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Barriers to Care and Quality of Primary Care Services in Children with Sickle Cell Disease

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Abstract

Aims.—To 1): identify barriers to care in children with sickle cell disease; 2) examine the quality of primary care services received by these children; and 3) examine the relationship between barriers to care and quality of primary care services in children with sickle cell disease.

Background.—Effective management in children with sickle cell disease requires early access to a comprehensive range of preventive screenings, urgent care treatments for vaso-occlusive pain crisis and ongoing prophylactic treatments.

Design.—A cross-sectional survey of parents of children with sickle cell disease was conducted between April - September 2011.

Methods.—Parents of children with sickle cell disease completed the Barriers to Care Questionnaire and Parent's Perceptions of Primary Care.

Results.—Parents of children with sickle cell disease (n=38) reported health system barriers such as inability to contact doctors or clinics, extended wait times and inconvenient clinic hours. Some barriers were reported more frequently among children with concurrent sickle cell disease and asthma, compared with those children without a concurrent asthma condition. Parents who reported more barriers were least likely to perceive their care as accessible, comprehensive and coordinated.

Conclusions: Minimizing healthcare barriers may improve the quality of primary care services received by children with sickle cell disease and consequently prevent complications associated with sickle cell disease.

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- 1) substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data;
- 2) drafting the article or revising it critically for important intellectual content.

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Implications for Nursing Practice: Nurses and other care providers need to identify healthcare barriers, so that access, coordination, comprehensiveness and overall quality of primary care services may be improved in children with sickle cell disease.

Keywords

Sickle cell disease; barriers to care; quality of primary care; nursing; children

INTRODUCTION

Sickle cell disease (SCD) is an inherited blood disorder characterized by recurring episodes of vascular occlusion and inflammation (Rees *et al.* 2010). Children with SCD experience episodes of extreme pain and are at-risk for a wide range of medical complications such as asthma, acute chest syndrome, kidney disease and cerebrovascular disease (Modell & Darlison 2008). SCD is one of the most common genetic disorders in the world, primarily affecting populations of African descent (Modell & Darlison 2008, Piel *et al.* 2013). As many as 25% of inhabitants of Central and West Africa have the sickle cell trait and nearly 1–2% of newborns in these countries experience some form of the disease (Diallo & Guindo 2014).

Worldwide, approximately 3.5% of deaths of children under the age of 5 can be attributed to SCD (Makani *et al.* 2011, Piel *et al.* 2013). Life expectancy has increased for children with SCD, particularly in developed nations, as a result of access to quality healthcare.

Furthermore, early diagnosis of this disease through newborn screenings and the use of prophylactic treatments have helped to dramatically increase life expectancy and minimize preventable complications for children with SCD. Despite these advances, the healthcare needs of those with SCD are still chronic and there are wide variations in the quality of primary care services received by individuals with SCD. Thus, not every child has access to such high quality healthcare and, as a result, may experience a shorter life expectancy and lower quality of life (Makani *et al.* 2011, Piel *et al.* 2013).

Children with SCD in the United States have healthcare needs that frequently require use of outpatient and inpatient services for vaso-occlusive pain crises and other disease related complications (Boulet *et al.* 2010, Dale *et al.* 2011). Effective SCD management requires early access to a comprehensive range of preventive screenings, urgent care treatments for vaso-occlusive pain crisis and ongoing prophylactic treatments such as hydroxyurea or blood transfusions (Grosse *et al.* 2009, Kanter & Kruse-Jarres 2013, Leschke *et al.* 2012). While it is clear that children with SCD have ongoing healthcare needs, recent evidence suggests that not all children with SCD are receiving optimal primary care services (Liem *et al.* 2014, Raphael *et al.* 2008).

Using data from a large national survey, Raphael and colleagues (2013) estimated that only two-thirds of children with SCD receive comprehensive care and only 20% experience effective care coordination between their primary care and specialist providers. In another study, Boulet and colleagues (2010) found that children with SCD experienced more limited access to care (e.g. extended wait times, problems with transportation and difficulties in reaching providers over the telephone) compared with children without SCD. Limited access

to high quality primary care services may contribute to more severe SCD complications (Jacob *et al.* 2005) and subsequent hospitalizations (Bundy *et al.* 2012, Fosdal & Wojner-Alexandrov, 2007, Leschke *et al.* 2012, Raphael *et al.* 2013). Given these identified gaps in access to primary care services, it is important to determine if there are particular healthcare barriers that cluster in SCD populations and prevent optimal treatment in the primary care setting.

Background

‘Barriers to care’ refers to difficulties in accessing a clinic or provider (i.e. not having regular provider, lack of health insurance coverage) as well as processes of care that prevent patients from receiving needed treatments, such as perceived maltreatment in the clinic setting and difficulties with provider knowledge or communication (Valenzuela *et al.* 2014). Prior studies of healthcare barriers have tended to focus on populations of children with special healthcare needs and those with specific conditions such as persistent asthma or diabetes (Seid *et al.* 2009, Seid *et al.* 2004, Valenzuela *et al.* 2014). Some of the barriers that are associated with poor quality care for children with special healthcare needs include insurance restrictions, conflicting expectations or beliefs about the delivery of services, lack of skills to navigate a complex healthcare system and maltreatment or marginalization from healthcare providers. These barriers interfere with the care-seeking process and prevent children with special healthcare needs from receiving high quality services (Newacheck *et al.* 2000, Seid *et al.* 2004). When children with special healthcare needs encounter barriers to care, they report more negative perceptions of primary care and poorer health-related quality of life (Seid 2008, Seid *et al.* 2009).

Crosby and colleagues (2012) asked parents of children with SCD about barriers to attending clinic appointments. Parents frequently reported logistical barriers, such as transportation difficulties and inability to take time off of work as barriers to attending clinic appointments (Crosby *et al.* 2012). In another study, Bollinger *et al.* (2011) found that caregivers’ lack of knowledge was a greater barrier to children’s receipt of transcranial doppler screenings, than practical barriers such as financial barriers, transportation and clinic hours.

Parents of children with SCD may encounter different barriers than parents of non-SCD children such as children with asthma (Seid *et al.* 2008) and other children with special health care needs (Seid *et al.* 2004) as they navigate through the healthcare system. More research is needed to consider how healthcare barriers may contribute to previously identified gaps in primary care services for children with SCD. Although some research has been conducted about barriers to clinic appointments for children with SCD (Crosby *et al.* 2009), there was no information about the quality of primary care services and the relationship between barriers to care and the quality of primary care services in children with SCD.

THE STUDY

Aims

The specific aims of the study were to: 1) identify barriers to care in children with SCD; 2) examine the quality of primary care services received by children with SCD; and 3) examine the relationship between barriers to care and quality of primary care services in children with sickle cell disease (SCD).

Design

We conducted a cross-sectional study to examine barriers to care and quality of primary care services provided for children with SCD. Parents of children with SCD completed questionnaires related to barriers to care (Seid *et al.* 2004, Seid 2008; Seid *et al.* 2009) and primary care experiences (Seid *et al.* 2001, Seid *et al.* 2003).

Participants

Parents of children with SCD were recruited from a community based organization (CBO) in Southern California that provides educational workshops and social service activities for individuals with SCD. The CBO offers services to an estimated 5,000 children and adults with SCD. Most of the CBO's clients reside in a greater metropolitan area of Southern California spanning approximately 75 miles. The population served consists of more than ninety five percent African-American and less than five percent Latino and other ethnic groups. Parents of children with SCD were approached by a research associate as they waited for their children who were participating in the programs offered by the CBO.

Parents were included if they were: 1) able to read and write in English (as questionnaires were available only in English); and 2) willing to complete the questionnaires. They were excluded if they: 1) had physical or cognitive impairments that precluded completion of the questionnaires; 2) did not have time to complete the questionnaires; and/or 3) they were not willing to participate. A convenience sample was used and sample size was determined by the number of parents that were available during the data collection period (April - September, 2011) on the Saturdays when children with SCD were participating in the programs offered by the CBO. A sample size of 38 parents would allow a prevalence estimate that more than 50% (with a 95% confidence interval of $\pm 5\%$) of the parents would report barriers to care.

Data Collection

Parents were asked to complete questionnaires that inquired about their child's healthcare experiences (The Barriers to Care Questionnaire and The Parent Perceptions of Primary Care Questionnaire). Parents also completed a demographic questionnaire that included: 1) age of children with SCD; 2) gender of children with SCD; 3) SCD genotype; 4) number of times child was hospitalized for SCD-related conditions the previous 12 months; and 5) history of common SCD related conditions (e.g. acute chest syndrome, splenic sequestration, cerebrovascular accident, etc.). In the event that parents had limited reading ability, a protocol was established for the research associate to read the study measures to parents.

None of the parents in the study required such protocol. Parents were given a \$30 gift card after completion of the study measures.

Barriers to Care Questionnaire (BCQ).—The Barriers to Care Questionnaire (BCQ) has previously been used for assessment of barriers to care for children with asthma and other special healthcare needs (Seid *et al.* 2009, Seid *et al.* 2004). It consists of 39 items that measure specific healthcare barriers to care in five domains: 1) pragmatic barriers, such as logistic and cost factors that may prevent or delay care use (i.e. ‘having to wait too long in the waiting room’); 2) skill barriers, including deficits in acquired or learned proficiency to navigate the healthcare system (i.e. ‘getting referrals to specialists’); 3) expectation barriers, such as pessimistic care outlooks (i.e. ‘worrying that doctors and nurses will not do what is right for your child’); 4) marginalization barriers, including negative experiences in the healthcare system (i.e. ‘impatient doctors’); and 5) knowledge and belief barriers which included circumstances where the patient knowledge differs from that of the healthcare provider (i.e. ‘disagreeing with the doctor’s orders’).

Each item on the BCQ was preceded by ‘how often was each of the following barriers a problem in the past 3 months when trying to get care for your child’. Parents rated each item on a Likert scale with responses as: ‘no problem’ [scored as 100], ‘almost never’ [scored as 75], ‘sometimes’ [scored as 50], ‘often’ [scored as 25], or ‘almost always’ [scored as 0]. These scoring procedures were used previously (Seid, et al. 2009) and were computed to generate a continuous variable ranging from 0–100. Higher scores indicated more positive healthcare experiences (i.e. fewer barriers to care).

Parent Perceptions of Primary Care (P3C).—The Parent Perceptions of Primary Care (P3C) is a measure of six aspects of pediatric primary care which corresponded to the Institute of Medicine’s definition of high quality care (Institute of Medicine 1996, Seid *et al.* 2003). The six dimensions were continuity, access, contextual knowledge, communication, comprehensiveness and coordination. In the P3C, ‘continuity’ represented the length of time that parents brought their child to the same provider. ‘Access’ referred to parent’s ability to receive timely care. The access dimension included items such as ‘If your child is sick, can you see the doctor within one day?’). The third dimension, ‘contextual knowledge’, referred to parent’s perceptions of how well the provider knows the parent and child. An example of a contextual knowledge item was, ‘Do you feel the doctor knows your child’s medical history?’ ‘Communication’ referred to parent-provider communications. For example, the P3C item included ‘Do you feel comfortable asking the doctor questions?’ The fifth dimension, ‘comprehensiveness’, included items related to the breadth of care provided by the child’s provider, such as, ‘Does the doctor talk to you about your child’s growth?’ Last, ‘coordination’ indicated items related to the provider’s ability to refer and coordinate care with others. For example, ‘When necessary, can the doctor arrange for other health care for your child?’

For each of the P3C items, parents were asked how frequently they experienced each indicator of high quality care. The questions were asked on a Likert scale and the response choices were ‘never’ [scored as 0], ‘sometimes’ [scored as 25], ‘often’ [scored as 50], ‘almost always’ [scored as 75], or ‘always’ [scored as 100]. The items for the six quality

dimensions (continuity, access, contextual knowledge, communication, comprehensiveness and coordination) were scored and computed to produce a range from 0–100, with higher scores indicating more positive primary care experiences (Sobo et al. 2006).

Ethical Considerations

A research associate explained the details of the study in terms the parents could understand and answered their questions or concerns. When the parents agreed to participate, the parent signed the consent form. All parents who were approached participated; no parents refused. All study procedures were approved by the affiliated university Institutional Review Board.

Data Analysis

Data were entered by two research assistants and checked for accuracy. All analyses were conducted using Stata version 10.0. Descriptive statistics (means, standard deviations, frequencies) were used to describe barriers to care (pragmatic, skills, expectations, marginalizations, knowledge and beliefs). Descriptive statistics (means, standard deviations, frequencies) were also used to describe the quality of primary care services (continuity, access, coordination, communication, comprehensiveness and contextual knowledge). Pearson's correlation coefficients were calculated to examine the relationship between barriers to care and primary care experiences among parents of children with SCD.

Validity and Reliability

The BCQ was previously assessed for feasibility, reliability and validity in a diverse and multiethnic sample of children with asthma (Seid *et al.* 2009) and children with special healthcare needs (Seid, *et al.* 2004). Feasibility was evidenced by a low percentage of missing items. Reliability was indicated by high internal consistency ranging from 0.75 to 0.95 for each of the BCQ subscales (Seid *et al.* 2004, Seid *et al.* 2001). The BCQ subscales completed by parents of children with SCD in this study exhibited high internal consistency ($\alpha = 0.85\text{--}0.97$), indicating that the scales were reliable and acceptable for between-group comparisons.

The P3C has previously been assessed for feasibility, reliability and validity (Seid *et al.* 2003). Prior studies have demonstrated feasibility by revealing a low percentage of missing values overall and for parents with low education (Seid *et al.* 2001). In addition, the P3C has previously been reported to have high internal consistency with α ranging from 0.90 to 0.95 in a study of children with special healthcare needs (CSHCN) varying in racial/ethnic heritage and primary language (Seid *et al.* 2003). Previous studies also demonstrated validity of the P3C measure by showing that P3C scores were higher for CSHCN with health insurance compared with those without (Seid *et al.* 2003). Thus, the P3C has been determined to be culturally appropriate for use in our multi-ethnic sample of children with SCD. In the current study, less than one percent of the P3C items completed by parents of children with SCD had a missing value. Moreover, each of the P3C subscales reported by parents of children with SCD exhibited moderate to high internal consistency ($\alpha = 0.70\text{--}0.95$).

RESULTS

During the data collection period (April - September 2011), 38 parents were available who received information and all parents participated; no parents were excluded. Parents were mostly female (76%), middle-aged (76% below age 50 years) and cared for one or two children with SCD at home (64%). Nearly all parents reported having insurance coverage: 1) 53% reported a private insurance carrier, 2) 39% had public insurance; and 3) 8% were uninsured or unsure of their insurance status at the time of data collection. The gender, age and hemoglobin genotypes of their children with SCD are presented in Table 1.

Approximately half of their children carried the HgbSS genotype, which is often a more severe form of sickle cell disease (Rees, *et al.* 2010). The majority (71%) of their children experienced at least one pain crisis every year requiring hospitalization and most (87%) had history of medical conditions typically observed in SCD. The most frequently reported conditions were acute chest syndrome (58%), asthma (26%) and other SCD-related conditions (26%).

Barriers to Care in Children with SCD

The majority of healthcare barriers were reported as occurring ‘never’ or ‘almost never,’ resulting in high BCQ scores. The average total BCQ was 79.2 (SD 19.7). There were variations in average ratings across the healthcare barriers subscales. Parents of children with SCD had the highest scores in the domain of Knowledge and Beliefs with a mean of 84.5 (SD 21.4) and Marginalization with a mean of 82.2 (SD 18.5), suggesting that parents had positive experiences in these domains. Parents of children with SCD had the lowest scores in the domain of Pragmatics with a mean of 73.8 (SD 23.3), suggesting there were some difficulties in accessing clinic services due to transportation, wait times and competing family priorities. The Skills domain and Expectations domain had scores in the middle range with means of 79.3 (SD 23.0) and 78.3 (SD 21.4), respectively), suggesting that parents had both positive and negative experiences in these domains.

Approximately one third to one half of parents reported that they ‘often’ or ‘almost always’ experienced difficulties in ‘having to take time off work,’ ‘having to wait too long in the waiting room,’ ‘getting hold of the doctor’s office or clinic by telephone’ and ‘meeting the needs of other family members’ (Table 2). In addition to these pragmatic barriers, approximately one third of parents reported that they experienced barriers in navigation skills, such as ‘knowing how to make the healthcare system work for you,’ ‘getting referrals to specialists’ and ‘needing to be more savvy or knowledgeable about getting health care’ (Table 2). Additionally, approximately one third of parents reported ‘lack of communication between different parts of the healthcare system,’ ‘feeling like the healthcare system is trying to give as little service as possible,’ ‘getting the doctor to listen to you’ and ‘getting your questions answered’ (Table 2).

We explored associations between demographic characteristics (age, gender, disease severity, medical conditions) and BCQ scores to determine if there are particular subpopulations of children with SCD who may be at-risk of experiencing more barriers to care. Parents reported that children with concurrent asthma (n=10) often experienced more

barriers to care compared with children without concurrent asthma (n=28). Independent t-tests showed a significant difference between children with SCD without concurrent asthma and children with SCD with concurrent asthma who experienced greater barriers to care on the dimensions of 1) skills -- 87.8 (SD 16.9) vs 71.6 (SD 22.9), $t(36)=2.9$, $p=0.004$, 2) expectations-- 87.9 (SD 17.4) vs 66.8 (SD 23.0), $t(36)=3.7$, $p<0.0003$, 3) marginalization -- 88.9 (SD 16.9) vs 75.3 (SD 17.5) $t(36)=2.5$, $p=0.012$ and 4) knowledge and beliefs -- 91.1 (SD 19.9) vs 75.6 (SD 19.2), $t(36)=2.4$, $p=0.017$ (Figure 1). Specifically, parents of children with SCD and concurrent asthma reported more barriers in skills to navigate the healthcare system (e.g. 'Knowing how to make the healthcare system work for you'), divergent expectations (e.g. 'Worrying that doctors and nurses will not do what is right for your child'), marginalization in the healthcare setting (e.g. 'Being judged on your appearance, your ancestry, or your accent') and discordant knowledge and beliefs ('Disagreeing with the doctor's orders'). None of the other measured demographic variables (age, gender and disease severity) were associated with barriers to care.

Quality of Primary Care Services in Children with SCD

The overall P3C total resulted in a mean of 79.1 (SD 20.0) and included continuity, access, coordination, communication, comprehensiveness and contextual knowledge domains. Parents frequently reported that their children had received care in the same setting and with the same provider for five or more years. This resulted in reported high scores with mean of 90.1 (SD 20.5) on the continuity dimension of the P3C. Communication with the healthcare provider also resulted in reported high mean of 86.2 (SD 20.9), suggesting that parents are satisfied with their provider interactions. However, there were difficulties reported in the dimensions of access, comprehensiveness and coordination of services with a mean of 70.1 (SD 28.4), 75.1 (SD 23.8) and 76.9 (SD 29.9), respectively. Approximately 25% of parents expressed difficulties with the following: 'is it easy for you to travel to the doctor,' 'can you see the doctor within one day,' 'can you get help on evenings/weekends,' and 'can you see the doctor as soon as you want for routine care for your child' (Table 3). The contextual knowledge domain had a mean of 81.1 (SD 26.58), suggesting that parents have both positive and negative experiences.

Relationships between Barriers to Care (BCQ) and Quality of Primary Care Services (PC3).

Children who reported more healthcare barriers (lower BCQ scores) were less likely to receive care consistent with standards of high quality (lower PC3 scores) in the primary care setting, Pearson $r = 0.64$, $p < 0.01$. Overall, the inter-correlations between the BCQ and P3C subscales were of moderate strength and statistically significant (Table 4). Pragmatic barriers to care, which were most frequently reported for children with SCD, were highly correlated with parental perceptions of access and comprehensiveness of services (Pearson $r = 0.67$ and 0.57 , respectively). Other barriers related to services comprehensiveness, while less frequent, were experiences of marginalization and misaligned healthcare expectations (Pearson $r = 0.64$ and 0.57 , respectively).

DISCUSSION

Parents of children with SCD experienced a range of healthcare barriers including pragmatic barriers in accessing appointments, barriers in skills to navigate the healthcare system, gaps in healthcare expectations, negative experiences with healthcare providers and circumstances where patient knowledge differed from that of the healthcare provider. Parents of children with SCD reported more barriers to care when compared with barriers reported for children with other chronic conditions such as asthma (Seid 2008) and children with special healthcare needs (Seid *et al.* 2004). Importantly, barriers to care were significantly correlated with gaps in primary care, such as comprehensiveness of services that have been previously identified as an area in need of improvement for children with SCD (Raphael *et al.* 2013).

While barriers can occur at many stages, parents of children with SCD most frequently reported health system-related barriers such as difficulties in getting in contact with the doctor or clinic, extended wait times and inconvenient clinic hours. Parents of children with SCD often had favorable experiences with healthcare providers once access was obtained. Findings from this study therefore suggest that inconveniences in appointment scheduling and visit duration may help to explain why some children with SCD experience significant delays in help-seeking for SCD-related symptoms (Jacob *et al.* 2005). Because treatment delays have such negative consequences for SCD complications and hospitalizations, our study findings indicate that expanding clinic hours to include evening or weekend services may be one strategy to improve care for children with SCD. Moreover, because expanded access is a central goal of the Patient Centered Medical Home (Raphael *et al.* 2009), there is a compelling need to determine whether the Medical Home is an effective model of primary care services delivery to improve the health services experiences and health outcomes of children with SCD.

It is interesting to note that children with concurrent SCD and asthma experienced even more barriers to care than children with SCD alone. A growing body of literature suggests that poor management of respiratory symptoms may be associated with more frequent pain episodes among children with SCD (Glassberg *et al.* 2006, Jacob *et al.* 2010). Reducing healthcare barriers that interfere with the management of respiratory symptoms may be one way to improve health outcomes in this vulnerable population. Additional research is needed to determine whether implementing practice guidelines and quality measures for asthma symptom management are effective strategies to improve the delivery of primary care services and to achieve better health outcomes for children with SCD-related respiratory symptoms (Crabtree *et al.* 2011, Wang *et al.* 2011).

Although we found significant differences in barriers experienced by children with SCD compared with children with asthma, there were no significant differences in the average reporting of healthcare barriers in our sample of children with SCD compared with a larger sample of children with special healthcare needs (Seid *et al.* 2004). It may be the case that children with SCD and other children with special healthcare needs recruited from specialized settings in Southern California experience similar systemic barriers to care. Findings of this study indicate a need to develop policies and programs that reduce service barriers for vulnerable populations with special healthcare needs overall, particularly for

those with SCD. For example the use of satellite Hematology clinics with extended hours, telehealth hematology services and/or mobile hematology health vehicles (Riebschleger *et al.* 2014) offering comprehensive healthcare services for patients who live outside major medical centers may increase access to care for those having difficulties with transportation. The use of smartphone technology (Jacob *et al.* 2012) and wireless intervention programs (Jacob *et al.* 2013a, McClellan *et al.* 2009) may allow remote monitoring of pain and symptoms (Jacob *et al.* 2013a) and facilitate screening, education, consultations and communications between parents and primary care providers (Jacob *et al.* 2013b). Use of services by nurse navigators with expertise in hematology may minimize delays in treatments and improve coordination of primary care services (Wagner *et al.* 2014). Mass counseling sessions may also improve parent's knowledge, attitudes and health-seeking and preventive behaviors (Kulkami & Kulkami 2014).

Our findings build off others which demonstrates significant gaps in primary care services for children with SCD (Liem *et al.* 2014, Raphael *et al.* 2008). We found that when children with SCD experience more healthcare barriers (such as difficulties in access or marginalization in the healthcare system), they are afforded fewer opportunities to receive high quality care that is accessible, comprehensive and well-coordinated. Comprehensive approaches to treatment and effective care coordination are needed to effectively manage pain symptoms and to prevent recurring hospitalizations for SCD-related conditions (Ballas 2011). Findings from our study therefore indicate that interventions are particularly needed to minimize access-related barriers and to improve the comprehensiveness and coordination of services for children with SCD.

The expanded use of wireless technologies and electronic medical records present an opportunity to enhance provider communication and services coordination for children with SCD (Jacob *et al.* 2013a, Jacob *et al.* 2013b). Our findings suggest that interventions to enhance provider communication and services coordination will be particularly critical for SCD populations with co-occurring conditions, particularly for those children with SCD who also have concurrent asthma. In the changing landscape of healthcare reform, patient-centered medical homes should also be evaluated for their potential as integrated models of primary care services delivery to improve the comprehensiveness and coordination of services for children with SCD (Raphael *et al.* 2013a). A patient centered medical home represents a collaborative partnership between the child, family and a primary provider, where the child and family are the focal point. It is an important mechanism for uniting the many segments of a child's care and emphasizing an ongoing relationship with a personal primary care provider trained to provide continuous and comprehensive care. The primary care provider leads a team, collectively taking responsibility for the coordinated and integrated care across all elements of the complex health care system (e.g., preventive care in hematology clinics, acute care in hospitals and chronic care in the community involving schools and other community-based services). Care is facilitated by information technology to assure that children get the indicated care when and where they need in a culturally and linguistically appropriate manner (Raphael *et al.* 2013a; Raphael *et al.* 2013b).

Limitations

The results of our study should be considered in light of several limitations. First, this study included a convenience sample of children living in Southern California who participated in Saturday activities offered by the community based organization. Results from this study may not be generalizable to other populations of children with SCD, such as those with limited English proficiency or those living in other geographic regions of the U.S and other areas in the world. This study should be replicated with nationally- and internationally-representative samples of children to provide a more complete picture of healthcare experiences for children with SCD within and outside of the US.

As a second limitation, this sample size was small restricting our power to detect statistical effects that otherwise might have been observed. For example, only 10 children in this study had concurrent SCD and asthma conditions. It is not possible to draw conclusions about the high rates of healthcare barriers observed in this vulnerable sub-population. Future studies would benefit from a larger sample size to describe processes of care for children with SCD and asthma.

Third, the study did not control for potentially confounding factors which may explain the relationship between healthcare barriers and perceived quality of primary care services for children with SCD. Parent's perceptions of healthcare quality may be influenced by several factors including healthcare literacy, attitudes and preferences and healthcare needs. While the objective of this study was to identify healthcare barriers for children with SCD, subsequent research studies would benefit from use of a health systems framework, such as the Behavioral Model of Healthcare Use for Vulnerable Populations (Gelberg *et al.* 2000, Stein *et al.* 2007), to further elaborate the relationship between access and health service outcomes for children with SCD.

Fourth, the questionnaires were available only in English. Therefore, non-English speaking parents may have additional barriers that were not addressed in this study. In addition, we did not have parents that require additional assistance with reading the questionnaires. However, there may be parents whose literacy levels are lower than the parents in our sample and may have additional barriers that were not addressed in this study. Future studies are needed to examine language and cultural barriers in non-English speaking parents and additional barriers in those with low literacy levels.

Finally, because the study design was cross-sectional, it was not possible to establish a temporal relationship between barriers to care and perceived primary care experiences. Prospective studies are warranted, particularly those that include direct process measures, to enhance our knowledge regarding healthcare barriers and how these barriers influence the delivery of high quality healthcare for children with SCD.

CONCLUSIONS

Our study identified specific barriers to care, such as inconvenient clinic hours and lengthy wait times for services, which may be targeted to improve access to care for children with SCD. In addition to these healthcare barriers, we found evidence of gaps in the

comprehensiveness and coordination of services for children with SCD in the primary care setting. Eliminating healthcare barriers for children with SCD is critically needed to improve the delivery of pediatric services and to enable this vulnerable and high-risk population to achieve optimal health outcomes. Additional research is warranted to determine whether specific interventions, such as expanding clinic hours, employing practice guidelines to improve the management of respiratory and pain symptoms and expanding the availability of integrative delivery models are effective strategies in preventing frequent hospitalizations among children with SCD.

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SUMMARY STATEMENT**Why is this research needed?**

- Children with sickle cell disease have ongoing healthcare needs and often receive inadequate primary care services.
- Barriers to care may consequently lead to preventable complications and costly emergency room visits and hospitalizations in children with sickle cell disease.
- Identifying specific healthcare barriers may improve access, coordination, comprehensiveness and overall quality of primary care services in children with sickle cell disease.

What are the key findings?

- Parents reported lack of skills in navigating the healthcare system, gaps in healthcare expectations, negative experiences with providers and general lack of knowledge about disease management.
- Children with sickle cell disease and concurrent asthma had more barriers to care and lower quality of primary care services compared with children with sickle cell disease only.
- Pragmatic healthcare barriers (transportation access, communication with doctors) were associated with lower quality of primary care services (access and comprehensiveness of services).

How should the findings be used to influence policy/practice/education?

- Findings support the need to develop and test strategies that minimize access-related barriers to care for children with sickle cell disease.
- Use of patient-centered medical homes may improve access to comprehensive, well-coordinated and improved quality of primary care services for children with sickle cell disease.
- The expanded use of wireless technologies and electronic medical records present an opportunity to enhance provider communication and coordination of services for children with sickle cell disease.

Figure 1.
Barriers to Care in children with SCD only, and SCD with concurrent asthma.

Table 1:

Demographics of Children with Sickle Cell Disease (N=38)

	N (%)
Age	
10–13 years	23 (61%)
14–17 years	15 (39%)
Gender	
Male	19 (50%)
Female	19 (50%)
Ethnicity	
African American	32 (84%)
Other	6 (16%)
Sickle Cell Genotype	
HgbSS	17 (45%)
HgbSC	12 (32%)
Other	9 (24%)
Pain Crises Requiring Hospitalization in Past Year	
None	11 (29.0%)
1 to 2 times/year	18 (47.7%)
3 times/year	9 (23.7%)
History of Medical Complications	
None	5 (13%)
1 to 2	26 (69%)
3	7 (18%)

Table 2:**Barriers To Care in Children with Sickle Cell Disease (N=38)**

Barriers To Care Questionnaire Items	N (%)
Pragmatics	
1. Access/transportation to the doctor's office	7 (18%)
2. Ease of communication with the doctor's office or clinic by telephone	12 (32%)
3. Ease of getting an appointment in a timely manner	10 (26%)
4. Access to care after hours or on the weekends	11 (19%)
5. Needing to take care of household responsibilities	14 (37%)
6. Needing to take time off work	19 (50%)
7. Long wait times in the waiting room	16 (42%)
8. Needing to the needs of other family members	18 (47%)
9. The cost of health care	11 (19%)
Skills	
1. Understanding of how to make the health care system work for you	14 (37%)
2. Doctors or nurses not fluent in primary language	4 (11%)
3. Doctors or nurses who speak in a way that is too technical or medical	7 (18%)
4. Obtaining referrals to specialists	12 (32%)
5. Understanding doctor's orders	6 (16%)
6. Access to enough information about how the health care system works	11 (29%)
7. Needing to be more "savvy" or knowledgeable about getting health care	13 (34%)
8. Receiving enough help with paperwork or forms	9 (24%)
Expectations	
1. Offices and staff that are not child-friendly	8 (21%)
2. Mistakes made by doctors or nurses.	6 (16%)
3. Worrying that doctors and nurses will not do what is right for your child	10 (26%)
4. Doctors treating the symptom without finding out the cause of the illness	9 (24%)
5. Getting a thorough examination.	9 (24%)
6. Lack of communication between child's provider and others in health care	8 (21%)
7. Lack of communication between different parts of the health care system	16 (42%)
Marginalization	
1. Feeling like providers are trying to give as little service as possible	9 (24%)
2. Feeling like the health care system is giving as little service as possible	14 (37%)
3. Impatient providers	5 (13%)
4. Intimidating providers	6 (16%)
5. Rude office staff.	6 (16%)
6. Uncaring office staff	4 (11%)
7. Provider listens to parent/child effectively	12 (32%)
8. Questions are answered	12 (32%)
9. Not knowing what to expect from one visit to the next	10 (26%)
10. Judgment based on appearance, ancestry, or accent	6 (16%)
11. Providers rushing parent/child through the visit	6 (16%)

Barriers To Care Questionnaire Items	N (%)
Knowledge and Beliefs	
1. Disagreeing with the provider's orders.	9 (24%)
2. Doctors not believing in home or traditional remedies.	6 (16%)
3. Providers giving instructions that seem wrong.	5 (13%)
4. Doctors or nurses that have different ideas about health than patient/parent	6 (16%)

^{**} Number and percent of parent's response to "how often were the following barriers a problem in the past 3 months when trying to get care for your child" as "often" or "almost always".

Table 3:

Quality of Primary Care Services in Children with Sickle Cell Disease (N=38)

Parents Perceptions Of Primary Care (P3C) Items	N (%)**
Access	
1. Ease of traveling to the doctor	26 (68%)
2. Timeliness of routine care (checkups, physicals) for child	24 (63%)
3. Availability of same day appointments when child is sick	24 (63%)
4. Accessibility of help or advice on evenings or weekends	26 (68%)
Contextual Knowledge	
1. Provider's knowledge of child's medical history	31 (82%)
2. Provider's knowledge of parent's concerns about child	28 (74%)
3. Provider's knowledge of parent/child's values and beliefs about health	25 (66%)
4. Provider's knowledge of child overall	29 (76%)
Communication	
1. Comfort in asking the provider questions	33 (87%)
2. Satisfaction with the provider's explanation of things	31 (82%)
3. Satisfaction with the amount of time spent with parent and child	30 (79%)
4. Provider listens to parent/child effectively	32 (84%)
Comprehensiveness	
1. Provider's ability to treat almost any problem child might have	30 (79%)
2. Provider discusses ways to keep child healthy	7 (18%)
3. Provider discusses child safety safety (car seats, seat belts, bike helmets, accidents, etc.)	19 (50%)
4. Provider discusses child's growth	25 (66%)
5. Provider discusses child's behavior in general (having friends, citizenship at school, etc.)	29 (76%)
Coordination	
1. Provider's ability to arrange other health care for child when necessary	31 (82%)
2. Provider follows up on visits to other health care providers when necessary	30 (79%)
3. Provider communicates with other health providers about child's health when necessary	30 (79%)
4. Provider and child's school work together for child's health when necessary	23 (61%)

**

Number and percent of parents response to "how frequently they experienced each indicator of high quality care" as "almost always" or "always"

Table 4:

Correlations between Barriers to Care and Perceptions of Primary Care in Children with SCD (N=38)

Barriers To Care	Parent's Perceptions of Primary Care (P3C)						
	Total P3C	Continuity	Access	Coordination	Communication	Comprehensiveness	Knowledge
Total BCQ							
r	.64	.54	.64	.34	.48	.58	.52
p	.00001	.0005	.00001	.037	.002	.00001	.0009
Pragmatics							
r	.64	.41	.67	.36	.49	.57	.51
p	.00001	.010	.00001	.025	.002	.0002	.001
Skills							
r	.43	.46	.46	.16	.34	.37	.35
p	.007	.003	.004	.338	.035	.02	.03
Expectations							
r	.61	.56	.54	.35	.41	.57	.53
p	.00001	.00002	.0004	.03	.01	.0002	.0007
Marginalization							
r	.69	.57	.68	.39	.50	.64	.53
p	.00001	.0002	.00001	.015	.001	.00001	.0006
Knowledge & Beliefs							
r	.61	.53	.62	0.3	.49	.53	.47
p	.0001	.0007	.0001	.06	.002	.0005	.003

r: Pearson correlations; p: p-value