Title: The financial and travel burden to access care for Canadians with scleroderma: results of a cross-sectional survey

Author names and affiliations:

Logan Trenaman, PhD[1,2] (ORCID: 0000-0002-6399-5741)
Julia Kaal, PhD[1]
Tracey-Lea Laba, PhD[3] (ORCID: 0000-0002-5182-9092)
Abdollah Safari, PhD[4,5] (ORCID: 0000-0001-9715-3086)
Magda Aguiar, PhD[1]
Tiasha Burch, BSc[6,7]
Jennifer Beckett[8]
Sarah Munro, PhD[2,9]
Marie Hudson, MD, MPH[10,11]
Mark Harrison, PhD[1,2,11]

[1] Collaboration for Outcomes Research and Evaluation, Faculty of Pharmaceutical Sciences, University of British Columbia, Vancouver, BC, Canada.
[2] Centre for Health Evaluation and Outcome Sciences, Vancouver BC, Canada.
[3] Centre for Health Economics Research and Evaluation. University of Technology Sydney.
[4] Data, Analytics, Statistic and Informatics (DASI), University of British Columbia, Vancouver BC, Canada.
[5] Department of Mathematics, Statistics and Computer Science, University of Tehran, Iran.
[6] Scleroderma Association of British Columbia, North Vancouver, BC, Canada.
[7] Patient Partner, Vancouver, BC, Canada.
[8] Patient Partner, Kamloops, BC, Canada.
[9] Department of Family Practice, University of British Columbia, Vancouver, BC, Canada.
[10] Division of Rheumatology, Jewish General Hospital and Lady Davis Institute, and Department of Medicine, McGill University, Montreal, QC, Canada.
[11] Arthritis Research Canada, Vancouver BC, Canada.

Corresponding author: Dr. Mark Harrison: mark.harrison@ubc.ca
Word count: 2,396
Acknowledgements: We are grateful to the SPIN cohort for sending the survey to their members. Preparation of this article was possible through the financial support from the BC SUPPORT Unit Health Economics and Simulation Modelling Methods Cluster, which is part of British Columbia's Academic Health Science Network (Award number: HESM-001). The BC SUPPORT Unit receives funding from the Canadian Institutes of Health Research and the Michael Smith Foundation for Health Research. Logan Trenaman holds a CIHR postdoctoral fellowship in patient-oriented research. Mark Harrison is supported by a Michael Smith Foundation for Health Research Scholar Award 2017 (#16813). Sarah Munro is supported by a Michael Smith Foundation for Health Research Scholar Award in partnership with the Centre for Health Evaluation and Outcome Sciences 2019 (#18270).
Competing interests: The authors have no competing interests to declare.
Data sharing statement: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.
Author contributions: LT and AS conducted the statistical analysis. LT and MHa prepared the manuscript. JK, AS, T-LL, MA, TB, JB, SM, MHu provided input into the study design and the revision of the manuscript. The corresponding author attests that all listed authors meet authorship criteria and that no others meeting the criteria have been omitted.

Patient Engagement in Research: This is a patient-oriented research study. Two patient partners were involved in designing the survey of scleroderma patients. Notably, the patient partners defined the objectives of this study at the outset. They requested that the survey include questions related to out-of-pocket costs and travel distance for care and treatment and were involved in designing the survey questions. Further, it was their suggestion to quantify the magnitude of this burden for those in rural and remote communities which is they focus of this analysis. Thus, they have played a central role throughout this project, including identifying the research questions, choosing how the data were analyzed, and writing this manuscript.

ABSTRACT

Background: In Canada's publically funded health care system, patients may still face a substantial financial and travel burden in accessing care. The purpose of this study was to quantify travel distance and out-of-pocket costs borne by Canadians with scleroderma and compare this burden for those living in larger compared to smaller communities.

Methods: We analyzed responses to a web-based survey of people living in Canada with scleroderma. Respondents reported travel distance to their rheumatologist and annual out-of-pocket medical, travel and accommodation, and other non-medical costs. Descriptive statistics were used to describe travel distance and out-of-pocket costs. A log-transformed linear regression model and a two-part model was used to estimate the impact of living in a smaller community on travel distance and out-of-pocket costs respectively.

Results: The survey included 120 Canadians with scleroderma. The mean travel distance was 139 km (SD = 369 km) while the mean annual total out-of-pocket costs were $3,245 (SD=$5,619). Respondents living in smaller communities travelled four times further to their rheumatologist (e^{β}=3.76, 95% CI: 2.22-6.37), and had increased odds of reporting any (OR=2.72, 95 % CI: 1.06-7.42), medical (OR=4.29, 95% CI: 1.83-10.69), and travel costs (OR=2.34, 95% CI: 1.06-5.27). Among those reporting travel and accommodation costs, those in smaller communities reported three times the costs (e^{β}=2.96, 95% CI: 1.16-7.04).

Interpretation: Many patients with scleroderma incur considerable out of pocket costs and this burden is exacerbated for those living in smaller communities.

1 INTRODUCTION

In Canada's publicly funded health care system, the costs borne by patients for accessing care can still be substantial. Patient-borne costs include out-of-pocket payments for drugs, travel, paid caregivers, premiums paid to private insurers, time spent travelling to and receiving treatment, and lost time at paid or unpaid work for the patient or carers.[1] Patient borne costs may impact whether and how often patients access care or the treatment they choose[2,3] and also have direct social and psychological impacts.[4] Cost-related non-adherence to prescriptions was reported by nearly 10% of respondents to the Canadian Community Health Survey,[5] and the introduction of copayments has been shown to decrease prescription use and lead to increased emergency department visits.[6]

Quantifying patient-borne costs is important to elucidate the economic and equity impacts of health system interventions and policies. Patient-borne costs may be a driver of observed inequities in access to care and outcomes, and fall disproportionately on those with lower socioeconomic status, those living in rural and remote communities, and those with chronic conditions.[7–9] For example, a recent survey of 381 British Columbians living in rural and remote regions found that the average travel distance and costs for one episode of care were 1,966km and $777, respectively,[10,11] while a recent systematic review found that annual out-of-pocket medication costs for individuals with a single chronic condition are 2.7 times that of those with none.[12]

Patients with scleroderma, also known as systemic sclerosis, require a lifetime of treatment and frequent contacts with rheumatologists and other health care professionals, making it a condition where patients may incur substantial patient-borne costs. Scleroderma is a rare, chronic, multisystem autoimmune disease characterized by thickening and fibrosis of the skin and internal organs such as the lungs, heart, and gastrointestinal tract[13], leading to significant morbidity and negative impacts on quality of life.[14] Approximately 17,000 Canadians have scleroderma, and the disease is four times more common in women than men.[15] A 2009 study by the Canadian Scleroderma Research Group estimated that the annual productivity loss from missing paid and unpaid work was over $8,000 per patient, substantially more than the direct health care costs.[16] While significant, productivity costs represent just one category of patient-borne costs, and patients may face additional costs for accessing care and treatment.

The purpose of this study is to determine travel distance and out-of-pocket costs borne by Canadians with scleroderma. We also sought to elucidate equity implications in access and costs as it relates to those living in smaller compared to larger communities.

2 METHODS

We collected information on the out-of-pocket costs incurred by people with scleroderma as part of an international online survey which aimed to understand the preferences of people with scleroderma for autologous stem-cell transplant treatment (results reported elsewhere).[17] Participants were recruited to a 'by invitation' open survey sent to the mailing list of the Scleroderma Patient-centered Intervention Network (SPIN) cohort (a group of people with scleroderma from around the world who participate in SPIN's online studies),[18] and two patient organizations, the Scleroderma Association of British Columbia and the Sclérodermie Québec. Participants were eligible if they reported having a diagnosis of scleroderma. Surveys were administered between September 2019 and February 2020. Ethics approval was obtained from the University of British Columbia Behavioural Research Ethics Board (H18-02389).

2.1 Variables

The surveys collected a detailed set of demographic characteristics including age, gender, identity (combinations of aboriginal or indigenous, black, Asian, Caucasian, Hispanic or Latino, South Asian, or free text response), province/territory of residence, whether they live in a remote, rural, or small (<30k population), medium (30k to 100k) or large (>100k) urban centre, and annual net household income. Clinical variables included the type of scleroderma (limited/diffuse/other), age at diagnosis, and type of health insurance (public/private) and overall health. Cost and travel variables assessed the travel distance to their rheumatologist and annual out-of-pocket costs related to their scleroderma for a) medical expenses, b) travel and accommodation, and c) other non-medical expenses. The survey defined other non-medical expenses as costs for alternative medicine or wellbeing treatments and childcare to attend medical appointments.

2.2 Statistical Analysis

Data were analyzed in R version 4.0.4 (Vienna, Austria). This analysis focused specifically on the subset of Canadian respondents with complete data. The independent variable of interest was a binary variable that indicates whether the respondent lived in a large (>100k population) or small community (rural, remote, or small/medium urban centre with <100k population). The dependent variables of interest included travel distance to the respondent's specialist and four categories of annual out-of-pocket costs, including: total, medical, travel/accommodation, and other out-of-pocket costs. Total out-of-pocket costs were the sum of the three other categories. Missing values were assumed to represent zero costs if respondents completed all other survey questions.

Travel distance and cost data were summarized using descriptive statistics. Regression models were used to control for variables that may confound the relationship between the independent and dependent variables. This included demographic (age; gender; household income) and clinical variables (type of scleroderma). Multiple imputation using predictive mean matching was used to impute missing values (R package *mice)*. With respect to modelling, distance and cost data have unique properties. For example, cost data often have a high mass of observations at zero and are right-skewed meaning that traditional, linear regression is ill suited to model the distribution.[19] There are several methods for analyzing such data, including transforming the data, discretizing the data, using a tobit model, and using a two-part model.[20] Data were investigated to determine whether the data were normally distributed (QQ plots and Shapiro-Wilk test) and the proportion of observations at zero.

Travel distance had minimal observations at zero and was modeled using: a log transformation using a standard linear regression, and a generalized linear model (GLM) which assumed a Gamma error distribution with a log-link. The best model was chosen based on the result of the Shapiro-Wilk test and by examining the distribution of the fitted models' residuals. Ultimately travel distance was modelled using a linear model with log transformation given that the Shapiro-Wilk test suggested that the assumption of normality for the linear model with log transformation was reasonable ($p=0.06$) while it was not for the GLM ($p<0.01$). Estimated marginal mean travel distances were computed using the *emmeans* R package. Cost variables also failed the Shapiro-Wilk test and had a high number of observations at zero, ranging from 26 (of 120, 22%) for total costs to 64 (of 120, 53%) for other costs. Thus, costs were modelled using two-part models which included 1) a logistic regression model to predict the probability of reporting any amount of the outcome (e.g., any costs), and a 2) GLM (Gamma, log-link) for nonzero values. Model coefficient estimates were exponentiated and reported as odds ratios (OR) with 95% confidence intervals (logistic regression) or multiplicative increases with 95% confidence intervals ($\exp^{\beta}$). This approach has been used

previously to evaluate out-of-pocket costs in health care. Combined predictions of costs from two-part models were estimated using predictive margins as described by Buttner et al.[21]

3 RESULTS

This survey included 120 Canadians with scleroderma (see Table 1). The median age was 59 (IQR 50, 66), most respondents were female (n= 104, 87%), Caucasian (n=97, 81%), and nearly half were from Ontario (n = 59, 49%) (Table 1). About a third of the sample reported living in rural/remote regions (n=35, 29%) and half reported living in a large metropolitan area (n=59, 49%). In terms of clinical characteristics, the sample was split between reporting diffuse (n = 57, 48%) and limited (n = 58, 48%) types of disease. Respondents from smaller communities were more likely to be female and from British Columbia or Quebec. Table 2 explores travel distance and out-of-pocket costs for all respondents and is disaggregated by the size of their community (three respondents were missing this data).

Forty-three respondents were missing data on household income (Table 1). Respondents living in smaller communities' travel on average almost four times further to see their healthcare provider than those in larger communities (e^{β} = 3.76, 95% CI: 2.22 - 6.37) (Table 3). On average, respondents from larger communities travelled 17 km (95% CI: 9 km - 32 km) to visit their specialist, compared to 65 km (95% CI: 34 km - 124 km) for respondents living in smaller communities (Table 4). On average, those in smaller communities had increased odds of reporting any (OR = 2.72, 95% CI: 1.06 to 7.42), medical (OR = 4.29, 95% CI: 1.83-10.69), and travel costs (OR = 2.34, 95% CI: 1.06-5.27). For those that did report costs, those in smaller communities reported, on average, triple the travel and accommodation costs than those in larger communities (e^{β} = 2.96, 95% CI: 1.16 to 7.04). Table 4 presents mean estimates of out-of-pocket costs for individuals by the size of their community. On average, individuals in large communities report out-of-pocket travel costs of $331 [95% CI: $196-$466] compared to $1,154 [$828-$1,480] for those in smaller communities (Table 4).

4 INTERPRETATION

This patient-oriented research study is the first study to estimate the magnitude of the financial and travel burden faced by Canadians with scleroderma and highlights the extent of inequity in accessing care despite universal health insurance. We have estimated that Canadians with scleroderma travel an average of 139 km to access care and spend an average of $3,245 out-of-pocket every year to manage their condition. Individuals living in smaller communities are disproportionately impacted, with our data suggesting they travel approximately four times further on average to visit their specialist and are at increased odds of reporting any, medical, and travel or accommodation related out-of-pocket costs.

A systematic review found six studies that had described the economic burden of scleroderma.[22] This review included two Canadian studies that describe health system and productivity costs,[16,23] however neither estimated the travel burden or other patient-borne costs. Three international studies included relevant cost categories (e.g., travel, informal care) however in is hard to determine whether these costs were borne by patients.[24–26] There is literature on patient-borne costs for other rheumatic conditions,[27–29] but it is difficult to make comparisons given differences in health conditions and health care systems. Despite this, while one might expect that universal health insurance system would mitigate the impact of out-of-pocket costs, our analysis suggests that Canadians with scleroderma still face a considerable financial burden.

Nearly a quarter of Canadians with rheumatoid arthritis report that out-of-pocket medication costs were never discussed during their consult, despite most patients and providers viewing these costs as "quite" or

"very" important.[30] Dedicating time during the clinical encounter to discuss the burden of out-of-pocket costs could help mitigate this impact. This could involve discussing a lower cost medication or care plan, or changing the time or frequency of follow-up appointments to mitigate the travel burden.[31] Virtual care is another potential solution. A recent study of veterans with rheumatic conditions living in rural areas found that those using virtual care travelled 330 miles fewer and saved $114 per visit compared to those in usual care.[32] Despite this there was no difference in patient satisfaction or health outcomes. A systematic review of virtual care for people with rheumatic conditions has found that it was feasible, patients report high rates of satisfaction, and effectiveness was comparable or higher than face-to-face consultations.[33] It is important to acknowledge that virtual care is not a solution in all circumstances or acceptable to all individuals. In such cases, policy options include supporting specialists to travel and provide care in smaller communities through outreach visits or providing funds to subsidize the cost of travel and accommodation for people who must travel.[11]

While this analysis suggests Canadians with scleroderma incur substantial costs in access care, our estimates are an underestimate of the true burden. The costs reported here do not account for foregone wages due to time off work due to illness. Previous Canadian research has estimated that productivity loss from paid and unpaid work is more than $8,000 annually per patient.[16] Further, our analysis does not account for the impact on friends, family members, and caregivers. Recent research from BC found that 85% of rural residents reported having a travel companion when accessing care, some of whom incur additional financial costs.[10] Lastly, our analysis did not consider the value of the time spent by Canadians with scleroderma and their family, friends, and/or caregivers in accessing care. As stated by Russell, "Patient time is a resource that is essential to the production of health and medical services… Yet patient time is rarely included in costing studies... By excluding it, analysts treat it as though it were free and had no value. As we all recognize in our daily lives, this is not the case. Time is a scarce resource."[34] Methodological work is needed to determine how to value patients time and incorporate these estimates into economic analyses.

4.1 Limitations

This analysis has limitations. Given the cross-sectional nature of the survey, we were not able to disentangle the relationship between travel burden, out-of-pocket costs, and health outcomes. In exploring the impact of community size on these estimates we adjusted for self-reported health status to control for the impact on travel or cost estimates based on community size. That said, the distribution of self-reported health in our sample was comparable between those in smaller and larger communities. Costs were self-reported and may be subject to recall bias. Patients tend to underreport health care resource use,[35] though it is unclear whether this holds for patient-borne costs. If it does our cost estimates would be conservative. When comparing patient-borne costs based on travel distance and out-of-pocket expenditures, we dichotomized our sample as living in large versus smaller communities. This is a broad categorization and likely masks important heterogeneity in patient-borne costs. Our analysis considered the travel distance and out-of-pocket costs but did not account for frequency of physician visits. Due to the added burden for those in smaller communities it is possible that they are accessing care less frequently – a finding that has been observed in people with rheumatoid arthritis.[37]

4.2 Conclusion

Many patients with scleroderma incur considerable out of pocket costs to receive the care they need, and this burden is exacerbated for those living in smaller communities. There is a need for larger studies to

quantify the burden of costs borne by Canadians with scleroderma and other chronic conditions to understand cost drivers and identify potential solutions to ensure equity in access to treatment.

Table 1. Participant characteristics

		Total (n=120)	Large Community (>100k) n=59	Small Community (<100k) n=58
Age, median [IQR]		59.50 [50.00, 66.00]	58.00 [49.00, 65.00]	62.00 [55.00, 66.00]
Gender, n (%)	Female	104 (86.7)	48 (81.4)	53 (91.4)
	Male	16 (13.3)	11 (18.6)	5 (8.6)
Province, n (%)	Alberta	3 (2.5)	1 (1.7)	2 (3.4)
	BC	25 (20.8)	6 (10.2)	16 (27.6)
	Manitoba	3 (2.5)	2 (3.4)	1 (1.7)
	Nova Scotia	1 (0.8)	1 (1.7)	0 (0.0)
	Ontario	59 (49.2)	38 (64.4)	21 (36.2)
	Quebec	28 (23.3)	10 (16.9)	18 (31.0)
	Saskatchewan	1 (0.8)	1 (1.7)	0 (0.0)
City, n (%)	Large	59 (49.2)	59 (100.0)	0 (0.0)
	Medium	16 (13.3)	0 (0.0)	16 (27.6)
	Small	7 (5.8)	0 (0.0)	7 (12.1)
	Rural/Remote	35 (29.2)	0 (0.0)	35 (60.3)
	Missing	3 (2.5)	0 (0.0)	0 (0.0)
Race / ethnicity, n (%) [a]	Indigenous	2 (1.7)	0 (0.0)	1 (1.7)
	Asian	4 (3.3)	4 (6.8)	0 (0.0)
	Caucasian	97 (80.8)	44 (74.6)	51 (87.9)
	Hispanic	4 (3.3)	4 (6.8)	0 (0.0)
	Southeast Asian	2 (1.7)	2 (3.4)	0 (0.0)
	Not listed	8 (6.7)	6 (10.2)	2 (3.4)
	Prefer not to say	1 (0.8)	1 (1.7)	0 (0.0)
Household Income, median [IQR]		$85,000 [$47,000, $120,000]	$85,000 [$50,000, $140,000]	$85,000 [$42,500, $105,000]
	Missing	43 (35.8)	23 (40.0)	19 (32.8)
Scleroderma type, n (%)	Limited	58 (48.3)	24 (40.7)	34 (58.6)
	Diffuse	57 (47.5)	32 (54.2)	22 (37.9)
	Other	5 (4.2)	3 (5.1)	2 (3.4)
Age at diagnosis, median [IQR]		47.00 [35.00, 55.00]	44.00 [35.00, 54.00]	50.00 [36.00, 56.00]
Disease duration, mean (SD)		13.17 (9.46)	12.46 (7.93)	13.91 (11.01)
General health, n (%)	Excellent	4 (3.3)	3 (5.1)	1 (1.7)
	Very Good	16 (13.3)	7 (11.9)	9 (15.5)
	Good	45 (37.5)	23 (39.0)	22 (37.9)
	Fair	41 (34.2)	20 (33.9)	18 (31.0)
	Poor	14 (11.7)	6 (10.2)	8 (13.8)

SD: standard deviation; IQR: interquartile range; [a] participants can report more than one category

Table 2. Travel distance and out-of-pocket costs by community size

	Total (n=120)	Large Community (>100k) n=59	Small Community (<100k) n=58
Distance (km)			
Mean (SD)	139 (369)	81 (389)	201 (349)
Median (IQR)	27 (10-100)	15 (9-30)	70 (24-245)
Range	0-3000	0-3000	4-2200
Costs ($): Travel & Accommodation			
Mean (SD)	$679 ($1,690)	$370 ($981)	$1,019 ($2,180)
Median (IQR)	$15 ($0-$500)	$0 ($0-$200)	$100 ($0-$1,000)
Range	$0-$12,000	$0-$5,000	$0-$12,000
Costs ($): Medical Costs			
Mean (SD)	$1,812 ($3,565)	$1,622 ($4,109)	$2,025 ($3,030)
Median (IQR)	$500 ($0-$1,625)	$0 ($0-$1,000)	$1,000 (63$-$2,500)
Range	$0-$20,000	$0-$20,000	$0-$15,000
Costs ($): Non-Medical Costs			
Mean (SD)	$754 ($2,140)	$732 ($2,660)	$671 ($1,422)
Median (IQR)	$0 ($0-$600)	$0 ($0-$500)	$108 ($0-$903)
Range	$0-$20,000	$0-$20,000	$0-$9,000
Costs ($): Total			
Mean (SD)	$3,245 ($5,618)	$2,724 ($6,250)	$3,716 ($5,037)
Median (IQR)	$1,025 ($175-$3,500)	$780 ($0-$1,650)	$2,035 ($525-$5,000)
Range	$0-$34,000	$0-$34,000	$0-$26,000

SD: standard deviation; IQR: interquartile range

Table 3. Exponentiated regression coefficients and confidence intervals [CI]s for models with travel distance and out-of-pocket costs as the dependent variable

	Travel Distance	Total Costs		Medical Costs		Travel Costs		Other Costs	
	OLS: exp$^{\beta}$ [CI]	*logistic: OR [CI]*	*glm: exp$^{\beta}$ [CI]*	*logistic: OR [CI]*	*glm: exp$^{\beta}$ [CI]*	*logistic: OR [CI]*	*glm: exp$^{\beta}$ [CI]*	*logistic: OR [CI]*	*glm: exp$^{\beta}$ [CI]*
Community Size: Small	**3.76**	**2.72**	1.45	**4.29**	0.82	**2.34**	**2.96**	1.86	1.13
	[2.22 to 6.37]	**[1.06 to 7.42]**	[0.65 to 3.18]	**[1.83 to 10.69]**	[0.34 to 1.98]	**[1.06 to 5.27]**	**[1.16 to 7.04]**	[0.85 to 4.19]	[0.54 to 2.26]
Age	**1.03**	0.95	0.99	**0.94**	1.01	0.96	0.99	0.97	0.97
	[1.01 to 1.05]	[0.91 to 1.00]	[0.95 to 1.02]	**[0.90 to 0.98]**	[0.97 to 1.05]	[0.93 to 1.00]	[0.95 to 1.02]	[0.94 to 1.01]	[0.94 to 1.01]
Gender: Male	0.77	0.91	0.79	0.79	1.07	0.42	3.09	0.48	1.88
	[0.36 to 1.63]	[0.26 to 3.76]	[0.30 to 2.49]	[0.24 to 2.68]	[0.39 to 3.53]	[0.12 to 1.32]	[0.79 to 17.39]	[0.14 to 1.52]	[0.58 to 7.42]
Household Income	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00
	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]	[1.00 to 1.00]
Scleroderma: Diffuse	1.19	1.31	1.35	1.10	1.59	0.84	1.28	1.28	0.93
	[0.70 to 2.02]	[0.51 to 3.43]	[0.68 to 2.69]	[0.47 to 2.59]	[0.80 to 3.22]	[0.38 to 1.87]	[0.59 to 2.82]	[0.58 to 2.89]	[0.49 to 1.77]
Scleroderma: Other	1.96	1.07	0.45	0.84	0.72	1.11	0.60	2.28	0.23
	[0.53 to 7.20]	[0.12 to 24.15]	[0.11 to 3.66]	[0.10 to 8.91]	[0.15 to 6.45]	[0.15 to 10.09]	[0.13 to 6.01]	[0.3 to 21.89]	[0.06 to 1.29]
Health: Fair or Poor	0.66	0.99	0.46	0.67	0.45	1.22	0.87	0.48	0.40
	[0.33 to 1.33]	[0.3 to 3.67]	[0.19 to 1.28]	[0.22 to 2.04]	[0.18 to 1.36]	[0.42 to 3.59]	[0.41 to 2.71]	[0.15 to 1.41]	[0.13 to 1.45]
Observations	116	117	91	117	73	117	59	117	53

bold indicates statistical significance at $p < 0.05$

Table 4. Predicted mean and 95% confidence interval for travel distance and out of pocket costs

	Large Community (>100k) n=59	Small Community (<100k) n=58
Travel distance [a]	17 km [9 km - 32 km]	65 km [34 km - 124 km]
Cost (travel) [b]	$331 [$196 - $466]	$1,154 [$828-$1,480]
Cost (medical) [b]	$1,662 [$1,164 - $2,160]	$2,013 [$1,637-$2,388]
Cost (other) [b]	$759 [$394 - $1,124]	$1083 [$647-$1,520]
Cost (total) [b]	$2,562 [$2,052 - $3,072]	$3,975 [$3,367-$4,583]

[a] predictions from emmeans R package

[b] combined predictions from two-part models

5 REFERENCES

[1] Canadian Agency for Drugs and Technologies in Health. Guidelines for the Economic Evaluation of Health Technologies: Canada, 3rd Edition 2006. https://www.cadth.ca/media/pdf/186_EconomicGuidelines_e.pdf (accessed July 27, 2018).

[2] Huang J, Zhou S, Groome P, Tyldesley S, Zhang-Solomans J, Mackillop WJ. Factors affecting the use of palliative radiotherapy in Ontario. J Clin Oncol Off J Am Soc Clin Oncol 2001;19:137–44. https://doi.org/10.1200/JCO.2001.19.1.137.

[3] Dodd R, Palagyi A, Guild L, Jha V, Jan S. The impact of out-of-pocket costs on treatment commencement and adherence in chronic kidney disease: a systematic review. Health Policy Plan 2018;33:1047–54. https://doi.org/10.1093/heapol/czy081.

[4] Fitch M, Longo CJ. Exploring the impact of out-of-pocket costs on the quality of life of Canadian cancer patients. J Psychosoc Oncol 2018;36:582–96. https://doi.org/10.1080/07347332.2018.1486937.

[5] Law MR, Cheng L, Dhalla IA, Heard D, Morgan SG. The effect of cost on adherence to prescription medications in Canada. CMAJ Can Med Assoc J J Assoc Medicale Can 2012;184:297–302. https://doi.org/10.1503/cmaj.111270.

[6] Tamblyn R, Laprise R, Hanley JA, Abrahamowicz M, Scott S, Mayo N, et al. Adverse events associated with prescription drug cost-sharing among poor and elderly persons. JAMA 2001;285:421–9. https://doi.org/10.1001/jama.285.4.421.

[7] Paszat LF, Mackillop WJ, Groome PA, Zhang-Salomons J, Schulze K, Holowaty E. Radiotherapy for breast cancer in Ontario: rate variation associated with region, age and income. Clin Investig Med Med Clin Exp 1998;21:125–34.

[8] Alter DA, Naylor CD, Austin P, Tu JV. Effects of socioeconomic status on access to invasive cardiac procedures and on mortality after acute myocardial infarction. N Engl J Med 1999;341:1359–67. https://doi.org/10.1056/NEJM199910283411806.

[9] Dunlop S, Coyte PC, McIsaac W. Socio-economic status and the utilisation of physicians' services: results from the Canadian National Population Health Survey. Soc Sci Med 1982 2000;51:123–33. https://doi.org/10.1016/s0277-9536(99)00424-4.

[10] Centre for Rural Health Research. Out-of-Pocket Costs for Rural Residents When Traveling for Health Care. University of British Columbia; 2020.

[11] Kornelsen J, Khowaja AR, Av-Gay G, Sullivan E, Parajulee A, Dunnebacke M, et al. The rural tax: comprehensive out-of-pocket costs associated with patient travel in British Columbia. BMC Health Serv Res 2021;21:1–17.

[12] Sum G, Hone T, Atun R, Millett C, Suhrcke M, Mahal A, et al. Multimorbidity and out-of-pocket expenditure on medicines: a systematic review. BMJ Glob Health 2018;3:e000505. https://doi.org/10.1136/bmjgh-2017-000505.

[13] Calderon LM, Pope JE. Scleroderma epidemiology update. Curr Opin Rheumatol 2021;33:122–7. https://doi.org/10.1097/BOR.0000000000000785.

[14] Hudson M, Thombs BD, Steele R, Panopalis P, Newton E, Baron M. Health-related quality of life in systemic sclerosis: A systematic review. Arthritis Care Res 2009;61:1112–20. https://doi.org/10.1002/art.24676.

[15] Bernatsky S, Joseph L, Pineau CA, Belisle P, Hudson M, Clarke AE. Scleroderma prevalence: demographic variations in a population-based sample. Arthritis Care Res 2009:400–4.

[16] Bernatsky S, Hudson M, Panopalis P, Clarke AE, Pope J, Leclercq S, et al. The cost of systemic sclerosis. Arthritis Care Res 2009;61:119–23. https://doi.org/10.1002/art.24086.

[17] Aguiar M, Laba T-L, Munro S, Burch T, Beckett J, Kaal KJ, et al. Co-production of randomized clinical trials with patients: a case study in autologous hematopoietic stem cell transplant for patients with scleroderma. Trials 2021;22:1–13.
[18] Kwakkenbos L, Cumin J, Carrier M-E, Bartlett SJ, Malcarne VL, Mouthon L, et al. Factors associated with patient-reported likelihood of using online self-care interventions: a Scleroderma Patient-centered Intervention Network (SPIN) cohort study. BMJ Open 2019;9. https://doi.org/10.1136/bmjopen-2019-029542.
[19] Basu A, Manning WG. Issues for the Next Generation of Health Care Cost Analyses. Med Care 2009;47:S109–14.
[20] Boulton AJ, Williford A. Analyzing Skewed Continuous Outcomes With Many Zeros: A Tutorial for Social Work and Youth Prevention Science Researchers. J Soc Soc Work Res 2018;9:721–40. https://doi.org/10.1086/701235.
[21] Büttner M, König H-H, Löbner M, Briest S, Konnopka A, Dietz A, et al. Out-of-pocket-payments and the financial burden of 502 cancer patients of working age in Germany: results from a longitudinal study. Support Care Cancer Off J Multinatl Assoc Support Care Cancer 2019;27:2221–8. https://doi.org/10.1007/s00520-018-4498-1.
[22] Fischer A, Zimovetz E, Ling C, Esser D, Schoof N. Humanistic and cost burden of systemic sclerosis: A review of the literature. Autoimmun Rev 2017;16:1147–54. https://doi.org/10.1016/j.autrev.2017.09.010.
[23] McCormick N, Marra CA, Aviña-Zubieta JA. Productivity Losses and Costs in the Less-Common Systemic Autoimmune Rheumatic Diseases. Curr Rheumatol Rep 2017;19:72. https://doi.org/10.1007/s11926-017-0698-9.
[24] López-Bastida J, Linertová R, Oliva-Moreno J, Serrano-Aguilar P, Posada-de-la-Paz M, Kanavos P, et al. Social/economic costs and health-related quality of life in patients with scleroderma in Europe. Eur J Health Econ HEPAC Health Econ Prev Care 2016;17 Suppl 1:109–17. https://doi.org/10.1007/s10198-016-0789-y.
[25] Chevreul K, Brigham KB, Gandré C, Mouthon L, BURQOL-RD Research Network. The economic burden and health-related quality of life associated with systemic sclerosis in France. Scand J Rheumatol 2015;44:238–46. https://doi.org/10.3109/03009742.2014.976653.
[26] Belotti Masserini A, Zeni S, Cossutta R, Soldi A, Fantini F. [Cost-of-illness in systemic sclerosis: a retrospective study of an Italian cohort of 106 patients]. Reumatismo 2003;55:245–55. https://doi.org/10.4081/reumatismo.2003.245.
[27] Hülsemann JL, Mittendorf T, Merkesdal S, Handelmann S, von der Schulenburg J-M, Zeidler H, et al. Direct costs related to rheumatoid arthritis: the patient perspective. Ann Rheum Dis 2005;64:1456–61. https://doi.org/10.1136/ard.2004.031880.
[28] Nathan N, Nguyen AD, Stocker S, Laba T-L, Baysari MT, Day RO. Out-of-pocket spending among a cohort of Australians living with gout. Int J Rheum Dis 2021;24:327–34. https://doi.org/10.1111/1756-185X.13979.
[29] Shenoi S, Horneff G, Cidon M, Ramanan AV, Kimura Y, Quartier P, et al. The burden of systemic juvenile idiopathic arthritis for patients and caregivers: an international survey and retrospective chart review. Clin Exp Rheumatol 2018;36:920–8.
[30] Kaal KJ, Bansback N, Hudson M, Anis A, Koehn C, Harrison M. Patient-provider communication about medication cost in rheumatoid arthritis. Clin Rheumatol 2021;40:93–100. https://doi.org/10.1007/s10067-020-05188-z.
[31] Hunter WG, Zhang CZ, Hesson A, Davis JK, Kirby C, Williamson LD, et al. What Strategies Do Physicians and Patients Discuss to Reduce Out-of-Pocket Costs? Analysis of Cost-Saving Strategies in 1,755

Outpatient Clinic Visits. Med Decis Making 2016;36:900–10. https://doi.org/10.1177/0272989X15626384.
[32] Wood PR, Caplan L. Outcomes, Satisfaction, and Costs of a Rheumatology Telemedicine Program: A Longitudinal Evaluation. JCR J Clin Rheumatol 2019;25:41–4. https://doi.org/10.1097/RHU.0000000000000778.
[33] Piga M, Cangemi I, Mathieu A, Cauli A. Telemedicine for patients with rheumatic diseases: Systematic review and proposal for research agenda. Semin Arthritis Rheum 2017;47:121–8. https://doi.org/10.1016/j.semarthrit.2017.03.014.
[34] Russell LB. Completing Costs: Patients' Time. Med Care 2009;47:S89–93.
[35] Evans C, Crawford B. Patient self-reports in pharmacoeconomic studies. Their use and impact on study validity. PharmacoEconomics 1999;15:241–56. https://doi.org/10.2165/00019053-199915030-00004.
[36] Bassel M, Hudson M, Taillefer SS, Schieir O, Baron M, Thombs BD. Frequency and impact of symptoms experienced by patients with systemic sclerosis: results from a Canadian National Survey. Rheumatology 2011;50:762–7. https://doi.org/10.1093/rheumatology/keq310.
[37] Sørensen J, Linde L, Hetland ML. Contact Frequency, Travel Time, and Travel Costs for Patients with Rheumatoid Arthritis. Int J Rheumatol 2014;2014:e285951. https://doi.org/10.1155/2014/285951.