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ORIGINAL RESEARCH

''I'm Talking About Pain'': Sickle Cell Disease Patients With Extremely High Hospital Use

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BACKGROUND: A small minority of sickle cell disease patients accounts for the majority of inpatient hospital days. Admitted as often as several times a month, over successive years, this cohort of patients has not been studied in depth despite their disproportionate contribution to inpatient hospital costs in sickle cell disease.

OBJECTIVE: To characterize the subjective experience of extremely high hospital use in patients with sickle cell disease, and generate hypotheses about the antecedents and consequences of this phenomenon.

DESIGN: Qualitative study involving in-depth, open-ended interviews using a standardized interview guide.

SETTING: A single urban academic medical center.

PARTICIPANTS: Eight individuals, of varying age and gender, identified as the sickle cell disease patients who are among the highest hospital use patients over a 3-year period.

RESULTS: A common narrative emerged from the interview transcripts. Participants were exposed to the hospital environment and intravenous (IV) opioids at a young age, and this exposure was associated with extremely high hospital use in adulthood, evident in descriptions of multiple dimensions of their lives: pain and opioid medication use, interpersonal relationships, and personal development.

CONCLUSIONS: Our results suggest a systematic, self-reinforcing process of isolation from mainstream society, support structures, and caregivers, based on increasing hospitalization, growing dependency on opioid medications, as well as missed developmental milestones. Further study and interventions should be geared towards breaking this spiraling cycle with long-term strategies in disease management and social integration. *Journal of Hospital Medicine* 2013;8:42–46. © 2012 Society of Hospital Medicine

Sickle cell disease (SCD) accounts for approximately 113,000 hospital admissions annually in the United States, at a cost of approximately $500 million.[1] The majority of these hospital admissions are due to painful episodes, vaso-occlusive crises, often triggered by a psychological or physical stressor.[2] Most individuals manage these crises at home,[3] with sporadic admissions occurring, on average, 1.5 times per year.[4] However, a minority of patients are admitted as often as several times per month, persistent over successive years,[5,6] a phenomenon we call extremely high hospital use (EHHU). These patients account for a disproportionate share of total costs, and may suffer worse health outcomes. Three or more hospital admissions per year has been correlated with a lower 5-year survival rate,[7] and high emergency room utilization was found to be associated with more reported pain, and more opioid use at home.[8]

To improve patient quality of life and to decrease healthcare costs in the management of SCD, there has been increased focus on predicting high utilization[9] and identifying strategies to decrease hospitalization rates, especially among patients with EHHU.[10] Although SCD patients with EHHU have been identified as a small group of outliers,[5] the psychosocial factors associated with EHHU in adults with SCD have not been investigated. The objective of this qualitative study is to characterize the subjective experience of patients with sickle cell disease and EHHU, and generate hypotheses about its antecedents and consequences.

METHODS

The institutional review board (IRB) of Yale University School of Medicine, New Haven, CT, approved the research protocol.

Participants

We accessed the Yale-New Haven Hospital administrative database to identify the number of patients with SCD who demonstrated EHHU that did not remit over successive years.[5] We identified the 10 highest inpatient utilizing individuals with sickle cell disease over the period January 1, 2008–December 31, 2010; 8 individuals consented to participate. We collected the following data on each participant through chart review: hemoglobinopathy, length of stay, primary diagnosis for each admission, and SCD-related comorbidities (eg, avascular necrosis, leg ulcer, etc). No research team member was involved in the care of any of the participants.

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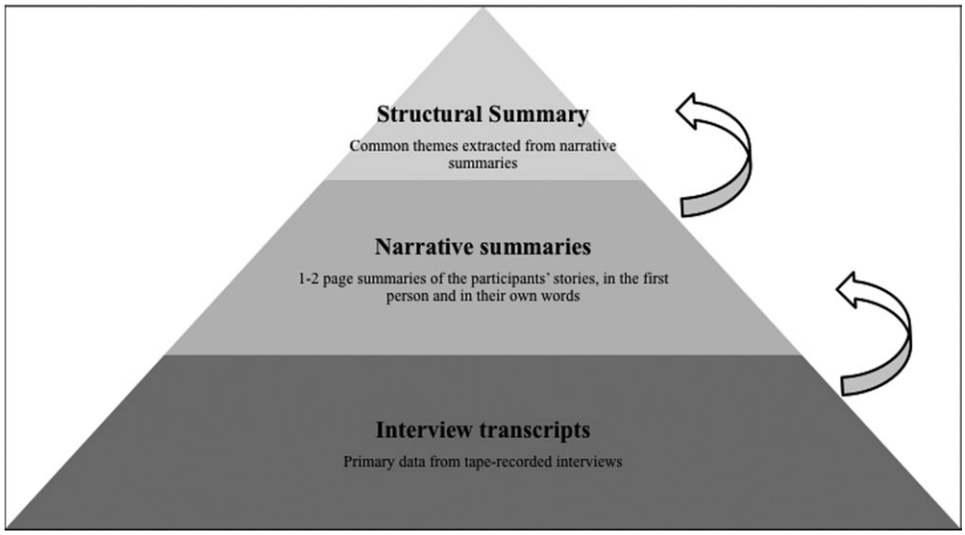

FIG. 1. Narrative and analysis model.[13]

Interviews

Based on literature review of other qualitative research in SCD, we created an interview guide to include the following themes: 1) disease, pain, and medication; 2) hospitalization; 3) support structures; 4) daily life; and 5) personal relationships (see Supporting Information, Appendix I, in the online version of this article). Applying Grounded Theory in qualitative research, the interview guide underwent several minor modifications based on field-testing interviews with 4 interviews of patients not enrolled in the study and early interviews with study participants.[11] Tape-recorded interviews, each lasting at least an hour,[12] were conducted by 1 researcher (D.W.) during inpatient hospitalizations, at least several days after admission to ensure that participants were comfortable enough to participate. When the interview exceeded an hour, it was continued at a later time. Recordings were transcribed by a professional transcription service and verified for accuracy by the interviewer. Participants were compensated $25 for completed interviews.

Narrative Analysis

The analysis team consisted of 2 psychiatrists (1 with additional training in internal medicine), 1 medical student, and 1 internist with additional training in addiction medicine. Analysts read each transcript, became thoroughly familiar with its content, and met to discuss preliminary findings. Then, we created "patient experts" among the group, assigning each analyst 2 interviews with which s/he prepared a detailed summary in the first person, using the participant's own words, according to an established process in phenomenological research[13] (Figure 1). These "narrative summaries" allowed for the development of a holistic view of the participant, the creation of a narrative structure, and the fostering of an "empathic bridge,"[13] a connection between the experiences of the participant and those of the reviewer. The summaries were read aloud at research meetings allowing for discussion, and the content of the summaries were modified based on the consensus of the group.

Next, we randomly rotated the narrative summaries so that each of the 4 analysts became an "expert" for 2 additional participants in order to critically evaluate the compiled narratives, and develop a "structural summary"[14]—a summary of the prevalent themes. We extracted content from the narrative summaries based on these common themes, and returned to the transcripts as needed for relevant quotations. This inductive process allowed unique participant narratives to come through unconstrained by a predetermined coding structure.

The team reached consensus on organizing themes following the chronology of childhood to adulthood. This model was utilized to preserve the narrative basis of the methodology, and to ultimately elucidate the antecedents, subjective experience, and consequences of EHHU. An audit trail was maintained throughout the data analysis process.

RESULTS

Table 1 displays demographic, clinical, and hospitalization data for the 8 participants. These patients represented approximately 8% of the population with sickle cell disease at the study institution, but accounted for 57% of hospital days among sickle cell disease patients over a 3-year period, with cumulative hospital days near or above 100 days per patient each year. Greater than 90% of admissions were for vaso-occlusive crises without other SCD-related complications. However, many participants had complicated medical histories, including avascular necrosis, acute chest syndrome, and leg ulcers.

Participant interviews presented a common narrative of the evolution of EHHU from a young age, culminating in a universally negative description of

TABLE 1. Demographic and Clinical Data

Participant	Age at Interview Date	Gender	Hemoglobin Diagnosis*	Average Hospital Days per Year 2008–2010†
1	34	F	HbSS	171
2	27	F	HbSS	263
3	26	M	HbS-B thalassemia	151
4	34	M	HbSC	111‡
5	37	F	HbSS	202
6	25	F	HbS-B thalassemia	104
7	24	F	HbSS	123
10	32	M	HbSS	94

*HbSS denotes homozygosity for the sickle cell gene (*HBB* glu6val), sickle cell anemia. HbS-B thalassemia denotes heterozygosity for the sickle cell gene (*HBB* glu6val) and one of the B-thalassemia gene mutations. HbSC denotes heterozygosity for the sickle cell gene (*HBB* glu6val) and the hemoglobin C gene (*HBB* glu6lys), sickle-hemoglobin C disease.
† Hospitalization data only applies to hospital days at Yale-New Haven Hospital.
‡ Participant spent 1 y in prison—this interval was not included in his hospitalization rate.

hospitalization: "It's like jail." (participant 2); "It's like a massacre, coming in the hospital; I get tortured." (participant 1). Saturation was reached on major themes, which fit into 3 general categories: pain and opioid medication use, interpersonal relationships, and personal development.

Pain and Opioid Medication Use

Participants reported hospital use dating back to childhood, which was the first exposure to intravenous opioid medications and the beginning of a trajectory of accelerating use, tolerance, and dependence: "You know, I came in the hospital when I was two years old . . . I stayed 'til I was like five and a half. I started school here." (participant 1); "I started taking that medicine when I was on the pedi side. . . . So my body's already used to it . . . it doesn't really touch me. They're gonna have to up my doses." (participant 7).

As adults, participants expressed awareness of the potential problems of opioids. During interviews, many participants exhibited side effects of these medications, such as itching and somnolence. Moreover, participants expressed awareness of the skepticism and mistrust from providers, and acknowledged that such sentiments may be justified toward certain patients: "That's all our body knows, is meds, meds, meds. And because your body is addicted to this level, you gotta go up another level, but some doctors think we're taking too much. How can we be taking too much when we need it?" (participant 5); "The oxy which I'm on [oxycontin 240 mg per day] . . . when you take that, you going to sleep. . . . And then some of them will say 'when you go home you're not taking the medicine like you should.'" (participant 5).

Opioids were taken in and out of the hospital by all participants, and were identified as necessary in combating debilitating pain. Many participants expressed a reluctance to try other forms of therapy, such as hydroxyurea: "These new chemicals, you come across doctors who say 'there's this new medicine out, and it's been out for such and such amount of time—I think it would be good, can we try it with you?' No. I'm not a guinea." (participant 2).

While all participants described unpredictable pain crises, some also described an underlying, constant pain syndrome: "You know, like, I could be fine right now; the next minute I could be 'Oh, my God' crying, so much pain. . . . You never know when you're gonna have a crisis." (participant 1); "There's never no pain. There's always pain. It's just a fact of life. . . . I wake up and I can deal with the pain, it's not that bad today. But then when the crisis hits, that's when it gets unbearable." (participant 10); "I'm not in pain every day, every second. . . . To me, I don't think that any sickle cell patient is in pain every day. They make theirselves to [be] . . . it saddens me sometimes." (participant 6).

Interpersonal Relationships

In childhood, participants developed close relationships with the staff of the children's hospital and an attachment to this institution: "I loved pediatrics. It's the adult side I can't stand. They treat you better." (participant 7); "Some people in the ER, they know us; and I call them my family . . . they already know what I need." (participant 5).

In contrast, the hospital experience during adulthood was often punctuated by bitter relationships with staff, and distrust over possible excessive use of opioids. Moreover, participants raised the possibility of racism in their interactions with hospital staff. Overall, participants highlighted a lack of empathy among caregivers: "Some doctors, they're rude, like, they're rough. . . . They'll just pull out the scope . . . bang it onto my back, or just push on my body or areas where it hurts." (participant 6); "'I'm your doctor. And I say I think you're doing a lot better, how would you feel about going home today?' And you can say . . . 'I don't think I'm ready.' And I can say 'Well, you can't live here in the hospital. Why do you think you're not ready for home yet? . . . You're never, ever going to be pain free,' and that's when you turn around to me and say 'I know that. I've been dealing with this for all my life. I know I'm never gonna be pain free!'" (participant 2).

Such negative interactions extended to friends and family members, leading to a sense of social isolation and a reluctance to discuss their disease with others: "I just don't think people will understand where I'm coming from. . . . So I just don't talk to anybody, I keep it inside. Or I write it in my diary." (participant 5); "I don't have any friends. I have associates. I'm always by myself." (participant 1).

Even though participants expressed dismay at dysfunctional relationships within the hospital, they also voiced affection for staff members. Participant 1 described hospital stays that were "loving," and participant 3 described his hematologist as his "brother from another mother," and a nurse practitioner as his "aunt."

Personal Development

Hospitalization in childhood was linked to EHHU as an adult by the derailment that participants described in their personal development. Prolonged hospitalization and illness were barriers to education, interfering with the development of social as well as academic skills: "I couldn't spell … me being in a hospital for so much … I was like, no, I don't want that bookwork like everybody else." (participant 3); "I stopped going to school. I told [my mom] that I was not going back to school because the kids made fun of me … 'Oh, she has a disease. Be careful, you might get the cooties.'" (participant 2).

Participants also described a sense of foreshortened future. Many were told that they would not survive their teens: "They told my mother I would die before I was 12 years old. And I would be scared to go to sleep, because I would think I was gonna die in my sleep." (participant 2)

As an adult, numerous and/or prolonged hospitalizations interfered with participants' ability to remain employed, and they experienced strains on fulfilling family roles: "I would love to work again, but who gonna hire somebody that's always out, more than you're working. Nobody." (participant 5); "Sometimes I feel like I'm neglecting my son, being here…. You have to take care of yourself in order for you to be there for him. But it just stresses me out." (participant 6).

The struggles of hospitalization and pain management took their toll on participants' mental health. Participants described difficulty sleeping, depression, and suicidal thoughts: "Sorrow. That's what [sickle cell] means to me. Unhappy.… Everything's depressing. It takes over your body and your mind and your soul." (participant 5); "I really was gonna kill myself 'cause it's like, sometime, the pain, you be in so much pain, you be like, 'fuck this, man.' … My pain was bothering me so much, I laid down on the highway, wishin' a car would run me over." (participant 4).

Despite fragile mental and physical health, many participants described feelings of strength and resilience, and some described hope in the future for employment, education, travel, and family: "I don't know why God picked me, but for some strange, mysterious reason he picked me.… I still don't know what that reason may be, but I ain't gonna give up…. Maybe he got some kind of plan in store." (participant 2).

DISCUSSION

Our study population represents a unique and understudied group among patients with SCD. While several themes from prior research on individuals with SCD were present—reciprocal mistrust between patients and providers surrounding opioid analgesics and pain reporting,[15,16] racial and disease-related bias,[17] patient dissatisfaction with clinical services,[18]—the common narrative of thwarted personal development in the setting of a long history of hospitalization and opioid use was striking.

The developmental perspective posits that age-appropriate tasks govern basic capacities and skills (academic, interpersonal, affective, and cognitive) honed through institutional interactions (family, school, and community), which allow individuals to develop autonomy that guides them into effective participation in social groups and civil society, and eventually to becoming guarantors for the next generation. Our participants described problems such as social isolation, depression, and dependence on medications, all linked to their description of recurrent stays in the hospital during childhood and adolescence, where missed vocational and social opportunities left their indelible mark. Participants expressed an awareness of their inability to lead productive lives, and the perception that they were burdensome to their caregivers and the hospital.

While previous research has correlated high hospital utilization in SCD with factors like poor coping strategies,[19–21] high levels of stress,[22] and inadequate support,[17] our interviews suggest that such psychosocial difficulties may be consequences as well as causes of hospitalization, creating an accelerating downward spiral of dysfunction. At the center of this spiral is participants' ongoing experience of pain, which may be related to SCD, medications, or neither.[23] Additionally, chronic anemia, opioid exposure, airway disease, and cerebrovascular disease are all implicated in impaired neuropsychological functioning in children and adolescents with sickle cell disease.[24,25]

Clinical features of SCD remain relevant to hospitalization in adulthood. Chart review revealed that the vast majority of inpatient admissions were due to vaso-occlusive crises uncomplicated by SCD-related pathology, such as aseptic necrosis of bone or acute chest syndrome. This finding is consistent with previous work,[9] which correlated new onset of high hospital use with SCD-related complications, but not reliably with persistent EHHU, our study population.

The double-edged sword of opioid use[26] was starkly evident in participants' narratives. Crippling vaso-occlusive crises were competently treated with opioids starting in childhood, but then a cycle of increasing outpatient doses of opioids and more frequent and longer courses of inpatient intravenous opioids followed. Participants felt judged and stigmatized for seeking one of the few treatment options they had been offered, resulting in confusion and bitterness at times. Other potential complications of long-term opioid therapy less well known to patients and providers —such as hypogonadism and hyperalgesia[27]—may have played a role in the patient experience and should be examined in future research. In addition, undertreatment of pain may lead to pseudoaddiction,[28] underscoring the complexity of delineating the

pathologies of dependence, addiction, withdrawal, acute pain, and chronic pain.

Our study is limited primarily by the fact that it was conducted with a small number of participants. It is also possible that institutional variation, especially with regard to pain management, makes it difficult to generalize our hypotheses. Similarly, our participants grew up in similar environments outside the hospital, which may differ significantly from environments of other individuals with EHHU. Lastly, participants were all interviewed as inpatients, and the acuteness of their illness may have influenced responses. Despite these limitations, we achieved saturation on the major themes, and there was substantial agreement in their experiences of their illness.

Breaking the cycle of alienation from the external world and dependence on the hospital necessitates an acknowledgement of the role of the hospital, pain, and opioid use in the long-term development of individuals with EHHU. Further research should test this developmental hypothesis, and focus on early interventions and the critical transition from pediatric to adult care.[25] Longitudinal quantitative analysis could include psychosocial variables in SCD in the attempt to predict EHHU as has been accomplished in the chronic pain literature.[29] Additionally, a comprehensive qualitative approach including the perspectives of caregivers, family members, and comparison to low hospital utilizers will better inform interventions aimed at ameliorating EHHU. It is particularly important to understand the similarities and differences in the long-term development of patients with SCD who demonstrate EHHU versus low hospital use. The optimal strategy for opioid use in the long-term management of pain in patients with SCD remains to be determined. Alternatives to opioids should be investigated in a controlled trial, and institutional differences should be examined as they relate to EHHU and pain-management strategies. Lastly, our results suggest that psychosocial and skill rehabilitation may mitigate EHHU, and that multidisciplinary resources proactively directed towards this population will reduce hospitalization.[30]

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