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Research

The financial burden of accessing care for people with scleroderma in Canada: a patient-oriented, cross-sectional survey

Logan Trenaman PhD, K. Julia Kaal PhD, Tracey-Lea Laba PhD, Abdollah Safari PhD, Magda Aguiar PhD, Tiasha Burch BSc, Jennifer Beckett, Sarah Munro PhD, Marie Hudson MD MPH, Mark Harrison PhD

Abstract

Background: Patients with scleroderma require a lifetime of treatment and frequent contacts with rheumatologists and other health care professionals. Although publicly funded health care systems in Canada cover many costs, patients may still face a substantial financial burden in accessing care. The purpose of this study was to quantify out-of-pocket costs borne by people with scleroderma in Canada and compare this burden for those living in large communities and smaller communities.

Methods: We analyzed responses to a Web-based survey of people living in Canada with scleroderma. Respondents reported annual out-of-pocket medical, travel and accommodation and other nonmedical costs (2019 Canadian dollars). We used descriptive statistics to describe travel distance and out-of-pocket costs. We used a 2-part model to estimate the impact on out-of-pocket costs of living in a large urban centre (≥ 100 000 population), compared with smaller urban centres or rural areas (< 100 000 population). We generated combined mean estimates from the 2-part models using predictive margins.

Results: The survey included 120 people in Canada with scleroderma. The mean, annual, total out-of-pocket costs were $3357 (standard deviation $5580). Respondents living in smaller urban centres and rural areas reported higher mean total costs ($4148, 95% confidence interval [CI] $3618–$4680) and travel or accommodation costs ($1084, 95% CI $804–$1364) than those in larger urban centres (total costs $2678, 95% CI $2252–$3104; travel or accommodation costs $332, 95% CI $207–$458).

Interpretation: Many patients with scleroderma incur considerable out-of-pocket costs, and this burden is exacerbated for those living in smaller urban centres and rural areas. Health care systems and providers should consider ways to alleviate this burden and support equitable access to care.

Plain language summary: People with scleroderma require a lifetime of treatment. Although many costs are covered, patients often incur out-of-pocket costs. We know very little about how much Canadians with scleroderma spend out of pocket to access care and treatment, and whether costs vary for those living in larger and smaller communities. We asked Canadians with scleroderma how much they spend out of pocket each year and compared out-of-pocket costs between those living in larger and smaller communities. We found that the average Canadian spends over $3000 out of pocket each year for medical care, travel and accommodation and other costs related to their scleroderma. We also found that people living in smaller communities spend nearly $1500 more out of pocket each year compared with those in larger communities, primarily for travel and accommodation. We have shown that Canadians with scleroderma spend a considerable amount out of pocket and that costs are higher for people living in smaller communities.

In Canada's publicly funded health care systems, 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 from paid or unpaid work for the patient or carers.[1] Patient-borne costs may affect whether and how often patients access care or the treatment they choose[2,3] and may also have direct social and psychological impacts.[4] Cost-related nonadherence 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 increase visits to emergency departments.[6]

Quantifying patient-borne costs is important to elucidate the economic and equity impacts of health system interventions

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Correspondence to: Mark Harrison, mark.harrison@ubc.ca

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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 people living in rural and remote regions of British Columbia who travelled to access health care found that the average travel distance and costs for 1 episode of care were 1966 km and $777, respectively;[10] 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.[11]

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 for which 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,[12] leading to substantial morbidity and negative impacts on quality of life.[13] About 17 000 people in Canada have scleroderma, and the disease is 4 times more common among women than men.[14] A 2009 study by the Canadian Scleroderma Research Group estimated that the annual productivity loss from missing paid and unpaid work was more than $13 000 per patient, substantially more than the direct health care costs.[15] Although substantial, productivity costs represent just 1 category of patient-borne costs, and patients may face additional costs in accessing care and treatment.

The purpose of this study was to estimate out-of-pocket costs borne by people in Canada with scleroderma. We also sought to elucidate equity implications as it related to those living in smaller urban centres and rural areas compared with those living in larger urban centres.

Methods

We collected information on the out-of-pocket costs incurred by people with scleroderma as part of an international online survey (primarily Canada, the United States, France and the United Kingdom), which aimed to understand the preferences of people with scleroderma for autologous stem-cell transplant treatment (results reported elsewhere).[16] Participants were recruited to an 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)[17] and 2 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.

Data collection

We developed the survey using focus groups with people with scleroderma, and pilot tested it with the study team, which included 2 patient partners with scleroderma. The survey was developed in English and translated into French using a translation service approved by the SPIN.

Survey respondents reviewed and signed an informed consent form before participation. The form described the research team, the study objectives, why they had been invited and the approximate length of the survey. The survey was hosted on secure servers at the University of British Columbia and files were encrypted and password protected. We invited people who accessed the online survey to enter a prize draw for an iPad (or gift card of equivalent value). Participation in the draw was not contingent on completing the survey. The survey asked 1 question per page. Questions were not randomized, and skip logic was used to ensure that questions were asked only if relevant. Participants could change their answers and skip questions if they preferred not to answer. We did not use cookies to assign unique identifiers to participants; however, we checked Internet Protocol addresses to identify potential duplicate entries.

The survey 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 or territory of residence, annual net household income and whether the participant lived in a rural area, or a small (< 30 000 population), medium (30 000–99 000) or large (≥ 100 000) urban population centre. The latter is based on Statistics Canada's Population Center and Rural Area Classification framework.[18] Clinical variables included the type of scleroderma (limited, diffuse, other), age at diagnosis, type of health insurance (public, private), overall health status and annual out-of-pocket costs for medical expenses, travel and accommodation, or other expenses. The survey question asked, "How much do you normally pay out-of-pocket each year toward the cost of your scleroderma/CREST syndrome for any [medical expenses/travel and accommodation/other expenses]?" The survey described other costs as "additional, medical and non-medical expenses related to your scleroderma/crest such as alternative medicine, wellbeing, or childcare to attend medical appointments." The survey is presented in Appendix 1, Section 1, available at www.cmajopen.ca/content/11/4/E630/suppl/DC1.

Outcomes

The exposure of interest was the size of the respondent's community. Given our sample size, we created a binary variable (community size) that indicated whether the respondent lived in a large urban population centre (≥ 100 000 population) or smaller urban centres or rural areas (< 100 000 population). The dependent variables of interest were annual scleroderma-related, out-of-pocket costs (in 2019 Canadian dollars) for medical expenses, travel and accommodation, other non-medical expenses and total costs. We calculated total costs by adding medical, travel and accommodation and other non-medical expenses.

Statistical analysis

We analyzed data in R, version 4.0.4. We did not weight survey responses to adjust for the nonrepresentative sample. This analysis focused specifically on the subset of respondents living in Canada. We summarized cost data using

descriptive statistics. We used regression models to control for variables that may confound the relationship between community size and out-of-pocket costs. Potential confounders included demographic (e.g., age, gender, household income) and clinical variables (e.g., type of scleroderma, self-reported health status). We used multiple imputation with predictive mean matching to impute missing values (R package mice). This included 3 respondents who were missing data on community size, 43 who were missing data on household income and 7 who were missing data on out-of-pocket costs. Cost data typically have a high mass of observations at 0 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 2-part model.[20] Investigation of data determined that our cost variables had a high number of observations at 0, ranging from 22% (26 of 120) for total costs to 53% (64 of 120) for other costs. Thus, we modelled costs using 2-part models, which included a logistic regression model to predict the probability of reporting any amount of the outcome (e.g., any costs), and a generalized linear model (γ, log-link) for nonzero values. Model coefficient estimates were exponentiated and reported as adjusted odds ratios (ORs) with 95% confidence intervals (CIs) (logistic regression) or multiplicative increases with 95% CIs ($\exp^{\beta}$). We estimated combined predictions of costs from 2-part models using predictive margins, as described by Buttner and colleagues.[21]

Patient partner involvement

The patient-oriented approach in this research study has been described elsewhere.[16,22] Briefly, this project began with a conversation between patients, clinicians and researchers, with the aim of understanding how the patient perspective could be elicited and integrated into the design of randomized controlled trials. Two patient partners (T.B., J.B.) were members of the research team and contributed at all stages of the research process. This included identifying the research questions, writing the funding application, designing the survey, recruiting respondents and interpreting and disseminating the results. With respect to the current analysis, the patient partners identified a need to better understand the financial burden in accessing care and treatment, and whether this burden is exacerbated for those living in smaller communities. To address this research question, the patient partners codeveloped the survey questions related to out-of-pocket costs, reviewed and provided critical feedback on the survey before data collection, supported the analysis and interpretation of the data and contributed to the final manuscript. Further information describing the involvement of patient partners and their contribution is available in Appendix 1, Section 2.

Ethics approval

Ethics approval was obtained from the University of British Columbia Behavioural Research Ethics Board (H18-02389).

Results

The full survey had an overall completion rate of 71.5% (278/389) and a completion rate of 80.5% for respondents in Canada (120/149). The final sample comprised 120 people in Canada with scleroderma (Table 1). The median age was 59.5 (interquartile range [IQR] 50.0–66.0) years; most respondents were women (n = 104, 86.7%) and White (n = 97, 80.8%), and nearly half were from Ontario (n = 59, 49.2%) (Table 1). About a third of the sample reported living in rural regions (n = 35, 29.2%) and half reported living in a large urban centre (n = 59, 49.2%). In terms of clinical characteristics, the sample was split between reporting diffuse (n = 57, 47.5%) and limited (n = 58, 48.3%) types of disease. Respondents from smaller communities were more likely to be women and from British Columbia or Quebec. Respondents reported a median of $1425 (IQR $488–$3500) in out-of-pocket costs (Table 2). Boxplots of the distribution of costs by community size are presented in Appendix 1, Section 3.

Combined estimates from the 2-part models found that that, on average, people living in smaller urban centres or rural areas reported higher mean total costs ($4148, 95% CI $3618–$4680), compared with those in large urban centres ($2678, 95% CI $2252–$3104) (Table 3). Further, those in smaller urban centres or rural areas also reported higher mean out-of-pocket travel and accommodation costs ($1084, 95% CI $804–$1364) than those in large urban centres ($332, 95% CI $207–$458) (Table 3). Analysis of exponentiated model coefficients (Appendix 1, Section 4) found that, compared with participants in large urban centres, those in smaller urban centres or rural areas had increased odds of reporting any medical costs (adjusted OR 3.56, 95% CI 1.51–8.86) and any travel and accommodation costs (adjusted OR 2.17, 95% CI 0.99–4.87). Furthermore, those who incurred out-of-pocket travel and accommodation costs reported, on average, nearly triple the costs (e^{β} = 2.77, 95% CI 1.14–6.27).

Interpretation

We have estimated that people in Canada with scleroderma spend an average of $3300 out of pocket every year to manage their condition. People living in small urban centres or rural areas are disproportionately affected, with our data suggesting they reported greater total costs and travel and accommodation costs.

A systematic review found 6 studies that had described the economic burden of scleroderma.[23] This review included 2 Canadian studies that described health system and productivity costs;[15,24] however, neither study estimated the travel burden or other patient-borne costs. Three international studies included relevant cost categories (e.g., travel, informal care); however, it is hard to determine whether these costs were borne by patients.[25–27] There is literature on patient-borne costs for other rheumatic conditions. Hülsemann and colleagues[28] found that total annual out-of-pocket costs for patients with rheumatoid arthritis were €417.20. Nathan and colleagues[29] estimated that median annual out-of-pocket costs for Australians with gout

Table 1: Participant characteristics

Characteristic	No. (%) of participants*†		
	Total *n* = 120	Large urban centre *n* = 59‡	Small urban centre or rural *n* = 61‡
Age, yr, median (IQR)	59.5 (50.0–66.0)	58.0 (49.0–65.0)	62.0 (55.0–66.0)
Gender			
Woman	104 (86.7)	48 (81.4)	56 (91.8)
Man	16 (13.3)	11 (18.6)	5 (8.2)
Province			
Alberta	3 (2.5)	1 (1.7)	2 (3.3)
British Columbia	25 (20.8)	6 (10.2)	19 (31.1)
Manitoba	3 (2.5)	2 (3.4)	1 (1.6)
Nova Scotia	1 (0.8)	1 (1.7)	0 (0.0)
Ontario	59 (49.2)	38 (64.4)	21 (34.4)
Quebec	28 (23.3)	10 (16.9)	18 (29.5)
Saskatchewan	1 (0.8)	1 (1.7)	0 (0.0)
City§			
Large	59 (49.2)	59 (100.0)	0 (0.0)
Medium	16 (13.3)	0	16 (26.2)
Small	7 (5.8)	0	7 (11.5)
Rural	35 (29.2)	0	38 (62.3)
Race and ethnicity¶			
Indigenous	2 (1.7)	0	2 (3.3)
Asian	4 (3.3)	4 (6.8)	0 (0.0)
White	97 (80.8)	44 (74.6)	53 (86.9)
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 (3.3)
Prefer not to say	1 (0.8)	1 (1.7)	0 (0.0)
Household income, $, median (IQR)	95 000 (56 250–120 000)	95 000 (57 000–125 000)	95 000 (54 000–120 000)
Scleroderma type**			
Limited	58 (48.3)	24 (40.7)	34 (55.7)
Diffuse	57 (47.5)	32 (54.2)	25 (41.0)
Other	5 (4.2)	3 (5.1)	2 (3.3)
Age at diagnosis, yr, median (IQR)	47.0 (35.0–55.0)	44.0 (35.0–54.0)	50.0 (36.0–56.0)
Disease duration, yr, mean ± SD	13.17 ± 9.46	12.46 ± 7.93	13.85 ± 10.76
General health			
Excellent	4 (3.3)	3 (5.1)	1 (1.6)
Very good	16 (13.3)	7 (11.9)	9 (14.8)
Good	45 (37.5)	23 (39.0)	22 (36.1)
Fair	41 (34.2)	20 (33.9)	21 (34.4)
Poor	14 (11.7)	6 (10.2)	8 (13.1)

Note: IQR = interquartile range, SD = standard deviation.
*Unless indicated otherwise.
†Data were imputed for 3 respondents who were missing data on community size and 43 respondents who were missing data on household income.
‡We defined large urban centres as those with 100 000 population or more, and small urban centres or rural areas as those with less than 100 000 population.
§Participants were asked to self-report whether they lived in a rural or metropolitan area. Those living in a metropolitan area were asked to specify the population, which we subsequently grouped into large (≥ 100 000 population), medium (30 000–99 000 population) or small urban centres (< 30 000 population).
¶Participants could report more than 1 category.
**Limited type generally involves the fingers and, potentially, the hands, forearms or face; the diffuse type generally involves more of the body, including organs such as the gastrointestinal tract, kidneys, lungs and heart.

Table 2: Out-of-pocket costs by community size*

Variable	Total *n* = 120	Large urban centre *n* = 59†	Small urban centre or rural *n* = 61†
Total, $			
Mean ± SD	3357 ± 5580	2837 ± 6220	4095 ± 4882
Median (IQR)	1425 (488–3500)	975 (225–1900)	2500 (675–5001)
Range	0–34 000	0–34 000	0–26 000
Medical costs, $			
Mean ± SD	1884 ± 3550	1710 ± 4095	2053 ± 2954
Median (IQR)	500 (0–2000)	400 (0–1100)	1000 (250–2500)
Range	0–20 000	0–20 000	0–15 000
Travel and accommodation costs, $			
Mean ± SD	682 ± 1689	372 ± 981	983 ± 2131
Median (IQR)	0 (0–500)	0 (0–200)	100 (0–1000)
Range	0–12 000	0–5000	0–12 000
Other costs,‡ $			
Mean ± SD	790 ± 2146	755 ± 2655	825 ± 1522
Median (IQR)	8 (0–625)	0 (0–500)	300 (0–1000)
Range	0–20 000	0–20 000	0–9000

Note: IQR = interquartile range, SD = standard deviation.
*In 2019 Canadian dollars. Cost data was imputed for 7 respondents.
†We defined large urban centres as those with 100 000 population or more, and small urban centres or rural areas as those with less than 100 000 population.
‡Other costs were defined as additional medical and nonmedical expenses related to scleroderma/CREST such as alternative medicine, well-being or child care to attend medical appointments.

Table 3: Predicted out-of-pocket costs from 2-part models*

Variable	Mean (95% CI)	
	Large urban centre† *n* = 59	Small urban centre or rural† *n* = 61
Total costs, $	2678 (2252–3104)	4148 (3618–4680)
Medical costs, $	1818 (1367–2269)	2024 (1704–2343)
Travel and accommodation costs, $	332 (207–458)	1084 (804–1364)
Other costs,‡ $	720 (464–977)	852 (609–1095)

Note: CI = confidence interval.
*In 2019 Canadian dollars. Combined estimate which controls for age, gender, household income, scleroderma type, and self-reported health status.
†We defined large urban centres as those with 100 000 population or more, and small urban centres or rural areas as those with less than 100 000 population.
‡Other costs were defined as additional medical and nonmedical expenses related to scleroderma/CREST such as alternative medicine, well-being or child care to attend medical appointments.

were AU$200 (IQR $60–$750). Shenoi and colleagues[30] conducted an international survey of 61 patients with systematic juvenile idiopathic arthritis and found mean annual travel and treatment expenses of $984 (standard deviation [SD] $1610) and $969 (SD $800), respectively. Comparisons between health conditions and systems are inherently difficult; however, our estimates for travel and treatment costs are in line with those reported by Shenoi.[30] Notably, although one might expect that publicly funded health care would mitigate the impact of out-of-pocket costs, our analysis suggests that people with scleroderma in Canada still face a considerable financial burden.

In Canada, nearly a quarter of people 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.[31] 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.[32] Virtual care is another potential solution. A recent study of veterans with rheumatic conditions living in rural areas in the United States found that those using virtual care travelled 330 miles fewer and saved $114 per visit, compared with those in usual care.[33] Despite this, there was no difference in patient satisfaction or health outcomes. A systematic review of virtual care for people with rheumatic conditions found that it was feasible, patients report high rates of satisfaction and effectiveness was comparable or higher than face-to-face consultations.[34] Virtual care is not a solution in all circumstances or acceptable to all patients. 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.[10]

Although this analysis suggests that people in Canada with scleroderma incur substantial costs in accessing care, our estimates are an underestimate of the true burden. The costs reported here do not account for forgone wages from time off work because of illness. Previous Canadian research has estimated that productivity loss from paid and unpaid work is more than $13 000 annually per patient with scleroderma.[15] 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 patients with scleroderma and their family, friends 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."[35] Methodological work is needed to determine how to value patients' time and incorporate these estimates into economic analyses.

Limitations

Our analysis recruited patients using the mailing lists of Canadian organizations for patients with scleroderma, and used data from an international survey focused on the preferences of people with scleroderma for autologous stem-cell transplant. Therefore, our sample may not be representative of the broader population of people in Canada with scleroderma, such as those not affiliated with scleroderma patient organizations. Given the cross-sectional nature of the survey, we were not able to disentangle the relationship between 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 cost estimates. Costs were self-reported and may be subject to recall bias. Further, we did not provide examples of medical or travel and accommodation costs, which means that respondents used their discretion when deciding what costs to report. Patients tend to under-report health care resource use,[36] although it is unclear whether this holds true for patient-borne costs. If it does, our cost estimates would be conservative. We imputed missed data for several variables, notably costs (n = 7) and community size (n = 3), which assumes that any differences in the missing data (compared with those observed) can be completely accounted for by the other variables collected in the survey. Scleroderma is a heterogenous disease, and disease severity may affect out-of-pocket costs. Our sample included an equal number of participants with limited and diffuse scleroderma and, thus, our estimates likely underestimate the out-of-pocket costs of patients with more advanced, diffuse scleroderma. When comparing patient-borne costs, we dichotomized our sample as living in large urban centres and small urban centres or rural areas. This is a broad categorization and likely masks important heterogeneity in patient-borne costs. Our analysis considered out-of-pocket costs but did not account for frequency of physician visits. Given the added burden for those in smaller urban centres or rural areas, it is possible that they are accessing care less frequently — a finding that has been observed in people with rheumatoid arthritis.[37]

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 small urban centres or rural areas. Larger studies are needed to quantify the burden of costs borne by patients in Canada with scleroderma and other chronic conditions to understand cost drivers and identify potential solutions to ensure equity in access to treatment.

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Affiliations: Centre for Health Evaluation and Outcome Sciences (Trenaman, Munro, Harrison), Vancouver, BC; Department of Community Health and Epidemiology (Kaal), Dalhousie University, Halifax, NS; Clinical and Health Sciences Unit (Laba), The University of South Australia, Adelaide, Australia; Centre for Health Economics Research and Evaluation (Laba), University of Technology, Sydney, Australia; College of Science (Safari), School of Mathematics, Statistics and Computer Science, University of Tehran, Tehran, Iran; Collaboration for Outcomes Research and Evaluation (Aguiar, Harrison), Faculty of Pharmaceutical Sciences, University of British Columbia, Vancouver, BC; Scleroderma Association of British Columbia (Burch), North Vancouver, BC; Patient partner (Burch), Vancouver, BC; Patient partner (Beckett), Kamloops, BC; Department of Obstetrics and Gynaecology (Munro), University of British Columbia, Vancouver, BC; Division of Rheumatology (Hudson), Jewish General Hospital and Lady Davis Institute; Department of Medicine (Hudson), McGill University, Montréal, Que.; Arthritis Research Canada (Hudson, Harrison), Vancouver, BC

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