Increasing Documentation and Disclosure of Sickle Cell Trait Status: An Implementation Science Approach

NCT05387564

Document Date: September 1, 2023
Updated cover page: October 21, 2025

Increasing Documentation and Disclosure of Sickle Cell Trait Status: An Implementation Science Approach: SCTalk

COMPLIANCE STATEMENT

This study will be conducted in full accordance with all applicable Nemours Research Policies and Procedures and all applicable Federal and state laws and regulations including 45 CFR 46. All episodes of noncompliance will be documented.

The investigators will perform the study in accordance with this protocol, will obtain consent, and will report unanticipated problems involving risks to subjects or others in accordance with Nemours IRB Policies and Procedures and all federal requirements. Collection, recording, and reporting of data will be accurate and will ensure the privacy, health, and welfare of research subjects during and after the study.

STUDY OBJECTIVES

A two-arm, randomized, interrupted time series design will compare an "all-in" and "add-in" approach to a suite of intervention strategies to increase PCP's EHR documentation and disclosure of NBS results and SCT status during infancy. This is a pilot study. The main goal of this pilot study is to establish a reliable estimate of the effect size of a suite of intervention strategies.

Primary Objectives (Aim)

Using an implementation framework we will assess and compare the acceptability, feasibility, self-efficacy, and time costs of using the intervention strategies when delivered "all-in" versus "add-in" by measuring the following outcomes.

- Acceptability: via PCP survey for each toolkit component.
- Feasibility: Percent of PCPs who use individual toolkit components.
- Adoption/Self-efficacy**:** PCP survey. Intention and confidence to document/discuss NBS/SCT result.
- Cost**:** PCP survey: Perceived time (in mins) to document and discuss NBS/SCT result, effect on other discussion topics during the visits.
- Penetration (via interrupted time series)**:** via EHR extraction. Rate of scanned NBS results available in the EHR. Rate of documentation and disclosure of NBS and SCT result at < 2 months of age in each of the following: the problem list, the medical history section, and progress note. Evaluate monthly: retrospectively (pre intervention) and prospectively (post intervention).

Secondary Objective (Aim)

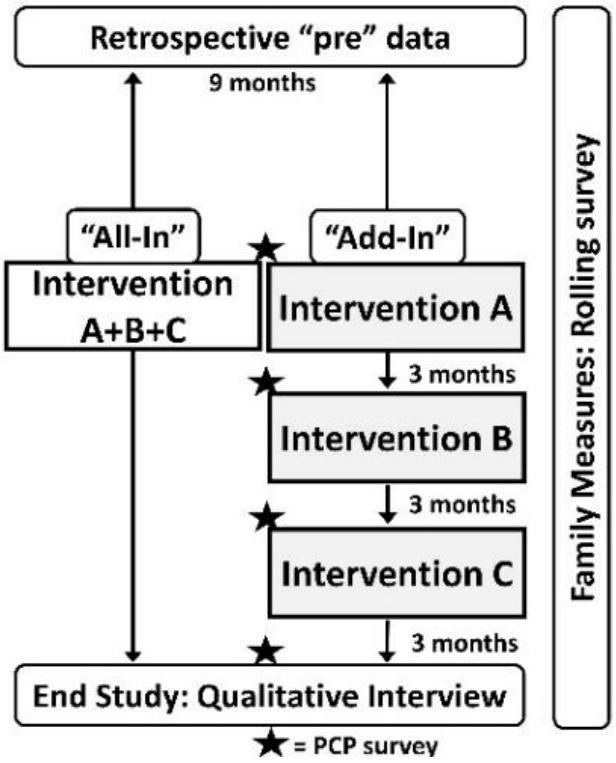
Assess the effects of the intervention strategies when delivered "all-in" versus "add-in" by measuring the following outcomes.

- Dispersion: Via EHR extraction- Proportion of children over 2 months of age who have NBS/SCT newly documented (as above).
- Knowledge: Caregiver survey- Proportion of caregivers who accurately reported their child's NBS result.
- Inequities in documentation**:** Via chart review. Demographic data for each infant: primary care site, sex, race, ethnicity, and insurance type.
- Satisfaction: Caregiver survey.

INVESTIGATIONAL PLAN: Overview

General Schema of Study Design

A two-arm, randomized, interrupted time series design will compare an “all-in” and “add-in” approach. In the “all-in” arm, participants receive all toolkit components at once. In the “add-in” arm, participants will have “multiple interruptions” with sequential addition of components in 6 week increments (**Figure – replase 3 month label with 6 week label**). This will allow for exploration of the added benefit of certain components as compared to the whole toolkit.



Toolkit Components:

1) Intervention A: EHR “nudge” within the birth history and/or well-child progress note template

2) Intervention B: “lunch-and-learn” sessions (in person or pre-recorded per office preference)

3) Intervention C: written educational materials (i.e. post cards, poster, PDF files) to alert PCPs to the importance of SCT documentation and “talking points” to facilitate disclosure;

Duration of Study Participation

Duration of study participation for a site will last a total of approximately 5 months. During the 5 months, PCPs will participate in 4 surveys (15 minutes each) and 1 qualitative interview (30 minutes). Study duration of caregivers will be for survey completion (5-10 minutes).

Total Number of Study Sites/Total Number of Subjects Projected

1) Sites: At least 2 sites per arm of the study with at least 10 PCPs in each arm are required.
2) PCPs: All PCPs from a given office will be eligible for participation. This will result in at least 20 Nemours PCPs at 4 or more office locations in the Delaware Valley. PCP offices will be purposively chosen to include a wide racial distribution within their patients.
3) Caregivers: 500 caregivers of infants with SCT or a normal NBS who are cared for within the specified office location.

Study Population

Participants will be Nemours primary care offices, Nemours PCPs, and caregivers of children seen for infant primary care well visits at the enrolled office.

Inclusion Criteria:

- Sites: Nemours primary care locations within the Delaware Valley that provide well child care to infants
- PCPs: All Nemours PCPs (physicians and nurse practitioners) at included office locations.
- Caregivers: English speaking caregiver of an infant seen at a participating office location for a well visit before 2 months of age (2 months = 60 days) with an email address available within the EHR.

Exclusion Criteria:

- Caregivers and PCPs: Unable to converse in English.
- Caregivers: Caregivers of infant seen for a first well visit at 61 days of age or older

Subjects that do not meet all of the enrollment criteria may not be enrolled. Any violations of these criteria must be reported in accordance with IRB Policies and Procedures.

STUDY PROCEDURES

Site selection: Emails will be sent to the physician-in-charge of Nemours outpatient office locations within the Delaware Valley with a description of the study and an invitation to participate. For those interested in participating as a site, a REDCap interest survey will be included asking for site specific information (i.e. location, name/contact information of physician-in-charge (PIC), name/contact information of office manager or other person designated by the PIC to handle communication about the trial, Number of PCPs at site). Sites will be matched and paired to create dyads/triads of locations with similar racial demographics of patients (i.e. a dyad with the majority of patients documented as Black or Hispanic, etc). For sites selected for participation in the trial, name/contact information will be collected from the PIC or office manager for all PCPs at the office and anyone else they would like to receive communication about the trial (i.e. nursing staff). An introductory email will be sent to all staff identified by the PIC/office manager explaining the study.

Randomization: Sites will then be randomized within each dyad (i.e. one clinic randomized to the “all-in” arm and the other to the “add-in” arm). All PCPs from the same location will be assigned to the same trial arm.

Practice level data from the medical record. For each practice location: Patient level demographics (based on having been seen for a well child exam in the 3 years prior to data extraction) will be linked with the primary practice location at the start of the trial. Practice level data will be obtained from a retrospective evaluation of available EHR data and focus on.

- Age and gender distribution
- Race and Ethnicity distribution
- Insurance make-up (Private, Medicaid, other)
- Zipcode to assess median household income of patients seen at that location (census data), Childhood Opportunity Index (COI)
- Number and portion of the patient population with SCT documented in the problem list, medical history section, and diagnosis codes

PCP Consent and Demographic Survey:
Toolkit components will be available to PCPs and office staff in all trial sites regardless of their participation in the research portion of the trial. Use of the toolkit components is as an educational aid and to facilitate routine patient care.

PCPs will initially be approached for consent for the research portion of the trial prior to toolkit initiation. For PCPs who elects to participate in the longitudinal research portion of the trial, a general demographic survey will be completed at time of initial consent. The demographic survey will be facilitated by REDCap and administered electronically. PCPs may enroll at any time during the trial though first attempt at consent will be before toolkit initiation at with each toolkit component rollout and survey point.

"Roll-out" of toolkit components:

- "All-in" arm: The "all-in" trial arm will have access to and be educated regarding the intervention components at one time point via email, via discussion at lunch & learn, via office meeting, or via virtual meeting or recording (depending on site preference).
- "Add-in" arm: Each PCP in the "add-in" arm will receive the intervention components in the same sequence (within a 1 month timeframe of other offices in the "add-in" arm) with 1 component added every 6 weeks.

With each toolkit component rollout an email will be sent to the office staff reminding them about the project, alerting them to the new component, and notifying them about a virtual Teams "office hours" where the study PI will be available for questions or concerns. The study PI will also be available for any virtual or in-person education an office site may request (ie at an office meeting, etc)

PCP Implementation Surveys: PCPs will receive surveys by email (through REDCap) every 6 weeks. Survey is expected to take 15 minutes to complete. For PCPs in the "all-in" arm, the first survey will be sent prior to the intervention "roll-out" and every 6 weeks after that. For PCPs in the "add-in" arm, the first survey will be sent prior to intervention A "roll-out", intervention B "roll-out", intervention C "roll-out", and then 6 weeks after. The survey is created specifically for this study and focused on the acceptability of using the intervention component, feasibility of using the intervention component, the opportunity costs of using the intervention component (the time it takes to use the intervention, the perceived effect on other discussion topics with well child visits), and the intention and confidence (i.e. self-efficacy) to document and discuss NBS and SCT results.

PCP Qualitative Interviews: At completion of the Intervention period, PCP participants will be contacted to schedule the qualitative interview. Interviews will be expected to last ~30 minutes. Interviews will be completed over the telephone or using a video conferencing platform. If video conferencing is used, participants will be instructed to keep their camera turned off, so their image will not be recorded. The participant will be notified verbally when the recording has started and when it has stopped.

The qualitative interviews will assess their experience with the components and intention to document and disclose NBS/SCT results in the future.

Infant Identification:
Infants seen at a participating site during the retrospective and intervention period for a well child visit before 2 months of age will be identified through EHR data extraction on a rolling basis (i.e. once a month during the trial) based on ICD10 code for a new patient/well child exam (ICD 10: Z00.110, Z00.111, Z00.121, Z00.129, Z00.00, Z00.01 or CPT: 99381, 99382, 99383, 99384, 99385, 99391, 99392, 99393, 99394, 99395).

Documentation and Discussion of NBS/SCT results for all infants identified: via chart review. For each infant identified, the following data will be collected via EHR extraction and manual chart review:

- Email
- Preferred language
- MRN (to facilitate manual chart review)
- Gender
- Race, ethnicity
- Insurance
- Zipcode (to assess Childhood Opportunity Index)
- Phase of study they were first seen for a WCC (retrospective, “all-in” A/B/C, Intervention A, Intervention B, Intervention C)
- Presence/absence of NBS
- Result of hemoglobinopathy NBS
- Presence/absence of NBS result documented in 1) problem list, 2) medical history, 3) progress notes
- Presence/absence of a disclosure conversation about NBS result in progress notes (including topics discussed: inheritance patterns, medical implications, reproductive implications, hematology/genetics consultation)
- Presence/absence of NBS results in After Visit Summary communication

Caregiver Survey from retrospective period:

Caregivers of three groups of infants will be surveyed if they are identified as speaking English (through EHR demographic data) and with an email address in the EHR:

- Group 1: All infants with SCT seen before 2 months of age within the 6 month retrospective period
- Group 2: All infants identified as Black or Hispanic seen before 2 months of age within the 6 month retrospective period

- Group 3: Infants seen before 2 months of age within the 3 month retrospective period and not included in group 1 or 2

Caregiver Survey during Intervention Period:
Caregivers from the participating office locations of three groups of infants will be surveyed if they are identified as speaking English (through EHR demographic data) and with an email address in the EHR:

- Group 1: All infants with SCT
- Group 2: All infants identified as Black or Hispanic
- Group 3: Infants seen in the first half of the month and not included in group 1 or 2

Caregivers will receive a survey at 2 and 6 months of age (+/- 1 month) delivered by email (through REDCap). Survey is expected to take less than 10 minutes to complete. Consent and survey responses will be obtained electronically. The survey will inquire about results of the NBS, preferences for information receipt, and satisfaction with well child visit.

Recruitment of Participants

- **See site selection under Study Procedures**

- **PCP Recruitment**. An introductory email (through REDCap) will be sent to all staff identified by the PIC/office manager for any site participating in the trial explaining the study before the Intervention period. The PI will be available to attend any office meetings or question and answer sessions that the PIC or PCPs request. PCPs will be informed that participation in the research components is voluntary, will not affect their employment, and that they will receive access to the toolkit components without participation. The study coordinator and/or PI will follow-up with PCPs by telephone, email, virtual meeting, or in person to answer additional questions.

  They will be given the contact information for the PI and study coordinator for any questions or withdrawal from participation. Three follow-up requests will be sent to non-responders at ~1 week intervals and then at 6 week intervals (ie when interventions are rolled out and surveys distributed) during study time period.

- **PCP Retention**. Trial procedures will include every 6 week on-line REDCap facilitated surveys and a final qualitative interview. Surveys will include an introductory expression of appreciation for their continued participation and reminder of reason for the trial. Incentive compensation will be given after each study procedure (survey or qualitative interview) is completed. Three reminder e-mails and/or telephone calls will be provided for each survey. For surveys, we will also include the option of completing the survey by phone or on paper if the participant prefers. We will provide flexibility in timing for the final qualitative interview to accommodate their work schedule and maximize participation. PCPs will be compensated $25 per survey they complete and $100 for the qualitative interview. PCPs will receive an additional $25 if all four surveys are completed.

- **Caregiver Survey Recruitment:** Infants seen at a participating site for a well child visit before 2 months of age will be identified through EHR data extraction on a rolling basis

(i.e. at start of the Intervention phase and then every month during the intervention phase) and NBS result reviewed if available in the EHR. Caregivers identified as speaking English (through EHR demographic data) and with an email address in the EHR will receive a survey at the start of the intervention phase if seen during the retrospective phase or at 2 and 6 months of age (+/- 1 month) if seen during the intervention phase. Email will be delivered through REDCap. Consent and survey responses will be obtained electronically.

- They will be told that participation is voluntary. They will be given the contact information for the PI and study coordinator for any questions or withdrawal from participation. The on-line survey will be ~5 minutes in length. Three follow-up requests will be sent to non-responders at ~1 week intervals. Responses will be collected within REDCap. Participants be informed that completion of each survey will enter them into a lottery for a $100 gift card. Twenty lottery gift cards will be given in total; 10 at 4 month intervals after initiation of the trial. Each caregiver that completes a survey will also receive a $10 gift card.

- **Caregiver Survey Retention:** The only study procedure is completion of a 2 and 6 month survey. Each time the caregiver completes a survey they will be entered into a lottery for a $100 gift card. Each caregiver that completes a survey will also receive a $10 gift card.

Informed Consent/HIPAA Authorization

For PCPs: One consent form, called an information sheet, has been developed for this study that incorporates consent and HIPAA information. It has been designed to not require a signature. We are applying for a waiver of documentation of informed consent and HIPAA and alteration of consent. The phone numbers of the PI, research coordinator, and Nemours IRB are provided on the information sheet for potential participants to call or email with any questions that might arise regarding the research. This information sheet will be included in the REDCap survey link from the study team and attached to the recruitment email.

An individual REDCap survey link will be emailed to potential participants. The information sheet will be the first item to appear once clicked on. After reading the page, the participant will be instructed to select an “I Agree” or “I Disagree” button as acknowledgement of consent. Participants that consent will then click a “submit” button to continue to the first questions of the survey. Those that select “I Disagree” will be thanked for their time and the survey window will close.

As informed consent is an on-going process, prior to the qualitative interview, all participants will be asked to consent to use of a recording device and will be asked to reaffirm their consent/assent to participate in the study. These statements will be documented on the interview recording.

For caregiver surveys: One consent form, called an information sheet, has been developed for this study that incorporates consent and HIPAA information. It has been designed to not require a signature. We are applying for a waiver of documentation of informed consent and HIPAA and alteration of consent. The phone numbers of the PI, research coordinator, and

Nemours IRB are provided on the information sheet for potential participants to call or email with any questions that might arise regarding the research. This information sheet will be included in the REDCap survey link from the study team and attached to the recruitment email.

An individual REDCap survey link will be emailed to potential participants. The information sheet will be the first item to appear once clicked on. After reading the page, the participant will be instructed to select an "I Agree" or "I Disagree" button as acknowledgement of consent. Participants that consent will then click a "submit" button to continue to the first questions of the survey. Those that select "I Disagree" will be thanked for their time and the survey window will close.

Protection against risk

The study procedures within this application meet the definition of Behavioral Human Subjects research. Human subjects approval from the Institutional Review Board of Nemours will be obtained prior to recruitment or any study procedures occurring. All study procedures will take place at Nemours. All study personnel will have undergone human subjects training for the protection of human subjects. Every effort will be made to assure confidentiality.

For all procedures: The PI and co-investigators will be responsible for data management and accuracy of records. The PI may assign designated qualified individuals to perform screening, facilitate surveys, conduct interviews, and communicate with study sites. Only the PI and assigned research staff under direct supervision from the PI will have access to study data. All information collected in this study will be kept confidential as required by law. Upon enrollment, subjects will be designated a unique 3-digit identification (ID) study number. Any written demographics, recordings, and transcripts will be labeled with the subject's unique identifier number. A master list of participant name or MRN and ID study number will be kept in a secure Nemours OneDrive or Sharedrive location only accessible by study investigators and purged upon completion of the study. Demographic data of PCPs will be collected directly into REDCap and then saved on a secure Nemours OneDrive or Sharedrive location for analysis. Any paper based originals will be kept in a locked office. Survey requests will be sent directly via email through REDCap or from the study coordinator to the PCP's Nemours email address or to the caregivers' email provided with the Nemours' electronic health record. Data extracted from the EHR and chart review will be kept on a secure Nemours OneDrive or Sharedrive location accessible only by study staff. All data and records generated during this study will be kept confidential in accordance with Institutional policies and HIPAA on subject privacy. The Investigator and other site personnel will not use such data and records for any purpose other than conducting the study. No identifiable data will be used for future study without first obtaining IRB approval. Data from this study will be used for research purposes only.

For qualitative interviews: During qualitative interviews, a semi-structured format with the interviewer employing a discussion guide composed of open-ended questions will be used. The PI may review selected audiotapes and transcripts of the interviews to monitor the interviewers' performance and to provide further guidance as needed. All interviewers will be trained to handle any emergencies that may arise and to be sensitive to the participants. Each semi-structured interview will be digitally audio recorded and stored on a secure Nemours OneDrive or Sharedrive location only accessible to study staff. Recordings will be labeled with the participant's 3-digit identification code. In the event a name or other identifying information is mentioned during the interview, it will be removed during the transcription process. Digital audio

recordings will be kept until transcription is complete, checked for accuracy and verified, and will then be destroyed.

Control of Bias and Confounding
We anticipate that responses could vary based on social desirability. We will test our survey and interview scripts beforehand in attempt to limit any leading questions. Additionally, we will utilize two coders for qualitative data during analysis to further reduce any reporting or interpretation bias.

Alteration of Consent, Waiver of Documentation of Consent and HIPAA Authorization

We are asking for an alteration of the consent process. Study procedures are brief and minimal/no risk. Requiring inperson communication of busy physicians to provide consent would be distruptive and time intenstive during patient care hours (ie business hours) and would be equally burdensome to parents of young infants. To decrease the burden of participation, a virtual, asynchronous consenting process will allow participants to consider study participation while away from patient care hours or child care duties and leisurely consideration of consent documents. All potential participants will have the option to speak with a study team member (coordinator or PI) before consent if desired.

We are requesting to waive a signature for consent, as the participants will be asked to document consent/assent via REDCap survey, and reaffirm consent/assent verbally prior to the qualitative interview. The surveys and qualitative interview presents no more than minimal risk to the participants. Additionally, we are not asking for any additional personal health information or identifiers apart from Greenphire information. The consent document would be the only other document connecting the survey responses and the PCP. The participants will be provided with an information sheet in recruitment emails and prior to starting the REDCap survey.

We are asking for a waiver of HIPAA authorization. The survey study involves no more than minimal risk to the participant. Additionally, the HIPAA form would be the only other document connecting survey response and the participant. Because we want the participants to be as honest as possible, we do not want the participant to feel that we can connect them to their responses and that connection could include the HIPAA documentation.

PHI will be used 1) to ensure eligibility of participants; 2) to contact for recruitment; and 3) to appropriately describe our sample. We will collect identifiable information (i.e. DOB and address) for those who receive a gift card to process study payment through Greenphire. This data will not be provided to or used for anything aside from payment. As this study will be conducted entirely remotely, it cannot be conducted without access to and use of PHI such as

Subject Completion/Withdrawal

contact information.

Subjects may withdraw from the study at any time without prejudice to their care or employment. The PI may withdraw a participant from the study if it is learned that they did not fulfill eligibility criteria. It will be documented whether or not each subject completes the study.

STATISTICAL CONSIDERATIONS

Evaluation of the Toolkit will focus on Proctor's framework of IS outcomes:[47]

Primary Endpoint

- Penetration (via interrupted time series)**:** via EHR extraction. Rate of scanned NBS results available in the EHR. Rate of documentation and disclosure of NBS and SCT result at < 2 months of age in each of the following: the problem list, the medical history section, and progress note. Evaluate monthly: retrospectively (pre intervention) and prospectively (post intervention).

Secondary Endpoint

- Acceptability: via PCP survey for each toolkit component.
- Feasibility: Percent of PCPs who use individual toolkit components.
- Adoption/Self-efficacy**:** PCP survey. Intention and confidence to document/discuss NBS/SCT result.
- Cost**:** PCP survey: Perceived time (in mins) to document and discuss NBS/SCT result, effect on other discussion topics during the visits.
- Dispersion: Via EHR extraction- Proportion of children over 2 months of age who have NBS/SCT newly documented (as above).
- Knowledge: Caregiver survey- Proportion of caregivers who accurately reported their child's NBS result.
- Inequities in documentation**:** Via chart review. Demographic data for each infant: primary care site, sex, race, ethnicity, and insurance type.
- Satisfaction: Caregiver survey.

Statistical Methods

Unit of analysis will be the individual PCP (i.e. pre/post) and by trial arm with the primary end point being the rate of documentation and disclosure of NBS and SCT (penetration).

Data will be summarized in numeric and graphic forms. Mean and standard deviation or median and interquartile range will be used for summarizing numerical variables and frequencies and percentages will be used for categorical variables. PCP's EHR documentation and disclosure of newborn screen (NBS) and sickle cell trait (SCT) carrier results will be presented by pre-post intervention as well as by intervention groups. The primary end point of the study is to compare the proportion of PCP's/ EHR documentation and disclosure of NBS and SCT between pre- and post- intervention periods.

Generalized linear mixed effects model will be used for the analysis of the primary end-point. Odds ratio with confidence interval will be provided for the documentation and disclosure. Generalized linear mixed effects model will also be used to compare the proportion of post-intervention documentation and disclosure over time (monthly) between two intervention groups. Mean score of acceptability for each intervention (A, B, C) as well as mean total score for all three interventions will be compared between two intervention groups. Similarly, other survey results such as Self-Efficacy of PCP to document NBS and time cost will be compared between two intervention groups. Also, dispersion, caregiver knowledge and caregiver satisfaction will be

compared between two groups. Two sample t-test, Mann-Whitney U-test, Chi-square test, analysis of variance (ANCOVA), liner mixed effects model, logistic regression, and mixed effects logistic regression will be used, for the above comparisons, as appropriate. PCP nested within clinic will be used as the random effects in the model as needed. All tests will be two-tailed with an overall level of significance of 0.05. Benjamini-Hochberg method will be used to adjust the level of significance for multiple comparisons. Statistical software packages SPSS, SAS (version 9.4) or latest version of R will be used for the analysis.

Sample Size Justification: This is a pilot study and there is no reported effect size on the intervention of this study in the literature. Therefore, the main goal of this study is to explore reliable effect size that would assist designing future studies with formal hypothesis testing. The primary aim of the study is to increase PCP's EHR documentation and disclosure of NBS and SCT carrier results during infancy (age less than 1 month). It has been found in a chart review that 71% of newborns over the last 3 years had a newborn screen result scanned into their chart. The goal of this study is to raise this to 90%. A sample size of 74 in each of the pre- and post-intervention period can detect the targeted difference in newborn screen of 19% with 86% power at the level of significance of 0.05. PCP documentation has been reported as 20% in the literature. We aim to raise this 50%. Again, a sample size of 43 in each of the pre- and post-intervention period would be adequate to detect the targeted difference in the PCP documentation with a power of 85% at the level of significance of 0.05. Currently 53% sickle cell traits has been documented. Once again, the goal is to raise this to 90%. A sample size of 21 infants with sickle cell traits (SCT) in each of pre- and post-intervention periods would be adequate to detect this difference in this documentation with a power of 80% at the level of significance of 0.05. In DE, the prevalence of SCT is 2.1% in overall children and about 7% in African Americans. Considering 2.1% SCT in overall children, we need 1000 infants to screen in each of the pre-and post-intervention periods, to detect the targeted difference with a power of 80% at the level of significance of 0.05. The study plans to recruit 10 PCPs in each of the intervention arm (20 PCP in both arms). About 6-10 infants are expected to be seen by a PCP in a month and 54-90 new infants are expected to be seen by a physician during 9 months of post intervention period. We expect that minimum of 540 x 2 = 1080 patients (540 for each intervention) to be seen by 20 PCPs. Therefore, a minimum sample size of 1080 infants is planned for each of the pre-and post-intervention periods for this study. The planned minimum sample size of 1080 in the post intervention period would also be adequate to determine the effect size for survey results and documentation and disclosures for each intervention arm separately.

Analysis of Qualitative Data:

Interviews will be digitally recorded and transcribed by GMR Transcription, a company vetted through Nemours that specializes in transcription of medical data, or via Teams. Transcripts will be analyzed by two reviewers. We will develop an a priori coding scheme and dictionary prior to initiation of coding based on relevant literature and the researcher's clinical expertise. The coding scheme will be revised over the first several transcripts and as needed after that. We will utilize a grounded theory approach to build a theoretical framework for understanding the interview data. We will utilize a constant comparative analysis to assess and develop themes. We will assess inter-rater reliability. Any differences in coding will be discussed and negotiated. We will utilize Dedoose (an on-line qualitative data software) to assist our analysis. Content analysis will begin shortly after interviews are initiated and continue throughout accrual.

SAFETY MANAGEMENT

Clinical Adverse Events

Some information collected from the study participants is sensitive information (attitudes, information giving preferences, etc) and may cause the subjects emotional discomfort. This risk is considered minimal and no more than what is normally associated with being a physician or nurse practitioner or routine well child care. Should the participant experience psychological distress during or after participation in this study, they will have contact information for the PI and study coordinator to connect with appropriate services.

Adverse event reporting

Since the study procedures are not greater than minimal risk, significant adverse events (SAE) are not expected. If any unanticipated problems related to the research involving risks to subjects or others happen during the course of this study (including SAEs) these will be reported to the IRB. AEs that are not serious but that are notable and could involve risks to subjects will be summarized in narrative or other format and submitted to the IRB at the time of continuing review.

STUDY ADMINISTRATION

Data Collection and Management

All study related documentation will be accessible only to research team members. All study material will be collected solely for the purpose of this investigation and will be kept in secured Nemours OneDrive or ShareDrive locations only accessible by the study staff. Only the PI and other necessary team members will have access to the human subjects' data and identifying information. All data will be identified only be Subject ID number and the master list linking the subjects' identities to the ID numbers will kept in a separate file.

A master list of subjects who participate and who refuse will be kept within Excel for study duration to assess participation rate and prevent re-contact. Names/email addresses will not be attached to actual study responses.

The Primary and co-investigators will be responsible for data management and accuracy of records. The PI may assign designated qualified individuals to perform screening and initiate survey distribution. Only the principal investigator on this protocol and assigned research staff under direct supervision from the principal investigator will have access to study data.
All information collected in this study will be kept confidential as required by law. Upon enrollment, subjects will be designated a unique study number. All evaluable data will be identified only by Participant ID number and the master list linking the participants' identities to the code numbers will be kept in an Excel/Word file separate from where the data is stored.
A master list of subjects who participate and who refuse will be kept within Excel and REDCap for study duration to assess response rate and assure appropriate compensation. Names and email addresses will not be attached to actual study responses within the evaluable dataset.

Demographic data and survey responses will be collected via participant self-report directly into the Nemours' REDCap[29] secure password-protected server unless requested differently. If a participant wished to answer questions verbally, their responses will be directly entered into

REDCap. The database will be designed with validation rules to facilitate entry of data and completion of all questions but will allow for skipping of questions per the respondents choice.

Qualitative Interviews: Qualitative interviews will be conducted over the telephone or using a video conferencing platform such as Teams. Qualitative data will consist of recordings that will be downloaded to the secure Nemours server, labeled with a study ID number, and deleted from the recording device. Recordings will then be transferred electronically via a secure method to GMR Transcription for processing. The recordings will be transcribed verbatim into Microsoft Word and stripped of any identifying information by the transcribing service, which is knowledgeable about qualitative data preparation techniques and emphasizes privacy and confidentiality. The transcripts will then be sent securely back to the study team, stored on a secure Nemours server, and checked against the recordings to ensure accuracy. The recordings will then be deleted. The PI may review selected audio files and transcripts of the interviews to monitor the interviewers' performance and to provide further guidance as needed. All interviewers will be trained to handle any emergencies that may arise and to be sensitive to the participants.

Confidentiality

All data and records generated during this study will be kept confidential in accordance with Institutional policies and HIPAA on subject privacy. The Investigator and other site personnel will not use such data and records for any purpose other than conducting the study. No identifiable data will be used for future studies, without first obtaining IRB approval.

The Investigator and other site personnel will not use such data and records for any purpose other than conducting the study. Name and email address will not be included in the final identifiable dataset and will only be used to recruit participants and provide compensation.

REGULATORY AND ETHICAL CONSIDERATIONS

Data and Safety Monitoring Plan

As this study poses no more than minimal risk to participants, the Principal Investigator will monitor study progress, ensure the accuracy and security of the data and ensure subject safety. Additionally, the Data Safety Monitoring Board of the Sickle Cell COBRE grant will be available.

Safeguards to maintain subject confidentiality and data security are described in "Data Collection and Management" above.

Risk Assessment

This project is not greater than minimal risk. There is no physical risk to the PCP, study staff, or caregiver participant in the surveys or interviews. The knowledge gained through the interviews and surveys will help assure we are providing NBS and SCT results in ways PCPs and caregivers prefer. The major risk to this study is the discussion of sensitive subject matter. We will conduct interviews virtually via telephone or Teams at a time that is convenient to the participant. In addition, each interview script will be reviewed by the Stakeholder groups to minimize any confrontational or judgmental terminology and assess for face validity and comprehension.

Since the study procedures are not greater than minimal risk, significant adverse events (SAE) are not expected. If any unanticipated problems related to the research involving risks to subjects or others happen during the course of this study (including SAEs) these will be reported to the IRB in accordance with Nemours IRB policies. AEs that are not serious but that are notable and could involve risks to subjects will be summarized in narrative or other format and submitted to the IRB at the time of continuing review.

Potential Benefits of Study Participation

The PCPs within the implementation science pilot trial may receive the following direct benefits: improved knowledge of the importance of NBS and SCT results, enhanced self-efficacy (i.e. confidence) in the disclosure of NBS and SCT results to their patients, improved rates of documentation of NBS and SCT results. If the implementation strategies are found to be acceptable, feasible, and enhance penetration of NBS and SCT result documentation and disclosure they will be further tested within a multi-site implementation trial.

The caregiver participants may not receive any direct benefit to completing the survey. They may receive direct benefit because their PCP is a participant since they may receive additional education from their PCP regarding their child's NBS results. Results from the caregiver participants will help to improve the delivery of care for others in the future.

Payment to Subjects/Families

All compensation will be provided via Greenphire gift card: Name, address, and date of birth will be required to facilitate gift cards.

- Primary Care Provider (PCP) Survey Incentives (1 every 6 weeks during the intervention phase x 4). $25 per survey. An additional $25 if all four surveys are completed.
- PCP Qualitative Interviews: $100 per interview
- Caregiver Participant Survey Incentives: Each caregiver who completes a survey will be entered into a drawing for a $100 gift card for each survey they complete. There will be 20 lottery gift cards given. Each caregiver that completes the survey will also receive a $10 gift card.

PUBLICATION

The results of this study may be presented at professional meetings or published in a professional journal. Any personal identifying information will not be included in these presentations or publications.