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Scleroderma Caregiver Advisory Com; Canedo-Ayala, Mara; Rice, Danielle B.; Levis, Brooke; Carrier, Marie-Eve; Cumin, Julie; Malcarne, Vanessa L.; Hagedoorn, Mariet; Thombs, Brett D.

Published in:
Disability and Rehabilitation

DOI:
10.1080/09638288.2018.1500647

Publication date:
2020

Disability and Rehabilitation

ISSN: 0963-8288 (Print) 1464-5165 (Online) Journal homepage: https://www.tandfonline.com/loi/idre20

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To cite this article: Mara Cañedo-Ayala, Danielle B. Rice, Brooke Levis, Marie-Eve Carrier, Julie Cumin, Vanessa L. Malcarne, Mariët Hagedoorn, Brett D. Thombs & the Scleroderma Caregiver Advisory Committee (2020) Factors associated with symptoms of depression among informal caregivers of people with systemic sclerosis: a cross-sectional study, Disability and Rehabilitation, 42:3, 394-399, DOI: 10.1080/09638288.2018.1500647

To link to this article: https://doi.org/10.1080/09638288.2018.1500647

DISABILITY AND REHABILITATION
2020, VOL. 42, NO. 3, 394–399
https://doi.org/10.1080/09638288.2018.1500647

Taylor & Francis
Taylor & Francis Group

RESEARCH PAPER

Factors associated with symptoms of depression among informal caregivers of people with systemic sclerosis: a cross-sectional study

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ABSTRACT

Purpose: Our study aimed to identify caregiver characteristics (gender, age, occupational status, educational attainment, relation to care recipient), care recipient characteristics (age, disease subtype), and caregiving factors (hours of care, perceived caregiving burden) associated with symptoms of depression among informal caregivers of persons with systemic sclerosis (also known scleroderma).

as **Materials and methods:** A questionnaire was developed and administered online from December 2016 to June 2017 to informal caregivers of people with scleroderma, including the Patient Health Questionnaire-9 to assess depressive symptoms. Multiple linear regression was used to identify factors associated with depressive symptoms.

Results: Caregivers ($n = 202$) were 60.9% male. Average age was 57.2 years (standard deviation = 14.4 - years). Most caregivers were partners (72.3%), children (11.9%), or parents (7.4%), of care recipients. Self-reported caregiving burden (standardized regression coefficient $\beta = 0.54$, $p < 0.001$) and hours of care per week ($\beta = 0.17$, $p = 0.005$) were significantly associated with greater symptoms of depression.

Conclusions: Depressive symptoms were primarily associated with caregiving burden among a sample of scleroderma caregivers. There is need to develop interventions targeting caregivers in scleroderma. Rehabilitation professionals should consider the specific needs of scleroderma caregivers and should provide or refer to support services as appropriate.

➤ IMPLICATIONS FOR REHABILITATION

- Most people diagnosed with scleroderma are cared for by an informal caregiver and the majority of these caregivers experience mild symptoms of depression.
- For caregivers that experience moderate to severe symptoms of depression, the ability to provide care to a loved one with scleroderma may be more difficult.
- Rehabilitation professionals should be aware of the burden faced by caregivers of persons with scleroderma and provide support services or referrals as appropriate.

ARTICLE HISTORY
Received 9 January 2018
Revised 9 July 2018
Accepted 10 July 2018

KEYWORDS
Caregivers; rare disease; depressive symptoms; scleroderma; systemic sclerosis

Introduction

Informal caregivers are people who provide unpaid care for a friend or family member with a medical condition [1]. Although informal caregivers typically do not receive training for this role, the care and support they provide can be substantial [2]. Caregiving involves providing both emotional and practical support. Taking on a caregiver role often involves significant adjustments to daily life, including having to manage additional tasks, such as household chores, transportation, or assistance with activities of daily living [3,4]. Due to caregiving responsibilities, many informal caregivers have to abandon or reduce the amount of time they can devote to paid employment, which can cause financial strain [5]. For some people, the burden associated with providing informal care and support can lead to emotional, social, and physical health consequences [3,4].

Informal caregiving has been associated with increased depressive symptoms [6–8], which can lead to difficulties with caregiving tasks, such as making decisions about patient care [9]. Given the impact on the patient, considering the wellbeing of caregivers is important when planning the ongoing care and rehabilitation for patients with chronic diseases. A meta-analysis of 84 studies that compared caregivers of people with dementia, stroke, cancer, and other physical and cognitive conditions to non-caregivers found that depressive symptoms were more common in caregivers than non-caregivers [10]. Multivariate analyses with caregivers of people with dementia and Alzheimer's disease have found higher levels of depressive symptoms in caregivers compared to

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non-caregivers, even after controlling for caregiver socio-demographic characteristics [11–13].

Both socio-demographic and caregiving variables have been associated with depressive symptoms. Previous meta-analyses have examined correlates of depression among people providing care for older adults, stroke survivors, and persons with dementia, cognitive, or physical impairments [14–16]. These reviews have identified a number of care recipient (sex), caregiver (younger age, sex, race, education level, employment status, social support, and physical health), and caregiving (hours of care provided per week, duration of caregiving, relationship with the care recipient) factors that were significantly associated with greater depressive symptoms [14–16]. Characteristics that were found to be associated to greater depressive symptoms in more than one meta-analysis included relationship to care recipient and hours of care provided [14–16].

Informal caregivers of people living with a rare disease likely experience similar challenges as informal caregivers of people with more common diseases, but they may also experience additional stressors. Caregivers of people with rare diseases typically have limited access to information and resources on the rare disease and its treatment [17–19]. Many informal caregivers of a person with a rare disease have not met other caregivers of somebody with the same disease, and many do not have a support network of other caregivers [20]. Most existing evidence on the mental health of informal caregivers, however, has focused on caregivers of people with common health conditions, and there is little information available on the experiences of informal caregivers of people with rare diseases [21–23].

Systemic sclerosis, also known as scleroderma, is a rare chronic autoimmune disease, in which abnormal fibrotic processes and excessive collagen production result in skin thickening, damage to internal organs, including the lungs, kidneys, and gastrointestinal tract, and vascular implications [24]. Scleroderma prevalence has been reported to be between 150 and 300 cases per million [25]. Disease onset is most common between 30 and 50 years of age with approximately 80% of people affected being women [25]. Patients with scleroderma experience a range of problems that affect quality of life, including gastrointestinal symptoms, respiratory problems, fatigue, pain, pruritus, symptoms of depression, and disfiguring changes in appearance [26].

Many people with scleroderma are cared for by a family member or friend [20,27]. To date, however, only one study, an unpublished thesis, has examined the experiences of informal caregivers of people with scleroderma. Based on qualitative interviews with 13 informal caregivers, the study reported that emotional distress was an important challenge for caregivers [20]. No studies have examined factors associated with depressive symptoms among informal caregivers in scleroderma. The objective of the present study was to identify factors associated with depressive symptoms among informal caregivers of patients with scleroderma, including caregiver characteristics (gender, age, occupational status, educational attainment, and relation to care recipient), care recipient characteristics (age and scleroderma subtype), and caregiving factors (hours a week providing care and perceived caregiving burden).

Materials and methods

Participant sample and procedure

Participants who were informal caregivers of a person diagnosed with scleroderma were recruited to complete study questionnaires anonymously *via* the online survey tool *Qualtrics* between December 2016 and June 2017. To be eligible for the study, participants had to indicate that they currently or previously provided unpaid care for a friend or family member with scleroderma, were 18 years or older, and were fluent in English or French. Participants were recruited through scleroderma patient organizations, including Scleroderma Canada, the Scleroderma Foundation of the USA, Scleroderma and Raynaud's UK, the Association des Sclérodermiques de France, and the Scleroderma Association of New South Wales, Australia. In addition, participants were recruited *via* emails and posts on scleroderma-related websites and other social media venues (e.g., Twitter, Facebook). Advertisements were also posted in newsletters and *via* emails to people with scleroderma participating in an ongoing internet-based cohort [28]. All participants gave their informed consent prior to their inclusion in the study.

Respondents who accessed the survey website were able to complete the survey online in English or French. After clicking on the survey link and selecting the preferred language, respondents were shown a consent form that briefly described study objectives and survey instructions. Respondents were given the option to provide consent by clicking an arrow to continue the survey or to choose to close their browser and not participate. The survey was set up using cookies to prevent respondents from completing the survey more than once so as to reduce the possibility of duplicate responses. This study was approved by the research ethics committee of the Jewish General Hospital.

Measures

Caregiver, care recipient, and caregiving characteristics were reported by caregivers who completed the survey. Caregiver variables were gender, age, current occupational status, educational attainment, and relationship with the care recipient. Care recipient characteristics were scleroderma subtype (diffuse or limited) and age.

Caregiving variables were hours per week of care and caregiving burden. Burden was measured with the self-report 12-item Zarit Burden Interview (ZBI-12) [29]. The ZBI-12 items are Likert-type items with a 0–4 response format (0 = never and 4 = nearly always). The ZBI-12 is a validated instrument for measuring caregiver burden. Higher scores represent a higher amount of burden. Strong validity and internal consistency have been found for the ZBI-12 among informal caregivers of several diseases, including dementia and cancer [29].

Caregiver symptoms of depression were measured using the 8-item Patient Health Questionnaire (PHQ-8) [30]. The PHQ-8 measures symptoms of depression over the last 2 weeks. Items have a 0–3 response scale (0 = not at all and 3 = nearly every day) with higher scores indicating more depressive symptoms. The PHQ-8 performs similarly to the PHQ-9 [30], and the PHQ-9 has been established as a valid measure of depressive symptoms in scleroderma [31].

Statistical analyses

Descriptive statistics were calculated for demographic and caregiving variables. Categorical variables were presented as percentages and counts, and continuous variables were presented as means and standard deviations.

Simple linear regressions were used to assess the associations between each independent variable and reported symptoms of depression, separately. Multiple linear regression was used to assess the independent associations between caregiver factors

(gender, age, current occupational status, educational attainment, and relationship with the care recipient), care recipient factors (scleroderma subtype and age), caregiving factors (hours of care per week and caregiver burden), and symptoms of depression (dependent variable). Variables to be included in the regression model were specified *a priori* and were selected based on previous studies in other caregiver groups [7,15,32,33].

Prior to conducting regression analyses, preliminary tests were performed in order to confirm that there were no violations of multiple regression assumptions. Assumptions of outliers, normality, linearity, homoscedasticity, and independence of residuals were checked by inspection of the normal probability plot of the regression standardized residual and the scatter plot.

All statistical analyses were two-tailed with a $p < 0.05$ significance level and were performed using SPSS Statistics, version 21.0 (Chicago, IL).

Results

Sample characteristics

A total of 262 people who indicated that they were past or current informal caregivers of a person with scleroderma accessed the survey. Of these, 202 (77.1%) completed the entire survey and were included in this study. Caregivers included 123 men (60.9%), and the average age was 57.2 years (standard deviation = 14.4 - years). Caregivers were partners (72.3%), children (11.9%), parents (7.4%), siblings (3.5%), or friends (5.0%) of the care recipient. Most were employed (49.0%) or retired (39.6%); there were also students (2.0%), and people who indicated they were homemakers, unemployed, on disability, or on a leave of absence (11.4%). Educational attainment included 28.2% who had pursued a postgraduate degree, 26.7% who completed a university degree, 23.8% who completed some university, and 21.3% who completed primary to high school education. Caregivers provided on average 14.3 h of care per week (standard deviation = 13.4), with 81.2% providing under 25 h of care per week, and 18.8% providing 25 h of care per week or more.

Care recipients were on average 58.0 years of age (SD = 13.4). Caregivers reported that the care recipients were diagnosed with diffuse scleroderma (49.5%), CREST or limited scleroderma (33.7%), or that they did not know the subtype (16.8%), see Table 1.

Table 1. Caregiver, care recipient, and caregiving variables among 202 scleroderma informal caregivers.

Variable	
Male sex, *n* (%)	123 (60.9%)
Age in years, mean (standard deviation)	57.2 (14.4)
Current occupational status, *n* (%)	
Employed	95 (47.0%)
Retired	80 (39.6%)
Students	4 (2.0%)
Other	23 (11.4%)
Educational attainment, *n* (%)	
Primary to high school	43 (21.3%)
Some college or university	48 (23.8%)
University degree	54 (26.7%)
Postgraduate degree	57 (28.2%)
Scleroderma subtype, *n* (%)	
Limited scleroderma or CREST	68 (33.7%)
Diffuse scleroderma	100 (49.5%)
Unknown or not specified	34 (16.8%)
Age of person with scleroderma, mean (standard deviation)	58.0 (13.4)
Relation to care recipient, *n* (%)	
Parent	15 (7.4%)
Child	24 (11.9%)
Partner	146 (72.3%)
Sibling	7 (3.5%)
Friend[a]	10 (5.0%)
Hours of care per week, mean, (standard deviation)	14.3 (13.4)
Zarit Burden Inventory-12, mean, (standard deviation)	13.5 (9.8)
Patient Health Questionnaire-8, mean, (standard deviation)	4.6 (5.4)

[a]Includes one cousin and one coworker.
CREST: calcinosis; Raynaud's phenomenon; esophageal dysmotility; sclerodactyly; and telangiectasia.

Assumptions of normality, linearity, homoscedasticity, and independence of residuals

In tests of multicollinearity, predictor variable tolerance values ranged from 0.28 to 0.84, and the variance inflation factors ranged from 1.19 to 3.52, indicating that multicollinearity was not an issue [34]. There was no deviation in the assumption of normality based on the inspection of the normal probability plot of the regression or evidence of violations of assumptions of outliers, linearity, homoscedasticity, and independence of residuals based on the standardized residual and scatter plot inspections.

Correlates of PHQ-8 scores

The mean PHQ-8 score was 4.6 (range 0–24) with 86.6% scoring under 10 and 13.4% scoring 10 or greater. In simple linear regression analyses, greater caregiver age (standardized regression coefficient [β] = −0.15, $p = 0.033$), completing a university degree (reference = postgraduate degree, $\beta = -0.15$, $p = 0.029$), and greater care recipient age ($\beta = -0.25$, $p < 0.001$) were statistically significantly associated with lower PHQ-8 scores (lower depressive symptoms). Higher caregiver burden as measured by the ZBI-12 ($\beta = 0.63$, $p < 0.001$) and more hours of care per week ($\beta = 0.31$, $p < 0.001$) were significantly associated with higher PHQ-8 scores (higher depressive symptoms) (Table 2). In the multiple linear regression analysis, only higher self-reported caregiver burden ($\beta = 0.54$, $p < 0.001$) and more hours of care per week ($\beta = 0.17$, $p = 0.005$) were significantly independently associated with higher PHQ-8 scores (Table 2). Variables in the model accounted for 45.6% of the variance in PHQ-8 scores.

Discussion

The main finding of this study was that hours spent providing care each week and perceived caregiver burden were independently associated with symptoms of depression among a sample of informal caregivers of persons living with scleroderma. In the unadjusted linear regression analyses, caregiver age, care recipient age, and education level were also significantly associated with depression symptom scores, but these variables were not statistically significant in the multiple linear regression.

This is the first study to assess depressive symptoms among caregivers for people with scleroderma using a validated measure of depressive symptoms in a large sample of caregivers. Based on interviews with 13 caregivers of people with scleroderma, the author of a previous study suggested that stress experienced due to caregiving may result in decreased physical and mental health [20]. Our findings show that, consistent with this proposal, hours of care provided and perceived caregiver burden are robustly associated with symptoms of depression.

Our main finding that perceived caregiver burden may be an important factor associated with depressive symptoms in caregivers is consistent with results from other studies of informal caregivers of people with rare and chronic diseases. Studies done on caregivers of people with amyotrophic lateral sclerosis, dementia, and elderly individuals with medical comorbidities, also

Table 2. Caregiver, care recipient, and caregiving variables with symptoms of depression as measured by the PHQ-8 ($N = 202$).

	Unadjusted (simple) linear regression analyses		Multiple linear regression analyses				
Variables	Standardized β	p Value	Raw B	SE	Standardized β	t	p Value
Caregiver factors							
Female sex	0.10	0.152	0.48	0.77	0.04	0.63	0.532
Age in years	−0.15	0.033	−0.03	0.04	−0.09	−0.89	0.374
Current occupation (reference group = employed or student)							
Retired	−0.10	0.172	0.77	0.82	0.07	0.95	0.346
Other	0.01	0.957	−0.64	1.02	−0.04	−0.63	0.530
Educational attainment (reference group = postgraduate degree)							
Primary to high school	−0.02	0.788	−0.58	0.88	−0.04	−0.66	0.509
Some college or university	0.10	0.173	−0.30	0.84	−0.02	−0.36	0.721
University degree	−0.15	0.029	−1.24	0.81	−0.10	−1.52	0.130
Relation to care recipient (reference group = partner)							
Parent	0.10	0.157	0.16	1.39	0.01	0.12	0.907
Child	−0.12	0.080	−0.98	1.34	−0.06	−0.73	0.465
Sibling or friend	0.09	0.219	1.07	1.21	0.06	0.88	0.381
Care recipient factors							
Scleroderma subtype (reference group = limited scleroderma or CREST)							
Diffuse scleroderma	0.05	0.512	−0.58	0.68	−0.05	−0.85	0.399
Unknown or not specified	−0.01	0.870	−0.13	0.89	−0.01	−0.14	0.889
Age in years	−0.25	0.001	−0.04	0.04	−0.10	−1.19	0.235
Caregiving factors							
Hours of care per week	0.31	0.001	0.07	0.02	0.17	2.88	0.005
Zarit Burden Inventory-12 score	0.63	0.001	0.30	0.03	0.54	8.97	0.001

$R^2 = 0.456$; adjusted $R^2 = 0.412$; $p < 0.001$.
CREST: calcinosis; Raynaud's phenomenon; esophagea; dysmotility; sclerodactyly; and telangiectasia.

reported that caregiver burden, measured by the ZBI, was significantly associated with depressive symptoms [35–37].

As in our study, previous studies have also reported that hours of care provided per week are associated with lower levels of emotional wellbeing. In rare diseases, a study of 770 caregivers of people with Duchenne muscular dystrophy, a chromosomal recessive neuromuscular disorder, found a significant association between hours of care per week and anxiety and depression scores on the EuroQol-5D-3L. In that study, 62% of caregivers provided 25 or more hours of care per week [22], which is substantially greater than the 19% reported by scleroderma caregivers in our study. Another study, which involved 67 caregivers of people with Pompe disease, a rare progressive muscular disorder, found that more hours of care per week were associated with greater mental and physical health problems. Caregivers in that study provided an average of 17.7 h of care per week [23], which is greater than the mean of 14.3 h of care per week in our scleroderma caregiver study.

A study of 198,678 Americans sampled from the US population found that 8.6% scored 10 or greater on the PHQ-8 [30], which is less than the 13.4% of scleroderma caregivers in the present study. The mean PHQ-8 score of 4.6 in the present study, on the other hand, was lower than the mean score of 6.6 among 1170 adults with chronic diseases from the US who were enrolled in a study of a chronic disease self-management program [38]. This suggests that caregivers of people with scleroderma may experience higher symptoms of depression than the general population, but lower symptoms than people living with a chronic disease.

The findings of our study have implications for integrating support of scleroderma caregivers into rehabilitation. Specifically, our findings suggest that there is a need for interventions that target caregiver burden. Support groups are interventions in which people going through a similar situation help one another by providing both practical and emotional support [39]. A meta-analysis of 30 studies of support group interventions for caregivers of persons living with dementia reported that both caregiver burden and symptoms of depression decreased with social support provided by the support groups [40]. Developing support groups for caregivers of persons with scleroderma may be one way to decrease the burden associated with caregiving, and in turn, reduce symptoms of depression, which may improve caregiving capabilities [9]. Other interventions may also be useful for scleroderma caregivers. A meta-analysis of 40 studies that examined interventions for caregivers of people with dementia reported that several types of interventions, including support groups, cognitive behavioural interventions, and educational interventions, were found to increase psychological wellbeing. The authors suggested that interventions seem to be effective as long as they include social components or a combination of social and cognitive components [41]. Previous research has also shown that dyadic interventions improve communication between caregivers and their care recipients, which may provide another option to reduce caregiver distress [42,43]. One important consideration is the degree to which interventions can be feasibly delivered in a rare disease context. Thus, an important step will involve research that will help to understand caregiver preferences and implementation challenges in scleroderma. Specifically, due to the rarity of the disease, few caregivers live in any given geographic area, making it difficult to gather caregivers in one place. Consequently, online-based support tools might be a feasible way to implement interventions for scleroderma caregivers.

Limitations

Several limitations should be considered when interpreting the results of our study. First, given the cross-sectional study design, cause and effect cannot be inferred from our findings. It is possible that caregivers with high levels of depressive symptoms may have experienced them prior to taking on a caregiving role. Second, social support was not assessed. Social support has been found to be a protective factor that is associated with decreased burden and symptoms of depression [40,44]. Although this has not been studied specifically in scleroderma, it would be reasonable to assume that social support is also important in this context. Third, care recipient distress was not assessed, which is another factor that has been found to impact caregiver wellbeing

[23,45]. Fourth, the survey was conducted online. Therefore, all participants had to have access to internet services and a computer, which may have influenced the characteristics of our sample. Finally, it is likely that our sample was influenced by the sampling process as recruitment was completed through scleroderma patient organizations. Caregivers who completed our survey may represent those who are more actively involved in or connected to their care recipient's health. Previously formed ties with patient organizations might create an additional support system, lessening the distress experienced by the caregivers in our sample.

Conclusions

In conclusion, multiple linear regression analysis showed that greater caregiver burden and more hours spent providing care per week were independently associated with symptoms of depression among caregivers of persons diagnosed with scleroderma. Rehabilitation professionals should be aware of the mental health burden that is sometimes associated with caring for persons with scleroderma and should provide support or refer, as appropriate. Our findings emphasize the need for scleroderma caregiver interventions. Due to the rarity of the disease, online-based interventions may provide an option to lessen the burden associated with caregiving in scleroderma. Future studies should examine the unique needs of caregivers to persons with scleroderma and their preferences for support services.

Acknowledgements

We thank our Scleroderma Caregiver Advisory Team Members: Marcia Greiten, BA, Goleta, USA; Christy McCaffery, Philadelphia, USA; Judi McDonald, Bedford, Canada; John Michalski, Macomb, USA; Mathieu Ross, Quebec City, Canada; Lisa Spinney, Berlin, USA; Terry Stacey, Oshawa, Canada; Linda Tarantino, Larkspur, USA.

Disclosure statement

No potential conflict of interest was reported by the authors.

Funding

This research was supported by a grant from the Rare Disease Foundation and the BC Children's Hospital Foundation (Funding Reference Number 173-16; PI Rice). Ms. Rice was supported by a Vanier Graduate Scholarship. Ms. Levis was supported by a Canadian Institutes of Health Research Frederick Banting and Charles Best Canadian Graduate Scholarship doctoral award. Dr. Thombs was supported by a Fonds de recherche du Québec - Santé researcher salary award.

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