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Access to Patient-Centered Medical Homes in Children with Sickle Cell Disease

Robert I. Liem · Chibuzo O'Suoji · Paris S. Kingsberry · Stephanie A. Pelligra · Soyang Kwon · Maryann Mason · Alexis A. Thompson

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Abstract To determine the proportion of children with sickle cell disease (SCD) followed in a subspecialty clinic with access to a primary care provider (PCP) exhibiting practice-level qualities of a patient-centered medical home (PCMH). We surveyed 200 parents/guardians of children with SCD using a 44-item tool addressing PCP access, caregiver attitudes toward PCPs, barriers to healthcare utilization, perceived disease severity, and satisfaction with care received in the PCP versus SCD clinic settings. Individual PCMH criteria measured were a personal provider relationship and medical care characterized as accessible, comprehensive and coordinated. Although 94 % of respondents reported a PCP for their child, there was greater variation in the proportion of PCPs who met other individual PCMH criteria. A higher proportion of PCPs met criteria for coordinated care when compared to accessible or comprehensive care. In multivariate models, transportation availability, lower ER visit frequency and greater PCP visit frequency were associated favorably with having a PCP meeting criteria for accessible and coordinated care. Child and respondent demographics and disease severity had no impact on PCMH designation. Average respondent satisfaction scores for the SCD clinic was higher, when compared to satisfaction scores for the PCP. For children with SCD, access to a PCP is not synonymous with access to a medical home. While specific factors associated with PCMH access may be identified in children with SCD, their cause and effect relationships need further study.

Keywords Sickle cell disease · Healthcare access · Primary care provider · Medical home

R. I. Liem (✉) · C. O'Suoji · P. S. Kingsberry · S. A. Pelligra · A. A. Thompson
Division of Hematology, Oncology and Stem Cell Transplant, Ann & Robert H. Lurie Children's Hospital of Chicago, 225 East Chicago Avenue, Box 30, Chicago, IL 60611, USA
e-mail: rliem@luriechildrens.org

S. Kwon · M. Mason
The Mary Ann and J. Milburn Smith Child Health Research Program, Children's Hospital of Chicago Research Center, Chicago, IL, USA
e-mail: skwon@luriechildrens.org

Introduction

Recent national efforts have focused on increasing the proportion of children with access to a patient-centered medical home (PCMH), an ideal health delivery model that promotes continuous primary care from childhood to adulthood [1, 2]. Various definitions of a PCMH exist, but most promote having a personal provider and ensuring comprehensive, accessible, compassionate, culturally effective and coordinated care at the practice level. Although longterm data are not available, proponents of the PCMH cite decreased healthcare utilization, enhanced patient and family satisfaction and overall improved health outcomes as major benefits of this care delivery model [3–7]. Despite these recommendations and proposed goals, a substantial proportion of children in the United States lack adequate access to a PCMH. Importantly, children with chronic medical conditions and special healthcare needs are less likely to have access to a PCMH when compared to children without special healthcare needs, as reported in Strickland et al.'s National Survey of Children with Special Health Care Needs [8]. Moreover, socio-economic and socio-demographic disparities may further widen the gap between children with and without access to a PCMH [9–11].

Children with sickle cell disease (SCD) represent a candidate patient population likely to benefit from long-term primary care espousing the qualities of the PCMH model. SCD is not only characterized by a chronic hemolytic anemia but also marked by severe painful events as well as acute and chronic complications, including life threatening infections, stroke and cardiopulmonary disease. Among affected children and adults, SCD remains a lifelong, potentially debilitating condition associated with high acute care utilization and decentralized care [12, 13]. Within this context, dissatisfaction with the medical establishment is common and frequently confounded by perceived racial disparities regarding access to medical care [14]. Although it is assumed that the principles endorsed by proponents of the PCMH are necessary to address these barriers to care in SCD and other chronic medical conditions, it is not entirely clear if this model, either in its entirety or specific tenets, is best implemented through a subspecialist or primary care provider (PCP) [15–17].

Examining PCMH access in SCD is important for several reasons. Little data currently exist regarding the frequency with which children with SCD in the United States are cared for by providers exhibiting qualities of a PCMH [18]. The "comprehensive" SCD clinic is a multi-disciplinary care model adopted by some institutions, including ours, to improve delivery and coordination of health care services. Although this model is ideal for children with SCD, how it impacts their access to a PCMH has not been well studied. Whether or not subspecialty clinics that provide "comprehensive" care can fulfill the role of the PCMH in chronic medical conditions such as SCD is also not known. More importantly, pediatric SCD is marked by high utilization of the ED and other acute care services given the frequency of acute complications from this disease. Recently, Raphael et al. [19] have shown an association between ED encounters/hospitalizations and lack of access to a PCMH. Lastly, data related to medical home access from the National Survey of Children with Special Healthcare Needs did not include SCD in its list of conditions, making it challenging to extrapolate findings to children with SCD.

The objectives of our pilot study were to (1) determine the proportion of children with SCD in a large, tertiary care hospital with access to a PCP, (2) explore whether or not these PCPs exhibited practice-level qualities expected of a PCMH, and (3) investigate socio-demographic variables as well as parent/guardian attitudes, perceptions and barriers associated with access to a PCP. Lastly, we compared parent/guardian satisfaction with care received from a PCP to care received from a subspecialty clinic setting as a surrogate measure of how well each setting adopted PCMH practices. We hypothesized that the majority of children followed in our Comprehensive Sickle Cell Program do not receive routine care from providers who meet criteria for a PCMH.

Methods

To test our hypothesis, we conducted a cross-sectional survey of parents and caregivers of children with SCD followed in our institution's Comprehensive Sickle Cell Disease Program. This study was conducted in accordance with standard ethical principles. Our Institutional Review Board approved the study, and informed written consent was obtained from all participants prior to survey administration and medical chart review.

Study Population and Sampling Frame

Our study population comprised a convenience sample of parents or primary caregivers with legal guardianship of children from birth to 16 years old with SCD (hemoglobin SS, SC, S/β^0 or S/β^+ thalassemia) followed at Ann & Robert H. Lurie Children's Hospital of Chicago (formerly Children's Memorial Hospital). Ours is a large urban, tertiary-care, free-standing pediatric facility caring for approximately 350 active children and young adults with SCD, from which our sample was derived. The survey was administered without any monetary incentive during routine clinic visits, all of which occurred in the main hospital. Parents and caregivers were approached and recruited by research staff or a clinician not routinely involved in the care of the child. Only one parent or caregiver completed each survey. For families with more than one child with SCD in the program, we asked respondents to keep in mind only one affected child when answering questions. Parents and caregivers of children with SCD on a chronic transfusion program were excluded due to differences in the complexity of care and follow-up required in this group of children.

Survey Development

We developed a 44-item survey tool after reviewing several existing tools and resources, including the 2005 version of the Pediatric Medical Home Family Index and Survey (Center for Medical Home Improvement, Concord, NH, USA), the American Academy of Pediatrics policy statement on The Medical Home and the National Survey of Children with Special Health Care Needs. Our survey was composed of yes/no and multiple response questions. Standard patient and parent/caregiver demographic information was collected as part of the survey. Survey questions were developed to address the following domains of

Table 1 General domains evaluated in study survey

Domain	Topics addressed by survey questions
Access to primary care provider (PCP)	Access
	Type and setting
	Frequency of visits
	Timing of last visit
	Reasons for visits
Child's sickle cell disease (SCD) severity	Presence of other medical conditions
	Caregiver perception of disease severity
Barriers to PCP access and healthcare utilization	Cost of appointments
	Transportation
	Difficulty getting appointments
	Insurance issues
	Location of available providers
Attitudes toward and perceptions related to PCP	Perceived importance of PCP for the following:
	Overall health
	SCD care
	SCD knowledge
	Non-SCD care
	Listening and communication skills
	Coordination of care
Satisfaction with care in PCP versus sickle cell clinic settings	Level of satisfaction with the following:
	Overall care
	Visit waiting time
	After hours access
	Getting referrals
	Addressing school issues
	Advice about SCD and non-SCD concerns

interest and variables (Table 1): access to a PCP, defined for the respondent as "a doctor who provides regular and ongoing care for your child, is usually a pediatrician or family doctor, and is not your child's sickle cell doctor"; attitudes toward and perceptions related to PCP; barriers to PCP and healthcare utilization; parental/caregiver perception of their child's SCD severity; and satisfaction with care received from PCP versus Comprehensive Sickle Cell Disease Clinic. We tested our survey tool in 20 subjects during a pilot phase to ensure respondent comprehension and content validity. Given that no major modifications were made to the survey tool, data from this pilot phase were included in the final analysis.

Criteria for Determining PCMH

A primary objective of this study was to examine the proportion of children in our program with access to a PCP who demonstrated characteristics of a PCMH. We developed an algorithm determined by responses to survey questions evaluating level of parental/caregiver satisfaction or agreement with PCP characteristics or services reflecting four major criteria of the PCMH, including (1) being a personal provider, (2) providing care that is accessible, (3) providing comprehensive care, and (4) facilitating care coordination. Access to a personal provider and care coordination was determined by responses to a single question each, while accessible and comprehensive care represented composite criteria based upon responses to six survey items for each (Table 2).

Statistical Considerations

Summary statistics were used to report frequency and to evaluate distribution of all continuous data (SAS® Analytics, V9.4). We used logistic regression and calculated odds ratios to evaluate the internal consistency of individual survey items comprising our composite criteria. The proportion of PCPs who met all criteria for a PCMH was reported as a range using various cut points for the number of supportive responses required to meet the composite criteria for accessible and comprehensive care. PCPs were excluded from this calculation for any of the following: (1) missing response to question about care coordination, (2) $\geq$3 missing responses to questions about accessible care, or (3) $\geq$3 missing responses to questions about comprehensive care.

Standard bivariate analysis was performed using Pearson's Chi square or Fisher's exact test where appropriate to examine variables associated with meeting each PCMH criterion. We used the median split of supportive responses to define meeting criteria for accessible and comprehensive care given the composite nature of these criteria. Some variables were also collapsed and dichotomized given the small numbers associated with some categories. Variables with a P value of 0.2 or smaller were included in a logistic regression model to determine those variables that were independently associated with meeting each PCMH criterion.

Categorical responses to satisfaction questions were assigned numerical ratings (1 = very dissatisfied, 2 = dissatisfied, 3 = satisfied and 4 = very satisfied) so that mean satisfaction scores could be calculated for the PCP versus sickle cell clinic settings. Cronbach's α coefficients were calculated to assess agreement among items used to determine satisfaction. The relationship between PCP and sickle cell clinic satisfaction was evaluated using

Table 2 Criteria required for patient-centered medical home (PCMH)

PCMH criterion	Corresponding survey questions	Required for supporting criterion
PCP is *personal* provider	Q1. Based on the definition above, does your child have a primary care provider?	Answer yes
PCP provides care that is *accessible*	*Barriers to primary care provider:*	Answer no or satisfied/very satisfied to at least 3 of 6 questions
	Q15. Cost of the appointments (e.g. deductible or co-pay, lack of insurance, etc.	
	Q16. Lack of transportation to get to appointments	
	Q17. Difficulty getting appointments	
	Q18. Problems with insurance (e.g. provider is not covered by your insurance plan)	
	Q19. No provider conveniently located in your area	
	Level of satisfaction with PCP:	
	Q30. Getting help from one of the office staff when you call on the phone, including after hours	
PCP provides care that is *comprehensive*	*Reason for past visits to the PCP:*	Answer yes to at least 3 of 6 questions
	Q6. For regular checkup (including school physicals)	
	Q7. For immunizations (shots, vaccines)	
	Q8. Any illness related to sickle cell (e.g. fever or pain)	
	Q9. Any illness not related to sickle cell (e.g. a cold)	
	Q10. Growth and development monitoring (e.g. weight checks)	
	Q11. Counseling (e.g. behavioral problems, nutrition)	
PCP provides care that is *coordinated*	*Level of satisfaction with PCP:*	Answer satisfied/very satisfied
	Q31. Getting referrals to other specialists when needed	

Pearson's correlation coefficient. Mean satisfaction scores were also compared by paired t test between the two settings. A two-tailed, P value <0.05 was considered statistically significant.

Results

Respondent and Patient Characteristics

We collected completed surveys from 200 respondents between May 2010 and August 2011. Only 2 parents who were approached declined participation, citing lack of time as the primary reason. The majority of respondents were female (87 %) and over 30 years old (69 %). A total of 6/200 (71 %) respondents reported obtaining some education beyond high school, defined as technical training, college or post-graduate studies. More than three quarters drive a vehicle to most of their children's medical appointments (78 %) and have medical insurance of their own (83 %).

In total, 188/200 (94 %) respondents reported having a PCP for their child with SCD, described most commonly as a pediatrician (68 %) or a family practice physician (21 %) affiliated most frequently with either a private medical practice (34 %) or a clinic that is part of a community health center (28 %). The majority of respondents described their child's SCD as mild (61 %) or moderate (29 %) in severity. More than half of these children saw their PCP at least 1 to 3 times (48 %) or 4–6 times (28 %) in the previous 2 years. Age of these children did not influence whether or not they saw their PCP at least once in the previous 2 years. PCP visits occurred most commonly either within the past 2 months (43 %) or the past 6 months (28 %). Only 3 % of children had not visited their PCP at all in the previous 2 years, and only 7 % last saw their PCP over 12 months ago at the time of survey completion. The most common reasons for past PCP visits were regular check-ups (88 %), immunizations (84 %) or non-SCD related illness (60 %). Just over one-third (33 %) of past PCP visits were for SCD-related illness.

Overall, more than a quarter of parents and caregivers (28 %) cited at least one or more major barrier associated with either finding a PCP for their child or taking their child to the PCP. Of reasons given, lack of providers in their immediate area (11 %), difficulty getting appointments made (11 %), and lack of available transportation to the PCP (9 %) were the most frequently reported barriers. The majority of respondents believed that PCPs play an integral part of their child's healthcare. Almost all respondents agreed or strongly agreed that having a PCP was important for their child's overall care (95 %) and sickle cell care (90 %). Among the PCP's general responsibilities as a provider, being knowledgeable about SCD was cited as a quality almost all respondents (95 %) believed their child's PCP should possess.

PCMH Criteria and Primary Care Providers

Of the total 188 identified PCPs, 18/188 (10 %) were excluded from our analysis of PCMH criteria due to missing responses to our questions about care coordination. Of the remaining 170 PCPs, another 14/170 (8 %) were excluded due to ≥3 missing responses to questions that assessed either accessible or comprehensive care, resulting in a final sample of 156 PCPs included in this analysis. In general, we found that the internal consistency was good among almost all of the items that comprised our composite criteria, with odds ratios ranging from 1.37 to 4.62 for questions about accessible care and 1.33–2.75 for questions about comprehensive care. However, question 30, which assessed satisfaction with getting help when calling the PCP, had low internal consistency (OR 1.03, [95 % CI 0.29, 3.34]) compared to the other items for accessible care.

Although all of the identified PCPs were considered personal providers by parents and caregivers, there was greater variation in whether or not these PCPs demonstrated characteristics suggestive of a PCMH. We found that 159/170 (94 %) of PCPs, for whom the question was answered, met the criterion for providing coordinated care. PCPs also appeared to be mostly accessible, with supportive responses to all six items comprising the criterion observed for 107/156 (69 %) PCPs. The distribution of supportive responses for comprehensive care was greater, ranging from 51/156 (33 %) for 4 items to only 7/156 (5 %) for all six items. The range of PCPs who met all criteria for a PCMH using various cut points in the number of responses supporting accessible and comprehensive care each is shown in Table 3.

Factors Associated with PCMH

We examined if child and respondent demographics (sex, age and insurance status), transportation availability, perceived disease severity, frequency of ED visits, and frequency of PCP visits were associated with whether or not PCPs met each criterion for a PCMH (Table 4). For the criterion accessible care, question 30 (i.e. satisfaction with getting help from office staff when calling the PCP, even after hours) was evaluated separately given its low internal consistency with the other items used to assess accessible care. By logistic regression modeling, there was an independent association between transportation modality and accessible care. Respondents who drove their own car were more likely to have a PCP for their child who provided accessible care (OR 2.46 [95 % CI 1.17, 5.19], $P = 0.02$; Table 5). For question 30, making fewer than four visits to the ER in the past year for SCD-related issues was significantly associated with greater respondent satisfaction with getting help from staff when calling the PCP (OR 5.15 [95 % CI 1.86, 14.27], $P < 0.01$). We also found that PCP visit frequency was independently associated with having a PCP who met the criterion for coordinated care. Respondents who had not taken their child to see the PCP at all in the past 24 months were less satisfied with their ability to get referrals to other specialists when needed (OR 0.03, [95 % CI 0.00, 0.58], $P = 0.02$). However, there were no significant relationships between factors we examined and whether or not PCPs met the criterion for comprehensive care.

Satisfaction with PCP Versus Sickle Cell Program

Overall, there was good agreement among the items in our survey that assessed respondent satisfaction with their child's PCP (Cronbach's $\alpha = 0.79$) and the sickle cell clinic (Cronbach's $\alpha = 0.86$). There was a significant correlation between mean satisfaction scores for the PCP versus sickle cell clinic (Pearson's $r = 0.43$, $P < 0.01$). On average, respondent satisfaction scores for the sickle cell clinic was significantly higher, when compared to satisfaction scores for the PCP (3.9 ± 0.8 vs. 3.5 ± 0.6, $P < 0.01$).

Discussion

In this study of children with SCD followed in a large urban Comprehensive SCD Program, we demonstrate that

Table 3 Proportion of PCPs meeting all criteria for PCMH by cut points for 2 composite criteria

	Comprehensive care			
	3 responses	4 responses	5 responses	6 responses
Accessible care				
3 responses	76 %, $N = 118$	56 %, $N = 88$	24 %, $N = 38$	5 %, $N = 7$
4 responses	76 %, $N = 118$	56 %, $N = 88$	24 %, $N = 38$	5 %, $N = 7$
5 responses	67 %, $N = 104$	48 %, $N = 75$	21 %, $N = 32$	4 %, $N = 6$
6 responses	54 %, $N = 84$	38 %, $N = 59$[a]	15 %, $N = 23$	2 %, $N = 3$

Criteria for PCMH requires primary care physician who provides accessible, comprehensive and coordinated care. Cut points determined by number of supportive responses out of 6 questions each for the composite criteria of accessible and comprehensive care

[a] Median split for responses for each composite criterion

Table 4 Bivariate analysis of factors and their relationship to meeting individual PCMH criteria

	PCP	No PCP	*P*	ACCESS	Non-ACCESS	*P*	After hours[a]	No after hours	*P*	COMP	Non-COMP	*P*	COORD	Non-COORD	*P*
Respondent sex															
Male (%)	24 (92)	2 (8)	0.66	20 (77)	6 (23)	0.35	24 (96)	1 (4)	0.21	10 (38)	16 (62)	0.21	23 (100)	0 (0)	0.36
Female (%)	164 (94)	10 (6)		118 (68)	56 (32)		133 (86)	22 (14)		90 (52)	84 (48)		136 (93)	11 (7)	
Respondent age															
≤30 years old (%)	57 (98)	1 (2)	0.29	36 (62)	22 (11)	0.15	45 (83)	9 (17)	0.33	33 (57)	25 (43)	0.29	49 (94)	3 (6)	1.00
>31 years old (%)	129 (93)	9 (7)		100 (72)	38 (28)		110 (89)	14 (11)		67 (49)	71 (51)		108 (93)	8 (7)	
Child age															
<6 years old (%)	80 (99)	1 (1)	0.03	58 (72)	23 (28)	0.51	67 (88)	9 (12)	0.75	47 (58)	34 (42)	0.06	68 (97)	2 (3)	0.13
≥6 years old (%)	108 (91)	11 (9)		80 (67)	39 (33)		90 (87)	14 (13)		53 (45)	66 (55)		91 (91)	9 (9)	
SCD severity															
Mild to moderate (%)	170 (95)	9 (5)	0.23	123 (69)	56 (31)	0.78	142 (87)	21 (13)	1.00	92 (51)	87 (49)	0.56	144 (94)	9 (6)	0.23
Severe (%)	14 (88)	2 (13)		12 (75)	4 (25)		12 (86)	2 (14)		7 (44)	9 (56)		12 (86)	2 (14)	
ED frequency															
0–3 times/year (%)	160 (95)	8 (5)	0.22	118 (70)	50 (30)	0.70	138 (91)	13 (9)	**<0.01**	84 (50)	84 (50)	0.74	138 (96)	6 (4)	**0.04**
≥4 times/year (%)	27 (90)	3 (10)		20 (67)	10 (33)		19 (68)	9 (32)		16 (53)	14 (47)		21 (84)	4 (16)	
Respondent insurance															
Yes (%)	155 (94)	10 (6)	0.68	113 (68)	52 (32)	0.85	131 (87)	19 (13)	1.00	88 (53)	77 (47)	0.05	132 (93)	10 (7)	1.00
No (%)	25 (93)	2 (7)		19 (70)	8 (30)		20 (87)	3 (13)		9 (33)	18 (67)		21 (95)	1 (5)	
Child insurance type															
Private (%)	43 (93)	3 (7)	1.00	37 (80)	9 (20)	0.06	42 (100)	0 (0)	**<0.01**	29 (63)	17 (37)	**0.04**	39 (100)	0 (0)	0.07
Non-private (%)	143 (94)	9 (6)		100 (66)	52 (34)		113 (83)	23 (17)		70 (46)	82 (54)		118 (91)	11 (9)	
Transportation to visits															
Own car (%)	151 (97)	5 (3)	**<0.01**	116 (74)	40 (26)	**<0.01**	126 (88)	18 (13)	0.78	82 (53)	74 (47)	0.16	130 (94)	8 (6)	0.42
Other transport (%)	35 (83)	7 (17)		21 (50)	21 (50)		29 (85)	5 (15)		17 (40)	25 (60)		27 (90)	3 (10)	
PCP visits															
0 times/past 2 years (%)	6 (100)	0 (0)	NA	3 (50)	3 (30)	0.35	3 (60)	2 (40)	0.12	1 (17)	5 (83)	0.10	2 (40)	3 (60)	**<0.01**
≥1 time/past 2 years (%)	181 (100)	0 (0)		131 (72)	50 (28)		151 (88)	20 (12)		99 (55)	82 (45)		155 (95)	8 (5)	
Last PCP visit															
2 mos to 1 year ago (%)	173 (100)	0 (0)	NA	124 (72)	49 (28)	1.00	143 (88)	20 (12)	0.65	94 (54)	79 (46)	0.39	147 (95)	8 (5)	**0.03**
>1 year ago (%)	13 (100)	0 (0)		9 (69)	4 (31)		10 (83)	2 (17)		5 (38)	8 (62)		9 (75)	3 (25)	

ACCESS accessible care, *COMP* comprehensive care, *COORD* coordinated care, *NA* not calculated given distribution of responses

Bold values are statistically significant ($P < 0.05$)

[a] Single item (Q30) referring to getting help from staff at PCP office, even after hours

Table 5 Factors independently associated with meeting individual PCMH criteria

Variable	Comparison	OR [95 % CI]	*P* value
Accessible care			
Respondent age—≤30 years old	>30 years old	0.75 [0.38, 1.50]	0.42
Child insurance—private	Other	1.77 [0.73, 4.27]	0.21
Getting to appointments—drive own car	Other	2.46 [1.17, 5.19]	**0.02**
Accessible care—getting help from staff			
Respondent gender—male	Female	3.53 [0.44, 28.65]	0.24
ER visits for sickle cell in past 12 mos—0–3 times	≥ 4 times	5.15 [1.86, 14.27]	**<0.01**
Visits to PCP for any reason in past 24 mos—0 times	1 to >6 times	0.25 [0.03, 1.99]	0.19
Comprehensive care			
Child age—<6 years old	≥6 years old	1.69 [0.91, 3.15]	0.10
Respondent insurance status—has insurance	No insurance	2.22 [0.90, 5.51]	0.08
Child insurance—private	Other	1.94 [0.90, 4.18]	0.09
Getting to appointments—drive own car	Other	0.87 [0.39, 1.95]	0.73
Visits to PCP for any reason in past 24 mos—0 times	1 to >6 times	0.17 [0.02, 1.49]	0.11
Coordinated care			
Child age—<6 years old	≥6 years old	2.17 [0.37, 12.82]	0.39
ER visits for sickle cell in past 12 mos—0–3 times	≥4 times	3.18 [0.68, 14.94]	0.14
Visits to PCP for any reason in past 24 mos—0 times	1 to >6 times	0.03 [0.00, 0.58]	**0.02**
Last time saw PCP for any reason—2 mos to 1 year ago	>1 year ago	0.78 [0.05, 13.14]	0.86

Bold values are statistically significant ($P < 0.05$)

the majority of children with SCD have an identified PCP. Fewer, however, have access to a PCP who meets criteria for a PCMH. Applying even the least conservative cut points (i.e. the fewest number of required supportive responses) to our two composite criteria only resulted in 76 % of identified PCPs in our study meeting all criteria for a PCMH. We also evaluated the impact of socio-economic variables, disease severity and frequency of PCP visits on whether or not a child's PCP met each criterion for a PCMH. In our models, we found that being able to drive one's own car was associated with having a PCP who provided accessible care. Fewer ED visits for SCD-related issues were specifically associated with getting help from the PCP's office, even after hours. Importantly, children who saw their PCP at least once in the previous 2 years were significantly more likely to have a PCP who met the criterion for coordinated care. This suggests that caregivers took their children to see their PCP more frequently if their PCPs provided coordinated care. An alternative explanation might be that caregivers whose children required more frequent visits had more opportunity to engage with their providers in ways that allowed them to view their ability to coordinate care more positively. Finally, we found that respondents were on average more satisfied with care received from our comprehensive SCD program when compared to that received from their children's PCPs.

It is difficult to compare the range of children in our study who had access to a PCMH to that reported in other published studies. In their landmark study, Strickland et al. [20] reported just over 50 % of a national sample of children with special health care needs had access to a PCMH. In contrast, only 11 % of children with SCD, whose caregivers were surveyed, were found in a recent study to have PCPs who met criteria for a PCMH [18]. Several reasons may account for differences in these study results. The most important distinction is the number and type of domains used to define a PCMH, which vary and overlap among studies of PCMH access in the existing literature. In their Medical Home Policy Statement, the American Academy of Pediatrics sums up their definition of a PCMH using seven descriptive core elements, including accessible, continuous, comprehensive, family-centered, coordinated, compassionate, and culturally effective [21]. Only a subset of these elements, however, are operationalized in current survey tools commonly used, including the National Survey of Children with Special Health Care Needs, the National Survey of Children's Health or the Consumer Assessment of Healthcare Providers and Systems (CAHPS) Patient-Centered Medical Home Item Set. The lack of a standardized, validated tool for patients with SCD represents a fundamental challenge to the assessment of the "medical homeness" of providers in this population. Despite the complexity and chronic nature of this disease, SCD was not listed in the National Survey of Children with Special Health Care Needs, making it difficult to generalize findings from even this important study to children with

SCD. In our survey, we focused on four core elements of the PCMH model, namely access to a personal provider and provider care regarded as accessible, comprehensive and coordinated. Although our questions were developed after reviewing several tools, we focused primarily on the Pediatric Medical Home Family Index and Survey, a validated tool developed by the Center for Medical Home Improvement [7, 22].

Several aspects of our survey distinguish our study from others that have evaluated PCP access in children with chronic medical conditions, including SCD. Recognizing the methodological challenges associated with defining PCMH access, we reported a range for the proportion of PCPs who met criteria for a PCMH based on various cut points for our composite criteria for accessible and comprehensive care rather than arbitrarily determining the number of survey items required to support each of those criteria. Future work should be focused on testing the sensitivity and specificity of various cut points for our criteria once a "gold standard" tool is available for comparison. We also sought to explore the variables that predicted whether or not PCPs met each criterion comprising our definition of a PCMH rather than all of them simultaneously. This hypothesis-generating approach not only provides more insight into modifiable risk factors for deficiencies related to each domain in this population but also lays the foundation upon which interventions aimed at improving these domains may be developed. For example, addressing transportation issues may be essential for improving access to quality PCP care, given our finding that the ability to drive one's own car to appointments was associated with having a PCP who provided more accessible care. Access to a car, versus reliance on other modes of transportation, may be a surrogate for higher socioeconomic status in our patient population. Likewise, improving after hours access to one's PCP may reduce ED utilization for SCD-related complications. Finally, encouraging frequent contact and follow through with PCP visits may actually improve coordination of care for complex medical issues.

In our Comprehensive Sickle Cell Disease Program, the majority of caregivers have favorable attitudes toward their child's PCP, and few cited logistical barriers associated with access to PCPs. Fewer PCPs met the individual criterion for providing comprehensive care, when compared to coordinated or accessible care. This suggests that working with PCPs to improve this element of the PCMH model may be particularly important. That we evaluated caregiver satisfaction with care received in the subspecialty versus primary care setting represents another unique aspect of our study. Our institution's Comprehensive SCD Program encourages patients to have a PCP and with the exception of the 23-valent pneumococcal polysaccharide vaccine, routine childhood vaccinations are not administered. Although satisfaction scores for the two settings correlated well with each other, respondent satisfaction on average was higher for the sickle cell clinic when compared to that for the PCP. This is a potentially important finding given current arguments by some that comprehensive subspecialty care alone may adequately serve as a PCMH for individuals affected with chronic medical conditions such as SCD [15–17]. However, small differences in mean satisfaction scores, though statistically significant, may not be clinically important. Still, even if families are mostly satisfied with their PCP and SCD clinic care, there may be opportunities to strengthen the interactions between the two settings to reduce service gaps and to enhance effective case management.

Several limitations of our study warrant discussion. First, we did not attempt to operationalize all of the ideals of the PCMH model in our survey questions. However, the domains we chose to evaluate represent some of the most important core elements endorsed by advocates of the medical home. There is no one standardized tool for measuring PCMH qualities, and no consensus regarding which existing tools are most appropriate. Current tools, like the Medical Home Family Index and the National Survey of Children with Special Health Care Needs, differ in respondent burden and may not accurately reflect the sickle cell patient experience. Second, our relatively small sample size might have limited our ability to accurately determine the relationship between various factors and whether or not PCPs met criteria for a PCMH, although it is comparable to that of other similar studies in SCD [18]. In our analysis of variables associated with coordination of care, for example, the total number of respondents who reported their children did not see their PCP in the past 2 years was small. Third, although we compared satisfaction among caregivers with respect to both their PCP and our Comprehensive SCD clinic, we did not compare whether or not the two care models in fact differ in their adoption of the PMCH criteria. A direct comparison may have provided additional information about the adequacy of the two settings to model their practices according to PCMH ideals. Finally, we did not routinely survey caregivers during their child's hospitalizations. Although most of them would have later had the opportunity to complete our survey in clinic, we might have missed some caregivers of frequently hospitalized patients with poor follow-up. These individuals may comprise a less motivated, more challenging group with poorer access to a PCP or PCMH.

In summary, we demonstrate that access to a PCP is not synonymous with access to a PCMH for children with SCD followed in a large, tertiary-care Comprehensive SCD Program. At our institution, the availability of one's own transportation, fewer ED visits for SCD-related issues and

more frequent visits to the PCP were favorably associated with accessible and coordinated care, although the cause and effect relationships supporting these findings are not clear. The application of a common, validated tool within multiple sickle cell programs that care for children with SCD from diverse socio-demographic and socio-economic backgrounds is essential to gain additional insight into the barriers to PCP and PCMH access in this population. The development of strategies to address the unique challenges faced by individual patients in each setting would be a necessary next step. Examining the impact that PCP and PCMH access have on clinical outcomes would also be useful.

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