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Implementation of Two Developmental Screening Programs in Sickle Cell Disease Specialty Care

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Abstract

Objective: Developmental screening is a critical component of care for children with sickle cell disease (SCD), who are at elevated risk for neurodevelopmental disorders. This report describes the implementation of two related developmental screening programs implemented in different SCD specialty care settings with the purpose of describing screening protocols, outcomes, and lessons learned.

Methods: Program One reviewed medical records for 201 children with SCD screened at ages 2 and 4 years. Program Two reviewed program tracking and visit notes for 155 screenings across 67 children screened between 9 and 66 months of age. Key outcomes included characteristics of children screened, screening results, concordance between parent concerns and screening outcomes, and access to evaluation and intervention services.

Results: Each program identified a substantial number of children with developmental concerns, including 42% of screenings in Program One and 36% of unique children screened in Program Two. Program One resulted in 56% of identified children receiving follow-up developmental services and 62% receiving developmental monitoring. Program Two resulted in 58% of identified children receiving further evaluation following developmental screening, with 67–75% of children with neurodevelopmental diagnoses receiving intervention services following evaluation. While parent concerns were related to screening outcomes, screening instruments detected many children whose parents did not express developmental concerns.

Conclusions: Routine developmental screening is a feasible, acceptable, and effective method for identifying concerns in children with SCD in specialty care. Flexible and collaborative care

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There is overlap for patients from Program One and two publications noted below; however, the program evaluation and outcomes data described are unique to this paper:

Schatz, J., Reinman, L., Bills, S., & Johnston, J. (in press). Sociodemographic and biomedical correlates of developmental delay in two- and four-year-olds with sickle cell disease. *Journal of Developmental and Behavioral Pediatrics*.

Schatz, J., Schlenz, A., Reinman, L., Smith, K., & Roberts, C. W. (2017). Developmental screening in pediatric sickle cell disease: Disease-related risk and screening outcomes in 4 year olds. *Journal of Developmental and Behavioral Pediatrics*, *38*(8), 654-662. https://doi.org/10.1097/DBP.0000000000000486

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and sustainability are key considerations for effective programming, with pediatric psychologists uniquely positioned to provide optimal integrated care.

Keywords

sickle cell disease; developmental screening; integrated care

Introduction

Routine developmental screening is a recommended strategy for improving detection and early intervention for neurodevelopmental conditions. Early detection programs have been developed across pediatric clinics, social service programs, and early childhood programs with the goal of creating flexible delivery systems that address factors such as population-specific needs and local resources (Bricker et al., 2013). This strategy has been recommended for all children in the United States through guidelines from the American Academy of Pediatrics (AAP) that emphasize primary care as a critical location for this service. The most recent guidelines have placed additional emphasis on screenings for children at higher risk for neurodevelopmental concerns due to chronic health conditions, which mirrors other efforts to implement screening with high risk populations (Knutson et al., 2016; Lipkin et al., 2020).

The AAP developmental screening guidelines have not yielded high implementation rates in the United States, with a national average of approximately 30%. Potential factors attributed to variable screening rates include having a usual source of care/provider, socio-economic factors that influence care access, culturally competent care and language barriers, and state-level policies, practices, and incentives (Hirai et al., 2018). Prior efforts to encourage pediatricians to obtain screenings for children at elevated risk due to a chronic health condition have not significantly impacted the likelihood of screening (Knutson et al., 2016). The current manuscript describes an alternate approach for implementing developmental screenings for a population at elevated risk for neurodevelopmental concerns, children with sickle cell disease (SCD).

SCD is a group of inherited blood disorders that affect approximately 1 in 400 to 500 newborns each year (Gustafson et al., 2006). Developmental difficulties in SCD, beyond what is expected from population norms, may emerge as early as infancy and worsen with increasing age leading to ~30–50% with evidence for delay by age 4 years using standardized developmental evaluation instruments (Drazen et al., 2016; Knight et al., 2021; Thompson et al., 2002). Among disease-specific risk factors, cerebrovascular complications and chronic anemia have been most consistently associated with poorer developmental outcomes (Knight et al., 2021; Prussien et al., 2020). Silent cerebral infarction is particularly common in young children with the most severe form of SCD, sickle cell anemia, with a potential onset in infancy and a prevalence of approximately one in four children by age 6 (DeBaun & Kirkham, 2016). Among environmental risk factors in SCD, studies have most consistently identified social and economic disparities in relation to poorer developmental outcomes (Knight et al., 2021; Prussien et al., 2020).

The American Society of Hematology (ASH) has recently developed guidelines recommending that physicians involved in SCD care pay increased attention to developmental and behavioral indicators of cerebrovascular disease, in support of improved detection and intervention for neurodevelopmental and neurocognitive deficits (DeBaun et al., 2020). These guidelines are intended to complement those of the AAP, who specify the use of structured, developmental and social-emotional/behavioral screening instruments at 9, 18, and 30 months of age for all children. Screening or careful surveillance for school readiness at four years of age is also recommended but has not been as rigorously pursued in practice or research to date (Lipkin et al., 2020).

For many children with chronic health conditions, their specialty care clinic may be a preferred location for routine developmental screening and care coordination, given the complexity of disease complications that must be monitored and managed (Van Cleave et al., 2016). In this report, we describe the implementation of two related developmental screening programs within SCD specialty care. Specifically, we present program protocols and evaluation data and lessons learned. Differences in program protocols were context-specific modifications that highlight points of flexibility to address local needs and leverage local resources.

Program One

Methods

Program Overview and Goals—Supplemental Table 1 provides an overview of key characteristics for Program One and Two. The screening program at the University of South Carolina involved a partnership between the Department of Psychology and the Children's Center for Cancer and Blood Disorders (CCCBD) at Palmetto Health (currently Prisma Health) within the Department of Pediatrics in Columbia, South Carolina. The program director (J.S.) held a primary appointment in the Department of Psychology but was also an embedded CCCBD psychologist. Prior to developing early childhood screening programs, routine cognitive screenings were offered to school-age children at SCD visits (Schatz, 2004; Schatz, Finke, & Roberts, 2004). Most children participating in school-age screenings with cognitive deficits had already experienced significant academic problems, resulting in demoralization for the child and frustration for parents (Schatz, 2004). The primary goal of the developmental screening program was to identify preschool age children with developmental delay prior to elementary school. Early childhood screenings were rarely available from other sources (e.g., primary care, day care). Thus, psychology time and effort was organized to provide screenings for preschool age children.

Setting, Time Period, and Staffing—Program One was implemented within the comprehensive SCD clinic at the CCCBD in September 2009 with evaluation of the program conducted through December 2016 (at which time minor protocol changes were made). The center served approximately 375–475 youth with SCD over the course of the program (birth to 21 years of age). The screening program was developed as a clinical service following our experiences from September 2004 through August 2008 implementing developmental screenings at regional preventative SCD clinics then offered by the South

Carolina Department of Health and Environmental Control (Schatz et al., 2008). The screening program was primarily staffed by a half-time clinical psychology doctoral student who completed psychological assessments for children with SCD or brain tumors and other consultation services in addition to developmental screenings. Support was intermittently provided by the supervising psychologist (10% effort) and by an additional part-time doctoral student as backup support.

Screening Protocol—The AAP recommended screening ages were modified due to several factors. First, the age at which children began regular engagement in SCD care varied such that recommended 9- and 18-month screenings could not be reliably implemented across patients. Second, the precise age and frequency of routine visits differed across patients (ranging from monthly to every six months) based on disease severity, genotype, and other factors such that following specific AAP 24- or 30-month target ages would also be difficult to implement. Family schedules and other co-occurring medical appointments indicated that having multiple routine visits within the targeted age window would increase our ability to complete screenings. Therefore, the goal was to screen children at least once as two-year-olds. Finally, a major motivation for the screening program was to identify children at high risk for school difficulties. Therefore, we also adopted a goal of formal screening for children at least once as four-year-olds.

The protocol included the *Ages and Stages Questionnaire, Second Edition (ASQ-2)* and a brief structured interview to elicit perceived child strengths, concerns about the child, and current parent or family stressors. Standard interpretation of the ASQ-2 was done per test manual. Parents were encouraged to not complete ASQ-2 items if they were unsure or had not seen their child complete the task at home. A materials kit was used to facilitate completion of the ASQ-2 when parents were unsure and to confirm items when a child fell below cut-off in one or more domains. Results were shared with the parent either at the clinic or through follow-up contact. Four-year-olds were encouraged to complete additional direct child assessment that included articulation and syntactic processing subtests from the *Fluharty Preschool Speech and Language Screening Test, Second Edition (Fluharty-2)*. These measures were chosen due to frequent parent concerns for articulation and data indicating that syntactic processing may be particularly sensitive to neurocognitive risks in SCD (Sanchez, Schatz, & Roberts, 2010).

The model for developmental follow-up was to encourage direct referrals for intervention services with these resources providing their own evaluation at service onset. For children with positive screenings (i.e., concern for developmental delay), developmental activities sheets were shared with parents and specific activities were highlighted based on area(s) of concern to promote home-based environmental support for development. Formal evaluation and developmental services were also discussed with parents as recommended follow-up. Referrals were made to medical center-based or community-based developmental services depending on parents' interest in pursuing formal services and preferred location for follow-up.

Program Evaluation—Paper and electronic medical records were reviewed to evaluate the program following approval from the medical center Independent Review Board (IRB).

The program evaluation focused on: 1) number of children screened among those with preventative care visits; 2) differences in sex and SCD genotype for eligible children who did or did not receive a screening; 3) concordance between parents' subjective concerns for development and screening outcomes and whether those concerns impacted referral outcomes; and 4) whether children who screened positive received additional monitoring or services following the screening.

Preventative health care visits were evaluated for number of children in the target age ranges who had at least one visit within the time frame of the screening program. Sex and SCD genotype proportions were recorded for eligible children who attended a visit, and Fisher's Exact test was used to compare characteristics of children who were or were not screened. Parent's subjective concerns about child language, motor, or general development were recorded from standard items soliciting these concerns on the ASQ-2 and our brief structured interview ("Do you have any concerns about your child, for example, with their learning, behavior, or development?"). Concordance between parent concerns and screening and referral outcomes was assessed using Fisher's Exact Test. Follow-up procedures were recorded from medical record notes documenting the screening itself or in subsequent follow-up notes for 12 months following the screening. If records indicated no follow-up services in those 12 months (either within or outside of the hospital system), it was coded as "did not receive services." We also reviewed the medical record for specific notes indicating psychology, medical, or social work staff engaged in additional check-ins to inquire about developmental concerns beyond what was typical for a standard visit. These were coded as "increased monitoring for neurodevelopmental concerns."

Results

Program Outcomes—Approximately three-quarters of all eligible children were screened and approximately two-fifths of children had positive screenings for a specific area of developmental delay (Tables 1–2 and Supplemental Figure 1). We noted potential bias among those who participated with 128 of 201 cases screened being male (64% of those screened) as compared with those eligible with 143 of 260 cases being male (55%; 95% confidence interval: 49–61%). Those screened were representative of the total group for SCD genotypes (e.g., 65% severe genotypes screened vs. 67% severe genotypes in the total group). Among 100 two-year-olds completing screenings, 43 were "repeaters" who later completed a four-year-old screening. Among the two-year-old group, those that became repeaters did not differ from non-repeaters in gender (67% male for repeaters; 56% for non-repeaters, Fisher's Exact $p = .303$), proportion of severe genotypes (67% for repeaters, 67% for non-repeaters, Fisher's Exact $p = .999$), or proportion of positive two-year-old screenings (37% for repeaters; 28% for non-repeaters, Fisher's Exact $p = .388$).

Parent-reported developmental concerns (expressed prior to screening outcome) were more frequent among those with positive than negative screenings, Fisher's Exact $p < .0001$. Parents with developmental concerns were more likely to choose a referral for formal developmental services following a positive screening (57%; 20/35) than those without concerns (28%; 14/50; Fishers Exact $p = .013$). Referrals for formal services were predominantly in the speech/language domain (22/34 referrals; 65%) with five referrals

to multi-domain intervention programs and one referral for occupational therapy due to fine motor concerns. Five children with positive screenings continued with prior developmental services (5/85 positive screenings). The success rate for engaging in formal services following a referral was 56% (19/34).

A positive screening led to increased monitoring for neurodevelopmental status for children with positive screens who did not receive formal developmental services. A majority of children with positive screenings who did not receive formal developmental services had non-routine check-ins by clinic staff about the child's development that were noted in medical records over the next 12 months of care (62%; 41/66). There were no non-routine check-ins by clinic staff about the child's development for these same children in the prior 12 months.

Program Two

Methods

Program Overview and Goals—The developmental screening program through the Medical University of South Carolina (MUSC) in Charleston, South Carolina was created based on the program director's experience working in Program One. The program director (A.M.S.) was hired in 2015 with a primary appointment in Developmental-Behavioral Pediatrics and as an embedded SCD team member. Prior to the program's development, there were multiple obstacles for the clinic in identifying children with developmental concerns and obtaining services. A key issue was the absence of developmental screening for the majority of children within primary care, which was the result of variation in which pediatricians chose to implement developmental screening and difficulty accessing screenings outside of target ages or past 30 months of age. Additional challenges included long evaluation wait-lists in Developmental-Behavioral Pediatrics and complex processes for Early Intervention and Child Find.

Based on these needs, the primary program goal was to improve access to developmental screening, evaluation, and intervention services for young children with SCD. The protocol was adapted from Program One to meet the needs of this specific population and to leverage resources from Developmental-Behavioral Pediatrics. Adaptations included infant and toddler screening, broader coverage of social-emotional functioning, and the addition of a risk stratification protocol to fast track children for evaluation. These adaptations were made due to the SCD team's desire to provide support to as many ages as possible (particularly for infants and toddlers who are eligible for Early Intervention), to expedite evaluations for children with pre-existing needs, and to have a psychologist establish rapport with families from infancy as there had not been an embedded psychologist in the clinic for many years.

Setting, Time Period, and Staffing—The screening program was initiated in September of 2015 with program evaluation conducted through September of 2019. The MUSC Pediatric Sickle Cell Clinic serves approximately 500 children from birth to adulthood. Staffing included a dedicated psychologist (A.M.S.) who provided a range of evaluation and consultation services and who also had coverage from teaching and research funding

during some years of the program (15–39% effort from other sources). Screenings occurred throughout the week as part of routine SCD care. In 2016, predoctoral interns and postdoctoral fellows were introduced to the program (one day per week).

Referral, Scheduling, and Billing Procedures—A SCD clinic team member (e.g., hematologist, nurse practitioner, psychologist, or psychology trainee) provided education about developmental screening to families using a handout created by the psychologist and obtained consent for the appointment. Referrals for developmental screening were made by the child's hematology provider and the child was scheduled for an appointment to coincide with their next routine hematological visit. Visits were billed to the child's insurance in accordance with institutional and compliance billing policies.

Screening Protocol—Target ages included those from the 2001 AAP guidelines and also included annual screening for ages 3 to 5 years due to elevated risk past the 30-month target age in SCD (Schatz et al., 2022; Schatz et al., 2017). Children often attended appointments at other ages and were seen for screening even if they were not at a target age, particularly for their first screening or when a child needed to be re-screened. The psychologist then worked with the family to establish a frequency that approximated AAP guidelines (often every 6–9 months for children under age 2 and annually thereafter) or based on screening results if concerns were identified. In addition, a risk stratification protocol was used to fast track children for evaluation based on known medical risk factors (e.g., history of prematurity, stroke risk based on transcranial doppler screening) or for parents with longstanding concerns for a neurodevelopmental disorder.

The screening protocol included a clinical interview with developmental, medical, psychosocial, and school/intervention history; questions about feeding (including pica), sleep (including obstructive sleep apnea), and toileting; and parent concerns in each of these areas. The psychologist or psychology trainee then completed the *Ages and Stages Questionnaire – Third Edition (ASQ-3)* with the parent and child by interview and using a materials kit to assess specific skills that had not been seen at home and to obtain a semi-structured observation of the child. Parents also completed the *Ages and Stages Questionnaire – Social Emotional, First or Second Edition (ASQ-SE or SE 2)* and the *Modified Checklist for Autism – Revised/Follow-up (M-CHAT-R/F;* for ages 16 to 30 months of age) either by interview or questionnaire based on their preference. Scoring and interpretation were completed according to manual instructions. Parents sometimes expressed concerns for speech articulation, but the child did not fall below cut-off on the ASQ-3. The Caplan and Gleason (1988) method for rating expected speech intelligibility was used in these situations to determine a positive or negative screening.

Screening results and consultation around common concerns or SCD-specific topics (e.g., pica) were provided to the family. Developmental activity sheets were provided to all families. For scores in the monitoring range for the ASQ-3 or ASQ-SE 2, the psychologist recommended re-screening at the child's next visit and used activity sheets to recommend specific ideas to support development at home. For children who screened positive on any instrument, the provider recommended options for follow-up (i.e., evaluation and referral to intervention), but also supported families in making decisions based on their goals

and preferences. Screenings typically ranged from 30–60 minutes, including feedback and consultation.

Follow-up evaluations were provided by the psychologist or outside providers (based on family preference), including referrals to Early Intervention, Child Find, audiology, speech/language pathology, occupational therapy, physical therapy, and/or mental health. The psychologist continued to follow the family based on their preferences, with routine follow-up appointments, phone calls, or evaluations as needed. The psychologist also consulted with outside treating providers and school personnel when desired by the family for care coordination.

Program Evaluation—Based on IRB policies of MUSC, the program evaluation described below fell under the scope of quality improvement and was therefore not subject to IRB for review or approval. The following outcomes were evaluated: 1) percent of children who completed developmental screening out of those approached for the service; 2) concordance of parent concerns for development in any domain of the clinical interview and screening outcomes; 3) adherence to program target ages and AAP recommended ages; 4) percent of children who screened positive for developmental concerns on screening instruments; 5) percent of children who received evaluation and intervention services following identified concerns; and 6) reasons why children did not receive screening, evaluation, or intervention services. Information on outcomes for children who were risk stratified to evaluation were also collected.

Program records (tracking spreadsheets, screening and evaluation notes, and follow-up notes) were reviewed to obtain characteristics of children who participated in the program and key outcomes. Due to variation in number of screenings per child, screening results were determined *per unique child* for screening completion and results. To estimate adherence to target ages, we reviewed information on children who started receiving screenings in the first program year and evaluated whether they had received a screening +/− 3 months of a target age. These children were eligible for the highest number of target ages, so they provided the most representative estimate of adherence for the duration of the program. Concordance between parent concerns and screening outcomes was evaluated using chi-square and was basing on *screening outcome* (rather than unique child).

Results

Program Outcomes—Characteristics of children and several program outcomes can be found in Tables 1–2 and Supplemental Figure 2. About 120 children are estimated to have fallen between program ages at any point in time during the evaluation years. During this time, 100 children were approached for developmental screening ($n = 88$) or were risk stratified to developmental evaluation ($n = 12$). Of 88 families approached for developmental screening, 66 children (75%) were seen for developmental screening. The family of one child with medical risks for developmental concerns preferred screening rather than evaluation, for a total of 67 children with at least one developmental screening. The remaining 11 children were risk stratified for evaluation and all received evaluations. In total, 78 children received either developmental screening or evaluation.

A total of 155 screenings were completed across 67 children. Number of screenings completed per child ranged from 1 to 7 ($M = 2.33$, $SD = 1.28$). Of these children, 36% screened positive for developmental concerns during at least one screening, 36% fell in the monitoring range (but did not screen positive) during at least one screening, and 28% had consistently negative screenings. Average age at positive screenings was 3.06 years ($SD = 1.30$). Parent-reported concerns for development were more common during screening encounters that resulted in a positive screening versus those resulting in the monitoring range or that were negative (x^2 (2, $N = 155$) = 21.74, = $p < .001$). Among 11 children who started being screened in the first year of the program, adherence to all eligible age time points for the program was 77% (range of 40–100%; range of 3–5 eligible target ages). Among a subset of eight children who were 30 months of age or younger at the start of the program, adherence to recommended AAP infant and toddler ages was 96% (range of 67–100%; range of 1–3 eligible target ages).

Following developmental screening, 58% (14/24) of children with a positive screening received further evaluation, 86% (12/14) of whom received a diagnosis of one or more neurodevelopmental disorders (Language Disorder $n = 5$, Attention Deficit/Hyperactivity Disorder (ADHD) $n = 4$, Speech Sound Disorder $n = 2$, and Developmental Coordination Disorder $n = 1$). Of 12 children with a neurodevelopmental diagnosis, 9 (75%) received intervention services. Only one child who screened positive had received prior evaluation for developmental concerns. The remaining children were newly identified.

For children who were not screened ($n = 22$), only 3 families formally declined the service (overall refusal rate of 3.4% of all families approached). The remaining children were waiting to be scheduled ($n = 7$), had a deferred screening because they were being screened in primary care ($n = 5$), aged out of the screening range before they could be seen ($n = 5$), or moved away before they could be seen ($n = 2$). For children who were not evaluated following a positive screen ($n = 10$), reasons included family preference to monitor the child's development ($n = 3$), logistical challenges scheduling or getting to the evaluation appointment ($n = 3$), family declined ($n = 2$), insurance challenges ($n = 1$), and family moved prior to the evaluation ($n = 1$).

Of 11 children who were risk stratified and whose family opted for evaluation, primary indications were history of prematurity ($n = 5$), longstanding parent concerns for neurodevelopmental disorder ($n = 4$), and stroke risk ($n = 2$). Of these children, 64% (7/11) were diagnosed with one or more neurodevelopmental disorders (ADHD $n = 3$, Speech/Language Disorder $n = 2$, Global Developmental Delay $n = 1$, Intellectual Disability $n = 1$, Autism Spectrum Disorder $n = 1$, Developmental Coordination Disorder $n = 1$) and an additional two children required further evaluation for obstructive sleep apnea prior to diagnosis. Of these 9 children, 6 received intervention services (67%), including school services ($n = 6$), outpatient therapies ($n = 1$), and mental health ($n = 1$). Two children had pre-existing services that continued. Among all children who did not receive intervention services after evaluation, reasons including parents opting for informal school supports ($n = 3$), insurance challenges ($n = 1$), child not eligible for services ($n = 1$), and family declined services ($n = 1$).

Discussion

This report describes two programs that embedded routine developmental screening into SCD specialty care. Below we outline several lessons learned from our experiences and program evaluation data in order to inform future efforts in this area.

Developmental screening is an acceptable, feasible, and effective approach.

We have found that developmental screening is acceptable to families (as determined by a low refusal rate among families), feasible, and an effective approach for identifying developmental concerns in the specialty care setting, consistent with a separate report supporting the utility of the ASQ-3 within SCD specialty care (Hahn et al., 2020). Rates of positive screenings using a structured protocol were consistent with previous estimates of developmental concerns in young children with SCD (Drazen et al., 2016; Thompson et al., 2002), with a rate of 42% of screenings in Program One and 36% of unique children in Program Two. The number of children with developmental concerns was even higher for Program Two when considering children who were risk stratified for evaluation.

Screening yields a higher detection rate than surveillance in children with SCD.

The recent ASH cerebrovascular guidelines recommend surveillance of developmental concerns as a feasible method for medical providers to identify children who have (or are at risk for) cerebrovascular morbidity (DeBaun et al., 2020). Consistent with prior literature, we found that surveillance methods showed moderate correspondence with formal screenings for Program One. The primary weakness of surveillance relative to formal screening has been a lower detection rate with surveillance, rather than concerns about specificity (AAP, 2001). Thus, clinicians should be aware that ASH surveillance guidelines may miss half or more of children with likely developmental delay compared to formal screening. This point is particularly important as individuals with SCD already face well-documented health inequities, with many children with SCD residing in areas of the country in which pediatrician adherence to developmental screening is particularly low (Hirai, et al., 2018; Lee, Smith-Whitley, Banks, & Puckrein, 2019).

Program Two included a more extensive interview and the results described included parent concerns in any interview domain (e.g., language, motor, adaptive skills) due to the manner in which data was collated. These differences likely explain the higher rate of concordance between parent concerns and screening results for Program Two and point to the importance of research on the validity of specific surveillance methods. For both screening programs, clinic resources and collaboration with other departments was required. Thus, for SCD specialty clinics without such resources, surveillance may be the only feasible option.

Sustainability considerations are important.

Each program of this type needs to determine age targets that balance feasibility and sustainability, including population size, psychologist effort, timing of specialty care visits, and other sources for screening and support. If fewer age targets are feasible and sustainable, we recommend targeting the preschool period (ages 2–4) at a minimum. As shown in Program Two, the average age of positive screening was about 3 with most children captured

between the ages of 2 and 4. In addition, AAP screening guidelines stop at 30 months of age, with a less firm recommendation for kindergarten readiness assessment at age 4 (Lipkin, et al., 2020); thus, children with SCD may not receive formal screening after 30 months in primary care when risk for developmental concerns increases. On the other hand, if it is feasible and sustainable to target more ages and children are not receiving high quality screenings and support elsewhere, we recommend adopting the AAP ages and completing annual screening in the preschool period.

The two programs do illustrate how different protocols, staffing, and financial models can work, including philanthropic or fee-for-service models. We recommend that programs meet with their institution's compliance officers to discuss planned services, billing, and documentation standards. Supplemental Table 2 provides considerations and resources to support this process. Part-time positions and multi-staff programs can be challenging when roles are spread out, and a single, embedded role may provide the greatest amount of flexibility and coordination with specialty care. These programs also involve an extensive amount of program tracking; thus, allocating administrative time from the psychologist and clinic staff is important, particularly if referral and scheduling are required for billing and documentation.

Collaborative care and flexibility with families are important.

Developmental screening should be a collaborative process with parents and programs should be aware of family preferences for screening and follow-up care. We found that parents were more likely to want screenings for boys than girls for Program One. This may reflect the larger population-level trend for higher rates of parent-reported developmental concerns for preschool-age boys than girls (Marshall et al., 2016). Some parents also indicated preferences for location or type of service or preferred re-screening at their next visit rather than evaluation or intervention. Providing options to families is important, and we have found it helpful to include re-screening for families who initially decide to forgo services. We have also found that ongoing engagement with parents who are reluctant to pursue formal services can be helpful as parents' interest in services may change over time. Notably, the structure of SCD care for preschool children provides multiple routine visits each year, which allows SCD staff to have more opportunities to monitor concerns and engage families in services versus routine primary care.

Similar to other settings, we encountered challenges connecting children and families with developmental services, including access to providers, insurance and logistical barriers, and eligibility requirements for Early Intervention and Child Find (Elansary & Silverstein, 2020; King et al., 2010). Partnerships with community partners, family navigators, and outpatient specialties can be beneficial for improving outcomes. In addition, having multiple service options available is important (e.g., community services and outpatient therapies).

Connection with developmental services is only one potential benefit that can follow the screening process. We also provided parents with targeted information about home-based activities to stimulate development, and clinic staff often increased their attention to potential developmental concerns following a positive screening. Anticipatory guidance about child development is frequently an area of parent interest that is missed in medical

encounters (Combs-Orme et al., 2011). Brief parenting interventions similar to reviewing developmental activities and strategies with the parent and accompanying written materials have been used as effective interventions for children with developmental disabilities (Tellegen & Sanders, 2013).

Pediatric psychologists are uniquely positioned to provide developmental screening.

Although developmental screening can be completed by a range of professionals, we have found specific benefits of pediatric psychologists and psychology trainees providing this service. Careful communication of screening results, their meaning, and negotiating parent concerns about follow-up can require skills and training beyond what is minimally necessary for valid administration of screening tools, as seen in other quality improvement efforts in this area (Talmi et al., 2014). Having psychologists involved in the screening process reduces the number of “hand-offs” needed across professionals when positive screenings occur, which can reduce barriers to developmental evaluation or consultation. Developmental screening also provides opportunities for psychologists to develop rapport with families from a very young age.

Limitations

This report has important limitations. Program data were collected at a specific point in time, and it is likely that additional children would have been evaluated or would have received services given additional time. We did not collect information on how much time occurred between screening and evaluation or intervention, which would have revealed additional information on access issues. Finally, our program evaluation data do not provide information on family perspectives or satisfaction with screenings, though we had a low refusal rate over time.

Conclusions

Detecting and addressing developmental concerns requires a flexible set of resources that brings screening, evaluation, and intervention resources to convenient access points for families. For children with chronic health conditions, specialty care can be a useful location to integrate developmental screening and follow-up care. In major medical centers, access to psychology, social work, and other supports for providing this care could address barriers noted in primary care (King et al., 2010). More broadly, service integration in medical settings often increases patient perceptions of a holistic view of their care and perceptions of quality and satisfaction, which may ultimately improve the overall effectiveness of specialty care (Baxter et al., 2018).

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Implications for Impact Statement:

Children with SCD are at high risk for neurodevelopmental disorders and benefit from early, high quality identification and support for these concerns. Routine developmental screening delivered by a psychologist in specialty SCD care is a feasible, acceptable, and sustainable approach for meeting these important needs.

Table 1

Characteristics of Children with Sickle Cell Disease by Program

Characteristic	Program One	Program Two	
	Developmental Screening (*n* = 201)	Developmental Screening (*n* = 67)	Evaluation (*n* = 11)
Age (years; *M* ± *SD*)			
First screening	3.21 ± .35	2.46 ± 1.36	--
Overall	3.47 ± .35	2.58 ± 1.27	4.17 ± 1.13
Gender (*n*, %) *			
Female	73 (36.3%)	31 (46.3%)	5 (45.5%)
Male	128 (63.6%)	36 (53.7%)	6 (54.5%)
Sickle Cell Genotype (*n*, %)			
HbSS	127 (63.2%)	34 (50.7%)	8 (66.7%)
HbSC	49 (24.4%)	21 (31.3%)	2 (18.2%)
HbSß$^{+}$	19 (9.5%)	11 (16.4%)	1 (8.3%)
HbSß^{0}	2 (1.0%)	0 (0.0%)	0 (0.0%)
HbSO-Arab	2 (1.0%)	0 (0.0%)	0 (0.0%)
HbSHPF	1 (0.5%)	1 (1.5%)	0 (0.0%)

Note.

* For Program One, we noted a potential bias among those who participated in screenings with 128 of 201 cases screened being male (64% of those screened) as compared with those eligible with 143 of 260 cases being male (55%; 95% confidence interval: 49–61%).

Table 2

Developmental Screening Program Outcomes

Program Outcome	Program One		Program Two		
Screening Completion	77% (201 / 260)		75% (66 / 88)		
Overall Screening Outcomes	Positive	Negative	Positive	Monitoring	Negative
	42% (85 / 201)[a]	58% (116 / 201)	36% (24 / 67)[b]	36% (24 / 67)	28% (19 / 67)
Parent-Reported Concerns by Screening Outcome	Positive	Negative	Positive	Monitoring	Negative
	41% (35 / 85)	9% (11 / 116)	73% (19 / 26)	43% (19 / 44)	24% (20 / 85)
Positive Screenings by Screening Measure					
ASQ-2/ASQ-3	36% (72 / 201)		21% (14 / 67)		
Fluharty-2					
Articulation	10% (8 / 77)		--		
Syntactic Processing	31% (24 / 77)		--		
ASQ-SE/ASQ SE-2	--		12% (8 / 67)		
M-CHAT-R/F	--		11% (medium risk; 1 / 35)[c]		

Note.

[a]For Program One, the overall rate of positive screenings differed across the two age groups with 32% (32/100) positive screening on the ASQ-2 for two-year-olds and 52% (53/101) positive screenings on either the ASQ-2 or the Fluharty for four-year-olds, Fisher's Exact $p = .004$.

[b]For Program Two, four children (6%) were classified as having a positive screening for articulation concerns alone based on parent-reported concerns and low speech intelligibility using the Coplan & Gleason method.

[c]M-CHAT-R/F administered only for ages 16–30 months.