

Burden of employment loss and absenteeism in adults and caregivers of children with sickle cell disease

Rachel D'Amico Gordon,¹ Rina Li Welkie,² Nives Quaye,² Jane S. Hankins,³ Adetola A. Kassim,⁴ Alexis A. Thompson,^{5,6} Marsha Treadwell,⁷ Chyongchiou J. Lin,⁸ and Robert M. Cronin¹

¹Department of Internal Medicine, and ²Division of Hematology, The Ohio State University College of Medicine, Columbus, OH; ³Departments of Global Pediatric Medicine and Hematology, St Jude Children's Research Hospital, Memphis, TN; ⁴Department of Hematology/Oncology, Vanderbilt University Medical Center, Vanderbilt-Meharry Center for Excellence in Sickle Cell Disease, Vanderbilt University Medical Center, Nashville, TN; ⁵Department of Pediatrics, Department of Medicine, Northwestern University, Evanston, IL; ⁶Division of Hematology, Children's Hospital of Philadelphia, Philadelphia, PA; ⁷Department of Pediatrics, Division of Hematology, University of California San Francisco, San Francisco, CA; and ⁸College of Nursing, The Ohio State University, Columbus, OH

Key Points

- Adults and caregivers of children with sickle cell disease have high rates of employment loss and absenteeism related to hospitalization.
- Not living with a partner is associated with increased employment loss and missed work for families affected by sickle cell disease.

Sickle cell disease (SCD) is a genetic disorder affecting 100 000 people with an estimated annual medical cost of \$3 billion in the United States; however, the economic impact on patients is not well described. We aimed to examine the indirect economic burden and test the hypothesis that socioeconomic status and greater social vulnerability risks are associated with increased absenteeism and employment loss. We surveyed adults and caregivers of children with SCD at 5 US centers from 2014 to 2021. Logistic regression models were used to examine the associations of employment loss and missed days of work with demographics and social determinants. Indirect costs were estimated by multiplying the self-reported missed days of work and job loss by 2022 average wages by the state of the participating institution. Of the 244 participants, 10.3% reported employment loss in the last 5 years, and 17.5% reported missing 10 or more days of work. Adults had 3 times more employment loss compared with caregivers of children with SCD (OR, 3.18; 95% CI, 1.12-9.01) but fewer missed days of work (OR, 0.24; 95% CI, 0.11-0.51). Participants who did not live with a partner reported increased employment loss (OR, 4.70; 95% CI, 1.04-21.17) and more missed days of work (OR, 4.58; 95% CI, 1.04-20.15). The estimated annual indirect economic burden was \$2 266 873 (\$9290 per participant). Adults with SCD and caregivers of children with SCD commonly report employment loss and missed days of work as important risk factors. The high indirect economic burden suggests that future economic evaluations of SCD should include SCD-related indirect economic burden.

Introduction

Sickle cell disease (SCD), a hereditary disorder of the hemoglobin structure within red blood cells, affects >100 000 Americans,¹ and leads to debilitating pain episodes, disability, and early mortality.² As the vast majority of individuals with SCD are Black/African Americans,¹ this compounds other existing health and socioeconomic disparities. The direct economic costs of SCD care have been studied extensively.³⁻⁷ The average annual medical costs are ~\$10 000 for a child and \$34 000 for an adult with SCD,³ with an estimated lifetime cost of \$1.6 million per person.⁴ Given the complexities of SCD,

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The full-text version of this article contains a data supplement.

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procedures as simple as blood transfusions have significantly increased associated costs,^{5,8} and costs exponentially increase as individuals develop end organ damage.⁷

Despite the extensive interest in direct health care costs, little is known about their full economic impact on people with SCD. The projected lifetime income of a person with SCD is \$500 000 lower than that of matched individuals.⁹ There are many ways in which SCD can impact personal productivity¹⁰; over half of the people with SCD report a reduction in work hours and school achievement related to their disease,¹¹ and studies have shown that vaso-occlusive crises (VOCs) significantly impact work productivity.¹² However, the effect of SCD on employment remain poorly understood. The unemployment rates in adults with SCD vary from 25% to 60%.¹³ More individuals in the United States report a negative impact of SCD on their employment/schooling compared with other high- and middle-income countries,¹⁴ and employment status in SCD is associated with decreased health care use.^{15,16} The indirect costs associated with SCD have been studied largely in single-center studies, are often model-based, and are usually limited to adults and children.¹⁷ Empirical studies on the productivity costs of families affected by SCD in the United States are currently lacking.¹⁸ It is critical to understand not only the burden of loss of work productivity for families affected by SCD but also the predictors of absenteeism and employment loss to allow targeted interventions to improve financial health in those most at risk for hardship.

Our study aimed to test the hypothesis that greater social vulnerability risks (eg, lower education, not living with a partner, and difficulty paying bills) are associated with increased absenteeism and employment loss. We also characterized the prevalence and economic burden of work absenteeism and employment loss in adults and caregivers of children with SCD and the indirect economic costs at multiple institutions across the United States.

Methods

This project included data from the Mid-South CDRN, established in 2014¹⁹ and funded by the Patient-Centered Outcomes Research Institute, and a multisite feasibility randomized controlled trial.²⁰ This study was approved by the Institutional Review Boards of all participating sites, and informed consent was obtained from all participants.

Setting and procedure

We surveyed adults aged 18 years and older with SCD and caregivers of children with SCD between 0-16 years old. The following 5 SCD centers across the United States participated in questions regarding employment loss and absenteeism: Lurie Children's Hospital of Chicago, University of Tennessee Health Science Center, University of San Francisco, The Ohio State University, and Vanderbilt University. Three institutions (Lurie Children's Hospital of Chicago, University of Tennessee Health Science Center, and University of California San Francisco) were surveyed between October 2014 and March 2016, and 2 institutions (The Ohio State University and Vanderbilt University) were surveyed between 2018 and 2022. The inclusion criteria for survey participants in both studies included being over the age of 18 years, being able to read and speak English, having a diagnosis of SCD (all genotypes included) or being parents/caregivers of a child with SCD, and receiving care at one of the contributing

centers. Individuals with SCD and their caregivers were recruited by research personnel during clinic visits or via clinic flyers. Participants completed the surveys on electronic tablets or paper-and-pencil, with members of the research team present if they had questions or required assistance. Participants were compensated for their time with a gift card upon survey completion.

Surveys

The surveys were designed in collaboration with various stakeholders, including individuals with SCD. Question domains included sociodemographic variables, social support systems, and employment loss. The full survey are described in detail elsewhere.²¹⁻²³ Participants were asked 2 questions about whether they had lost their jobs due to needing hospitalization: (1) employment loss in the last year and (2) employment loss in the last 5 years. They were also asked how many days of work they had missed in the last year because of hospitalization. Of note, the question did not specify whether the hospitalization was due to SCD or another illness. Given the small available sample, survey results from the 2 studies were pooled together; the studies had similar inclusion criteria, and all included questions had identical wording. The demographics of the included adults were reviewed and were found to be similar (supplemental Table 1). All institutions were geographically remote from each other, making the likelihood of repeated survey participation low.

Statistical analysis

The study data were collected, deidentified, and managed using the REDCap electronic data capture tools. Surveys were excluded if participants did not answer basic demographic information (age and gender) or any questions regarding absenteeism or employment loss. We reported descriptive statistics (mean, standard deviation, and distribution) for the demographics of the sample, frequency of job loss, and number of missed days due to hospitalization.

Bivariate associations among employment loss in the last year, employment loss in the last 5 years, and the number of missed days due to hospitalization were explored with demographics and social determinants of health using univariable logistic regression models. Participants who reported "Not applicable" to these questions were excluded from the statistical test. Models were created for all participants, and separate models were created for adults with SCD and caregivers of children with SCD to understand whether the risk factors differed between these 2 groups. Notably, race/ethnicity was not studied because most participants reported being Black/African American (Table 1). Sex was not studied in the caregiver model, as sex reported is that of the patient (or child of the caregiver) and was thought unlikely to be related to employment loss. We also created multivariable logistic regression models, including all studied variables as predictors, for all participants. Given the large proportion of participants reporting "not applicable," multivariable analyses for the adult and caregiver subgroups were not performed due to small sample size. Analyses were performed using Stata version 17.0 and SAS version 9.4.

Economic burden estimation

Income estimates were obtained via the 2021 Current Population Survey Annual Social and Economic Supplements conducted by the US Census Bureau, which includes pretax wages and fringe benefits.²⁴ Annual salaries were calculated based on educational

Table 1. Sociodemographic information of adults and caregivers of children with SCD

Total		Adults	Children*	Combined
		168 (68.9%)	76 (31.1%)	244
Age of patient in years, mean (SD)		33.2 (12.1)	8.1 (4.7)	25.4 (15.6)
Sex of patient	Male	64 (38.1%)	40 (52.6%)	107 (42.6%)
	Female	102 (60.7%)	36 (47.4%)	135 (56.6%)
	Other	2 (1.2%)	0 (0%)	2 (0.8%)
Race	Black/African American	165 (98.2%)	71 (93.4%)	236 (96.7%)
Highest degree or level of school	High school graduate or less	70 (42.4%)	28 (54.9%)	98 (45.4%)
	Some college or beyond	92 (55.8%)	13 (25.5%)	105 (48.6%)
	Prefer not to answer	3 (1.8%)	10 (19.6%)	13 (6%)
Relationship status	Married/living together	51 (30.4%)	12 (15.8%)	63 (25.8%)
	Unmarried	108 (64.3%)	50 (65.8%)	158 (64.8%)
	Prefer not to say	9 (5.4%)	14 (18.4%)	23 (8.4%)
Difficulty paying monthly bills	Not very/not at all	67 (39.9%)	38 (50%)	105 (43.0%)
	Somewhat/very	98 (58.3%)	38 (50%)	136 (55.7%)
	Prefer not to say	3 (1.8%)	0 (0)	3 (1.2%)
Site	Chicago	14 (8.3%)	76 (100%)	90 (36.9%)
	OSU	25 (14.9%)	0	25 (10.2%)
	UCSF	51 (30.4%)	0	51 (20.9%)
	VUMC	32 (19%)	0	32 (13.1%)
	UTHSC	46 (27.4%)	0	46 (18.8%)

*Data obtained from caregivers of children with SCD.

attainment–matched national averages. Daily wages were determined by assuming 240 workdays per year, after excluding weekends and leave.²⁵ For participants who did not answer educational attainment, the US average annual income²⁶ was used to estimate the income loss. Absenteeism costs were estimated by multiplying the number of reported days of work missed due to hospitalization in the last year by the estimated daily wage. Employment loss costs were estimated by including all participants who reported employment losses in the last 1 or 5 years and multiplying the number of participants by their estimated wage. Because we did not ask how long the participants were out of work, we reported this as an annual potential earning loss.

Results

Demographics

A total of 268 participants were surveyed: 171 adults with SCD and 76 caregivers of children with SCD. A total of 25 participants (9.3%) were excluded from the analysis because they did not answer basic demographic data (age and sex) or did not answer any of the employment questions, leaving 244 participants analyzed. The average age of the adults was 33.2 years (SD: 12.1) and the average age of the children was 8.1 years (SD: 4.7), with 38.1% of adults and 52.6% of children being male (Table 1). Notably, caregivers only reported the sex of their child; thus, the sex of the caregiver filling out the survey was not known. Caregivers reported lower education levels on average than adults with SCD. There were similar rates of participating adults and caregivers who reported not living with a partner (64.3% adults vs 65.8% caregivers).

Two institutions participating in the study collected data regarding employment from 57 adults with SCD (supplemental Table 2). A total of 48.3% of the adults with SCD surveyed reported current employment; 20.7% reported that they were out of work and only 1.7% reported being unable to work or disabled.

Employment loss and absenteeism are prevalent in families affected by SCD

In total, 6% (n = 13) reported employment loss in the last year, and 10.3% (n = 25) reported employment loss in the last 5 years due to emergency room visits or hospitalization (Table 2). Adults had a much higher rate of employment loss in the last 5 years than caregivers of children with SCD: 11.9% vs 6.7%, respectively. A total of 22.1% (n = 48) reported missing 5 or more days of work in the last year due to hospitalization. Twice as many adults with SCD missed 10 or more days compared with caregivers of children: 21.3% vs 10.7%, respectively.

Larger household size and not living with a partner were associated with employment loss and absenteeism

Bivariate logistic regression models for patient characteristics and employment loss are shown in Table 3. Notably, because of the low number of caregivers reporting job loss (<5), the regression models could not be fitted for caregivers and were not used. A larger household size was associated with lower rates of employment loss in the last year (odds ratio [OR], 0.65; P = .046). Employment loss in the last 5 years was more likely for adults than for caregivers of children (OR, 3.18; P = .031). Not living with a

Table 2. Employment loss and absenteeism as reported by adults and caregivers of children with SCD

	Adults	Children*	Combined
In the last year, have you lost job because you had to go to the emergency room or hospital?			
Yes	9 (6.4%)	4 (5.3%)	c13 (6%)
No	59 (42.1%)	56 (74.7%)	115 (53.5%)
N/A	72 (51.4%)	15 (20%)	87 (40.5%)
In the last 5 years, have you lost job because you had to go to the emergency room or hospital?			
Yes	20 (11.9%)	5 (6.7%)	25 (10.3%)
No	68 (40.5%)	54 (72%)	122 (50.2%)
N/A	80 (47.6%)	16 (21.3%)	96 (39.5%)
How many days of work in the last year were missed because of hospitalization(s)?			
0-2 days	13 (9.1%)	23 (30.7%)	26 (16.6%)
3-5 days	8 (5.6%)	8 (10.7%)	16 (7.4%)
6-10 days	3 (2.1%)	7 (9.3%)	10 (4.6%)
10 or more days	30 (21.3%)	8 (10.7%)	38 (17.5%)
N/A	88 (62%)	29 (38.7%)	117 (53.9%)

*Data obtained from caregivers of children with SCD.

partner was also associated with a higher rate of employment loss in the last 5 years in the combined model (OR, 4.70; $P = .044$).

Bivariate logistic regression models among patient characteristics and number of days missed at work are shown in Table 4. Adults had significantly lower missed day rates than caregivers (OR, 0.24; $P < .005$). A larger household size was associated with more frequent missed days (OR, 1.26; $P = .049$). Caregivers of children who did not live with a partner reported missing more days of work (OR, 4.58; $P = .044$).

In multivariable analysis (Table 3), adults were statistically less likely to report missed days of work (OR, 0.21; $P = .005$). Not living with a partner approached statistical significance as a predictor of employment loss in the last 5 years (OR, 4.88; $P = .051$).

The indirect economic burden of absenteeism and employment loss was ~\$2 266 873 annually

By using the number of days participants reported missing work due to SCD in the last year multiplied by the average wage of the corresponding educational attainment (supplemental Table 3), we estimated that participants reported a total of \$144 726, ranging from \$0 to \$4811 with mean of \$586 per participant, in lost wages due to missed days of work. Employment loss is estimated to cost \$2 122 147 annually in lost wages, yielding a total annual indirect economic burden of \$2 266 873. Importantly, this only represents lost wages for participants who reported working in the last 5 years and excludes participants who were not working due to disability or other reasons.

Discussion

In this study, we illustrated that employment loss and absenteeism due to hospitalization represent a common problem for adults with SCD and caregivers of children with SCD at multiple institutions across the United States. More than 10% of the participants reported employment loss in the last 5 years due to hospitalization,

and >20% reported missing >5 days of work in the last year due to hospitalization. Our participants reported a total of \$2 266 873 per year of missed income due to employment loss and absenteeism, or \$9290 per participant annually. This economic burden represents nearly the entirety of the average annual medical costs for a child and one-third of an adult's expected direct health care costs.³ Importantly, participants were only asked to report employment loss due to hospitalization and unrelated to other causes (eg, home management of vaso-occlusive pain episodes and medical appointments). Participants from 2 institutions demonstrated that unemployment rates are much higher for families impacted by SCD than for the general population²⁷; 20.7% of participants reported being out of work, compared with 4.7% of families that included an unemployed person in the United States. SCD has many impacts that may make maintaining employment significantly challenging, including the need for frequent appointments, home management of vaso-occlusive pain episodes,²⁸ cognitive deficits,¹³ and chronic disability.²⁹ Simultaneously, employment loss and absenteeism increase financial burdens on families, which are already high, given the high cost of medical care for SCD. Given the significant direct and indirect economic burden of SCD, these costs should be considered when assessing the value of new curative therapies that may have a high upfront cost but could significantly alter patients' ongoing health and productivity. If our per-participant estimated cost is extrapolated to >100 000 Americans living with SCD,¹ absenteeism and employment represent >\$900 million of lost wages, a large-scale loss of productivity, and economic growth that demands action. Notably, this disproportionately affects 2 groups that already struggle with economic disenfranchisement: people who are Black, given the demographics of SCD, and women who are more likely to be caregivers for family members.³⁰

Employment challenges impact adults with SCD and caregivers of children with SCD differently. Although adults were more likely to report employment loss in the last 5 years, caregivers and adults had different levels of missed days of work due to hospitalization. Although both lead to decreased earnings and financial hardships, our results suggest that adults and caregivers need separate support to improve their employment stability. Individuals living with a partner had significantly lower rates of employment loss for the total sample and missed days of work for caregivers. Notably, no other social determinants of health (including age, education, and ability to pay bills) were associated with employment loss or missed workdays, suggesting that this is a challenge for families impacted by SCD from various backgrounds. Increased household size was associated with less employment loss but more missed days of work, suggesting a need for further study on how social support systems impact employment in families affected by SCD.

We suggest several potential mechanisms for the associations between employment loss and absenteeism. Living with a partner may help decrease the burden of unpaid work at home and childcare, if applicable, which may cause difficulties in completing employment requirements. Nationally, married couples have lower unemployment rates than single-adult households,²⁷ suggesting that this remains true for families with and without SCD. However, living with a partner may also help prevent depression or worsening social stigma, 2 significant concerns for people with SCD.^{31,32} One study has shown that people with SCD were most likely to receive emotional support from those living with them, as opposed to support outside of the home.³³ This support may have a

Table 3. Logistic regression models for employment loss in the last 1 and 5 years

		Employment loss in last 1 year			
Combined (Adults & caregivers)		Odds Ratio	95% CI	Unadjusted P-value	Adjusted P-value
Participant type	Adult (\geq 18 y/o)	2.14	(0.62, 7.33)	.228	.571
Education	Some college or more	0.50	(0.12, 2.06)	.337	.177
Household size	By one unit increase	0.65	(0.43, 0.99)	.046*	.235
Children in home	By one unit increase	0.71	(0.44, 1.15)	.162	
Relationship status	Not living with a partner	1.69	(0.35, 8.21)	.516	.317
Bills	Somewhat or very difficult	0.73	(0.23, 2.31)	.593	.992
Adults					
Age		0.97	(0.90, 1.04)	.355	
Sex	Male	1.00	(0.23, 1.42)	.233	
Education	Some college or more	0.50	(0.11, 2.31)	.377	
Household size		0.57	(0.30, 1.09)	.089	
Children in home		0.58	(0.25, 1.34)	.201	
Relationship status	Not living with a partner	2.56	(0.29, 22.59)	.397	
Bills	Somewhat or very difficult	0.34	(0.08, 1.49)	.152	
		Employment loss in last 5 years			
Combined					
Participant type	Adult (\geq 18 y/o)	3.18	(1.12, 9.01)	.030*	.061
Education	Some college or more	1.04	(0.41, 2.62)	.932	.375
Household size		0.93	(0.72, 1.19)	.544	.670
Children in home		0.91	(0.68, 1.22)	.526	
Relationship status	Not living with a partner	4.70	(1.04, 21.17)	.044*	.051
Bills	Somewhat or very difficult	1.48	(0.60, 3.65)	.390	.504
Adults					
Age		0.97	(0.92, 1.02)	.224	
Sex	Male	0.94	(0.35, 2.56)	.375	
Education	Some college or more	0.76	(0.28, 2.07)	.587	
Household size		1.03	(0.74, 1.43)	.866	
Children in home		0.88	(0.57, 1.36)	.565	
Relationship status	Not living with a partner	4.49	(0.95, 21.27)	.059	
Bills	Somewhat or very difficult	1.31	(0.46, 3.74)	.616	

Unadjusted P-value with univariate logistic regression analysis for all participants and adult participants. Adjusted multivariable logistic regression analysis for all participants.
*: statistically significant ($P < .05$).

protective factor that is critical to consider, given the burden of employment loss due to SCD. It is critical to note that while healthy relationships may benefit productivity and provide support, toxic or abusive relationships have negative effects on health and well-being, including economic well-being.³⁴

Our data are strengthened by its multi-institutional reach, encompassing findings from 5 SCD centers throughout the United States. Given the regional differences in employment availability and disability patterns, this allows us to make further generalizations about US families impacted by SCD. Although the 2 survey periods that were pooled together were separated by a couple of years, we found no significant difference in the proportion of adults with SCD reporting employment loss or missed days between the 2 survey periods (supplemental Table 1), suggesting that transient economic changes or the COVID-19 pandemic did not change the proportion of employment loss.

There are limitations in our study. First, given the self-reported nature of this survey, recall bias could impact participants' reports of health care use or employment loss. Second, the survey was also administered in outpatient clinics; therefore, it was limited to individuals who attended outpatient visits and may not represent people who are high inpatient utilizers and are rarely seen in the outpatient setting. Third, many participants reported "not applicable" to job loss (39.5%) or missed work days (53.9%). Based on participants' employment status from 2 institutions, we expect around one-third of participants to report not applicable (out of work, homemaker, retired, or unable to work); however, even for this data, one-quarter of participants reported "prefer not to say." Due to our small sample size, we were limited to performing bivariate regression analysis. Although there may be confounding in the bivariate analyses, the associations observed in these analyses could be further evaluated in multivariate analyses of larger populations. Finally, caregiver data

Table 4. Logistic regression models for absenteeism

		Missed days			
Combined (Adults & Caregivers)		Odds Ratio	95% CI	Unadjusted P-value	Adjusted P-value
Participant type	Adult (> 18 y/o)	0.24	(0.11, 0.51)	< .005*	.005*
Education	Some college or more	0.51	(0.22, 1.17)	.110	.089
Household size		1.26	(1.00, 1.58)	.049*	.986
Children in home		1.19	(0.93, 1.53)	.176	
Relationship status	Not living with a partner	1.88	(0.71, 4.96)	.202	.115
Bills	Somewhat or very difficult	1.15	(0.55, 2.39)	.717	.290
Adults					
Age		1.01	(0.97, 1.06)	.68	
Sex	Male	0.62	(0.21, 1.79)	.375	
Education	Some college or more	0.65	(0.23, 1.88)	.428	
Household size		0.92	(0.64, 1.31)	.626	
Children in home		0.93	(0.61, 1.40)	.714	
Relationship status	Not living with a partner	1.53	(0.37, 6.38)	.556	
Bills	Somewhat or very difficult	1.80	(0.60, 5.38)	.294	
Caregivers					
Age		1.03	(0.91, 1.17)	.628	
Education	Some college or more	2.56	(0.54, 12.10)	.237	
Household size/		1.22	(0.83, 1.80)	.306	
Children in home		0.94	(0.63, 1.38)	.739	
Relationship status	Not living with a partner	4.58	(1.04, 20.15)	.044*	
Bills	Somewhat or very difficult	0.88	(0.29, 2.63)	.817	

Unadjusted P-value with univariate logistic regression analysis for all participants and adult participants. Adjusted multivariable logistic regression analysis for all participants. *: statistically significant (P < .05).

were obtained from one institution based on the demographics of patients served at the included SCD centers; this may make the data obtained regarding caregivers more difficult to generalize to other centers. Caregivers only reported their child's sex, not their own, so we were unable to analyze differences in productivity by caregiver sex, given that known sex disparities in caregiving,³⁵ employment loss, and missed days due to caregiving are likely to affect women disproportionately.

Future research is needed to further characterize the productivity loss in paid employment for people with SCD and the impact of SCD on unpaid work (eg, caregiving and household productivity) to characterize the economic impact of SCD on families comprehensively. As women complete three-quarters of unpaid work,³⁶ this would also help characterize sex inequities in the disease burden. The effect of SCD vs non-SCD populations on productivity requires further evaluation to determine the incremental costs attributable to SCD. Although the surveys in this study focused on attitudes regarding outpatient care and barriers to access, not on disease complications and who are actively on treatment and adherent to their regimen, future studies will be critical to further delineate the impact of therapies and complications on the indirect economic burden. Ultimately, the economic burden of SCD is an important consideration in cost analyses for future therapies to determine treatments that would have a positive economic impact on direct health care costs and indirect costs for patients.

Conclusions

Employment loss and absenteeism due to hospitalization represent prevalent issues in families affected by SCD, which lead to significant financial losses of >\$2 million annually. The impact of medical care costs compounded by losses of income opportunities perpetuate a vicious cycle of poverty among many patients with SCD and their families. Adults with SCD are more likely to report employment loss, and caregivers of children with SCD report more missed days of work, suggesting different and significant employment challenges for both populations. Not living with a partner was associated with both employment loss and missed days of work, suggesting that this is an important risk factor for health care providers to consider for those who need more financial resources. A larger household size was associated with lower rates of employment loss but more missed workdays, suggesting that social support systems play an important but conflicting role in employment maintenance for families impacted by SCD. Further work is warranted to understand how social support protects against employment loss and potential interventions for those with limited social support.

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Authorship

Contribution: R.D.G., C.J.L., and R.M.C. designed the study; N.Q., J.S.H., A.A.K., A.A.T., M.T., and R.M.S. collected the data; R.D.G.

and R.L.W. performed the analysis; R.D.G., C.J.L., and R.M.S. interpreted the results; and all authors reviewed and provided critical input into the final manuscript.

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ORCID profiles: R.D.G., [0000-0002-6684-4498](#); R.L.W., [0000-0001-9477-4581](#); J.S.H., [0000-0003-4439-7321](#); A.A.T., [0000-0003-4961-8103](#); M.T., [0000-0003-0521-1846](#); C.J.L., [0000-0001-9441-3508](#); R.M.C., [0000-0003-1916-6521](#).

Correspondence: Rachel D'Amico Gordon, The Ohio State University, 895 Yard St, Columbus, Ohio, 43212.

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