Journal of Pain Research Dovepress

Open Access Full Text Article ORIGINAL RESEARCH

Attitudes and Beliefs Regarding Pain and Discrimination Among Black Adults with Sickle Cell Disease: A Mixed Methods Evaluation of an Adapted Chronic Pain Intervention

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Purpose: This study sought to adapt a chronic pain group curriculum for adults with sickle cell disease (SCD). Given the association of experiences of racism and discrimination with health outcomes, this study also explored how such experiences for patients with SCD impact their interactions with clinicians and health-care systems.

Patients and Methods: This mixed methods study recruited patients (aged ≥18 years) in a sickle cell treatment program; all self-identified as Black or African American. Key informant interviews evaluated a chronic pain program curriculum and rated the curriculum sessions' importance for SCD pain management. A survey containing six validated instruments then measured experiences of pain and discrimination, level of emotional expressivity, and trust in physicians and health insurers.

Results: Of the 19 adult patients approached, 12 (63%) completed the interview and survey. Interview data analysis resulted in five themes comprising pain, treatment, mental/emotional, health-care clinicians, and differences in SCD versus other chronic conditions. Each curriculum session was rated as "very important" or "important" by participants. Most described at least one experience of racism or discrimination in the medical setting and identified clinicians' lack of knowledge as a barrier to appropriate SCD care. Participants identified the importance of non-pharmacological treatments for SCD pain and appropriately managing pain caused by comorbid chronic conditions. Conversely, survey responses reported moderate levels of pain, infrequent discriminatory experiences in the medical setting, and moderate trust in medical professionals, whereas most reported experiences of discrimination in general settings. Many participants reported they either accepted it or did nothing in response to discriminatory experiences.

Conclusion: Study findings emphasize the need to address experiences of racism and stigma in addition to experiences of emotional and physical pain among patients with SCD in the context of pain management. Findings from this study will inform the development of a SCD pain group curriculum for adult patients.

Keywords: hemoglobinopathy, chronic pain, acute pain, quality of life, racism

Introduction

According to the National Heart, Lung, and Blood Institute, sickle cell disease (SCD) affects over 100,000 people in the United States and about 20 million people worldwide.[1] It is the most common inherited hemoglobinopathy and is often characterized by a spectrum of micro and macrovascular complications, most commonly presenting as chronic pain or acute episodes of vaso-occlusive crises.[2] In both pediatric and adult patients with SCD, those with more frequent chronic pain report more frequent depressive symptoms, more frequent hospital admissions, and more pronounced functional

Received: 6 June 2024
Accepted: 18 September 2024
Published: 6 November 2024

disability.[3,4] Owing to inpatient hospitalizations and emergency department visits, average annual costs are approximately $10,000 per child and $34,000 per adult, not accounting for lost work due to absenteeism or lost productivity.[5]

The Pain in Sickle Cell Epidemiological Study (PiSCES) followed 232 patients with SCD and demonstrated chronic pain was more common than acute pain.[6] They found that approximately 50% of patients reported experiencing chronic pain, which led to poorer quality of life and higher health-care utilization.[6] Both acute and chronic pain are typically treated with opioid therapy due to a lack of alternative effective treatment options. Chronic opioid therapy is associated with significant risks including hyperalgesia and overdose, which can lead to emergency department visits and subsequent hospitalizations.[7] Managing acute and chronic SCD-related pain requires multimodal strategies utilizing both pharmacologic and non-pharmacologic interventions. However, there is limited evidence demonstrating the effectiveness of non-opioid pain management modalities.

A 2016 review identified 28 studies that addressed non-pharmacologic pain interventions for people with SCD.[8] Skills-based therapy interventions discussed included cognitive behavioral therapy, massage, mindfulness, and other skills-training resources facilitated by experts.[8] Practitioner utilization of non-pharmacologic strategies has been limited in part because of questions about effectiveness and generalizability. In addition to skills-based therapies, five studies in the 2016 review addressed interventions incorporating peer-support groups with three studies showing significant improvements in pain as measured by participant self-reported surveys.[8] The quality of the non-pharmacologic studies identified were noted as moderate by the authors. In addition, a majority of the studies focused on children as opposed to adults and none of the studies evaluated whether their interventions decreased acute care utilization. Given this, our study addresses gaps in the evidence regarding non-pharmacological treatments involving peer-support groups and associated outcomes for adults with SCD. Given concerns over the use of chronic opioid therapy and its lack of utility for many patients, it is timely to explore the use of peer-mentored groups to improve chronic pain outcomes.

Since there is currently no sickle cell-specific peer-led curriculum reported in the literature, the goal of this study was to adapt a general chronic pain group curriculum for individuals with SCD by engaging the potential end-users in the adaptation process. An important component of any pain support group is the provision of cultural and disease-sensitive information, especially acquired from the perspective of patients themselves.[9] Furthermore, some research suggests that patients' trust in the health-care system and experiences of racism can impact acute care utilization, but there is little understanding of how these experiences influence chronic pain in particular or how these issues should be addressed.[10–12] Given this, our study also explored how experiences of racism and discrimination play a role in working with health-care clinicians and systems to manage physical pain associated with SCD.

Study Aims

Our first aim was to assess study participant reactions to a chronic pain peer-led program for use with adult patients with SCD. This was achieved by using key informant interviews of adult patients with SCD to evaluate the chronic pain program curriculum as it pertains to patients' needs, self-management goals, learning styles, cultural adaptability/relevance and to identify cultural- and disease-specific elements to be added.

Our second aim was to use the informant interviews and validated survey tools to describe the interactions between experiences of racism and discrimination with SCD manifestations such as physical pain among adult patients with SCD. We hypothesized that responses would demonstrate low health-care clinician trust and high experiences of discrimination among adult patients with SCD.

Material and Methods

Study Design and Setting

This exploratory sequential mixed methods study was conducted in a sickle cell treatment program within a regional health system based in Wilmington, Delaware. Participants were approached during a regularly scheduled sickle cell clinic visit. The PI (SG) and a trained research assistant (AY) conducted the informed consent process to determine if patients were willing to participate in the study. Informed consent including permission to publish anonymized responses and direct quotes. The research assistant (AY) then contacted participants at a mutually agreed upon time via telephone or

video call. We convened a stakeholder advisory board to review the findings from the key informant interviews and the revised SCD-specific curriculum. This stakeholder advisory board consisted of the Executive Board of a local sickle cell community-based organization and was comprised of adult patients with sickle cell disease and caregivers.

Participants

Adult participants aged 18 years and older with SCD were recruited during a routine clinical visit at the comprehensive adult sickle cell program in an urban community hospital. We included patients with any genotype of SCD. We excluded patients who were not able to speak, read, and write in English and those who did not have access to a phone and email or text messaging.

Data Collection

Qualitative Interview Data

Participants were virtually interviewed by the research assistant (AY) by video call or phone and recorded using two audio recorders. After confirming consent, key informant interviews were performed using a semi-structured interview guide (see Supplement 1). Item two of the interview guide included a prompt asking participants to review the six session modules of the general chronic pain group program curriculum (Supplement 2) and rate the importance of each module in terms of managing SCD-related pain on a scale of 1 to 5 with 1 being "very important" and 5 being "not important at all". At the close of the interview, participants were asked to provide some socio-demographic information including age, sex, marital status, number of individuals living in their household, income, and sickle cell genotype (Supplement 1). Patients who completed the key informant interview received a $20 gift card. Audio recordings from the key informant interviews were transcribed using a transcription tool in Microsoft Word and reviewed for accuracy by the research assistant (AY).

Quantitative Survey Data

Following completion of the key informant interview, participants received an email with instructions and a link to complete the survey (Supplement 3). Survey data were collected using Research Electronic Data Capture (REDCap), a HIPAA compliant web-based software designed to support the collection of research data.[13,14] We utilized six validated instruments to measure constructs related to experiences of pain and discrimination, level of emotional expressivity, and trust in physicians and health insurers.

The Pain Catastrophizing Scale (PCS) is a 13-item instrument that examines the relationship between different approaches to catastrophizing – an exaggerated negative orientation to an event or experience – and experiences of painful stimuli. The instrument contains item responses reported on a 5-point Likert scale ranging from "not at all" (0) to "all the time" (4).[15] The original authors who validated this instrument reported a Cronbach's alpha of 0.87 and a test–retest reliability of 0.75.[15]

The Discrimination in Medical Settings (DMS) was adapted for utilization in medical settings from a modified version of the Williams Everyday Discrimination Scale.[16] The DMS is a 7-item instrument with responses reported on a 5-point Likert scale ranging from "never" (1) to "always" (5).[17] This validated instrument reported a Cronbach's alpha of 0.89 and a test–retest reliability of 0.58.[17]

The Berkeley Expressivity Questionnaire (BEQ) assesses the strength of individuals' emotional responses and the degree of these tendencies. It is a validated 16-item instrument with responses reported on a 7-point Likert scale ranging from "strongly disagree" (1) or "strongly agree" (7) with three items reverse coded that were negatively phrased.[18] Testing of this instrument demonstrated a Cronbach's alpha of 0.85 and a test–retest reliability of 0.86.[18]

The Everyday Discrimination Scale (EDS) is an instrument with nine sections.[19] The EDS includes the 10-item Experiences of Discrimination (EOD) measure, which was validated by Krieger et al where they reported a Cronbach's alpha of 0.74 and a test–retest reliability of 0.70.[19] The EOD contains two sub-sections. The *response to unfair treatment* sub-section contains two items assessing how participants usually respond when treated unfairly. The second sub-section, *discrimination*, asks participants to confirm if they have experienced discrimination in nine possible settings with a follow-up question asking participants to note how many times (on a scale of one [1], two or three times [2], or

four or more times [3]) they experienced discrimination in settings where they reported "yes". The remaining sections of the EDS included four "Worry" questions assessing the frequency that respondents worry about experiences of unfair treatment due to racism or discrimination, two "Global" questions where respondents report the frequency that they feel certain groups or they themselves experience racism or discrimination, one question assessing if respondents ever filed a formal complaint due to racial discrimination, and questions created by Williams et al for the Major and Everyday Discrimination measure.[16]

The Wake Forest Physician Trust (WFPT) scale is a validated instrument with 10 items with responses reported on a 5-point Likert scale ranging from "strongly disagree" (1) to "strongly agree" (5).[20] Assessment of this scale reported a Cronbach's alpha of 0.93 for their national sample and 0.92 for their regional sample with a test–retest reliability of 0.75 for their regional sample.[20]

The Interpersonal Trust Scale (ITS) consists of three validated sub-scales assessing trust in: physicians, the medical profession, and health insurers.[21] This study utilized the trust in medical professionals and health insurers sub-scales. Both sub-scales contain 5-items each with responses reported on a 5-point Likert scale ranging from "strongly disagree" (5) to "strongly agree" (1). One item in the trust in medical professionals sub-scale is negatively phrased and thus, reversed coded. Two items in the trust in health insurers section are negatively phrased and reversed coded. The trust in medical professionals sub-scale reported a Cronbach's alpha of 0.87 and test–re-test reliability of 0.71.[21] And the trust in health insurers sub-scale reported a Cronbach's alpha of 0.84 and test–re-test reliability of 0.73.[21]

Data Analysis

Qualitative Data Analysis

Qualitative analysis of the interview data were completed using two approaches. First, we used the rigorous and accelerated data reduction (RADaR) technique.[22] The RADaR technique involves using tables and spreadsheets from general-purpose word-processing software to develop all-inclusive data tables that undergo several revisions. Two members of the research team [OB and CJ] reviewed interview transcriptions with recordings to check for accuracy and authenticity, and then the transcripts were imported into Microsoft Word for data management and analysis by three coders [OB, CJ, and CW]. In terms of choosing the various table themes and then source material that would populate the tables in word processing, the Framework Analysis approach was used and includes five key stages: 1) familiarization, 2) identifying a thematic framework (Table 2 theme),3) indexing, 4) charting of source material, and 5) mapping and interpretation using thematic analysis.[23] We used an iterative team-based approach to work both individually and as a group to develop consensus on focused themes and codes. Ten percent of the interview transcripts were read by a fourth member of the research team [SG] to ensure consistency with the final coding hierarchy. Disagreements were resolved through discussion to achieve consensus. A thematic codebook was developed. Findings were discussed with a stakeholder advisory board and investigators for further interpretation. To ensure trustworthiness of data, we satisfied confirmability through the development and usage of an *audit trail* which highlights every step of data analysis and dependability through usage of an *inquiry audit* which requires an outside person to review and examine the process and data analysis to ensure findings are consistent and could be repeated.

Quantitative Data Analysis

Survey data were analyzed using descriptive statistics to assess the frequency, central tendency (mean, median), and dispersion of the data (standard deviation, interquartile range) and characterize participant responses to the Likert scale and multiple-choice survey items. Quantitative data collected from item 2 of the interview guide and the socio-demographic questions asked at the end of the interview were analyzed using descriptive statistics (frequency, mean, median). Analysis of the survey data were completed using R version 4.3 (R Core Team, 2023) and analysis of the quantitative data collected during the key informant interview were completed using STATA version 17 (StataCorp, 2021).

Ethics Board Approval

The study protocol and associated study materials were reviewed and approved by the ChristianaCare Institutional Review Board (IRB00000480). Additionally, this study complies with the Declaration of Helsinki.

Results

Participants

Of the 19 participants approached about the study, 14 consented to participate. Of the 13 participants who completed a key informant interview, one completed the interview but did not return the survey. Our study sample included 12 participants who completed both the key informant interview and survey, resulting in a 63% response rate. Table 1 reports the sociodemographic characteristics of the patients approached about the study (N = 19) and the final study sample of those who completed both the key informant interview and the survey (N = 12). The mean (standard deviation [SD]) age of the study participants was 28.4 (7.05) years, ranging from 20 to 45 years old. Half of the participants

Table 1 Participant Sociodemographics

	Total Invited to Study N=19	**Completed Interview & Survey N=12**
Age in years, mean (SD, range)	29.0 (6.93, 20–45)	28.42 (7.05, 20–45)
Sex, N (%)		
Female	9 (47.4)	6 (50.0)
Male	10 (52.6)	6 (50.0)
Marital status, N (%)		
Single	12 (63.2)	11 (91.7)
Married	1 (5.3)	1 (8.3)
Missing/not reported	6 (31.9)	0 (0)
Number people in household, mean (SD, range)	2.69 (0.95, 1–4)[a]	2.67 (0.99, 1–4)
Household income, N (%)		
$0-$24,999	4 (21.1)	3 (25.0)
$25,000-$49,999	3 (15.8)	3 (25.0)
$50,000-$74,999	2 (10.5)	2 (16.7)
$75,000-$99,999	1 (5.3)	1 (8.3)
$100,000-$149,000	1 (5.3)	1 (8.3)
Missing/not reported	8 (42.1)	2 (16.7)
Sickle cell genotype, N (%)		
Hb-SS	7 (36.8)	7 (58.4)
Hb-SC	3 (15.8)	2 (16.7)
Beta Thalassemia	3 (15.8)	3 (25.0)
Missing/not reported	6 (31.6)	0 (0)

Note: [a]N=6 missing/not reported.

identified as female (N = 6), and a majority identified their current marital status as single (N = 11). Of the 12 participants included in the final analysis, 91% (N = 11) self-identified their race as Black/African American and 9% (N = 1) self-identified as African/African American. They reported that the median (interquartile range [IQR]) number of people living in their home was 3 (2, 3) people, ranging from a minimum of one to a maximum of four people. Half of the participants identifying their household income between $0 and $49,999 (N = 6). In addition, a majority of participants reported their sickle cell genotype as Hb-SS (N = 7).

Qualitative Data Results

After analysis and thematic saturation was reached for the qualitative key informant interview data, five themes and 30 codes were developed (Table 2). Representative quotes from the key informant interviews were identified for each theme and presented below.

Pain Theme

Interview participants described multiple forms and domains of pain that they experience as result of SCD. They noted feeling physical, throbbing pain that could be either acute or chronic. In either case, participants identified the debilitating nature of experiencing such pain.

> … because whether it's chronic, whether it's acute, whether is what, it's pain. Me, for sickle cell pain is no, not like a joke for me, so it doesn't matter if acute or if … nobody wants pain.

> …'we just gave her so much Morphine, or so much Dilaudid and now she wants some more,' and they're looking at you like you're, you're a drug addict trying to, and you're just in pain, you just want the pain to stop……

> … sometimes when I try to relax, that still don't help. It seems like the pains still there even when I do try to like ___ the breathing….

Participants also identified how disruptive severe pain can be on their lives. This participant demonstrated how extreme and debilitating the pain can be when it reaches a certain point.

> I know like once I really start to feel in my legs, I have to I have to get to like the nearest hospital or something immediately because it's gotten to a point where I almost hadn't been able to walk before. Like I'm like, really like struggling to like stand or move like so like even to and I've I've walked through the pain

In addition, another participant referred to how some psychological techniques can help manage the pain in a positive manner.

> …. Um, a lot of times you associate pain with how you think. So sometimes if you know that the pain is coming, automatically you just bump it up that it's going to be the worst pain of your life, but if you're like 'Okay, I know this pain, I've been here before' it's kind of 'Okay, I've done this, it's not as painful as—.' In your mind you can always trick yourself to say, 'It's not as painful as last time, and here's what I did last time.' So, it works some cases, some cases it doesn't….

Table 2 Interview Themes and Codes

Themes	Codes
Pain	Physical pain, mental pain, chronic pain, acute pain, throbbing pain, why pain
Treatment	Non-pharmacological, pharmacological, health care clinicians, hospital, emergency room care, home care
Mental/emotional	Goals, coping, worry/stress, depression, relaxation, relaxation
Health care clinicians	Knowledge and care of sickle cell disease, medication bias, discrimination, stigma, racial bias
Differences	Triggers, management, color/race, caring, transition of care, knowledge, management

Treatment Theme

One participant noted utilizing both pharmacological and non-pharmacological treatments for SCD.

> Developing, um, individual and group goals, I have developed individual goals…. And I did, uh, at CHOP I learned deep breathing, um, practices. And, uh, healthy eating is important, and nutrition and sleep comes ….

Another participant commented on their reliance on non-pharmacological treatment resources in terms of its accessibility and being able to utilize such resources regardless of their location.

> …. Uh, you know, the pain can happen, um, any time in the world, so in case you're not in the house or you're not, uh, close to your medication, um, that would be something, uh, so normally I don't, normally depend on the medication for pain unless I'm having pain. Then, uh, I'll drink water, I'll get myself warm, or I'll take hot shower for the same, like, thirty minutes to an hour, before, uh, I'll go onto medication

Mental/Emotional Theme

Participants described the emotional impact of experiencing chronic pain and how stress sometimes plays a reciprocal role in the experience of pain. They also described their conception of the mind-body connection, if applicable.

> … I got used to the pain and I'm always in pain. It's normal. The more I get emo—like, as I'm saying, the more I listen to, like, those words and I get emotion. And when I start getting emotion, then I get stressed out, then I start feeling some … pains and, like, um, … I don't know, cause, like, I'm going so… just not anymore, 'cause I used to do it. For now, it doesn't affect my pain when I'm stressed.

One participant noted the psychological impact of pain and the mental toll it took on them.

> Yeah, the pain definitely is, uh. I don't wanna say like 50% mental 50% physical like there's many like ratios and aspects and and emotionally it can definitely…Uh, it could damage…. it can make you feel like you feel like that's it's a life like that. Yeah, all you have is either you're in pain or you're not and or you're just gonna be anticipating it for whenever

Health Care Clinicians Theme

This theme encapsulated participants' experiences with health-care clinicians and the health system overall as SCD patients.

> Uhm, if anything I have never had [a negative] experience, but I think a lot of Black people are uncomfortable just with the healthcare system in general

Another participant noted their reluctance to go to the hospital since they are only given pain medication and released without further exploring the patient's clinical concerns regarding their SCD symptoms.

> … and maybe they can talk to… they can actually talk about. Maybe or like it such as doctors like letting doctors know that sometimes a sickle cell patients don't really wanna come to the hospital to get narcotics because when we come to the hospital for pain that is our first action is, 'We're gonna give you more morphine [inaudible],' but sometimes you don't want that. Sometimes we actually wanna just get like blood work done to see what's going on. Ibuprofen, Tylenol we don't want to go shoot the pain medicine because pain medicine can put you in a very bad depression

One participant noted the difficulties of transition from a pediatric to adult SCD patient and the lack of structured resources available for adult SCD patients outside of going to the ED for treatment of acute pain. When they do present at the ED for their pain, SCD patients feel like their pain concerns are dismissed and treated as drug-seeking for illicit use.

> … "you're in chronic pain and your course of action, you know, as a child, was they knew, you know, you had this treatment plan and they, they just did it; and now you're an adult, and you come to the ER and it's, 'Oh, you know, she's just got, we just gave her so much Morphine, or so much Dilaudid and now she wants some more,' and they're looking at you like you're, you're a drug addict trying to, and you're just in pain, you just want the pain to stop".

Another patient reflected on the same experience of being treated as drug-seeking for illicit and believing that their race plays a role in the biased and discriminatory treatment they received.

> …. Well, sometimes I feel like at different hospitals they, um, I guess because you black, they try to assume that you coming there to get like pain medicine and stuff, when really you just wanna get treated for the pain

Differences in Sickle Cell versus Other Chronic Diseases Theme

One participant noted the importance of understanding the unique needs of SCD patients and how their treatment and services need to be adjusted to meet these needs.

> …. OK, yeah, I think that's important especially because you have to keep in mind that what's okay or not like patients or people in general, adults, sickle cell adults were not like everyone else and we can't do the same things that everyone else could be able to complete in a day or in a week or an hour, because our bodies can only take so much. Uhm, especially if you're not like 100% healthy, or like you're recovering from a crisis you just had.

This participant commented on how pain experiences can differ depending on the patients age and the extent that they have been living with the disease. Determining and modifying a patient's treatment plan throughout their life course is essential to ensure continuity and patient-centered care is adequately provided.

> …. Cause a lot of people don't know that you can have sickle cell from birth and be in pain from birth. It's not always the adults that have sickle cell or only young children that have sickle cell.

One participant noted the importance of identifying and distinguishing the nuances of different types of pain that SCD patients experience to identify the optimal course of treatment.

> …. some pain is just different. You know it's different for everybody, so it's I feel like it's crucial to learn. How do we experience and how everybody else experience so we can further, you know, learn different ways to treat it.

Interview Likert Scale Question

In addition to the themes identified from the key informant interviews, we calculated participant responses to item two of the interview guide where participants rated the importance of each session module. Table 3 presents the median (IQR) scores of the rated pain group curriculum sessions and a summary of each session topic. Overall, the median score for each session was rated as either "very important" (1) or "important" (2) to participants. Sessions 2 and 4 had the highest median score and thus were rated the most important among participants with a median (IQR) score of 1 (1,2), respectively. Session 5 had the lowest median score of importance but still rated relatively high with a median (IQR) score of 2 (1, 3.25).

Quantitative Data Results

Survey responses were tabulated for each battery instrument that was administered to participants. Table 4 presents a summary of the participant survey responses with a full report of survey responses presented in Supplement 4.

In terms of catastrophizing experiences of pain, patients on average reported experiencing a moderate degree of pain catastrophizing on the overall PCS in addition to the rumination, magnification, and helplessness PCS sub-scores. The overall DMS mean score on the DMS, indicated relatively infrequent self-reported experiences of discrimination in medical settings. BEQ mean scores were also within the mid-range and thus relatively neutral for participants' levels of overall emotional expressivity and for negative expressivity, positive expressivity, and impulse strength sub-scores.

Responses to the EOD sub-section of the EDS demonstrated that when participants feel they have been treated unfairly, half (N = 6) accepted it as a fact of life and half (N = 6) tried to do something about it. When explicitly treated unfairly, a majority of respondents (N = 7) reported talking to other people about it versus keeping the experience to themselves. A majority of participants reported not experiencing any form of discrimination due to their race, ethnicity, or color at school (N = 7), when getting hired or getting a job (N = 7), at work (N = 7), when getting housing (N = 11), or from the police or in the courts (N = 9). Half of participants (N = 6) reported that they

Table 3 Rating of Pain Group Curriculum Sessions (Question 2 of the Key Informant Interview Guide). Participants Were Instructed to: "Please Review the Modules from the Current Pain Group Curriculum. On a Scale of 1 to 5 with 1 Being 'Very Important' and 5 Being 'Not Important at All' Please Rate the Importance of Each to You"

General Chronic Pain Group Curriculum Sessions	N=12 Median (IQR) Score
Session 1: What is chronic pain? Define chronic pain, identify both individual and group goals for the course. Start building our toolbox of skills with guided deep breathing exercises. Will review related relaxation techniques and discuss proper sleep hygiene. Nutrition and rest is how we fuel our bodies and if not eating or sleeping well, it is bound to take a toll on our mood and experience of pain.	1.5 (1, 3)
Session 2: Why do I experience pain? Discussion of pain in relationship to physical disease as well as additional factors that affect pain like emotional responses, behaviors, and social supports. Review the gateway theory. We will practice breathing exercises, guided imagery, and progressive muscle relaxation. Begin to think about how to manage severe pain days because often the anticipation of pain keeps us from living our best selves.	1 (1, 2)
Session 3: Getting a grip on your thoughts. Discuss the relationship between emotions and pain. We will discuss how thinking traps can affect pain and learn about the ABC model (activating event, beliefs, and consequences). We will teach restructuring and de-fusion techniques. Learn to use our wise mind- "I feel this, I know this, and so I will do this…" the middle path create joy.	2 (1, 2.25)
Session 4: How can I relax? I am in pain! Discuss how chronic pain is affected by stress and anger management. We will explore self-care activities like yoga, meditation, massages, and distraction techniques. We will discuss positive self- talk to change negative thinking and practice mindfulness. If we put ourselves back in the driver's seat, the pain will no longer be in control.	1 (1, 2)
Session 5: What's important to me? Discuss the relationship between pain and your personal values. We will help you identify what is important to you and illustrate how that can be used to improve our perception of pain. In doing so it's important to learn good pacing skills and how to get moving on days when we just feel like we cannot.	2 (1, 3.25)
Session 6: That's a wrap! We will review all the skills we put in the "tool kit" over the last 5 weeks. We will discuss what was learned, how to implement it and then reflect back on it to see how we can make each day better than the last. Feedback will, of course, be welcomed as we are a new program and looking to improve it to best meet your needs	1 (1, 2.5)

Note. IQR: interquartile range.

Table 4 Summary Participant Survey Responses

	N=12
Pain Catastrophizing Scale (PCS)	
Rumination, mean (SD) [sub-score range 0–16]	9.1 (3.5)
Magnification, mean (SD) [sub-score range 0–12]	6.2 (2.1)
Helplessness, mean (SD) [sub-score range 0–24]	10.5 (5.1)
Overall PCS score, mean (SD) [sub-score range 0–52]	25.8 (8.5)
Discrimination in Medical Settings (DMS), mean (SD) [score range 0–4]	1.4 (1.2)
Berkeley Expressivity Questionnaire (BEQ)	
Negative expressivity, mean (SD) [sub-score range 1–7]	3.8 (2.2)
Positive expressivity, mean (SD) [sub-score range 1–7]	5.1 (1.9)
Impulse Strength, mean (SD) [sub-score range 1–7]	4.5 (2.2)
Overall BEQ score, mean (SD) [score range 1–7]	4.4 (2.1)

(*Continued*)

Table 4 (Continued).

	N=12
Everyday Discrimination Scale (EDS)	
Experience of discrimination – Response to unfair treatment	
If you feel you have been treated unfairly, do you usually, N *(%)*	
Accept it as a fact of life	6 (50.0)
Try to do something about it	6 (50.0)
If you have been treated unfairly, do you usually, N *(%)*	
Talk to other people about it	7 (58.3)
Keep it to yourself	5 (41.7)
Experience of discrimination – Discrimination *Have you ever experienced discrimination, been prevented from doing something, or been hassled or made to feel inferior in any of the following situations because of your race, ethnicity, or color?*	
At school, N *(%)*	
One or more times	5 (41.7)
Not at all	7 (58.3)
Getting hired or getting a job, N *(%)*	
One or more times	5 (41.7)
Not at all	7 (58.3)
At work, N *(%)*	
One or more times	5 (41.7)
Not at all	7 (58.3)
Getting housing, N *(%)*	
One or more times	5 (41.7)
Not at all	7 (58.3)
Getting medical care, N *(%)*	
One or more times	6 (50.0)
Not at all	6 (50.0)
Getting service in a store or restaurant, N *(%)*	
One or more times	6 (50.0)
Not at all	6 (50.0)
On the street or in a public setting, N *(%)*	
One or more times	7 (58.3)
Not at all	5 (41.7)

(*Continued*)

Table 4 (Continued).

	N=12
From the police or in the courts, N *(%)*	
One or more times	3 (25.0)
Not at all	9 (75.0)
Experience of discrimination – Worry questions	
When you were a child or teenager (up to age 18), how much did you worry about people in your racial/ethnic group experiencing unfair treatment because of their race, ethnicity, or color?, N *(%)*	
Most or some of the time	8 (66.7)
Rarely or never	4 (33.3)
When you were a child or teenager (up to age 18), how much did you worry about yourself experiencing unfair treatment because of your race, ethnicity or color?, N *(%)*	
Most or some of the time	7 (58.3)
Rarely or never	5 (41.7)
In the last year, how much did you worry about people in your racial/ethnic group experiencing unfair treatment because of their race, ethnicity, or color?, N *(%)*	
Most or some of the time	4 (33.3)
Rarely or never	8 (66.7)
In the last year how much did you worry about yourself experiencing unfair treatment because of your race, ethnicity, or color?, N *(%)*	
Most or some of the time	9 (75.0)
Rarely or never	3 (25.0)
Experience of discrimination – Global questions	
How often do you feel that racial/ethnic groups who are not white, such as African Americans and Latinos, are discriminated against?, N *(%)*	
One or more times	1 (8.3)
Never	11 (91.7)
How often do you feel that you, personally, have been discriminated against because of your race, ethnicity, or color?, N *(%)*	
One or more times	10 (83.3)
Never	2 (16.7)
Experience of discrimination – Filed complaint*, N *(%)	
Yes	0
No	12 (100.0)
Williams Questions	
Williams Questions – *Major discrimination.* ***Have any of the following has ever happened to you?*** N *(%)*	
Fired from job	6 (50.0)
Not hired for job	6 (50.0)

(*Continued*)

Table 4 (Continued).

	N=12
Denied promotion	5 (41.7)
Stopped, searched, questioned, physically threatened, or abused by police	5 (41.7)
Discouraged by teacher or advisor from continuing education	7 (58.3)
Prevented from moving into neighborhood because the landlord or realtor refused to sell or rent house or apartment	3 (25.0)
Moved into neighborhood where neighbors made life difficult	5 (41.7)
Denied bank loan	4 (33.3)
Received service from someone such as plumber or mechanic that was worse than what other people get	3 (25.0)
Williams Questions – *Major discrimination.* ***If yes, what do you think was the main reason for this experience?*** N *(%)*	
Ancestry/ national origins	2 (2.8)
Gender	0
Race	1 (0.9)
Age	4 (3.8)
Religion	3 (2.8)
Height/weight	0
Shade of skin color	8 (7.4)
Sexual orientation	2 (2.8)
Education/income level	1 (0.9)
Physical disability	8 (7.4)
Other	15 (13.9)
Williams Questions – Day-to-day unfair treatment. *In your day-to-day life, how often have any of the following things happened to you?*	
You have been treated with less courtesy than other people, N *(%)*	
One or more times	9 (75.0)
Never	3 (25.0)
You have been treated with less respect than other people, N *(%)*	
One or more times	9 (75.0)
Never	3 (25.0)
You have received poorer service than other people at restaurants or stores, N *(%)*	
One or more times	7 (58.3)
Never	5 (41.7)
People have acted as if they think you are not smart, N *(%)*	
One or more times	8 (66.7)
Never	4 (33.3)

(*Continued*)

Table 4 (Continued).

	N=12
People have acted as if they are afraid of you, N *(%)*	
One or more times	6 (50.0)
Never	6 (50.0)
People have acted as if they think you are being dishonest, N *(%)*	
One or more times	7 (58.3)
Never	5 (41.7)
People have acted as if they are better than you are, N *(%)*	
One or more times	9 (75.0)
Never	3 (25.0)
You have been called names or insulted, N *(%)*	
One or more times	8 (66.7)
Never	4 (33.3)
You have been threatened or harassed, N *(%)*	
One or more times	6 (50.0)
Never	6 (50.0)
You have been followed around in stores, N *(%)*	
One or more times	8 (66.7)
Never	4 (33.3)
Williams Questions – Day-to-day unfair treatment supplement. *You indicated that you have experienced at least one of the scenarios in the previous section. What do you think was the main reason for this/these experience(s)?*	
Ancestry or national origin, N *(%)*	1 (8.3)
Gender, N *(%)*	0
Race, N *(%)*	4 (33.3)
Age, N *(%)*	1 (8.3)
Religion, N *(%)*	0
Height or weight, N *(%)*	0
Shade or skin color, N *(%)*	2 (16.7)
Sexual orientation, N *(%)*	0
Education or income level, N *(%)*	0
Physical disability, N *(%)*	3 (25.0)
Other, N *(%)*	1 (8.3)
Williams Questions – Response to unfair treatment. *You indicated that you experienced at least one of the scenarios mentioned in the previous section, how did you respond to this/these experience(s)?*	

(*Continued*)

Table 4 (Continued).

	N=12
Tried to do something about it	
Yes, N (%)	5 (41.7)
No, N (%)	7 (58.3)
Wake Forest Physician Trust (WFPT), mean (SD) [score range 10–50]	37.1 (3.2)
Interpersonal Trust Scale (ITS)	
Patient trust: medical professional, mean (SD) [score range 5–25]	18.0 (3.3)
Patient trust: insurance, mean (SD) [score range 5–25]	16.0 (2.7)

experienced some form of discrimination when getting service in a store or restaurant, or when getting medical care, respectively. A majority (N = 7) of participants reported experiencing some form of discrimination due to their race, ethnicity, or color when they were on the street or in a public setting. The "Worry" questions or the EOD sub-section demonstrated that, when they were under the age of 18, a majority of participants (N = 8) worried about people in their racial/ethnic group experiencing unfair treatment due to their race, ethnicity, or color and a majority (N = 7) worried about themselves experiencing unfair treatment because of their race, ethnicity, or color. This was also the case over the last year, where a majority (N = 9) reported worrying about other people in their racial/ethnic group and all participants (N = 12) reported worrying about themselves experiencing unfair treatment due to their race, ethnicity, or color. Item 1 of the "Global" questions in the EOD sub-section revealed similar sentiments with a majority (N = 11) reporting that they feel racial/ethnic groups who are not white are "often" discriminated against. In terms of participants themselves, a majority (N=7) reported feeling that they have either "rarely" or "sometimes" been discriminated against due to their race, ethnicity, or color. All participants (N = 12) reported that they have never filed a formal complaint due to racial discrimination. Responses to the Williams sub-section of the EDS revealed that a majority of participants (N = 7) have been discouraged by a teacher or advisor from continuing education and half (N = 6) reported being fired from or not hired for a job due to discrimination, respectively. Participants reported the overall main reason for any discriminatory experience was primarily due to shade of skin color (N = 8), physical disability (N = 8), or other (N = 15). If participants reported more than one experience of discrimination, they were able to provide a reason for each, allowing for more comments than total participants. A majority of participants also reported that they have been treated with less courtesy (N = 9), been treated with less respect (N = 9), received poorer service in restaurants or stores (N = 7), experienced others acting as if the participant was not smart (N = 9), experienced others acting as if the participant was being dishonest (N = 7), experienced others acting as if they are better than the participant (N = 9), have been called names or insulted (N = 8), and have been followed around in stores (N = 8). In addition, half (N = 6) of participants reported experiencing others acting as if they were afraid of the participant, and half (N = 6) reporting being threatened or harassed. Participants reported the main reason for such experiences was primarily due to race (N = 4), physical disability (N = 3), or their shade of skin color (N = 2). A majority (N = 7) also responded that they did not try to do anything about it after experiencing these discriminatory behaviors or actions.

Overall, participants reported moderately high levels of trust in their own physicians according to the mean WFPT scores. Regarding interpersonal trust, participants on average also scored moderately high on the ITS in terms of their trust in medical professionals in general but scored slightly lower on average in terms of their trust in insurance companies.

Discussion

This mixed methods study assessed participants reactions to a chronic pain peer-led program that was adapted for adult patients with SCD. In addition, we explored interactions between personal experiences of racism and discrimination and clinical manifestations of SCD such as physical pain and emotional distress among participants. Although prior studies endorsed the influence of pain from SCD on emotional wellbeing,[24] our key informant interviews revealed that most participants identified the influence between their emotions and SCD pain as bidirectional. Meaning that chronic and acute SCD pain can cause an emotional response and impact their overall wellbeing, but also external factors – such as experiences with health-care clinicians and systems – can contribute to emotional responses and play a role in how they experience SCD pain. Most described at least one experience of racism or discrimination in the health-care setting and identified a perceived lack of clinician knowledge about SCD as a barrier to appropriate care. These findings are similar to those reported by other groups evaluating the care of adults with SCD.[25,26]

Participants also identified both current use of non-pharmacological treatments to manage SCD-associated pain and the desire for more education around these modalities in the future. Many participants reported currently using non-pharmacological therapies as adjuvants to pharmacological therapies, primarily for mild-to-moderate pain to avoid using medications. There was a desire to understand which therapies might be most effective for SCD specifically and acute versus chronic pain. This follows with prior studies that evaluated the prevalence of non-pharmacological pain management techniques in adults with SCD, but emphasizes the need for further research to evaluate the effectiveness of these modalities.[8] The American Society of Hematology guidelines for managing acute and chronic pain in SCD also point to the need for additional studies focusing on the effectiveness of non-pharmacological therapies.[27] The literature does describe a nonpharmacological curriculum teaching psychological and biobehavioral pain management strategies, but is specific to children and adolescents and is delivered by health-care professionals in the inpatient setting.[28]

Study participants also highlighted the differences between pain associated with SCD and other chronic conditions causing pain. SCD is a condition causing a spectrum of morbidity that includes not only vaso-occlusive crises but acute and chronic organ complications which are often unrecognized by caregivers and clinicians. This underscores the importance of tailoring future interventions to be disease specific in a way that addresses the complex treatment needs of patients with SCD.

In contrast to discriminatory experiences from health-care clinicians and systems that were described by participants during the key informant interviews, analysis of survey responses did not support the hypotheses that participants would report high levels of discrimination and low levels of trust in their physicians. In the survey, participants reported infrequent experiences of discrimination in medical settings and overall moderate trust in medical professionals. When the survey asked about occurrences of discrimination in more general settings, most participants did report these experiences, raising the possibility that they did not distinguish between the two experiences. Prior research has suggested that self-reports of discrimination might be limited by patient recall.[29–31] Specifically, strategies used to address experiences of discrimination or racism might contribute to individuals being less likely to acknowledge and/or report such experiences.[29–31] Another consideration is that some participants may have been hesitant to disclose or describe negative past experiences in this study since the research was conducted in the same health system as where participants received their SCD care. It is also possible that the recent development of a comprehensive SCD center in this institution has provided additional education about and exposure to adults living with SCD, decreasing experiences of discrimination or racism. These are key factors for future studies to consider.

One other reasoning for this discordance is that our study did not directly ask about experiences of stigma related to SCD, which has been reported to be high in other studies.[32] Stigma and discrimination are two distinct entities but interact to influence overall experience in health-care settings.[33,34] When asked about specific responses to experiences of discrimination, many participants reported they either accepted it or did nothing. This demonstrates a key area for future research; giving patients with SCD the necessary tools and resources to better navigate these experiences.

The findings from the key informant interviews along with the participants' ratings of the current chronic pain curriculum will be used to develop a SCD-specific curriculum that will be evaluated in future work. The findings also call

attention to the need to address experiences of racism and stigma among patients with SCD in the context of pain management, given the connection identified by patient participants in this study.

Limitations

The age range of our sample may have been skewed towards younger age groups since we restricted study participants to those with access to a phone and email or text messaging. However, no one who declined to participate in the study cited technology-related concerns as their reason for declining. This was a pilot study with participants recruited from a single institution. As such, our findings may not be generalizable to other settings. Participants in this study were all cared for by a specialized sickle cell program and thus, results such as physician trust may not be generalizable to patients who do not have a dedicated sickle cell medical home and clinicians. Additionally, patients consenting to and participating in research studies likely have a substantial preexisting degree of trust in their SCD clinicians, possibly skewing the data on physician trust. The pain group curriculum used in this study was informed by feedback from a convenience sample of participants which may not be generalizable to other patients with SCD, particularly those in other institutions or settings. Although participants were given a summary of the current chronic pain group curriculum, there was still some ambiguity in topic areas and skills to be taught specifically for SCD patients. This may have limited participant's ability to effectively evaluate the current curriculum as an example of what could be delivered in SCD pain programs. The sample size for this study is relatively small but provided nuanced and detailed insights about real-world interactions between experiences of racism and discrimination, and SCD manifestations such as physical pain for adult patients with SCD. Despite our limitations, this study was a direct response to a need identified by patients with SCD who are interested in peer-mediated group interventions to improve chronic pain and represents an opportunity to explore novel methods of pain management for patients with SCD. Additionally, it gives voice to the SCD community in designing novel interventions that are culturally relevant to a population that is often stigmatized due to frequently seeking treatment for chronic and acute pain.[35]

Conclusion

SCD is a complex disease that presents with chronic pain and acute episodes of vaso-occlusive crises. As such, it is vital to create pain management resources that address the unique treatment and cultural needs of adults with SCD. In adapting a general chronic pain curriculum to manage SCD-associated pain, the patient-centered approach of this study identified the need to address experiences of racism and stigma in addition to experiences of emotional and physical pain among patients with SCD. Findings from this study will inform the development of a peer-led SCD pain group curriculum for adult patients.

Acknowledgments

Work supported by an Institutional Development Award (IDeA) from the National Institute of General Medical Sciences of the National Institutes of Health under grant number U54-GM104941 (PI: Hicks). This work was also partially supported by the National Institute of General Medical Sciences of the national Institutes of Health under Award Number P20GM109021 (PI: Brousseau). The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health. The abstract of this paper was presented at the 2022 Annual Meeting of the American Society of Hematology as a poster presentation with interim findings. The poster's abstract was published in "Poster Abstracts" in *Blood*: https://ashpublications.org/blood/article/140/Supplement%201/10873/493034/Pain-Attitudes-in-Adults-with-Sickle-Cell-Disease.

Disclosure

SHG, KS, and CMJ received support for work on this manuscript by the National Institute of General Medical Sciences of the National Institutes of Health under the grant number 5P20GM109021-08 (PI: Brousseau). SHG has received funding through grants or contracts with Teen Cancer America, a Patient-Centered Outcomes Research Institute grant DI-2021C2-23827 (PI: Lanzkron), and a National Institutes of Health grant 1R01NR020781-01A1 (PI: Crosby); received payments for consulting fees provided by Novartis; provided unpaid services provided to the Medical Society of

Delaware, Delaware Foundation for Medical Services; and provided unpaid services as an Executive Board member of the Sickle Cell Association of Delaware, Delaware Children's Museum, and the FC Brandywine Soccer Club. KDW received support for work on this manuscript by an Institutional Development Award (IDeA) from the National Institute of General Medical Sciences of the National Institutes of Health under grant number U54-GM104941 (PI: Hicks). CSW has received support from the Patient-Centered Outcomes Research Institute under contract number AD-2021C3-24941 (PI: Jan). LMP has received an honorarium for serving as Editor-in-Chief of the *Journal of Developmental & Behavioral Pediatrics*. SL has received funding through grants or contracts with the Health Resources and Services Administration, the Patient-Centered Outcomes Research Institute, Imara, Novartis, Takeda, Merck, Alexion, Imara, GBT, HRSA, PCORI, and CSL-Behring; received payments for consulting fees provided by Novartis, Bluebird Bio, Agios, Novo Nordisk, and GlycoMimetics; received payments or honoraria for lectures, presentations, speaker's bureaus, manuscript writing, or educational events provided by Pfizer and Janssen; received payments for participation on a Data Safety Monitoring Board or Advisory Board provided by Pfizer and Magenta; has provided unpaid services to the National Alliance of Sickle Cell Centers; and is a trustee of a family trust with Pfizer and Teva stock or stock options. The authors report no other conflicts of interest or financial support for this work.

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