A Chronology of Pain and Comfort in Children With Sickle Cell Disease

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Objective: To examine the patterns of children’s and caregivers’ descriptions of pain and the comfort measures used to relieve the pain of sickle cell disease (SCD) at home and in the hospital.

Design: Qualitative and quantitative techniques were used to obtain data. Participants were seen twice, first with and then without a vaso-occlusive episode. Multiple simultaneous methodological triangulation was used to integrate the findings from ethnographic interviews and observations as well as limited quantitative findings about pain and comfort measures used.

Setting: A Midwestern children’s hospital with a regional SCD service.

Participants: Twenty-one African American children and adolescents with SCD, aged 6 to 15 years, and 21 family caregivers.

Results: An 8-phase chronology of pain and comfort was revealed from the data about pain and comfort in children with SCD. Although this chronology was an unexpected finding, it was consistent with the original aim of the investigation. Phase 1 (baseline) represented the usual state of the child's condition, which for most was free of pain. Phase 2 or the “pre-pain” state involved no vaso-occlusive pain but the child began to show prodromal signs and symptoms of painful episodes, such as yellowing of the eyes or fatigue. Phases 2 through 7 involved increasing then decreasing levels of pain, including the pain start point (phase 3), pain acceleration (phase 4), peak pain experience (phase 5), pain decrease start point (phase 6), and steady pain decline (phase 7). A trip to the emergency department usually occurred during phase 5. In phase 8 (pain resolution), the pain had decreased to a manageable level so that the child could be discharged from the hospital. As pain increased and decreased, so did the number and variety of comfort measures.

Conclusions: A chronology of the pain and comfort experiences for children and adolescents during a vaso-occlusive event of SCD emerged from the descriptive data of this study. Findings need to be examined further in larger, quantitative, longitudinal studies that examine more closely the duration, intensity, and character of pain at different times during vaso-occlusive episodes as well as the comfort measures used during specific phases of the pain event.


Editor’s Note: There are few things more heart-wrenching than observing a child or teenager in severe pain. This study is a nice step toward better understanding the progression of sickle cell crisis pain.

Catherine D. DeAngelis, MD

Sickle cell pain has been classified as an acute, recurrent, and chronic pain problem. Little information is available in the research literature that describes the nature or trajectory of the pain of a vaso-occlusive event (VOE) or comfort measures used for children with sickle cell disease (SCD). Little information is available about the frequency of the pain episodes, factors influencing the pain, or how effective pain interventions are at home, in the emergency department, and in the hospital. Furthermore, there is little information about the events preceding and during the treatment of the child with VOE in the emergency department. Gaps in understanding and knowledge about the pain associated with a VOE suggest that more research is needed to provide important information about children's sickle cell pain and the ways it can be managed most effectively. Despite the lack of a strong knowledge base in these areas, pain remains the most common and most troubling symptom experienced by children with SCD and the most frequent reason why families seek medical care.1,2

The original aim of this study was to explore the characteristics of the VOE of SCD at home and in the hospital as described by children and their caregivers and to examine the comfort measures used to relieve that pain. Unexpectedly, a descriptive model emerged from the data that suggested the existence of a multiphase chronology of pain and comfort during VOE for this sample of participants. This
PARTICIPANTS, MATERIALS, AND METHODS

This descriptive study was a part of a larger investigation of comfort measures used for children with SCD from the perspectives of the children and their family caregivers. Multiple simultaneous methodological triangulation was used to examine the pain and to determine the effectiveness of comfort measures used during VOE. Multiple simultaneous methodological triangulation is a method that allows the comparison of information from different sources to examine patterns and then the integration of findings to provide a complete description of the phenomenon under study. It is particularly useful when little is known about the topic of interest, and was chosen for this study of pain and comfort in children with SCD because neither quantitative nor qualitative approaches alone would have been sufficient to provide the information needed to meet our objectives. It was also used because it allowed the use of multiple sources of data, research approaches, and investigators to obtain the most accurate, enhanced description and most complete understanding of the pain and comfort experience through corroboration of evidence. Findings were supported when different sources suggested the same rather than different information. Because these methods were applied concurrently rather than sequentially, the method was classified as multiple simultaneous methodological triangulation.

Three types of methodological triangulation were used in this study. Data triangulation included information from several sources: 2 different people (children and caregivers) and 2 different points in time (during a VOE in the hospital and after discharge after the VOE). Methods triangulation meant that both qualitative and quantitative approaches to data generation were used. The primary qualitative method used was focused ethnography and involved interviews and participant observation. The primary quantitative method was simple descriptive. Several clinically validated measurement tools were used for obtaining scores from children. Investigator triangulation was also used. In addition to the principal investigator (J.E.B.), who collected qualitative interview data, 3 master’s level pediatric nursing student investigators helped to obtain quantitative data. Also, a doctoral student in nursing who specialized in qualitative methods (L.E.S.) worked with the principal investigator in the coding, analysis, and interpretation of the data. Analysis was conducted in phases, initially to provide separate results, then all findings were integrated. This report emerged as only a part of this integration.

The participants in this study were 21 children and adolescents with SCD aged 6 to 15 years and their family caregivers, including 20 mothers and 1 aunt. The sample was derived from a list of 242 patients with active SCD receiving care at 1 Midwestern children’s hospital, 8.6% of the population of patients who were included. Patients were obtained by referral from the staff of the inpatient hematology/oncology unit of the hospital. Participants were selected if the child or family would be able to provide the information needed for the study, were English-speaking, and were born in the United States. Exclusion criteria were a mental or developmental delay of the child or parent or life-threatening complications at the time of the interview. The children and caregivers were the key informants in this study. After informed consent was obtained, the families were interviewed twice: once during and once after a VOE. Interviews were audiotaped and lasted between a half-hour and 2 hours per family. A laptop computer was used to type field notes after the interviews were completed. Field notes were downloaded onto disks. Audiotapes and disks were then labeled with the participants’ names and the date and were stored in the investigator’s home. For confidentiality, the 21 children are referred to as A through U in this article. The audiotapes were sent to an experienced transcriber who then typed the interviews verbatim.

The transcriptions were read at the same time that the audiotape was played to edit for completeness and accuracy. In total there were 40 sets of caregiver/child interviews and 40 sets of field notes, totaling 80 files that were processed for the study. It was not possible to obtain second interviews for 2 of the 21 families. HyperRESEARCH, a qualitative data analysis software package, was used for coding the data. The second author (L.E.S.) worked with the principal investigator (J.E.B.) and the study consultants to identify initial codes for the linguistic data in the interviews. Initial coding yielded more than 200 preliminary codes, which were later collapsed into a set of approximately 100 codes. Categories of meaning units were then developed based on the codes. To determine the accuracy and reliability of the coding process, both the first and second authors coded separately, compared codes for similarities and differences, and arrived at agreement on codes and meanings. After the coding process was complete and categories were clearly identified, the investigators focused on thematic analysis. One of the most important findings was the identification of a chronology of pain and comfort in this sample of pediatric patients with SCD.

Sickle cell pain has been characterized as intermittent, severe, and unpredictable, and requires a variety of pain relief measures to alleviate and eliminate the pain. A minority of the population with SCD (20%) is thought to have a majority of the severe and frequent pain events requiring hospitalization. The mechanism underlying the pain of SCD is the vaso-occlusion from the sickled red blood cells that clump and block blood flow in the vascular system. Although the pathophysiology of VOE is well described and understood, literature descriptions of VOE pain from the perspectives of children and their caregivers are scant. In addition, several sources suggest that VOE is a condition in which patients receive suboptimal management for their pain.
No studies were found that examined the characteristics of pain and comfort during the entire course of a VOE.

Suggestions of inconsistency and unpredictability of the pain of SCD, coupled with the fact that little research has been done on it, leaves many unanswered questions about what can be expected with the pain of SCD and how family and professional caregivers can help children with it. Shedding light on the many unknowns would certainly help. This study was conducted to find out more information about pain from the people closest to the experience—the children with SCD and their families.

**RESULTS**

Twenty-one children and adolescents with SCD and their 21 primary family caregivers participated in this study (Table 1). Based on the descriptive data obtained, a chronology of pain and comfort emerged. Although there were many subtle variations, the data obtained from caregivers were consistent and representative of most participants’ expressed experiences. Eight distinct phases were identified. The progression of the vaso-occlusive pain and the use of concomitant comfort measures seemed to occur within the context of these phases (Table 2). Ferholt defined a chronology as the “onset, duration, periodicity, frequency, and course of a symptom.”

The pain of SCD may be compared with the phases of the moon (waxing, reaching a peak, waning gradually, and then disappearing). The waxing or increase in intensity of pain occurred at varying rates from very slow to very fast; the end point of the waxing during the VOE was often agonizing pain that remained at a plateau, unrelied for some period of time. The waning or decreasing of the pain during a pain event also occurred at varied rates, sometimes rapidly, sometimes slowly.

**PHASE 1**

Phase 1 (baseline) represented the usual state of the child’s pain condition. There was no vaso-occlusive pain, and therefore no comfort measures were used for it. Some children always had some level of pain not related to a VOE but instead to some complication of SCD (eg, avascular necrosis of a hip) and comfort measures may have been used for this.

Many children and caregivers were able to trace a precipitating event that occurred during phase 1 that they believed influenced the onset of a VOE. They reported that changes in the weather, cold weather, states of dehydration, or overexertion from physical activity preceded the occurrence of a VOE. Rapid changes in environmental temperature, usually from warm weather to abruptly cooler weather, were especially troublesome. Mother S reported, “When the weather changes like that, we’ve got to keep him in. It has a great amount to do with his sickness. . . . We see a trend . . . in February or March and in September and October . . . he usually gets sick.”

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**Table 1. Demographic Information**

<table>
<thead>
<tr>
<th>Patient Identifier</th>
<th>Age, y/Sex</th>
<th>Other Conditions</th>
<th>No. of Hospital Admissions in 1 y</th>
<th>Caregiver Age, y</th>
<th>Lifetime Complications</th>
<th>Early Pain Score*</th>
<th>Late Pain Score†</th>
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</thead>
<tbody>
<tr>
<td>A</td>
<td>14.7/F</td>
<td>Asthma</td>
<td>2</td>
<td>35</td>
<td>Enuresis, infection, splenic sequestration, acute chest syndrome</td>
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<td>70</td>
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<tr>
<td>B</td>
<td>10.1/M</td>
<td>None</td>
<td>0</td>
<td>31</td>
<td>Enuresis, serious infection</td>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>C</td>
<td>15.8/F</td>
<td>None</td>
<td>1</td>
<td>43</td>
<td>None</td>
<td>80</td>
<td>80</td>
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<tr>
<td>D</td>
<td>11.75/M</td>
<td>None</td>
<td>2</td>
<td>34</td>
<td>None</td>
<td>11</td>
<td>15</td>
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<tr>
<td>E</td>
<td>14.5/M</td>
<td>None</td>
<td>4</td>
<td>35</td>
<td>Aplastic crisis, avascular necrosis of hips, cholecystectomy</td>
<td>26</td>
<td>33</td>
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<tr>
<td>F</td>
<td>11.5/M</td>
<td>None</td>
<td>2</td>
<td>41</td>
<td>Serious infection, cholecystectomy</td>
<td>50</td>
<td>50</td>
</tr>
<tr>
<td>G</td>
<td>13.4/F</td>
<td>None</td>
<td>2</td>
<td>27</td>
<td>None</td>
<td>40</td>
<td>60</td>
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<tr>
<td>H</td>
<td>14.1/M</td>
<td>Asthma</td>
<td>12</td>
<td>31</td>
<td>Enuresis, serious infection, cholecystectomy</td>
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<td>30</td>
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<tr>
<td>I</td>
<td>15.1/M</td>
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<td>1</td>
<td>32</td>
<td>Enuresis, aplastic crisis</td>
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<td>35</td>
</tr>
<tr>
<td>J</td>
<td>14.2/F</td>
<td>None</td>
<td>3</td>
<td>30</td>
<td>Avascular necrosis of hips</td>
<td>40</td>
<td>40</td>
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<tr>
<td>K</td>
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<td>Asthma</td>
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<tr>
<td>L</td>
<td>6.3/F</td>
<td>Asthma, seizures</td>
<td>38</td>
<td>45</td>
<td>Enuresis, infection, acute chest syndrome, avascular necrosis of hips, stroke</td>
<td>3†</td>
<td>2†</td>
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<tr>
<td>M</td>
<td>8.7/M</td>
<td>None</td>
<td>1</td>
<td>34</td>
<td>Serious infection</td>
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<td>0</td>
</tr>
<tr>
<td>N</td>
<td>12.5/F</td>
<td>Asthma, sinusitis</td>
<td>7</td>
<td>39</td>
<td>Aplastic crisis, avascular necrosis of hips, stroke, acute chest syndrome, cholecystectomy</td>
<td>30</td>
<td>30</td>
</tr>
<tr>
<td>O</td>
<td>13.4/M</td>
<td>None</td>
<td>2</td>
<td>41</td>
<td>None</td>
<td>50</td>
<td>60</td>
</tr>
<tr>
<td>P</td>
<td>15.7/F</td>
<td>None</td>
<td>16</td>
<td>26</td>
<td>Enuresis, splenic sequestration, aplastic crisis, cholecystectomy, splenectomy</td>
<td>50</td>
<td>50</td>
</tr>
<tr>
<td>Q</td>
<td>14.0/F</td>
<td>Asthma</td>
<td>1</td>
<td>35</td>
<td>Serious infections (4-5), avascular necrosis of hips</td>
<td>85</td>
<td>85</td>
</tr>
<tr>
<td>R</td>
<td>14.1/M</td>
<td>Asthma</td>
<td>4</td>
<td>37</td>
<td>Cholecystectomy</td>
<td>40</td>
<td>40</td>
</tr>
<tr>
<td>S</td>
<td>11.1/M</td>
<td>Hayfever</td>
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<td>26</td>
<td>None</td>
<td>80</td>
<td>80</td>
</tr>
<tr>
<td>T</td>
<td>10.7/F</td>
<td>None</td>
<td>1</td>
<td>29</td>
<td>None</td>
<td>70</td>
<td>70</td>
</tr>
<tr>
<td>U</td>
<td>12.9/F</td>
<td>None</td>
<td>1</td>
<td>29</td>
<td>Splenic sequestration, aplastic crisis, acute chest syndrome, cholecystectomy, splenectomy</td>
<td>20</td>
<td>20</td>
</tr>
</tbody>
</table>

* Pain scores obtained early in the study interviews during the vaso-occlusive event in the hospital. All scores are on 0 to 100 scale except as indicated.
† Pain scores obtained late in the study interviews during the vaso-occlusive event in the hospital.
‡ Patient L used a 0- to 5-point scale.
N stated, “...if the weather changes, like it’s cold and raining... that really does it.”

Participants reported instituting interventions to avoid the weather factor. N said, “Make sure the heat is avoided... when the weather is very warm... it makes him sick.” Mother N said, “Her eyes get real dark like the whites of her eyes... just the whites of her eye... it makes her sick.”

Also reported were interventions to prevent states of dehydration, which participants linked to a VEO, especially during summer months. M reported taking swimming lessons in the summer, being in the sun, and becoming dehydrated, resulting in a hospitalization for a VEO in July, an unusual month for him to have a VEO. Most caregivers acknowledged that they were able to see the episode of VEO coming before the pain actually began. Caregivers then began to initiate interventions to prevent the development of the pain episode. These measures were minimal or maximal, depending on the preferences of the individual caregiver.

Caregivers reported that the early indicators of impending pain could be observed, often linked to a precipitating factor. Some children’s eyes noticeably changed, either vaguely, such as loss of usual appearance or luster, or dramatically, such as a yellowish hue that may be the jaundice caused by the breakdown of red blood cells due to their shortened life spans or sickling. Mother H reported, “They get really yellow. On a day-to-day basis, they’re kind of light yellow, but they get dark yellow and I know then that he’s feeling bad and it won’t be long he’ll come tell me, ‘Mama, I’m hurting.’” Mother N said, “Her eyes get real dark like mustard... .” Another mother said, “When there’s no twinkle in her eyes, she is getting sick, it [sclera] turns yellow, otherwise, it’s white.” Mother S said “That’s a good signal for us that he might go into crisis, that his eyes start turning like a dark... kind of yellow color... just the whites of his eyes. And that’s always a clue to us.” Several caregivers indicated changes in the eyes but did not specify what was different about them, such as “her eyes tell me” (patient C’s aunt) and “it’s in her eyes” (mother T). One mother noticed circumoral pallor in her son. Several others said they

<table>
<thead>
<tr>
<th>Phase</th>
<th>Pain Characteristics</th>
<th>Comfort Measures Used</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 (Baseline)</td>
<td>No vaso-occlusive pain; pain of complications may be present, such as that connected with avascular necrosis of the hip.</td>
<td>No comfort measures used.</td>
</tr>
<tr>
<td>2 (Pre-pain)</td>
<td>No vaso-occlusive pain; pain of complications may be present; prodromal signs of impending vaso-occlusive episode may appear, eg, “yellow eyes” and/or fatigue.</td>
<td>No comfort measures used; caregivers may encourage child to increase fluids to prevent pain event from occurring.</td>
</tr>
<tr>
<td>3 (Pain start point)</td>
<td>First signs of vaso-occlusive pain appear, usually in mild form.</td>
<td>Mild oral analgesic often given; fluids increased; child usually maintains normal activities.</td>
</tr>
<tr>
<td>4 (Pain acceleration)</td>
<td>Intensity of pain increases from mild to moderate. Some children may skip this level or move quickly from phase 3 to phase 5.</td>
<td>Stronger oral analgesics are given; rubbing, heat, and distraction are often used; child usually stays in school until the pain becomes more severe; then stays home and limits activities; is usually in bed; family searches for ways to control the pain, often combining a number of methods; tries to calm and psychologically comfort child; some families use prayer.</td>
</tr>
<tr>
<td>5 (Peak pain experience)</td>
<td>Pain accelerates to high moderate or severe levels and plateaus; pain may remain elevated for extended periods of time. Child’s appearance, behavior, and mood are significantly different from “normal.”</td>
<td>Oral analgesics are given around the clock, at home; combination of comfort measures is used; family may avoid going to the hospital; if pain is so distressing to the child, is increasing despite all efforts, or the child develops a fever, the caregiver will take the child to the emergency department. After child enters the hospital, families often turn over comforting activities to health care providers and wait to see if the intravenous fluids and analgesics work. Child often sleeps from the sedative effects of the analgesics. Family caregivers often collapse from exhaustion of caring for the child for several days with little or no rest.</td>
</tr>
<tr>
<td>6 (Pain decrease start point)</td>
<td>Pain finally begins to decrease in intensity from the peak pain level.</td>
<td>Family caregivers again become active in comforting the child but not as intensely as during phases 4 and 5.</td>
</tr>
<tr>
<td>7 (Steady pain decline)</td>
<td>Pain decreases more rapidly, becoming more tolerable for the child. Child and family are more relaxed.</td>
<td>Health care providers begin to wean the child from the intravenous analgesics; oral opioids given; discharge planning is started. Children may be discharged before they are pain free.</td>
</tr>
<tr>
<td>8 (Pain resolution)</td>
<td>Pain intensity is at a tolerable level, and discharge is imminent. Child looks and acts like “normal” self. Mood improves.</td>
<td>May receive mild oral analgesics.</td>
</tr>
</tbody>
</table>
Some caregivers told their children if they manifested warning signs. Other caregivers would not tell their children, choosing instead to focus on wellness and normal functioning as long as possible. At times, caregivers were able to avert the pain, and the child would be spared the VOE. Responsibility on the part of the child for preventing the pain episodes was noted to increase with the age of the child. With younger children, caregivers gave more supervision to ensure that actions to prevent the pain event would be taken.

PHASE 3
The first sign of pain signaled the start of phase 3, the pain start point. The pain first appeared in mild form and gradually or rapidly increased. The pain was described as mild or “ache-ish” and was usually located in one specific area. The caregiver started to use more definitive actions in an attempt to prevent the escalation of pain. At this point, children were given mild analgesics, such as acetaminophen and/or ibuprofen. Often, they were encouraged to be as active as possible during pain-free times and to maintain a normal pattern of activities. The caregiver and child would monitor activities together to ensure that the child was not over-engaging. At school, the children would go to the nurse (if there was one) for rest or analgesics and could leave the classrooms for fluids or to go to the restroom. Mother L said, “. . . the least little thing I pay attention to it so I can determine, find out, start asking her questions about how she's feeling, or start giving her medications so it doesn’t get to the point that she is in uncontrollable pain.” Another mother reported, “I have him take a supply [of medicine] to school and that seems to work for him.”

PHASE 4
If the pain continued to increase, the pain/comfort cycle progressed into phase 4, the phase of pain acceleration. During this phase, stronger medications were used, usually preparations of acetaminophen with codeine. Decisions were often made about the child's attendance at school. Caregivers were often called to remove the patient from the school setting during this phase. Efforts to avert a peak pain experience were intensified. Caregivers became extremely vigilant of their child's condition. With the child at home, the caregiver either had to arrange for work coverage or for sick child care. This situation often created additional problems, such as losing work time and wages and/or inconveniencing a friend or family member.

As the pain intensified, the child and caregiver began to use numerous methods to relieve the pain, calm the child, and prevent the pain from becoming out of control. Children would often go to bed and try to “sleep off” the pain. They would engage in quiet activities in their rooms, listen to music or the radio, do homework, read, or watch television. Oral fluid intake would be increased. Codeine and acetaminophen often made the child tired and constipated.

Children would get or ask for a heating pad, take a long warm bath, or use other aquatic devices. A few of the children were taught and used cognitive-behavioral pain relief methods, such as relaxation and imagery. Caregivers would stay with the child talking and rubbing their backs or painful body part.

Caregivers closely monitored the child's needs, often through the night, frequently reassessing pain using a verbal report from the child. Pain assessments most often involved ratings of pain on a 0 to 10 scale. Children's needs differed in how much attention they required. The older and more mature children and adolescents performed much of their own care, obtained supplies, entertained themselves, limited their own activities, and monitored their own use of medications. They frequently validated these self-care interventions with the caregiver.

In discussing this phase, Mother B said, “We couldn’t live without the heating pad . . . and a warm bath with bubbles.” Mother E used multiple methods to comfort the child during this phase: “. . . but I still have to give him his medicine when I rub his back . . . he’ll calm down for a little while, then might fall into sleep.”

PHASE 5
The peak pain experience was reached when the pain was at moderate to high levels; maximal efforts were being made to reduce it; the child or adolescent clearly demonstrated behavioral, appearance, and mood changes because of the intensity of the pain; and the pain did not seem to abate. In this phase, children often were incapacitated and unable to relieve their own pain in the home setting. Pain descriptors used by children and adolescents in this phase were “stabbing,” “drilling,” “pounding,” “banging,” or “throbbing.” Caregivers used words like “excruciating” and “unbearable.” The child would indicate that “I can’t take it anymore.” The caregivers recognized that they were unable to help their children further, and altogether they would make the decision to seek emergency department help for stronger analgesic medications and protection from complications. Pain ratings at this time ranged from 6 to 10 on a 10-point scale.

Arrangements were made to take the child to the emergency department by driving, calling a cab, or requesting transportation from someone. Often, transportation arrangements took some time, prolonging the peak period of pain for the child. On arrival in the emergency department, an intravenous line and fluids were started. A bolus of intravenous analgesic would be given if the event had the appearance of being “routine.” If concerns were present about a complicating factor of SCD, such as acute chest syndrome, splenic sequestration, or cholelithiasis, then medication was withheld pending additional examination and testing. Often, the pain cycle could be “broken” and pain could be eliminated or brought to a manageable level with 1 or 2 boluses of intravenous medication. If the pain was not controlled with
When discussing the peak pain experience and the decision to seek professional help, one mother said, “I feel like I can control it at home . . . this [the hospital] is where I come when nothing else works, when her pain is too unbearable or . . . there’s no more I can do, no more pills . . . the fluids she’s getting are not holding her . . .”. Mother J said, “. . . when she gets to where she’s like a 9 [on a 0-10 scale] . . . she’s at the age where she’ll tell you, ‘It’s time to go, I can’t take it anymore’. . .”. Mother L indicated that “Most every time we come to the ER, I’ve already done all in my power to do before it gets to this point.” There was some indication that families avoided going to the hospital until quite late in the pain cycle. Both children and caregivers verbalized that they did not want to go unless they absolutely had to. Mother G, said “I stalled time. I try to wait and go along and see how long the pain is going to last . . . before we make a trip to the ER.”

Numerous children and caregivers indicated that many times the pain did not decrease until 2 or 3 days after admission to the hospital, thus prolonging the peak pain experience even further. Regarding his peak pain experience, on day 3 of hospitalization, H said, “It’s like it is throbbing . . . like somebody keeps stabbing a knife or something in my back and it’s been like that since the day I came in.” Mother A said, “I didn’t think anything was going to work from the day I brought her in . . . she was in excruciating pain, it was the worst I’ve ever seen it . . . and it took about 3 days to bring it down.”

PHASE 6

The pain decrease start point was signaled by a decline in the pain intensity of phase 5. Definite effects could be seen from the use of analgesics and intravenous fluids, and the pain began to resolve. Analgesics often sedated the children, and they often fell into a deep sleep. They often remained in bed in a flat position and slept for long periods of time. The caregiver often slept at home or at the hospital on a cot or in a chair. Pain levels had noticeably dropped from the time of the peak pain experience. J said that the pain “was a 9 when I came in, and . . . is about a 6 now. . . .” When awake, the children took more interest in their surroundings, roommates, and visitors. Irritability diminished.

Once the pain began to decrease, the family seemed to relax. Family caregivers looked more rested, and they would leave the children alone more often. Children were more easily engaged in games and conversation. Mother T stated, “I spent the first couple of nights with her . . . then she wasn’t scared or hurting so much, so I went home and got rest that next night.”

PHASE 7

This phase began when the pain showed a steady decline, either slowly or rapidly. The child might be taken into the tub room for a warm bath, watch television, or get involved in games with other children or hospital volunteers. Mobility improved, pain levels were reported as lower, and behaviors became more animated. Pain intensity was described as “just a little.” The family caregiver would resume using some of the comfort measures, such as rubbing and distraction. During this time, the caregiver felt even freer to leave the child, go back to work, or return home to care for other children. Hospital caregivers would wean the child quickly from the intravenous analgesic and give acetaminophen and codeine in preparation for hospital discharge.

PHASE 8

The phase of pain resolution was characterized by low pain scores and increasingly “normal” behavior, appearance, and mood for each individual child. Comfort techniques designed to alleviate the sensory experience of pain were less frequent and seemed less necessary as the child recovered from a VOE. Some children moved into phase 1 (baseline) before leaving the hospital, while others indicated low to moderate pain levels (phase 7 or 8). Analgesic use was discontinued or reduced to mild forms. All indications were that the pain cycle had been broken and that resolution had occurred or was in the process of occurring. Often, the child seemed more excited about life in general. During this phase, the caregiver and child attempted to recapture the lives they left before the pain event occurred.

LIMITATIONS

Limitations can be found in both the quantitative and qualitative methods used. No data can be generalized beyond the small, nonrepresentative, and nonrandom sample of this study, drawn from a single children’s hospital in the Midwest. Although pain and mood scores came from validated research tools and thus provided more rigorous and precise data, the scores were limited in scope and superficial when compared with the depth and richness of the qualitative findings. These scores were only obtained during brief but separate times of contact with participants: during and after VOE. Thus, quantitative data were only available for a very small part of the chronology, for several hours rather than several days. Although the qualitative methods provided a “rich” description of a chronology model of pain and comfort from the “insider” perspectives of the participants, it was developed primarily through the linguistic data of the ethnographic interviews. Thus, the model was derived inductively in the context of theory discovery. Verification of the chronology model can only occur if subsequent quantitative research methods yield findings from additional and larger samples that support the concepts, themes, and patterns of the model.

In addition, all children in this study had available caregivers who provided support. An available caregiver was a requirement for enrollment in the study. There were many children who did not meet this criterion and thus could not be included in the study. Their stories may have been very different, and a model developed from their data may have an entirely different configuration than the chronology found in this study (Figure).
tuous finding in this investigation. The model emerged through triangulation of findings from extensive qualitative interview data, participant observations, and a limited amount of quantitative pain intensity data. The chronology provides a tentative descriptive model to examine the pain and comfort experiences of the children and family participants in the sample throughout the entire VOE, not just at the discrete times that the researcher interacted with participants.

The chronology presented here is not a definitive, confirmed representation of the VOE experience of children with SCD and their families, but instead a tentative description of patterns revealed by most, but not all, subjects in this sample. These patterns require further empirical exploration, particularly through larger quantitative studies.

**QUESTIONS RAISED FOR FURTHER STUDY**

Three sets of questions can be asked about the treatment of pain in uncomplicated VOE based on the findings of this study. First, is it possible to treat the child with VOE earlier than usual, that is, in a clinic setting rather than in the emergency department of a hospital? According to Yang et al., adult and child patients in a managed care program that involved teaching, monitoring, and coaching patients with SCD on a regular basis had significantly fewer hospitalizations and emergency department visits than the patients with SCD who went through the emergency department for most of their SCD care. Of 194 patients seen in their 1989 study, 33.5% did not use the comprehensive health care clinic program; this 33.5% of the patient population accounted for 71% of the emergency department visits and 42% of the hospitalizations. In addition, future research should investigate further the validity of the phenomenon of the prodromal signs that many caregivers reported in the current study. These may provide first and early warning of impending VOE and may serve as an impetus for interventions to begin.

Second, if treated earlier, is it possible to avoid the VOE all together, avoid the peak pain experience, or reduce the duration or intensity of the typical VOE? To our knowledge, no literature has addressed aggressive treatment of the child with VOE in the home setting by family caregivers, advanced practice nurses, or home health staff. Could hospital admission be prevented or length of stay decreased? Would the cost of care also be significantly altered? According to estimates obtained from national discharge surveys conducted by the National Center for Health Statistics, the average length of stay for patients with SCD was 6.1 days and the cost was approximately $6300 per hospitalization based on data collected from 1989 to 1993. The cost of care to patients in their 1989 study, 33.5% did not use the comprehensive health care clinic program; this 33.5% of the patient population accounted for 71% of the emergency department visits and 42% of the hospitalizations. In addition, future research should investigate further the validity of the phenomenon of the prodromal signs that many caregivers reported in the current study. These may provide first and early warning of impending VOE and may serve as an impetus for interventions to begin.

Third, can the pain itself be prevented or reduced by earlier intervention in the pain cycle and would there be a positive effect on patient outcomes? For example, would there be fewer cases of acute chest syndrome, stroke, and avascular necrosis of the hips? Does the intensity, duration, or characteristics of the pain episode itself somehow set the child up for more complications and sequelae?

**CLINICAL QUESTIONS RAISED BY THE STUDY**

More immediate clinical issues that need to be addressed are those about the long duration and high intensity of the pain experienced by these children, the best ways to resolve this pain, and the demands the pain places on the family system. Unrelieved pain produces suffering, which is perhaps much longer in duration than is necessary. Suffering in turn may have a negative effect on the lives of each family member in terms of school, marital relationships, employment, self-esteem, achievement, and family function. It seems necessary to revise previously held beliefs about the pain experiences of children with SCD and to develop new ways to deal with this old problem. Should the emergency department be the only option for VOE, or might there be some benefit to the walk-in clinics or 24-hour facilities that have appeared in a few areas across the United States? Are there more pain relief resources that can be offered to families at home because, according to a study by Shapiro et al., almost 90% of the pain events were cared for at home? What factors maintain the pain at severe levels, sometimes for several days after hospital admission? Why do families come to the emergency department so late in the pain cycle?

**PARTIAL VERIFICATION OF THE CHRONOLOGY MODEL**

After the data from this study had been analyzed and a draft report was written of the findings, a comprehensive book on sickle cell pain was published by Ballas. In that volume was a description of a specific pattern of pain primarily in adults with SCD, including 4 specific phases, pathophysiological signs during each phase, and a heuristic model of the fluctuations in the pain. The Ballas phases were supported by previous literature and research findings.

The figure in the book by Ballas reflecting the “typical profile” of the painful episode in adults matches almost identically our Figure of the chronology model for children and adolescents. The pain pattern reflected by both figures included a prodromal phase in which there were signs of vaso-occlusion but no pain, a phase in which the pain intensity increased sharply, a long period in which the pain peaked and remained at the peak level for sev-
eral days, a period in which the pain decreased steadily, and then pain resolution in which the patient was pain-free once again. The similarity of the 2 independently generated models is surprising and provides support for the validity of each. Both included a prodromal phase in which changes occurred that warned the patient or family of an imminent VOE. However, there were differences in the nature of the prodromal signs between the 2 models.

The findings of this study, using primarily qualitative research methods, have lead to the serendipitous discovery of a model depicting a consistent pattern of pain in a sample of pediatric patients with SCD. This finding suggests a “trajectory” or chronology which, if validated, may serve to help describe or predict pain intensity in young people with SCD. It might also help suggest and test interventions to enhance pain control in certain phases. The descriptive model was interpreted conservatively as a finding that cannot be generalized but instead must be examined through additional research, particularly larger quantitative studies. The identification of a similar model of pain in the adult SCD population provided partial support for the validity of the chronology for children and adolescents.

Many questions can be raised about the current pain management practices for children and adults with SCD in light of the patterns of pain revealed. More research is needed to uncover the patterns, factors affecting these patterns, and how sickle cell pain is viewed by caregivers. Can the pattern be modified by changes in pain management? Can peaks in and duration of pain be reduced? Would changing the configuration of the pain pattern change life-altering complications and sequelae? More attention must be paid to the pain experiences of the patient and family from their own perceptions to answer these many questions. Massive attention is currently focused on pharmacological and genetic “cures.” While cures are sought, increased effort directed at prevention and palliation of the pain experience itself may do much for the children and families who live with SCD every day.

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