easily categorized as present or absent, and thus, the study results may differ if a more robust measure of appearance concerns was used. Additionally, the general population sample comparison was done with the only general population data that were currently available. While it was useful to contextualize patterns of appearance concerns among patients with SSc, the degree of actual differences may have been influenced by factors not considered in the present analysis. Another limitation is that the categories for reporting features of concern that were used in the present study were somewhat broad and non-specific, which did not allow for a detailed description of the features listed by patients. Nonetheless, they provided a general understanding of the common types of appearance concerns among patients and underlined that those most important do not vary substantially across sex or racial/ethnic groups. Finally, we used self-reported race/ethnicity classifications. Race/ethnicity is a complex construct that may reflect sociocultural aspects of experience as much as biological aspects. Furthermore, the Other race/ethnicity category allowed us to separate out people who did not identify as White or Black, but its composition was heterogeneous and not necessarily informative in relation to the experience of any patient or group of patients classified in the Other group.

In summary, female SSc patients had significantly higher odds of reporting appearance concerns than male patients. Although Black SSc patients had higher odds of experiencing appearance concerns than White patients on an unadjusted basis, this result was not statistically significant when accounting for the influence of other sociodemographic and disease/medical characteristics. It may be the case that greater appearance concerns among Black patients reflect more significant appearance changes, such as skin involvement, including pigmentation changes. Both women and men with SSc were more likely to report appearance concerns than general population survey respondents. The difference in reporting of appearance concerns between SSc patients and the general population was greatest for men and for older patients. In both of these cases, rates in the general population were relatively low for these groups, but much less so in SSc. Despite differences in overall rates, across groups defined by sex or race/ethnicity, the most commonly reported features of appearance concern related to the face and head regions, followed by the hands and fingers. Replications of the current study with larger samples of Black and other racial/ethnic groups are needed. In addition, future studies should include assessment of appearance concerns with a continuously measured outcome variable or one that explores multiple domains of appearance concerns, as well as include factors, such as disease duration and disease and serological subtype, to explore their potential impact on appearance concerns.

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Table 1.

Sociodemographic and Disease Characteristics

	Total	Race/Ethnicity				Sex		
Variable		White	Black	Other	p value	Female	Male	p value
Age (years), *mean ± SD*	55.3 ± 12.1	56.5 ± 11.5[a,b]	47.7 ± 11.5[a]	48.1 ± 14.1[b]	<.001	55.1 ± 12.0	56.2 ± 12.8	.462
Education >12 years, *n (%)*	512 (79.5)	434 (78.9)	33 (80.5)	45 (84.9)	.579	451 (79.8)	61 (77.2)	.591
Currently employed (full or part-time), *n (%)*	268 (41.7)[c]	225 (41.0)[d]	21 (51.2)	22 (41.5)	.439	236 (41.8)	32 (41.0)[e]	.901
Married/living as married, *n (%)*	471 (73.1)	413 (75.1)[a]	21 (51.2)[a]	37 (69.8)	.003	412 (72.9)	59 (74.7)	.741
Time since onset of first non Raynaud's symptoms (years), *mean ± SD*	11.5 ± 8.7[f]	11.6 ± 8.9[g]	8.9 ± 5.6[h]	10.2 ± 7.8[i]	.067	11.7 ± 8.8[j]	10.4 ± 7.8[k]	.227
Body Mass Index (BMI) *mean ± SD*	26.04 ± 6.12	26.14 ± 6.14	26.14 ± 6.84	24.97 ± 5.33	.413	26.02 ± 6.26	26.19 ± 5.00	.816
Patients with diffuse SSc, *n (%)*	258 (40.1)	200 (36.4)[a,b]	28 (68.3)[a]	30 (56.6)[b]	<.001	226 (40.0)	32 (40.5)	.971

Telangiectasias					<.001			.058
None, *n (%)*	183 (28.4)	137 (24.9)[a]	26 (63.4)[a]	20 (37.7)		153 (27.1)	30 (38.0)	
Body and face, *n (%)*	298 (46.3)	273 (49.6)[a]	7 (17.1)[a]	18 (34.0)		262 (46.4)	36 (45.6)	
Body only, *n (%)*	163 (25.3)	140 (25.5)[a]	8 (19.5)[a]	15 (28.3)		150 (26.5)	13 (16.5)	
Pigmentation changes					<.001			.497
None, *n (%)*	438 (68.0)	396 (72.0)[a,b]	13 (31.7)[a]	29 (54.7)[b]		385 (68.1)	53 (67.1)	
Body and face, *n (%)*	105 (16.3)	71 (12.9)[a,b]	18 (43.9)[a]	16 (30.2)[b]		89 (15.8)	16 (20.3)	
Body only, *n (%)*	101 (15.7)	83 (15.1)[a,b]	10 (24.4)[a]	8 (15.1)[b]		91 (16.1)	10 (12.7)	
Hand contractures					.898			.988
No/mild (0-25%), *n (%)*	488 (75.8)	418 (76.0)	31 (75.6)	39 (73.6)		428 (75.8)	60 (75.9)	
Moderate (25-50%), *n (%)*	121 (18.8)	102 (18.5)	7 (17.1)	12 (22.6)		106 (18.8)	15 (19.0)	
Severe (>50%), *n (%)*	35 (5.4)	30 (5.5)	3 (7.3)	2 (3.8)		31 (5.5)	4 (5.1)	
Skin thickening on fingers, *n (%)*	360 (55.9)	293 (53.3)[a]	31 (75.6)[a]	36 (67.9)	.004	311 (55.0)	49 (62.0)	.242
Presence of appearance concerns, *n (%)*	463 (71.9)	392 (71.3)	35 (85.4)	36 (67.9)	.122	421 (74.5)	42 (53.2)	<.001

Note. N Total Sample = 644. *N White Sample* = 550. *N Black Sample* = 41. *N Other Sample* = 53. *N Female Sample* = 565. *N Male Sample* = 79. [a]White patients statistically significantly different from Black patients. [b]White patients statistically significantly different from patients from another racial/ethnic group. Due to missing values: [c] N = 643; [d]N = 549; [e]N = 78; [f]N = 595; [g]N = 510; [h]N = 40; [i]N = 45; [j]N = 519; [k]N = 76.

Table 2.

Appearance Concerns Reported by SSc Patients who Specified Feature of Concern

	Total	Race/Ethnicity			Sex	
Feature	(n, %)	White (n, %)	Black (n, %)	Other (n, %)	Female (n, %)	Male (n, %)
Body Area-related						
Face/mouth/head/ neck	180 (40.5)	149 (40.5)	14 (40.0)	17 (41.5)	159 (41.2)	21 (36.2)
Hands/fingers	119 (26.8)	103 (28.0)	7 (20.0)	9 (22.0)	99 (25.6)	20 (34.5)
Arms	5 (1.1)	3 (0.8)	1 (2.9)	1 (2.4)	5 (1.3)	0
Chest/stomach	9 (2.0)	9 (2.4)	0	0	8 (2.1)	1 (1.7)
Legs/feet	13 (2.9)	12 (3.3)	1 (2.9)	0	12 (3.1)	1 (1.7)
Skin-related						
Telangiectasias (any)	33 (7.4)	28 (7.6)	1 (2.9)	4 (9.8)	26 (6.7)	7 (12.1)

Skin pigmentation changes (any)	12 (2.7)	9 (2.4)	1 (2.9)	2 (4.9)	10 (2.6)	2 (3.4)
Skin tightening or hardening	7 (1.6)	5 (1.4)	1 (2.9)	1 (2.4)	5 (1.3)	2 (3.4)
Skin unspecified/other	20 (4.5)	16 (4.3)	1 (2.9)	3 (7.3)	17 (4.4)	3 (5.2)
Weight-related						
Loss	4 (0.90)	3 (0.8)	1 (2.9)	0	4 (1.0)	0
Gain/bloating/ swelling	22 (5.0)	17 (4.6)	2 (5.7)	3 (7.3)	22 (5.7)	0
Unspecified	20 (4.5)	14 (3.8)	5 (14.3)	1 (2.4)	19 (4.9)	1 (1.7)
Total	444 (100.0)	368 (100.0)	35 (100.0)	41 (100.0)	386 (100.0)	58 (100.0)

Table 3.

Unadjusted and Adjusted Comparisons of Appearance Concerns across Sociodemographic and Disease Variables

Variable	Unadjusted		Adjusted	
	OR (95% CI)	p value	OR (95% CI)[a]	p value
Race/ethnicity (reference = White)				
Black	2.64 (1.01-6.34)	.030	1.76 (.67 - 4.60)	.250
Other	.92 (.52 - 1.65)	.788	.59 (.31 - 1.14)	.117
Age	.97 (.96 - .98)	<.001	.98 (.96 - .99)	.007
Female sex	2.70 (1.73 - 4.22)	<.001	2.97 (1.78 - 4.95)	<.001
Education ≤ 12 years	.69 (.47 - 1.02)	.063	.74 (.48 - 1.14)	.174
Not married/living as married	1.36 (.92 - 2.00)	.121	1.05 (.68 - 1.60)	.841
BMI	1.00 (.92 - 1.10)	.951	1.01 (.98 - 1.05)	.421
Diffuse disease subtype	1.76 (1.24 - 2.50)	.002	1.18 (.76 - 1.83)	.456
Telangiectasias (reference = none)				
Body and face	.81 (.54 - 1.22)	.313	1.48 (.89 - 2.47)	.136
Body only	.70 (.44 - 1.09)	.116	1.20 (.77 - 1.86)	.420

Pigmentation changes (reference = none)				
Body and face	1.58 (.96 - 2.58)	.071	.91 (.52 - 1.57)	.730
Body only	1.50 (.92 - 2.47)	.108	1.02 (.51 - 2.06)	.947
Hand contractures (reference = none/mild)				
Moderate	2.33 (1.41 - 3.86)	.001	2.05 (1.17 - 3.60)	.012
Severe	2.03 (.88 - 4.70)	.099	1.96 (.75 - 5.15)	.170
Skin thickening on hands	1.64 (1.18 - 2.28)	.003	1.48 (1.00 - 2.19)	.050

Note. Abbreviations: OR = odds ratio; CI = confidence interval; BMI = Body Mass Index. [a]Adjusted for age, sex, race/ethnicity, education level, marital status, disease subtype, telangiectasias, pigmentation changes, hand contractures, and skin thickening on fingers.

Table 4.

Proportion of Women and Men who Reported Appearance Concerns Based on Age in General Population and SPIN Samples

	General Population Sample (n, %)[a]	SPIN Sample (n, %)	Relative Risk SPIN / General Population (95% CI)
Women			
Age bands (years)			
18-40	367/537 (68)	50/60 (83)	1.22 (1.07-1.38)
41-50	139/226 (62)	80/101 (79)	1.29 (1.12-1.49)
51-60	151/240 (63)	145/183 (79)	1.26 (1.11-1.42)
61+	58/174 (33)	146/221 (66)	1.98 (1.57-2.50)
Men			
Age bands (years)			
18-40	152/341 (45)	5/9 (56)	1.25 (0.69-2.26)
41-50	69/170 (41)	8/11 (73)	1.79 (1.20-2.69)
51-60	49/202 (24)	12/22 (55)	2.25 (1.43-3.54)
61 +	45/218 (21)	17/37 (46)	2.23 (1.44-3.44)

Note. [a]Based on values reported in Harris & Carr, 2001.

Chapter 6

General Discussion

Summary of Main Findings

The objective of the first study of this thesis was to develop and validate a new, brief, patient-reported outcome measure of disease-specific body image avoidance and concealment in SSc, the Body Concealment Scale for Scleroderma (BCSS), including an evaluation of its basic psychometric properties. Findings revealed that a two-factor model, which specified Concealment with Clothing and Concealment of Hands factors, fit the data well for the 9-item BCSS; however, analysis with a bifactor model identified a strong general factor that explained a large proportion of item covariance. Furthermore, reliability indices for the Concealment with Clothing and Concealment of Hands factors were very low, suggesting that they do not likely constitute reliable indicators of unique constructs and that little variance remains beyond that explained by the general factor. Finally, internal consistency reliability was high for the BCSS total score, and the total score demonstrated strong convergent validity correlations with another measure of body image distress, as well as discriminant validity correlations with measures related to depression and pain intensity. Taken altogether, results from this study highlighted body concealment as a significant mechanism of body image avoidance for patients with SSc and provided evidence that the BCSS should be scored as a single, summed total.

The first objective of the second manuscript of this thesis was to conduct a replication of the BCSS development study, including the evaluation of the measure's psychometric properties in a separate, large, international SSc cohort. The second objective of Study Two was to extend findings from the initial development study by examining the measurement equivalency of BCSS items across sex and age groups. These are characteristics potentially capable of influencing item responses, as it has been documented across groups with visible differences that younger individuals and those of the female sex tend to report more body image distress and appearance-related concerns (Clarke et al., 2014; Rumsey et al., 2004; Tiggeman, 2004; Thompson & Kent, 2001). Assessing the measurement equivalency of patient-reported outcome measures is of central importance in the context of rare diseases like SSc because researchers increasingly combine results across groups with distinct characteristics in order to attain large enough sample sizes; however, results should only be combined if measurement equivalency is established (Mokkink et al., 2010). The findings from Study Two replicated those from the BCSS development study (Study One). Specifically, a bifactor model fit BCSS data best, with the general factor explaining the majority of item covariance. Reliability indices for the specific factors of Concealment with Clothing and Concealment of Hands were very low, again suggesting that they do not reliably measure substantive variance beyond that explained by the general factor (Reise, 2012). Thus, consistent with the initial BCSS development study, evidence from the second study of this thesis supports the use and interpretation of a single summed score for the BCSS. Beyond this, findings revealed no significant differential item functioning (DIF) for BCSS items across female and male SSc patients, and, while three items had statistically significant DIF for younger versus older patients, the magnitude of the differences was small, and the influence on scores was negligible. In sum, this study replicated findings from Study One and further demonstrated that BCSS scores can be compared and combined across females and males with SSc and those of different ages without concern that measurement differences will influence results. As such, evidence from Study Two, in addition to findings from Study One, point to the utility of the BCSS as a clinical assessment tool and patient-reported outcome measure to assess body concealment behaviours among people with SSc.

The third manuscript of this thesis again sought to examine measurement equivalency, but in this case, of a measure commonly used in SSc samples to assess social discomfort and body image dissatisfaction relative to acquired disfigurements from the disease, the Satisfaction with Appearance Scale (SWAP). Specifically, the aim was to assess possible DIF for SWAP items across different linguistic groups in a Canadian SSc cohort. This study found that, while some SWAP items demonstrated DIF based on language, the magnitude of the differences was minimal and did not affect overall scores to a notable degree. As such, findings highlighted that SWAP items do not function differently across English-and French-speaking Canadian SSc patients, and that scores can be pooled across these language groups in future research studies.

Finally, the primary objective of the fourth study of the present thesis was to examine the associations of age, sex, and race/ethnicity with the presence of appearance concerns, controlling for factors that are known to influence appearance concerns, among SSc patients in a large international cohort. The associations of disfiguring disease characteristics with appearance concerns were also assessed. Thirdly, comparisons were made between rates of appearance concerns reported by SSc patients and those from a UK general population sample (Harris & Carr, 2001), stratified by sex and age groups. Main findings from Study Four revealed that female SSc patients had significantly higher odds of reporting appearance concerns than male patients, and that older age was significantly associated with a reduction in the odds of reporting appearance concerns. After accounting for the influence of sociodemographic and disease characteristics, there was no significant difference in the odds of reporting appearance concerns between Black and White SSc patients, although Black patients had higher odds of reporting appearance concerns on an unadjusted basis. Moderate hand contractures were significantly associated with greater odds of appearance concerns, among disease characteristics assessed.

Both women and men with SSc were more likely to report appearance concerns than women and men in the general population, and this difference was greatest for men and older patients. The most commonly reported body features of concern among SSc patients, irrespective of sociodemographic characteristics, were the face and head, followed by the hands and fingers.

Implications of Findings and Directions for Future Research

Overall, findings from the present thesis both extend and enhance existing research on the assessment of body image distress among people living with SSc. Firstly, this research expands the number of disease-specific measures that capture relevant body image constructs for patients and establishes evidence of the measurement equivalence and validity of both new and existing tools, including their utility across subgroups of patients. Secondly, the present research aids in the understanding of relationships among central sociodemographic and disease characteristics and body image concerns. As such, information gained from the current studies enhances the breadth of clinical tools that can be used as measures of important body image constructs in SSc and facilitates the ongoing assessment of appearance distress among patients, particularly within largescale cohorts where patients with many distinct characteristics are often combined (e.g., Kwakkenbos et al., 2013). In addition, the current research supports the development of intervention programs that target appearance distress and related psychosocial difficulties.

The development and validation of a new patient-reported outcome measure of body concealment-related avoidance behaviours specific to SSc, the BCSS, addresses an important gap in the measurement of body image distress among patients, as, prior to its development, no disease-specific measure existed to address this significant mechanism of social and behavioural avoidance related to appearance among patients. Furthermore, the development of the BCSS extends the concept of body concealment and body image avoidance to people with acquired disfigurements from a rheumatic condition, building off of more general measures and conceptualizations of body image distress, self-consciousness related to appearance, and body image avoidance (Carr et al., 2000; 2005; Rosen et al., 1999; Davidson & McCabe, 1995).

Beyond this, results of the psychometric testing and replication of findings from Studies One and Two, respectively, demonstrate the potential utility of the BCSS for both researchers and clinicians looking to develop and evaluate interventions designed to address body image concerns among people with SSc. A program is currently being developed within SPIN (Kwakkenbos et al., 2013) that targets body image distress specific to SSc, including body image avoidance and concealment. This program incorporates established strategies to reduce avoidance behaviours that have been recommended for individuals with a range of visible differences, including elements from social interaction skills training, cognitive behavioural therapy (CBT) and acceptance and commitment therapy (ACT; Bessell et al., 2012; Clarke et al., 2014; Kapp-Simon, Simon, & Kristovich, 1992; Robinson, Rumsey, & Partridge, 1996; Wade, George, & Atkinson, 2009). These elements include techniques to help those with a visibly different appearance effectively anticipate and manage the reactions of others, increase their own confidence and self-esteem in social settings, evaluate and reattribute unhelpful patterns of thinking related to appearance, reassess the importance of appearance for their self-worth, change the relationship among distressing thoughts and feelings about appearance, and accept aspects of their visible differences (Rumsey & Harcourt, 2012; Wade, George, & Atkinson, 2009). This SPIN program (Kwakkenbos et al., 2013), and others, have yet to be tested in SSc, and the BCSS could be used as an outcome measure to assess body concealment-related behaviours in the evaluation of such interventions.

The intervention program targeting body image distress currently under development within SPIN will be enacted within the organization’s international Cohort of patients, which spans across North America, Europe, and Australia. These types of largescale cohorts are becoming increasingly common in SSc, as well as in research among other chronic diseases, in order to attain adequate sample sizes, given that, in the context of such rare conditions, there are typically few patients with a particular illness, and patients tend to be spread substantially across geographic locations. In such cohorts, data is required to be integrated across individuals with distinct characteristics, including race/ethnicity, sex, age, and language background, among others. It is therefore important to establish that outcome data from these heterogeneous groups of patients can, in fact, be combined, without concern that any one characteristic may influence responses. In both the SPIN and CSRG samples utilized in the present research, many instruments are administered to SSc patients at regular intervals as part of assessments related to a variety of psychosocial outcomes, one of which includes the SWAP (Lawrence et al., 1998), a commonly used measure of social discomfort and dissatisfaction with appearance. The SWAP is planned to be used as a primary outcome in a forthcoming SPIN randomized controlled trial (RCT) to evaluate the intervention program for body image distress currently under development (Kwakkenbos et al., 2013). While the SWAP has been previously validated for use among SSc patients, prior to the research conducted in Study Three of this thesis, no information existed as to whether or not scores on the measure could be reasonably combined across English and French linguistic groups, which are languages represented in both the SPIN and CSRG Cohorts. Present results established that SWAP scores in English and French can be combined across patients, and, as such, provided initial evidence for the utility of the measure across diverse linguistic backgrounds. These findings can also be extended to the short version of the SWAP, the 6-item Brief-SWAP (Jewett et al., 2010), as it can be reasonably assumed based on present findings that the Brief-SWAP functions similarly in terms of DIF as the full-length version of the measure.

Findings from the present research also point to which SSc patients experience higher levels of appearance concerns and, subsequently, who may benefit more from interventions addressing these issues. For instance, female patients and younger individuals with the disease displayed greater odds of appearance concerns than males and older patients with SSc. These results support well-documented findings that women and younger individuals have a tendency to experience greater body image distress than men and older members of society (Rumsey & Harcourt, 2012). Similar results have also been found in previous SSc samples (e.g., Jewett et al., 2012). As such, it is likely that younger women with SSc may benefit most from a targeted intervention to address body image concerns due to appearance changes from the disease. Importantly, however, compared to members of the general population, the relative risk of reporting appearance concerns was greatest for men with SSc and older patients, suggesting that perhaps body image issues impact these subgroups relatively more compared to peers in the context of a disfiguring disease, even though overall risk is lower compared to others with SSc. As such, older patients, as well as men with SSc, would likely also benefit from programs addressing appearance distress. Beyond this, present findings also indicated that, among other disease characteristics, hand contractures were most significantly associated with increased odds of reporting appearance concerns; therefore, patients with significant hand involvement may also constitute a subgroup with heightened body image support needs. Relatedly, patients categorized the hands and fingers as one of the central features causing concern, preceded only by the face and head body regions. These results align with those from the BCSS development studies, which highlighted concealment of hands as a particularly salient mechanism of body image avoidance for patients. Finally, while the significantly greater odds of reporting appearance concerns among Black patients compared to White patients disappeared in multivariate analyses, Black patients did have significantly greater odds of appearance concerns in bivariate analyses. Taken altogether, present findings suggest that programs currently under development to address appearance concerns in SSc, and those potentially forthcoming, should take into consideration all of these diverse sociodemographic and disease characteristics that may contribute to greater body image distress.

The studies conducted as part of this thesis also point to specific directions for future research. Firstly, as highlighted above, intervention programs targeting the reduction of body image distress among SSc patients, such as the one currently being developed through SPIN (Kwakkenbos et al., 2013), should incorporate content that reflects the sociodemographic and disease characteristics of patients who may be at a greater risk of appearance concerns. Future research could examine differences in body image concerns across men and women, older versus younger patients, and those with different manifestations of appearance changes from the disease, in order to determine nuances in the experiences of body image distress, and subsequent idiosyncratic versus shared needs in terms of intervention content and design.

Additionally, body image constructs not assessed by questionnaires such as the BCSS and SWAP could be explored in future studies as other potentially relevant aspects that capture the uniqueness and complexity of appearance-related concerns among patients. Having direct feedback from patients as to what other body image areas would be helpful to explore, and how interventions could be targeted appropriately may also be helpful in future studies. Based on the cognitive-behavioural theoretical model, a possible area of further investigation could include constructs related to the schema of investment in appearance that was not explored in the present thesis.

Replications of the present research are also needed, as is reflected below in the Limitations section of this thesis. For instance, further examinations of the measurement equivalency of both the BCSS and SWAP, as well as other patient-reported outcome measures currently used in SSc research paradigms, is needed, based on characteristics of age, sex, language, and race/ethnicity. Relatedly, replication of the item-specific DIF results found in the present research for the BCSS and SWAP is necessary. Replications of the associations of race/ethnicity with appearance concerns is also needed with larger samples of Black patients and other non-White racial/ethnic groups, as this may permit clarification regarding whether or not patients from non-White racial/ethnic groups do endorse more appearance concerns. In addition, future studies should include assessment of appearance concerns with a continuous outcome measure, or one that explores multiple domains of body image, in order to gain a more detailed understanding of the types of appearance concerns most common or salient for patients. Future research could also explore the potential impact of disease duration, disease and serological subtype (e.g., limited and diffuse SSc), and disease severity on appearance concerns, as these characteristics may also play a role in the development of body image issues.

Limitations

A number of limitations should be considered when interpreting the findings of the presented manuscripts. Firstly, the SSc samples across all studies constituted convenience samples of patients receiving treatment at large centres; therefore, results may differ from what might be found in community settings and for different SSc patient groups. Additionally, patients sampled from the SPIN Cohort in Studies Two and Four completed all study measures via an online platform, further limiting the generalizability of findings to all SSc patients.

Another limitation to the body of present research is that White patients made up the majority of the SSc samples utilized. While racial/ethnic differences in the reporting of appearance concerns was examined in Study Four, it was not possible to explore differences based on race/ethnicity in the other studies for issues such as measurement equivalency, due to the small number of patients from non-White racial/ethnic groups. Furthermore, as mentioned above, a limitation of Study Four directly related to the small number of patients from non-White racial/ethnic backgrounds, which potentially affected the ability to detect significant associations between race/ethnicity status and the presence of appearance concerns. The growth of largescale international cohorts such as SPIN (Kwakkenbos et al., 2013) will aid in addressing the lack of racial/ethnicity diversity found within many SSc samples, as, over time, numbers of patients from non-White racial/ethnic backgrounds will increase and allow for greater representation in research studies.

Studies Two and Three that evaluated the measurement equivalency of BCSS and SWAP items, respectively, have unique limitations to the scope of findings. In particular, not all characteristics that could potentially influence item responses were examined in these studies; Study Two explored the influence of sex and age, and Study Three examined the impact of linguistic background, but neither assessed measurement equivalency based on race/ethnicity, for instance. The decision to include these sociodemographic characteristics in the manner conducted was partially based on the specific aims of each study, as well as on the distribution of these variables across the SSc samples. For instance, there were too few people from non-White racial/ethnic backgrounds in both the CSRG and SPIN Cohorts to assess potential DIF across

BCSS or SWAP items based on this characteristic. Next, for both Studies Two and Three, a MIMIC model was utilized to assess DIF. This model implicitly assumes configural invariance, which is a strong assumption (Bauer, 2017). However, compared to a multi-group CFA model, which can be used to fully test measurement invariance, including configural invariance, the MIMIC model can provide greater power to detect DIF, if present (Bauer, 2017). This was important to consider for both these studies, as there were only 115 males in Study Two and 208 French-speaking SSc patients in Study Three; therefore, coverage of some parts of the item response spectrum would have been sparse if these characteristics had been modelled separately using a method such as multi-group CFA.

A further limitation specific to Study Three, which assessed DIF across English-and French-speaking SSc patients, is that the sample was from Canada only. As such, findings were limited to this linguistic and cultural context and require replication in other samples of French-speaking patients. Relatedly, DIF was only assessed for English- versus French-speaking patients and not for any other language groups. However, SSc patients who are part of largescale Cohorts, such as SPIN, or who live in regions with more than one spoken language, may complete measures in languages other than English and French (e.g., Spanish, German, Italian).

Final noteworthy limitations specific to Study Four include that the outcome variable was dichotomous and, while most patients reported the presence of appearance concerns, it is possible that results would have differed if a continuous or more detailed measure of appearance concerns was administered. Furthermore, the majority of the variables utilized to assess disease factors consisted of indicators of the presence or absence of a particular characteristic and did not provide a measure of severity. This may have reduced the ability to identify associations, if present, between the severity of visible differences from SSc and the presence of appearance concerns. Additionally, the race/ethnicity category of “Other” that was used in the study was comprised of a heterogeneous group of individuals from a variety of racial/ethnic backgrounds who did not identify as White or Black, and it was not necessarily particularly informative in relation to the experience of any patient or group of patients classified in this group, limiting the generalizability to a wider range of racial/ethnic identities.

Conclusion

People living with visible differences, whether present from birth or acquired later in life as the result of an injury or illness, commonly experience substantial psychological and psychosocial difficulties related to their appearance. These can include negative reactions from others (e.g., stares, unwanted attention, disgust); unfavourable self-perceptions, such as body image dissatisfaction, shame, anxiety, and depression; as well as social discomfort, often leading to avoidance and concealment behaviours (Bessell et al., 2012; Cash & Pruzinsky, 2002; Newell, 1999; Rosser et al., 2010; Rumsey & Harcourt, 2004). Concerns about appearance may be particularly salient among people with acquired visible differences from a medical illness, as such conditions often carry with them indicators of personal health status and can sometimes worsen pre-existing body image distress as well as feelings of separation from societal appearance ideals (Bessell et al., 2012; Rumsey & Harcourt, 2004; 2012).

SSc is a chronic, multisystem autoimmune connective tissue disease that leads to substantial physical disability and drastic appearance changes (Gholizadeh et al., 2017; Kwakkenbos et al., 2015; Mayes, 2008). The appearance changes from the disease typically affect body parts that are highly visible and relevant to social interactions, such as the face, mouth, and hands (Gholizadeh et al., 2017; Kwakkenbos et al., 2015). As such, disfigurements stemming from SSc are associated with negative outcomes including poor appearance self-esteem, body image dissatisfaction, social discomfort, and general psychological distress (Benrud-Larson et al., 2003; Jewett et al., 2012; van Lankveld, Vonk et al., 2007). Given the potentially significant psychosocial impact of appearance changes from the disease, researchers have called for increased investigations into body image concerns in SSc (e.g., Thombs et al., 2010), as well as the development of interventions to meet the needs of patients living with visible differences (Kwakkenbos et al., 2015; Thombs et al., 2012). In order to address gaps in knowledge and support the design of intervention programs that target body image distress, this thesis aimed to enhance the scope and validity of disease-specific measures of important body image concerns in SSc and to better understand the associations of both sociodemographic and disease factors with appearance-related concerns among patients.

The findings of the present research highlighted concealment as a significant mechanism of body image avoidance for SSc patients through the development and validation of a new, brief, measurement tool of body concealment and avoidance specific to SSc, the BCSS. Results also demonstrated that BCSS scores can be compared and combined across females and males with SSc and those of different ages without concern that measurement differences will affect scores, highlighting the utility of the BCSS as a clinical assessment tool and patient-reported outcome measure of body concealment behaviours that can be administered to a diversity of SSc patients and in large, international cohorts. The current research enhanced psychometric data on another commonly used measure of body image dissatisfaction and social discomfort in SSc, the SWAP, by demonstrating that items do not function differently across English-and French-speaking Canadian SSc patients and that scores can be pooled across these language groups in future studies. Finally, the present thesis identified patients who may be at a higher risk of developing appearance concerns, including females, younger patients, and those with significant hand involvement. Compared to the general population, both women and men with SSc were more likely to report appearance concerns than members of the general population, and this difference was greatest for men and older patients. Patients from White and Black racial/ethnic groups did not differ significantly in terms of the presence of appearance concerns, after accounting for the influence of sociodemographic and disease characteristics; however, Black patients had higher odds of reporting appearance concerns on an unadjusted basis, thus meriting further investigation.

Altogether, present findings provide initial evidence that the BCSS and SWAP could be utilized as patient-reported outcome measures in studies examining body image issues in SSc, as well as in research that aims to evaluate interventions targeting disease-specific appearance concerns. These measures could also be integrated into routine clinical practice as part of regular healthcare appointments to assess and permit discussion of potential psychosocial issues stemming from disfiguring appearance changes from the condition. This thesis also emphasizes the need for body image intervention programs under development to consider the diverse sociodemographic and disease characteristics that may contribute to greater body image distress among patients with SSc.

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Appendix A – Sensitivity Analyses Excluding Canadian Scleroderma Research Group (CSRG) Sites: Study Two

Table 1.

Sensitivity Analysis: Sociodemographic and Disease Variables Excluding Patients from Canadian Scleroderma Research Group (CSRG) Sites

Sociodemographic Variables	
Age (*mean, SD)*	54.7 (12.1)[a]
Female *(n, %)*	593 (86.8)
White *(n, %)*	562 (82.4)[b]
Years of Formal Education (*mean, SD)*	15.5 (3.1)
Employed Full or Part-time *(n, %)*	281 (64.6)[c]
Married or Living as Married *(n, %)*	497 (72.8)
Disease Variables	
Diffuse Scleroderma *(n, %)*	298 (43.6)
Disease Duration in Years (*mean, SD)*	10.2 (8.2)[d]

Note. $N = 683$. Due to missing values: [a]$N = 681$; [b]$N = 682$; [c]$N = 435$; [d]$N = 620$.

Table 2.

Sensitivity Analysis: Confirmatory Factor Analysis Models and Parameter Estimates for BCSS[a] *Excluding Patients from Canadian Scleroderma Research Group (CSRG) Sites*

		Single-Factor Model	Two-Factor Model		Bifactor Model			
Item	Mean (SD)	Factor Loading[b]	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	Factor Loading:[b] General	Factor Loading:[b] Clothing[c]	Factor Loading:[b] Hands[d]	θ[e]
1. I wear clothes I do not like	0.73 (1.11)	0.68	0.71		0.75	-0.23		0.62
2. I wear long sleeves to hide skin changes	0.82 (1.34)	0.84	0.86		0.82	0.25		0.52
3. I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts)	1.68 (1.76)	0.76	0.79		0.77	0.17		0.62
4. I wear clothes that hide the changes to my skin	1.09 (1.46)	0.91	0.94		0.90	0.42		0.43
5. I wear clothes that will divert attention from my appearance	0.80 (1.27)	0.82	0.86		0.89	-0.10		0.45
6. I wear gloves to hide my hands	0.86 (1.33)	0.70		0.76	0.58		0.49	0.65
7. I avoid shaking hands with people	0.93 (1.39)	0.79		0.84	0.61		0.62	0.49
8. I hide my hands so that people don't see them	1.01 (1.33)	0.83		0.89	0.67		0.57	0.48
9. I avoid directly giving change or other items to people	0.72 (1.27)	0.82		0.88	0.68		0.54	0.49

Note. *N* = 683. [a]BCSS responses are scored on a six-point scale ranging from 0-5 (0 = never, 1 = rarely, 2 = sometimes, 3 = often, 4 = usually, 5 = always). [b]Factor loadings are unstandardized, raw factor loadings. [c]Concealment with Clothing factor. [d]Concealment of Hands factor. [e]θ = Square root of the error variance. Sum of error variances = 6.44. ECV (explained common variance) = .78. Coefficient ω (omega) = .89. Coefficient $\omega_{H\text{-}GEN}$ (omega hierarchical – general = .79. $\omega_{H\text{-}Clothing}$ = .01. $\omega_{H\text{-}Hands}$ = .35. PUC (percent of contaminated correlations) = 56%. Single-factor model fit: $\chi^2(27) = 596.41$, CFI = .93, TLI = .91, RMSEA = .18; two-factor model fit: $\chi^2(26) = 152.35$, CFI = .99, TLI = .98, RMSEA = .08; bifactor model fit: $\chi^2(18) = 79.43$, CFI = .99, TLI = .99, RMSEA = .07. Correlation between Concealment with Clothes and Concealment of Hands latent factors = .75.

Sensitivity Analysis: Assessment of the Reliability and Validity of the BCSS Total Score Excluding Patients from Canadian Scleroderma Research Group Sites (N = 683)

- Cronbach's alpha for the BCSS total score was 0.89.
- The BCSS total score correlated strongly with the Brief-Satisfaction with Appearance (Brief-SWAP) Social Discomfort subscale score (r = 0.61); and moderately with the Brief-SWAP Dissatisfaction with Appearance subscale score (r = 0.42), Brief-Fear of Negative Evaluation-II total score (r = 0.41), Social Interaction Anxiety Scale-6 total score (r = 0.42), PROMIS-29 Profile Version 2.0 Depression domain score (r = 0.46), and PROMIS-29 Profile Version 2.0 Pain domain score (r = 0.46).

Appendix B – Body Concealment Scale for Scleroderma (BCSS)

Circle the number that best describes how often you engage in these behaviours at the present time.

	Rarely	**Sometimes**	**Often**	**Usually**	**Always**	**Never**
Concealment with Clothing Factor						
1. I wear clothes I do not like.	0	1	2	3	4	5
2. I wear long sleeves to hide skin changes.	0	1	2	3	4	5
3. I avoid wearing revealing clothes (e.g., bathing suits, tank tops, or shorts).	0	1	2	3	4	5
4. I wear clothes that hide changes to my skin.	0	1	2	3	4	5
5. I wear clothes that will divert attention from my appearance.	0	1	2	3	4	5
Concealment of Hands Factor						
6. I wear gloves to hide my hands.	0	1	2	3	4	5
7. I avoid shaking hands with people.	0	1	2	3	4	5
8. I hide my hands so that people don’t see them.	0	1	2	3	4	5
9. I avoid directly giving change or other items to people.	0	1	2	3	4	5

Appendix C – Satisfaction with Appearance Scale (SWAP)

In each of the following statements, circle the most correct response for you according to this scale:

0 = strongly disagree
1 = disagree
2 = somewhat disagree
3 = neutral
4 = somewhat agree
5 = agree
6 = strongly agree

Social Discomfort Factor						
1. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my family.	0	1	2	3	4	5
2. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of my friends.	0	1	2	3	4	5
3. Because of changes in my appearance caused by my scleroderma, I am uncomfortable in the presence of strangers.	0	1	2	3	4	5
4. Changes in my appearance have interfered with my relationships.	0	1	2	3	4	5
5. I feel that my scleroderma is unattractive to others.	0	1	2	3	4	5
6. I don’t think people would want to touch me.	0	1	2	3	4	5
Dissatisfaction with Appearance Factor						
7. I am satisfied with my overall appearance.	0	1	2	3	4	5
8. I am satisfied with the appearance of my scalp.	0	1	2	3	4	5
9. I am satisfied with the appearance of my face.	0	1	2	3	4	5

10. am satisfied with the appearance of my neck.	0	1	2	3	4	5
11. I am satisfied with the appearance of my hands.	0	1	2	3	4	5
12. I am satisfied with the appearance of my arms.	0	1	2	3	4	5
13. I am satisfied with the appearance of my legs.	0	1	2	3	4	5
14. I am satisfied with the appearance of my chest.	0	1	2	3	4	5