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An Intervention to Decrease Stigma in Young Adults With Sickle Cell Disease

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Abstract

Young adults with sickle cell disease (SCD) are often stigmatized when they seek care for pain. The purpose of this pilot study was to test an intervention to decrease health-related stigma during care-seeking. Young adults with SCD ages 18 to 35 years (n = 90) were randomized to either the care-seeking intervention (CSI) or an attention control group that participated in life review interviews. The two groups were compared by t tests and longitudinal data analyses on the change from baseline to the last time point in total health-related stigma and health-related stigma by doctors. Findings suggest that the CSI was associated with significant increased awareness of perceived total stigma and stigma by doctors compared with the attention control group. These findings are promising in terms of lessons learned from a pilot intervention that focused on the role communication skills play in decreasing health-related stigma in young adults with SCD.

Keywords

stigma, sickle cell, care-seeking, communication

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Sickle cell disease (SCD), the most common inherited hematologic disorder, is a chronic disease that affects approximately 100,000 individuals in the United States with most of them being African American (Hassell, 2010). The hallmark of the disease is pain caused by vaso-occlusion, reperfusion injury, hypoxemia, and vascular inflammation (Rees, Williams, & Gladwin, 2010; Sparkenbaugh & Pawlinkski, 2013). Although advances in SCD treatment have decreased the pain and prolonged survival (Lanzkron, Carroll, & Haywood, 2013; Smith et al., 2005), adults with SCD often experience unpredictable painful crises or pain episodes requiring health care visits (Waters &Thomas, 1995; Yale, Nagib, & Guthrie, 2000).

Health-Related Stigma in Young Adults With SCD

Currently, adults with SCD who present in emergency departments with complaints of acute pain may wait an average of 90 min for the first analgesic to be given (Tanabe et al., 2007). Although the genetics of SCD are well defined, the delay may be due to the fact that the mechanisms of pain of SCD are poorly understood (Vincent et al., 2013), and it is difficult to objectively assess a pain crisis. Additional barriers to adequate pain management include the fact that most individuals with SCD in the United States are African American and many are of lower socio-economic status (Ely, Dampier, Gilday, O'Neal, & Brodecki, 2002). In a study to determine whether there is differential opioid prescribing in emergency departments by race, it was found that White patients are significantly more likely to receive opioid prescription than Black patients (Pletcher, Kertesz, Kohn, & Gonzales, 2008). Thus, when adults with SCD seek treatment for acute pain in an emergency department, there is great potential for racial stereotyping, mistrust, and problematic physician–patient communication (Todd, Green, Bonham, Haywood, & Ivy, 2006). These factors may result in a negative pain management experience. Young adults with SCD in particular have significant issues related to seeking care for the pain of SCD (Brousseau, Owens, Mosso, Panepinto, & Steiner, 2010; Jenerette, Brewer, & Ataga, 2013).

The timeliness and appropriateness of the treatment of a painful SCD crisis depend to a great extent on the health care provider, who assesses the SCD patient's presentation and ultimately decides whether the individual's report of pain is credible and deserving of treatment (Jacob, 2001; Jenerette & Lauderdale, 2008; Maxwell & Streetly, 1998; Maxwell, Streetly, & Bevan, 1999). In a sample of 69 young adults with SCD, ages 18 to 35 years, Jenerette and colleagues (2013) noted that the main reasons they delayed care-seeking for pain were as follows: trying to treat pain at home, avoiding the emergency department because of past treatment experiences, and the desire to avoid a hospital admission.

The three main reasons for delaying care-seeking may be related to stigmatization. Young adults with SCD try to manage pain at home to avoid the emergency department and a hospital admission because perceived stigma during care-seeking for acute pain (Jenerette et al., 2013). Stereotypical labels for adults with SCD include drug-seekers, substance abusers, "clock watchers," and "frequent flyers" (Jacob, 2001; Maxwell & Streetly, 1998; McCaffery & Pasero, 2001). Because the disease is poorly understood and adults with SCD often present at a time when pain is so acute that they cannot clearly articulate their evidence of pain, health care providers stereotype and stigmatize adult patients with SCD as addicts. Labels such as "clock watchers" are given to individuals who ask for pain medication prior to the time that it should be administered or as soon as it can be given (Jacob, 2001; McCaffery & Pasero, 2001). "Frequent flyers" are individuals who either frequent the emergency department, visit several emergency departments to obtain pain medication, or are thought to frequent the emergency department for non-urgent health concerns (McCaffery & Pasero, 2001; Michelen, Martinez, Lee, & Wheeler, 2006). These stereotypes are perpetuated by the fact that African Americans are at risk of poor pain management across many conditions (Green et al., 2003; Ng, Dimsdale, Rollnik, & Shapiro, 1996; Todd et al., 2006). Young adults with SCD, those aged 18 to 30 years, have the highest rates of acute care encounters and frequent hospitalizations (Brousseau et al., 2010); thus they are at a greatest risk of stigmatization by health care providers.

The aim of this pilot study was to obtain preliminary estimates of the efficacy of a self-care management intervention to lower self-perceived health-related stigma for young adults with SCD. We hypothesized that young adults with SCD who receive the self-care management intervention would have lower perceived health-related stigma compared with those in the attention control group. This study was guided by theoretical relationships proposed in the Theory of Self-Care Management for Sickle Cell Disease (Figure 1).

Theoretical Framework

The Theory of Self-Care Management for Sickle Cell Disease focuses on vulnerability factors and self-care management resources that influence health outcomes. The Theory of Self-Care Management for Sickle Cell Disease evolved from the first author's doctoral dissertation, follow-up studies, and review of the literature (Jenerette & Lauderdale, 2008; Jenerette & Murdaugh, 2008). In the model, vulnerability factors (lack of sickle cell crisis cue recognition/response, number of complications, number of acute pain episodes per year, and overprotection) negatively influence self-care management resources

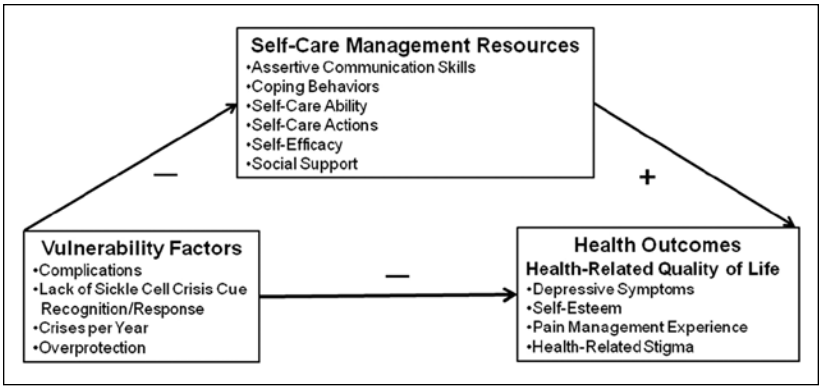

Figure 1. Revised Theory of Self-Care Management for Sickle Cell Disease.

(self-efficacy, coping behaviors, social support, self-care ability, self-care actions, and assertive communications skills) and health outcomes (pain management experience, depressive symptoms, self-esteem, and perceived health-related stigma). Self-care management resources positively mediate the relationship between vulnerability factors and health outcomes. As conceptualized in Figure 1, interventions can focus on decreasing vulnerability factors or increasing self-care management resources to improve health outcomes. Specifically, this research explores the influence of sickle cell crisis cue recognition/response (a vulnerability factor) and communication skills (a self-care management resource) to improve a health outcome—health-related stigma.

Method

Design

University institutional review board approval was obtained from The University of North Carolina at Chapel Hill prior to the start of the study. The pilot study used a prospective, longitudinal design where participants were randomized to either the Care-Seeking Intervention (CSI) group or an attention control group. Data were collected from August 2010 to September 2012.

Sample

A convenience sample of 90 young adults with SCD was recruited from the adult outpatient sickle cell clinic affiliated with the Comprehensive Sickle

Cell Program at the University of North Carolina at Chapel Hill. Inclusion criteria were a diagnosis of SCD; the ability to read, write, and understand English; and age 18 to 35 years. Individuals were screened by clinic staff and were excluded if they had any known cognitive impairment that would limit their ability to participate in study activities.

Rationale for Intervention and Intervention Description

Increased cue recognition and seeking care early, during the presentation during the prodromal or pre-crisis phase, would be more effective for young adults with SCD in preventing the evolution of an acute pain crisis or the necessity of an inpatient admission via the emergency department. Murray and May (1988) reported that 58% of 102 patients experienced a prodromal phase of an impending painful crisis up to 24 hr before developing features typical of their usual crisis. Akinola, Stevens, Franklin, Nash, and Stuart (1992) reported that 12 of 14 patients' feelings of impending crisis were followed by a typical painful crisis that required either home treatment ($n = 4$) or hospitalization ($n = 8$). It has been established that therapeutic intervention are more likely to be effective during the earlier phases of a crisis (Stuart, Stone, Akinola, Gallimore, & Pepys, 1994). Ballas (1998) described the SCD prodrome as "an intriguing entity that needs further study" (p. 85). Moreover, it may provide an avenue for the initiation of proactive or anticipatory analgesia to abort an evolving crisis (Katz et al., 1992; McQuay, 1992) and to prevent the evolution of painful crises. The earlier therapy is introduced, the better the clinical outcome for adults with SCD.

To increase the potential for appropriate and timely treatment, adults with SCD must be able to give the health care provider substantive information that leads the provider to arrive at the conclusion that this is a well-informed, credible individual, presenting early in the evolution of a sickle cell crisis. The adult with SCD needs to know what information the health care provider needs and how to communicate that information. Improved positive self-presentation is important for African Americans to get the best medical care (Malat, van Ryn, & Purcell, 2006). The more information the adult with SCD gives the provider, the greater the likelihood that the provider can devise an individualized treatment plan, in collaboration with the patient, while avoiding stereotypes of adults with SCD and their consumption of opioid analgesics (Ballas, 2005). Positive self-presentation can be improved through communications skills and therefore, adults with SCD may be more likely to get individualized, proactive pain strategies to improve the quality of their pain management experience (Malat et al., 2006).

Description of CSI

CSI consisted of two sessions delivered individually to participants by two doctorally prepared nurses. CSI sessions were delivered in a private area of the sickle cell clinic either before or after a scheduled appointment. CSI was introduced by showing participants an 8-min video that featured an adult hematologist and three adults living with SCD sharing the challenges that they often face during care-seeking for sickle cell pain (Haywood et al., 2011). Then using educational videos scripted and produced by the first author, Session 1 focused on describing the phases of a sickle cell crisis and the possible advantages of seeking care for a pain crisis earlier before the crisis fully evolves. In adults with SCD, a severe SCD crisis that results in the need for hospitalization usually evolves through four distinct physiologic phases: (a) prodromal, (b) the initial evolving, infarctive phase, (c) the established phase, and (d) the resolving, healing, recovery, post-crisis phase (Ballas, 1998). During prodromal phase (pre-crisis) patients develop symptoms of numbness, aches, and paresthesia in the sites that are subsequently affected by pain in advanced phases. Other reported signs and symptoms of the prodromal phase include fatigue, jaundice of sclera, nausea and vomiting, change of appetite, and stiffness in joints (Jacob et al., 2005). This phase can last up to 2 days, and individuals describe the pain as a low intensity ache. The ability of an adult with SCD to recognize the cues of onset of a SCD crisis as well as the transition through differing phases of the SCD crisis may make a significant difference in patient presentation and treatment. That is, if adults with SCD present for treatment during the 2 to 4 day period of low pain intensity, they may be able to avoid the need for aggressive pain management and inpatient hospitalization and have their prodromal phase pain adequately addressed.

Session 2 of CSI focused on assertive communication skills with videos titled “Speak Up to Get the Care You Need.” Brief videos scripted and produced by the first author depicted possible patient–provider interactions featured the most positive outcomes were the result of the patient seeking care early and using Situation, Background, Assessment, Recommendation (SBAR) communication technique. Woodhall, Vertacnik, and McLaughlin (2008) noted that SBAR has been found to assist with structuring and standardizing communication and is considered an easy-to-remember technique that provides for consistent, structured communication between members of the health care team. Jenerette and Brewer (2011) described SBAR and how it may benefit individuals who frequent emergencies departments such as individuals with SCD. In CSI, individuals with SCD were taught the SBAR communication technique to give structure to their communication with health care providers.

Attention Control Group

To be consistent with the two CSI sessions, individuals randomized to the attention control participated in two life review interviews either in person in a private location in the sickle cell clinic or via telephone. A life review is an interview that allows participants to respond to questions about their childhood, adolescence, family and home, adulthood, and overall life (Haight & Burnside, 1992; Jenerette & Lauderdale, 2008). Session 1 focused on a review of childhood memories to high school and Session 2 focused on the participants' lives as young adult living with SCD. A semi-structured life review interview using a modified Successful Aging with SCD Life Review Interview Guide that the first author previously used in life reviews with middle-aged and older adults with individuals with SCD (Jenerette & Lauderdale, 2008; Jenerette, Leak, & Sandleowski, 2011) was used to collect life review data.

Study Measures

Demographic questionnaire. The demographics questionnaire requested information to describe the sample by age, sex, education, and questions specific to SCD, such as number of crises per year that require hospitalization and the SCD genotype.

Communication skills. Assertiveness communication skills, defined as the ability to share thoughts, feelings, and information was measured with the Patient's Self-Competence Scale (Cegala, Coleman, & Turner, 1998). This 16-item Likert-type scale measures communication skills with four subscales (Information Giving, Information Seeking, Information Verifying, and Socio-Emotional Communication). The total score (range = 16-80) and subscale scores (ranges = 6-30, 3-15, 5-25, and 2-10, respectively) are obtained by summing responses, with higher responses indicating higher levels of communication skills (Cegala et al., 1998). Total scale and internal consistency reliabilities have been reported to range from .76 to .92 (Cegala et al., 1998). Indirect construct validity has been supported by using within- and between-sample comparison to support trends in patient–provider communication found in existing literature (Cegala et al., 1998). In the current study at baseline the Cronbach's alpha reliability coefficients for the total score and subscales were .66, .81, .89, .90, and .07 (2 items), respectively. Communication skills were measured at baseline (T1) and then at two follow-up time points (T2 and T3) post intervention or control sessions.

Health-related stigma. Health-related stigma, defined as the co-occurrence of its components—labeling, stereotyping, separation, status loss, discrimination, and emotional reactions (Link & Phelan, 2001; Link, Yang, Phelan, & Collins, 2004) in health-related situations was measured by modifying the Chronic Pain Stigma Scale (Reed, 2005). The Chronic Pain Stigma Scale was initially developed to measure the salience of perceived stigma for persons with significant pain. The scale consists of 30 items on a 6-point Likert-type scale ranging from *strongly agree* to *strongly disagree*. The scale has three subscales: General Public, Physicians, and Family. The total score of the Chronic Pain Stigma Scale is obtained by summing the mean score of the three subscales after required reverse coding (9 items), with higher scores indicating higher perceived stigma with a range of 30 to 180 for the total scale and subscales scores ranging from 10 to 60. The Chronic Pain Stigma Scale has reported total scale and subscale internal consistency reliabilities to be above .91 (Reed, 2005). The Chronic Pain Stigma Scale construct validity was supported by relationships with depression ($r = .29$, $p < .01$), and quality of life measures ($r = -.20$, $p < .05$; Reed, 2005). The Chronic Pain Stigma Scale was adapted, with permission, by changing the words *chronic pain* to *sickle cell pain* or *sickle cell disease* and is referred to here as the Sickle Cell Disease Health-Related Stigma Scale (SCD-HRSS; Jenerette, Brewer, Crandell, & Ataga, 2012). Higher total SCD-HRSS total scores or subscale scores indicate higher levels of perceived stigma. In the current study at baseline the Cronbach's alpha reliability coefficients for the total score and subscales were .84, .70, .69, and .81. Health-related stigma was measured at baseline (T1) and then at two follow-up time points (T2 and T3) post intervention or control sessions.

Procedures

All eligible participants received a letter and were approached for enrollment by the researcher or research assistant in the clinic. After providing written informed consent, participants completed a baseline questionnaire packet that included a demographic form as well as scales to measure pain, communication, and health-related stigma. After completion of the baseline data forms, participants were randomized into either CSI or the attention control group with both groups completing two post-baseline sessions to participate in either the CSI or the attention control activity. Then, all subjects completed the outcomes measure, health-related stigma, at two follow-up measurement occasions related to a care-seeking experience that varied by subject. Each group completed two group-specific sessions followed by two follow-up sessions for data collection (Time 2 and Time 3). From

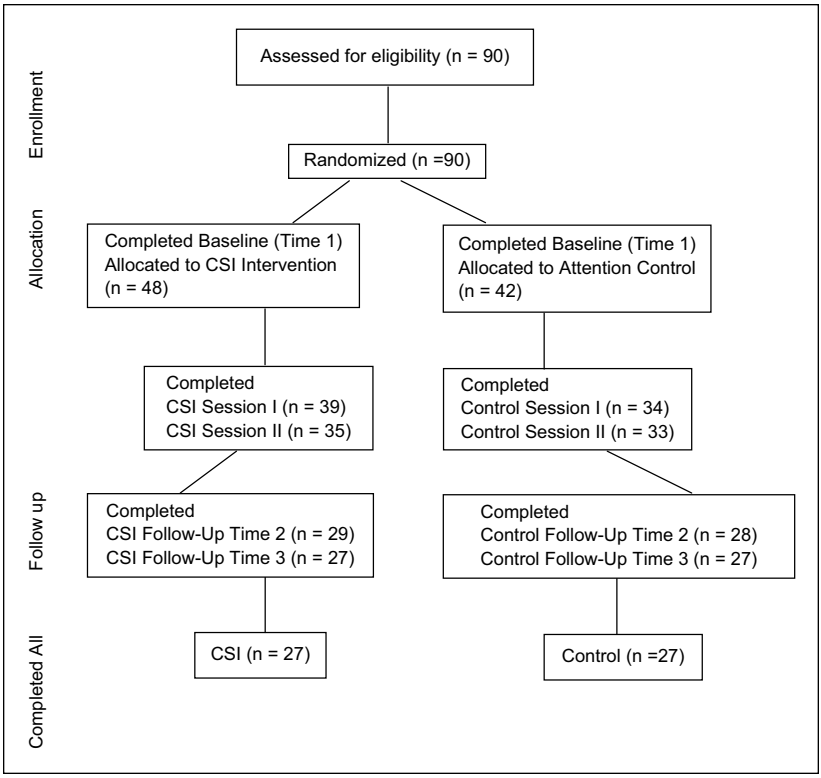

Figure 2. CONSORT diagram.

baseline (Time 1), 57 subjects took an average of 7.5 months (*SD* = 5.2) to complete the first follow-up (Time 2) while it took 54 subjects an average of 9.4 months (*SD* = 5.2) to complete the second follow-up (Time 3)—the end of the study. Figure 2, the CONSORT Diagram, depicts the randomization and study completion.

Analysis

The primary objective of this study was to compare the efficacy of the CSI and attention control interventions in decreasing overall health-related stigma. A second outcome consisting of the subscale measuring doctor's health-related stigma was examined because CSI specifically focused on communication with the health care provider. As this was a pilot study, a

sample size of $n = 38$ per group was calculated to detect a 0.5 standard deviation difference between group means change from baseline, with 80% power. A medium effect size and moderate autocorrelation are appropriate choices for a preliminary study in which little prior data are available as is the case of health-related stigma interventions in SCD.

Primary efficacy measures were the mean change from baseline (Time 1) on the health-related stigma total score and doctor subscale score at the end of Time 3. Repeated measures at Time 2 and Time 3 were evaluated by a generalized linear model (GLM) for longitudinal data analysis using a generalized estimating equation approach (GEE; Diggle, Heagerty, Liang & Zeger, 2002). The GLM is useful for analyzing longitudinal and other correlated outcome data with a continuous or discrete response with a focus on fixed effects, including incomplete longitudinal clinical trial data. It uses all available data from study completers and non-completers and is more robust in controlling the Type I and Type II error rates even under extreme departures from the Missing at Random (MAR) assumption compared with traditional methods using last observation carried forward (LOCF).

The full model GLM for each outcome included as fixed effects factors for demographics (age, sex, education, number of sickle cell crises per year, type of SCD, relationship status), care-seeking pain level, communication skills, time of measurement occasion since baseline (in months), the continuous effect of baseline, and intervention group (main effect of CSI group compared with attention control group). The correlation structure for the model was assumed to be exchangeable correlation. The models of primary interest were main effects models only. The test statistic from the intervention group parameter in the GLM model was used to assess comparisons between CSI and attention control groups. All tests of hypothesis were two-tailed with the Type I error rate fixed at 5%.

Sensitivity analyses were conducted for score change from baseline in the two measures (health-related stigma, and doctor subscale) using an ANCOVA model with last post-baseline measurement carried forward as a means of imputation for missing data. All analyses were performed using SAS software (version 9.3, SAS Institute, Cary, North Carolina).

Results

The sample of 90 young adults with SCD consisted of 61.1% females, 35.6% were in employed, 16.3% were a relationship, and 63.4% reported a sickle cell type of sickle cell anemia. Based on these demographic and disease characteristics, there were no significant differences between CSI and

control except with employment where 47.6% of CSI compared with 25.0% of the control group reported employment ($p = .0253$). Other basic demographic and other disease characteristics for study subjects are summarized in Table 1. A total of 90 young adults with SCD were randomized to either CSI or attention control. The mean age of subjects enrolled in the study was 25.8 years ($SD = 4.8$). The majority of subjects were female (61.1%), 35.6% were employed, and 16.3% reported being in a relationship. In addition, the majority of the respondents (63.4%) self-reported having homozygous SCD with a mean age of first sickle cell crisis at 4.1 years. Overall, the demographic and disease characteristics of study subjects were comparable between the CSI and attention control groups. The observed balance between the two groups in terms of baseline prognostic factors is an indication of the effectiveness of the randomization implemented in this study. A slightly higher proportion of SCD subjects completed the intervention in the attention control group (64.3%; 27/42) compared with the CSI group (56.3%; 27/48; Figure 2).

Efficacy Results

Based on the full model GLM analysis, baseline health-related stigma total score ($p < .0001$) and intervention group ($p = .0023$) were significant predictors. Statistically significant predictors of health-related stigma by doctors subscale in the GLM were baseline ($p < .0001$), intervention group ($p = .0020$), and care-seeking pain level ($p = .0353$). Separation from attention control on health-related stigma total score was observed from baseline but became more evident over time as the difference in means between the two groups widened (Tables 2 and 3). A similar pattern in mean change over time was detected for health-related stigma by doctors subscale. The patterns were assessed as interaction terms between intervention group and time in the GLM model. For health-related stigma total score, the interaction was not statistically significant ($p = .2297$) but was significant for health-related stigma by doctors subscale ($p = .0002$).

In a sensitivity analysis, an ANCOVA was performed on change from baseline to the end of Session 2 for the two outcome measures. In this analysis, CSI intervention was associated with statistically significant greater changes in the least square means for health-related stigma total score ($p = .0250$) and the health-related stigma by doctors subscale score ($p = .0330$). Overall, the results of the sensitivity analysis for both outcomes were similar to, and support the results of the primary analysis conducted through the use of the GLM analysis.

Table 1. Baseline Characteristics by Group and Overall of Young Adults With SCD in a Randomized Study of CSI Versus Attention Control, All Subjects Randomized.

	CSI Group $n = 48$		Attention Control Group $n = 42$		Total $n = 90$		
Continuous Baseline Variables	*M*	*SD*	*M*	*SD*	*M*	*SD*	*p* Value
Age in years	25.8	5.0	25.8	4.7	25.8	4.8	.9954
Education in years	13.0	2.4	13.4	2.2	13.2	2.3	.4344
Age in years at first sickle cell crisis	3.9	4.3	4.4	6.4	4.1	5.3	.7000
Typical care-seeking pain level	8.6	1.3	8.8	1.1	8.7	1.2	.5277
Information giving: Communication subscale	26.5	3.5	27.4	3.4	26.9	3.4	.0608
Information verification: Communication subscale	22.7	3.0	22.9	2.8	22.7	2.9	.2531
Information seeking: Communication subscale	13.4	2.2	13.6	1.9	13.5	2.1	.8158
Socio-Emotional: Communication subscale	10.1	7.4	9.3	1.4	9.7	5.5	.6833
Communication skills total scale	72.8	11.7	73.0	8.1	72.9	10.1	.4504
Health-related stigma: Public subscale	35.8	7.7	33.0	9.2	34.5	8.5	.9833
Health-Related stigma: Doctors subscale	30.7	8.0	29.0	7.3	29.9	7.7	.1263
Health-Related stigma: Family subscale	21.4	9.0	18.8	6.3	20.2	7.9	.3093
Health-Related stigma total scale	87.4	18.4	80.9	17.5	84.4	18.2	.1308

Note. *p* value from *t* test for continuous variables. SCD = sickle cell disease; CSI = communication skills

Table 2. Study Outcomes Means by Time.

					Times for Paired Comparisons								
		Baseline (T1)			T1[a]			T2			T3		
Group	Outcome	*n*	*M*	*SD*	*n*	*M*	*SD*	*n*	*M*	*SD*	*n*	*M*	*SD*
Control	Health-related stigma total scale	42	80.9	17.5	28/27	81.8/82.4	17.1/17.2	28	76.3	14.6	27	75.6	15.3
	Health-related stigma: Doctors subscale	42	29.0	7.3	28/27	28.8/29.1	6.2/6.2	28	26.8	5.9	27	27.1	7.1
CSI	Health-related stigma total scale	48	87.4	18.4	29/27	90.5/88.9	19.3/20.5	29	93.9	21.8	27	92.6	20.4
	Health-related stigma: Doctors subscale	48	30.7	8.0	29/27	31.4/31.1	8.5/9.1	29	33.1	8.1	27	32.7	10.0

Note. CSI = communication skills.

[a]#T2/#T3: Baseline values different for T2 and T3 for comparisons due to sample size.

Table 3. Change in Study Outcome Mean Comparison by Time.

			Times for Paired Comparisons							
			T2				T3			
Comparison	Group	Outcome	*n*	*M*	*SD*	*p* Value	*n*	*M*	*SD*	*p* Value
Change from baseline	Control	Health-related stigma total scale	28	−5.5	11.0	.0133	27	−6.8	14.7	.0232
		Health-related stigma: Doctors subscale	28	−2.0	6.3	.1040	27	−2.0	6.6	.1280
	CSI	Health-related stigma total scale	29	3.4	20.7	.3872	27	3.8	13.8	.1663
		Health-related stigma: Doctors subscale	29	1.7	7.2	.2274	27	1.7	5.1	.0876
Mean difference	Control vs. CSI	Health-related stigma total scale	57	−8.9	16.7	.0493	54	−10.6	14.2	.0085
		Health-related stigma: Doctors subscale	57	−3.7	6.8	.0467	54	−3.7	5.9	.0238

Table 4. Baseline Characteristics by Completion Status of Young Adults with SCD in a Randomized Study of CSI Versus Attention Control, All Subjects Randomized.

Continuous Baseline Predictor Variables	Completers $n = 54$		Non-completers $n = 36$		
	M	*SD*	*M*	*SD*	*p* Value
Age in years	26.4	4.8	24.8	4.8	.1267
Education in years	13.3	2.3	13.0	2.3	.5508
Age in years at 1st sickle cell crisis	4.9	3.2	2.8	1.5	.0590
Typical care-seeking pain level	8.6	1.3	8.78	1.0	.6972
Information giving: Communication subscale	26.9	3.4	26.9	3.5	.9605
Information verification: Communication subscale	22.6	3.0	22.8	2.7	.7331
Information seeking: Communication subscale	13.6	2.2	13.4	1.9	.7749
Socio-emotional: Communication subscale	9.1	1.3	10.6	8.6	.2188
Communication skills total scale	72.2	8.5	73.8	12.2	.4907
Health-related stigma: Public subscale	35.0	9.3	33.9	7.3	.5519
Health-related stigma: Doctors subscale	29.8	7.5	30.2	8.1	.7942
Health-related stigma: Family subscale	21.0	8.0	18.9	7.6	.2283
Health-related stigma total scale	85.3	18.7	83.0	17.5	.5538

Note. *p* value from *t* test for continuous variables. SCD = sickle cell disease; CSI = communication skills; SD = standard deviation.

Attrition

Attrition is always a concern in longitudinal study. Attempts to minimize attrition in the current study included monetary incentives, parking passes, and participant reminders through telephone calls and text messaging. Despite these attempts, 40% (36/90) of the sample did not complete the study. Completers differed from early withdrawers with regard to gender (females completed the study in far greater proportion, 75.9%, $p = .0004$), and type of SCD (66.7% of completers reported suffering from sickle cell anemia, $p = .0370$). Table 4 displays additional demographic and other baseline characteristics as well as *p* values about completers and non-completers.

Discussion

This is the first known published study to test an intervention aimed at decreasing health-related stigma in young adults with SCD. The study was based on relationships posited in the Theory of Self-Care Management for Sickle Cell Disease. As posited by the Theory of Self-Care Management for Sickle Cell Disease, individuals in CSI received training in the benefits of recognition/response to a sickle cell crisis and communication skills.

Although overall health-related stigma was major outcome, CSI targeted communication between the young adult with SCD (patient) and the provider—most often a doctor. Therefore, we examined the effect of the intervention on perceived health-related stigma from doctors as an outcome. The pattern of findings for these two outcomes was the same in a GLM analysis and an analysis of covariance: subjects in the CSI group reported higher levels of health-related stigma than the attention control group. This was contrary to the proposed purpose of the study- to decrease health-related stigma with the CSI. However, increasing awareness of health-related stigma may be a first important step in decreasing health-related stigma for individuals with SCD.

Individuals in the attention control group benefited from the therapeutic effect of the life review interviews as they reported significantly less stigma over time compared with CSI. As proposed in the Theory of Self-Care Management for Sickle Cell Disease, perhaps the life reviews interviews provided self-care management resources in the form of either a means to cope or a form of social support for individuals with SCD as the reminisced about the ups and downs of living with SCD and their current life as a young adult.

The vulnerability factor of "lack of cue recognition/response" as measured by pain score was not supported as easy to modify with intervention as there was no significant difference in pain scores at the time of care-seeking between the CSI group and the attention control group. This lack of support may be due to the fact that many young adults have already had negative encounters with health care providers when seeking care for SCD pain. This would make it more difficult for them to change behaviors. This would make it more important to begin interventions such as CSI early—perhaps before the transition from pediatric to adult care. The Theory of Self-Care Management for Sickle Cell Disease could be used to guide a similar study in an adolescent population of individuals with SCD as CSI skills may be useful as they begin to interact with health care providers in the adult health care system.

Health care providers should find the results of the pilot study useful as a first step increasing awareness about health-related stigma. The next step

may be actually recording interactions between patients with SCD and health care providers to see if they are actually using the CSI. If SBAR technique, frequently used between health care providers, were to be used one would expect less health-related stigma as it would improve communication between patients with SCD and health care providers. Patient use of SBAR, as proposed in CSI, would provide the patient and health care provider, doctor or nurse, with common language and may lead to more positive providers' perceptions of the credibility of individuals with SCD and thus more timely and appropriate treatment. This may then lead to less health-related stigmatization and better care-seeking experiences for individuals with SCD.

A potential limitation in this study could be the relatively high proportion of subjects who withdrew early. One of the key assumptions commonly used in the analysis of longitudinal repeated-measures data is that study outcomes would be similar for completers and non-completers, had non-completers stayed in the study. If this assumption is false, then the conclusions of the study could be biased. There is no direct way to verify whether this assumption has been met. It is worth pointing out, however, that no statistically significant associations were found between these two baseline factors and any of the two study outcomes. As this is a pilot study, the lessons learned should guide the design and conduct of future studies to prevent this problem. It will be imperative to put in place additional strategies aimed at improving retention and protocol compliance from study participants especially focusing on subjects who may have some degree of pain at the point when data needs to be collected. In a study of minority subjects with asthma, Zook et al. (2010) noted that retention can be challenging due to high mobility, inconsistent telephone service, and stressful life situations. In addition, they suggest identifying factors related to missed study activities is an important step in designing tailored studies that include strategies to retain subjects.

In this study, repeated-measures analysis of longitudinal data was conducted to capture the effect of CSI intervention on two outcome measures (health-related stigma, and doctor subscale), after adjusting for known covariates. Intervention effect was based on mean change from baseline at the end of Time 3 for each measure. The findings of this research indicate a promising first step toward decreasing health-related stigma in young adults with SCD. Future studies need to consider the dose of the intervention as it may have led to more awareness instead of decreased health-related stigma. Larger confirmatory investigations that take into account the lessons learned from this pilot study should be considered.

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