

Title: SCTaware: A Comprehensive Program to Increase Sickle Cell Trait Knowledge and Awareness Among Parents of Infants Identified by Newborn Screening

Version Number: 1.1

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1. Project Summary/Abstract

1.1. Background

Nearly 3 million people in the United States have sickle cell trait (**SCT**) and approximately 2,000 infants are born annually with sickle cell disease (**SCD**).¹ SCD is a chronic blood disorder that can lead to pain, stroke, and early mortality.² Individuals with SCT are typically asymptomatic, but to make informed reproductive decisions, they must be knowledgeable about their SCT status, SCD, and that if their reproductive partner also has SCT, because each of their children has a 25% chance of having SCD and a 50% chance of having SCT. Despite universal newborn screening (NBS) that reliably identifies infants with SCT, >80% of individuals of childbearing age with SCT do not know their status.^{3,4} The programs intended to notify and educate parents of infants with SCT identified by NBS and to promote parental testing vary by state and have not been rigorously evaluated. We identified at our own center that these parents frequently have low health literacy and low baseline SCT knowledge which are factors that may limit their ability to achieve high SCT and SCD knowledge with the existing education programs. We also found that despite a presumed high prevalence (50%) of SCT among these parents and their reported intentions to obtain testing on their own, these parents did not obtain SCT testing.⁵ It is unclear if low SCT knowledge, decisional conflict about whether or not to obtain testing, or testing costs are barriers or if offering on-site testing with education could increase SCT status awareness among these parents. There is a critical need to develop an effective education and testing program for these parents so that they ascertain their own SCT status, are equipped to inform their children of their status when they are older, so more individuals with SCT can make informed reproductive decisions in the future.

1.2. Rationale

Most individuals with SCT do not know their status. **The optimal strategy to increase SCT knowledge and status awareness remains unknown.** An effective strategy to increase SCT awareness may be to provide more effective education to parents of infants with SCT identified by NBS (SCTaware) and to encourage these parents to be tested. First, these parents can inform their affected children of their status when they are older. Also, these parents need to know their own status, since they are at imminent risk of having subsequent children with SCD because they have a 50% presumed prevalence of SCT and are already of childbearing age. Finally, SCTaware could have additional benefits for parents of children with SCT, since qualitative studies of parents with traits for other genetic diseases suggest that effective trait education decreases parents' anxiety, increases their preparedness, and increases their sense of control about the potential of having a child with a genetic disease.⁶ Finally, while some aspects of educating parents of children SCD may differ compared to parents of children with SCT (e.g., different lived experiences and motivations for learning), these parents have similar baseline demographic and cultural experiences and frequently have low health literacy and baseline knowledge about SCT and SCD. Developing the SCTaware program, therefore, could inform how to more effectively educate parents of children with SCD to improve outcomes for children with SCD. For example, parents of children with SCD need effective education to obtain required screenings and to be able to make informed decisions about whether to accept and adhere to SCD modifying therapies (e.g., hydroxyurea) for their children.

1.3. Aims

Aim 1. Develop an effective SCT education program for parents of infants with SCT identified by NBS.

1a. Determine the feasibility of implementing a SCT education program (SCTaware) that is appropriate for all parents, including those with low baseline knowledge and low health literacy. (Phase I)

1b. Determine if significantly more parents who receive SCTaware education have high SCT knowledge 6 months after receiving the education compared to baseline.

Hypotheses: SCTaware will require limited additional resources to implement compared to Ohio's current program and it will result in significantly more parents to have high SCT knowledge.

Approach: A comprehensive team will develop SCTaware education through iterative review and revision of the current in-person program. The time required to train the educator and to administer SCTaware to parents will be recorded. SCTaware will be feasible if educator training can be completed during administrative time and if SCTaware can be provided to parents within Ohio's existing appointment schedule (Phase I). The SCT Knowledge Assessment will measure parents' knowledge before, after, and 6 months post-SCTaware (Phase II).

Aim 2. Determine the barriers that prevent parents of infants with SCT from getting SCT testing.

2a. Determine the percentage of parents who obtain on-site SCT testing at time of SCTaware education. (Phase II)

2b. Determine the range of decisional conflict that parents may experience regarding SCT testing before and after receiving SCTaware education. (Phase II)

Hypotheses: Making on-site testing available will result in $\geq 50\%$ of parents who are educated to obtain testing. Parents will have decisional conflict about whether to obtain testing that will be reduced with SCTaware education.

Approach: Parents will self-report their SCT status before receiving education. The number of parents that obtain on-site testing will be recorded. We will use the Decisional Conflict Scale to measure parents' uncertainty about obtaining SCT testing before and after education. Parents who do not obtain testing will be asked to report their other testing barriers (e.g., cost, needle phobia, other).

1.4. Methods

This will be a two-phase prospective study.

1.5. Population

Biological parents of infants with SCT identified by NBS who are referred and present for in-person SCT education at NCH.

NCH is a pediatric, tertiary care center of The Ohio State University College of Medicine, Department of Pediatrics. Typically, 500 of the 800 infants with SCT each year in this region have at least one parent attend the one-time, in-person SCT education session at NCH that is funded by the Ohio Department of Health.

1.6. Expected Outcomes

This proposed work will provide important insights about how to effectively educate this underserved population about SCT, the SCT testing barriers that exist for parents of children with SCT, and if making on-site testing convenient increases SCT status awareness.

General Information

2.1. Title of Protocol

SCTaware: A Comprehensive Program to Increase Sickle Cell Trait Knowledge and Awareness Among Parents of Infants Identified by Newborn Screening

2.2. Date of Anticipated Enrollment

Phase I April 2019-October 2019

Phase II October 2019-April 2020

2.2. Anticipated Funding

R03 from NHLBI

2.3. Principal Investigator

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2.4. Co-Investigators

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2.5. Stakeholders

Toyetta Kirk
Latrice Johnson

2.6. SCT Educator

Ismahan Adan

2. Study Design

2.1. Type of Study

Two-phase prospective study

2.2. Population

Parents of infants with Hemoglobin S-trait identified by NBS who present for in-person SCT education will be recruited for both phases of the study

2.3. Comparison

Phase I

Feasibility	Implementation resources: time required to train SCT educator and deliver SCTaware education
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Phase II

Parental Knowledge	SCT Knowledge Assessment (SCTKA): administered before, immediately after, and 6 months after SCTaware
On-site testing Decision Conflict Other Potential Testing Barriers	Percentage of parents who obtain SCT testing Decisional Conflict Scale Parental anxiety, testing cost, SCT Knowledge

2.4. Inclusion

1. Adult biological parents of infants with Hemoglobin S-trait identified by NBS who present for SCT education at NCH.
2. English proficiency will be required

2.5. Exclusion Criteria

1. Parents who self-report that they do not have functional verbal English (report that they are not proficient) or if they request an interpreter for the education session.
2. Parents who have previously attended an education session about an abnormal hemoglobinopathy trait
3. Parents who self-report that they have a child with SCD
4. Parents who self-report that they have SCD.

Note: More than one parent of each child with SCT could be eligible for the study, if they meet all inclusion and exclusion criteria.

a. Justification for Inclusion/Exclusion Criteria:

This study requires participants to be able to understand communications in English and since the surveys have not been validated in other languages, participants or consenting adults will be required to speak English. Although other hemoglobinopathy traits besides S-trait are identified on newborn screening, S-trait is the most common SCT identified on the newborn screen and the content of these sessions is the most consistent.

b. Study Duration

Phase I- 1 session

Phase II- 6 months

c. Time to Achieve Planned Accrual

Phase I-1 month for initial review, 3-6 additional months to iteratively recruit to evaluate the revised program

Phase II-18 months

5. **Methods**

a. **Prospective Participant Identification (Screening)**

NCH is a pediatric, tertiary care center of The Ohio State University College of Medicine, Department of Pediatrics. Parents of patients with an abnormal newborn screen for S-trait are referred to NCH for hemoglobinopathy education by their primary care providers and typically, 500 of the 800 infants with SCT each year in this region have at least one parent attend the one-time, in-person SCT education session at NCH. The research team will coordinate with the hemoglobinopathy education team that schedules these sessions to identify prospective parents of children with S-trait and approach these biological parents to participate when they present for the education.

b. **Recruitment**

These education appointments are scheduled 3-4 weeks in advance by an administrative assistant, parents are mailed an in-person education appointment reminder after the appointment is made, and the administrative assistant calls to remind parents of this appointment approximately 1 week before the appointment. To increase the number of potential subjects that can be enrolled on a particular day, the administrative assistant will try and schedule multiple prospective parents on the same day of clinic.

Prospective parents will be identified in coordination with the hemoglobinopathy education team (Dr. Creary is a member of that team). Upon arrival to the SCT counseling session, the educator will inform the prospective parent(s) that there is a research study that they may be eligible for and a clinical research coordinator will then approach the parent(s) to explain the study and ask if the parent(s) would like to participate.

c. **Informed Consent**

All participants will receive a thorough explanation of the study and a copy of the consent for their records.

d. **Enrollment**

Phase I: Approximately 5-10 participants

Phase II: 128 participants

e. **Withdraw**

Participants will be able to withdraw from the study at any time.

f. **Study Procedures**

Phase I:

Study Design This will be a prospective study of parents of infants with SCT identified by NBS who are referred and present for in-person SCT education at NCH.

Program Evaluation To evaluate the current program, 5 parents who attend NCH’s in-person education session will be recruited to have their standard SCT education session video-taped, timed, and reviewed by the SCTaware Team which includes the includes: principal investigator, co-investigators, two parent stakeholders (a parent of a child with SCT and a parent of a child with SCD), and the SCT educator). Since >50% of parents had low health literacy and baseline knowledge in our preliminary study, we anticipate that this small sample will include at least 3 parents with low health literacy and 3 parents with low SCT knowledge, but if it does not, we will continue to enroll parents until at least 3 parents with these baseline characteristics are included in the initial program evaluation. Enrolled parents will self-report their SCT status, complete a demographic survey, and the sickle cell trait knowledge assessment (SCTKA) before the education. They will also complete the SCTKA, an SCT Testing Needs Assessment (STNA), and an Education Effectiveness Survey (EES) immediately after the education (Phase I. Study Measures).

Phase I. Study Measures

Surveys	Before	After
	Day 1	Day 1
Demographics	x	
Self-reported SCT status	x	x
NVS	x	
SCTKA	x	x
EES		x
STNA		X

The Team will review the participant videos and the SCTKA results to identify where the education was not clear or appropriate for those with low health literacy (i.e., too much medical jargon) or did not result in learners being able to correctly answer the corresponding SCTKA question. To determine if the existing print visual aids are suitable for those with low health literacy, multiple validated tools will be used. The Suitability Assessment of Materials (SAM) will assess the content, literacy demand, graphics, layout and typography, learning stimulation and motivation, and cultural appropriateness of the materials.⁷ The CDC’s Clear Communication Index (CDCCI) will evaluate the clarity of the materials⁸ and the print and audio visual versions of the Agency for Healthcare Research and Quality’s Patient Education Materials Assessment Tool (PEMAT) will evaluate the understandability and action-ability of the materials.⁹ Finally, the STNA, EES and parent stakeholder input will identify how to modify the education so that it applies Adult Learning Theory principles and promotes parents to want to learn about SCT and SCD, to obtain SCT testing, and to use their knowledge to make health decisions.

Creating SCTaware Education Once the five sessions are evaluated, the SCT educator will receive additional health literacy- and Adult Learning Theory- based education. The educator will complete an in-depth experiential health literacy training and the online Always Teach-back! Toolkit interactive learning module. From the videos, the Team will create a plain language glossary for the SCT educator to use to reduce medical jargon during the education. The Team will provide specific feedback on strategies to make the education more understandable (e.g., open-ended questions, teach-back method, and Chunk and Check methods).¹⁰ The educator and a health literacy expert will role-play to increase the educators’ comfort using these techniques. To ensure the educator consistently uses the health literacy strategies during all sessions, the TEAM will select the two SCTKA items that the educator will consistently use the teach-back method when educating parents about these items. Methods identified by stakeholders and on parents’ STNA and EES surveys will be employed to further encourage parental learning. The time required to complete this additional educator training will be recorded. The Team will convert the print visual aids to be appropriate for those with low health literacy and also to be more interactive when they develop the SCTaware ebook that will be accessible on internet-enabled devices. The ebook will also include short videos, image hotspots, and quizzes to promote knowledge retention and to provide parents with a take-home tool to use to promote self-care in the Adapted Health Literacy Conceptual Model (Figure 2) and negotiate conversations about SCT with their family. The visual aids and ebook will be revised until they achieve pre-determined SAM, CDCCI, and PEMAT scores.

Pilot Testing SCTaware Education Two new parents will be recruited to pilot test the SCTaware education, to have their SCTaware session video-taped and to complete the associated surveys, but additional parents will be recruited, if needed, so that at least one parent with low health literacy and baseline SCT knowledge pilot test the new program. The Team will the revise the SCTaware program again after reviewing their videos and surveys and pilot test the revised SCTaware program on two additional parents (at least one with low health literacy and baseline SCT knowledge). This process will continue iteratively until the Team agrees that the SCTaware program is finalized. The time required to deliver the SCTaware education during pilot testing will

be recorded. If it is not feasible to deliver SCTaware (i.e. SCTaware education cannot be delivered within the context of the current in-person SCT program), the current in-person SCT education will be tested in Phase II.

Parent stakeholders The NCH hemoglobinopathy team will help the Research Team identify the parent stakeholder of a child with SCT who has previously received the standard SCT education and the parent of a child with SCD who is currently followed by the NCH Hemoglobinopathy Team. These stakeholders will be compensated \$1000 each for their time to review the sessions and to provide their feedback, insights, and lived-experience that will inform the SCTAware education.

Phase II:

Study Design Phase II will be a six-month prospective study.

Recruitment Using the same recruitment strategy as in Phase I, we will recruit 128 parents.

Study Procedures Prior to receiving SCTaware, participants will self-report their SCT status and complete the demographic survey, SCTKA, State Trait Anxiety Index for Adults (STAI), and Decisional Conflict Scales (DCS).

Surveys	Before Day 1	After Day 1	Follow Up #1 1 mo	Follow Up #2 ≥6 mo
Demographics	x			
Self-reported SCT status	x		x	x
NVS	x			
SCTKA	x	x	x	x
DCS	x	x	x	
STNA		x		
STAI	x	x		
EES		x	x	x
ebook Survey			x	x

Table 2. Phase II Study Measures

SCTaware sessions will be timed. Participants will complete the DCS, EES, STNA, SCTKA, STAI immediately after their session and a research coordinator will contact parents by telephone one month and ≥6 months after their session to complete additional surveys (Table 2).

SCT testing Barriers that prevent parents from obtaining SCT testing must be identified in order to increase SCT status awareness. Unresolved decisional conflict about whether or not to obtain SCT testing is a potential barrier, since the test results can have high-stakes for individuals who are identified as having SCT (e.g., being at risk for having a child with SCD). Decisional conflict is the psychological distress experienced when making a choice and it can lead to delayed and ineffective decision-making. Because decisional conflict can occur when individuals are uninformed about their options, effective education has the potential to reduce conflict and increase decision satisfaction. We will, therefore, use the Decisional Conflict Scale (DCS) to explore if parents experience decisional conflict about whether or not to obtain SCT testing and if SCTaware reduces this conflict (*Aim 2b*). Another potential barrier is that most parents of these children are African American and may not have access to a consistent provider who can order this testing.³⁸ To address this potential barrier, SCT testing will be available on-site, but not paid for, to participants after the educational program. SCT testing includes a CBC and a hemoglobin electrophoresis, and a paper order, signed by Dr. Creary (the PI and hematologist), will be provided to participants who wish to obtain this testing. Parents will be provided contact information for common insurance companies to determine if the testing will be covered but also informed of the estimated cost if not covered (~\$250). Finally, parents will be asked to report if they have other testing barriers (e.g., needle phobia, already aware of SCT status). Dr. Creary will review and interpret all test results and the SCT results will be mailed to tested parents. The research coordinator, using the teach-back method, will confirm test result receipt when parents are contacted to complete follow-up surveys.

Interim analyses will confirm that SCTaware does not result in more parents to have lower knowledge at 6 months. Several random sessions during Phase II will also be recorded and reviewed to confirm SCTaware education fidelity and that the teach-back method was used for the critical items, or if educator re-training is required.

6. Measures

Phase I:

Demographic Survey Participants will self-report their age, SCT status, gender, race, ethnicity, primary language spoken, employment status, household income, and highest education level achieved.

NVS Participants will complete the NVS health literacy tool prior to their SCT education session. The NVS can be completed in less than three minutes and is highly sensitive for detecting adults with low health literacy.¹¹ Scores range from 0-6, and scores <4 will be considered low.

SCTKA Participants will complete the 8-item SCTKA (Table 2) before and after they receive education. This tool was used to quickly assess SCT knowledge during our preliminary study⁵ and to be understandable by those with low literacy levels. It was pilot tested in parents of children with SCT to ensure readability and seven of the items were previously published.¹² Scores <75% correct will be considered low.

STNA The SCTaware program in Phase II will make on-site SCT testing convenient to parents. To identify parents' testing needs and potential methods to use during SCTaware to promote parental learning and testing, parents will complete the STNA. It will ask parents about their knowledge of their SCT status and their intentions for obtaining SCT testing and it will be developed using the results of a qualitative study of pregnant women who identified the factors they found important when deciding whether or not to obtain SCT testing and will be pilot tested for readability on parents who come for standard in-person education and revised based on their feedback.¹³

EES Participants will complete a modified version of the education satisfaction survey that was used in the preliminary study.⁵ The EES will gauge parents' reported readiness and responsibility for learning about SCT, if the education they received met their learning goals, if they have the self-efficacy to apply their knowledge to make informed reproductive health decisions, and if they have (or plan to) notify family members or their communities about SCT and their SCT status. Participants will provide open-ended feedback and offer methods to improve and/or broaden the SCTaware program to increase SCT knowledge and awareness.

Phase II:

Demographic, NVS, SCTKA, STNA, and EES Same as in Phase I.

Self-Reported SCT Status and Obtaining SCT testing Parents will self-report their SCT status multiple times during the study to determine if self-reported SCT status is reliable and accurate and impacts their decision to obtain testing. The percentage of parents who obtain on-site testing will be recorded to determine if on-site testing increases SCT status awareness and if tested parents can accurately recall their status ≥6 months after they were tested.

STAID (short form) Qualitative studies suggest that SCT education has the potential to reduce parental anxiety.⁶ The STAID short form is a 20-item sensitive and reliable measure of state anxiety and will be used to quantify parents' anxiety before and after receiving SCTaware education (Table 3).^{14,15}

DCS Participants will complete the 16-item validated tool to determine if decisional uncertainty is a SCT testing barrier for parents and if SCTaware increases parents' perception that they made an informed decision about whether to obtain testing. DCS has predictive validity over a range of decisions, and in studies of decision-supporting interventions, effect sizes ranged from 0.4-1.2.¹³⁻¹⁵

ebook Survey While most parents will have an electronic device, the non-interactive ebook content will be printed. Parents will be asked to report the type of device(s) they have since readability may differ by device, and to provide feedback on the content and format. They will also be asked to report how often they accessed the ebook since their in-person education to support their learning or to share information about SCT with their families and communities.

Time to Deliver SCTaware Education and Notifications Although health literacy-based methods can marginally increase the time to deliver information, in return, more learners may comprehend the information presented.¹⁶ We will compare the time to provide SCTaware education (Phase II) to Ohio's current in-person SCT education (Phase I). We anticipate that familiarity with SCTaware will reduce delivery time. The time to deliver parents' test results will also be recorded to determine if this testing can be included within the SCTaware program.

7. Survey Completion- A research coordinator will read all survey items to participants and record their responses electronically. This format will be used because participants are only required to have functional verbal English and the NVS was validated using an interview format. This will also ensure that the method of survey completion is consistent in Phase II because the follow up surveys at 1 and 6 months will be over the telephone.

8. Study Compensation

Phase I: Participants will receive \$25 for completing Phase I.

Phase II: Participants will receive \$25, \$5, and \$10 for completing the initial, 1-month, and 6-month surveys, respectively.

9. Safety

a. Privacy

Participants' data will be de-identified to minimize the risk that participants' survey responses will be intercepted by a third party. The assessments will be completed electronically and stored in Redcap.

b. Data Storage

The survey results will be stored on a research drive that is maintained behind NCH secure firewall. The research data will be retained for 7 years after publication of the data, in accordance with standard research methods. After this time all collected data will be destroyed.

10. Data Management and Statistical Analysis

a. Data Monitoring

Study data will be collected, downloaded, and stored on RedCap. This database will only contain de-identified patient adherence data and will be linked by study ID number.

b. Statistical Methods and Analysis

Phase I: SCTaware will be deemed feasible if educator training and the time to deliver SCTaware can be delivered within the context of the current SCT program at NCH.

Phase II: During our preliminary study, ~40% of parents had high SCT knowledge prior to receiving education and 90% percent of parents achieved high SCT knowledge immediately after they received the current standard education program,⁵ however, it was unclear if this knowledge was sustained at 6 months because few parents completed the 6-month SCTKA. To determine if significantly more parents who receive SCTaware education have high SCT knowledge 6 months after receiving the education compared to baseline (Aim 1b, the primary aim for Phase II of the study), we will enroll 128 (n=98 evaluable) parents on Phase II of the study. This sample size was calculated using a two-sided, McNemar's test and has 80% power, with a 5% alpha, and assumes 30% attrition (Aim 1b).

Study Design		Knowledge Level at 6-month after SCTaware	
		Proportion with High Knowledge	Proportion with Low Knowledge
Knowledge Level pre-SCTaware Education	Proportion with High Knowledge	30%	15%
	Proportion with Low Knowledge	35%	20%

Similar to our preliminary results, this example assumes that 45% of participants will have high SCT knowledge before receiving SCTaware and 65% will have high SCT knowledge 6 months after receiving SCTaware education. Therefore, a sample of 98 evaluable participants will have 80% power to detect an odds ratio of 2.33 when the proportion of discordant pre and post-SCTaware pairs is 0.5.

11. Quality Assurance

a. Training of research Staff

Prior to enrollment, the research team will meet to review all protocol procedures, including screening, enrollment, and survey administration. The PI will insure that all research staff who consent, enroll, and complete study follow up assessments are adequately trained. The research team will meet at least monthly during active enrollment and to discuss any potential problems and then at least bi-monthly while participants are still actively participating in the study.

12. Dissemination of Results and Publication

a. Planned Journal for Submission

Phase I: J Genet Counseling

Phase II: Pediatrics

b. Authorship

Susan Creary, Mary Ann Abrams, Deena Chisolm, Sarah O'Brien, Kristin Zajo, John Mahan

13. Timeline

Dates	Phase
April 2019-October 2019	Phase I
October 2019-April 2021	Phase II

14. Anticipated Problems and Solutions

1. Meeting recruitment goals is a challenge of all studies, however, this study will use the same strategy that successfully enrolled 91% of eligible parents in <12 months. To further encourage participation, this study will also include participation incentives and we have also expanded the eligibility criteria to include parents with functional verbal English (do not request interpreter services).
2. Recruitment bias is possible, since only parents who present for in-person education will be included. Once SCTaware is developed, however, adapting the program for electronic delivery may increase the number of parents who can receive this strategy.
3. The primary aim of Phase II of the study is to determine if more parents achieve high SCT knowledge at 6-months. Since these parents do not necessarily have ongoing communication that is standard with the SCT educator and our prior experience suggests that study attrition may be higher than in other studies, we will enroll more parents in anticipation of higher attrition (30%). Also, to reduce attrition during Phase II, we will collect multiple contact methods for participants at enrollment (email, address, telephone number) and confirm these are still accurate at the one-month follow up. Small monetary compensation will also be provided throughout to encourage completion of all study activities.

4. The Ohio Department of Health only has resources to provide one-time SCT education for each parent and is aware and has endorsed this project (provided a letter of support with the R03 grant application). If participants request that they want an additional session, the Team will provide a group SCT session for participants after they complete their 6-month surveys.

5. The research team will inform parents of the potential that SCT testing could identify non-paternity. To ensure privacy, the research team will only disclose SCT test results to the tested parent.

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