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Promoting Sickle Cell Trait Awareness and Education: A Typology of Interventions in the United States to Inform Ongoing Efforts to Patients and Providers

Stacey Cunnington, Jacey Greece

Department of Community Health Sciences, Boston University School of Public Health, Boston, Massachusetts, USA
Email: staceyac@bu.edu, jabloom@bu.edu

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Abstract

Research Background: Sickle cell trait has no treatment or cure and predominantly affects people who are Black, but can affect anyone of any race or ethnicity. While commonly incorrectly considered benign by providers and the public, people with a sickle cell trait experience life-threatening outcomes that are exacerbated by extreme conditions. There is a severe lack of awareness and understanding of sickle cell trait and the associated health complications among sickle cell trait carriers and healthcare providers. **Purpose/Aim:** Interventions that aim to improve awareness of sickle cell trait differ in approaches and are not well documented in the literature. This typology aims to highlight current efforts to inform targeted interventions that raise awareness through consistent messaging, educate people and providers on sickle cell trait and the related health complications, and support the design and implementation of comprehensive sickle cell trait awareness initiatives. **Methods:** We conducted a scoping review of United States-based sickle cell trait interventions and performed a content analysis to identify the categories and characteristics of these efforts. We then organized the results into a typology according to established protocols. **Results:** Among 164 interventions, twenty-five (15%) met the typology inclusion criteria described above and were grouped into categories: Seven of twenty-five interventions were *Educational Interventions* (28%), three of twenty-five interventions (12%) were *Combined Screening and Educational-Based Interventions,* eight of twenty-five interventions (32%) were *Policy and Guideline-Based Intervention,* and six of twenty-five interventions (24%) were *Sickle Cell Trait Organization-Led Interventions.* **Conclusions:** There is a lack of consistency in messaging across interventions whether delivered by credible healthcare institutions or national

organizations, which can result in lack of education and awareness and confusion around sickle cell trait. Categorizing interventions through a typology allows clarity and informs consistency in messaging, which should be at the forefront of future sickle cell trait efforts.

Keywords

Sickle Cell Trait Awareness, Sickle Cell Trait Messaging, Sickle Cell Trait Intervention, Typology, Scoping Review

1. Introduction

In the USA, African Americans carry the burden of sickle cell trait with 3.6 million, or 1 in 13 people who are African American or Black affected [1]. However, anyone of any race or ethnicity can have a sickle cell trait and, in one study, the trait has been identified in 7.3% of Black newborns, 6.9% of Hispanic newborns, 2.2% of Asian American and Native Hawaiian/Pacific Islander newborns, and 0.3% of White newborns [1]. There is no treatment or cure for having a sickle cell trait with potential associated health outcomes treated on an individual basis.

Sickle Cell Disease (SCD), which is a different condition than sickle cell trait, is typically the focus of awareness and education efforts in this space. People with sickle cell trait carry only one copy of the altered hemoglobin gene and in contrast, people with sickle cell disease carry two copies of the altered hemoglobin gene, which often causes more devastating health complications than in people with sickle cell trait, but this is not always the case [2]. While commonly incorrectly considered benign by providers and the public, people with a sickle cell trait experience a myriad of debilitating and life-threatening outcomes [3] that are initiated or exacerbated by extreme conditions, such as intense physical exertion, heat, dehydration, and high or low altitude [4]. This can lead to outcomes such as renal medullary carcinoma (RMC) [5], hematuria (blood in the urine) [6], hyposthenuria (inability to properly concentrate urine) [7], chronic kidney disease (CKD) [8], venous thromboembolism [9], splenic infarction [10], exertional rhabdomyolysis [11], exercise-related sudden death [12], glaucoma and hemorrhage post-hyphema [13], and, likely others [14]. Having a sickle cell trait can lead to negative health outcomes; people with the trait have been found to have up to 95% protection against malaria [15]. Exacerbated by the incorrect belief that sickle cell trait is benign, systemic racism, and underfunding of sickle cell trait research, the condition is largely misunderstood and comprehensive and consistent communication is lacking [16] [17].

Sickle cell disease (SCD) was the first heritable disease to be identified and to receive federal funding for testing in 1972 [18], [19] but since the initial period, research on the health implications and efforts to raise awareness have been minimal for both sickle cell disease and sickle cell trait [16]. The United States adopted a voluntary and targeted sickle cell trait screening program in 1972,

which was replaced by a universal newborn blood screening program in 2006 [18] [20] making available sickle cell trait testing at birth to individuals born in the United States after 2006. There are generations before that time, however, who are still unaware of their status with only 16% of all individuals in the United States aware of their sickle cell trait status [21]. Testing for these individuals can occur at their doctor's office or various local organizations through a blood test (either capillary electrophoresis, high performance liquid chromatography, or genetic testing), but these methods are often prohibitively expensive and time-consuming [22] and result in underutilization of testing.

A variety of factors contribute to low awareness of sickle cell trait status including not being screened at birth (e.g. prior to screening program, immigration to the United States); lack of follow-up counseling after a positive newborn screen [23]; and, inconsistent recording of test results [24]. The lack of follow-up counseling for people with a sickle cell trait has led to what experts describe as a "neglected opportunity" to inform affected populations [25]. Additionally, people with a sickle cell trait are commonly misdiagnosed as having other health conditions due to lack of awareness by providers, which is compounded by structural racism and underfunding of research and awareness in this area [16] [17].

Many awareness and educational interventions for the sickle cell trait community have been developed for people who do not know their sickle cell trait status. These interventions have also been targeted toward people who do know their status but who are at high risk of experiencing related complications, such as athletes, or those in the military because high intensity exercise is a risk factor for experiencing certain complications if precautions are not taken [26]. Sickle cell trait interventions are implemented by either medical institutions such as hospitals, or organizations with a mission to address sickle cell trait. While there are many sickle cell trait-focused organizations, the tailored education and awareness about sickle cell trait is often overshadowed by the organizations focused on SCD, ultimately providing very limited information about sickle cell trait and its related health complications. There are efforts by existing organizations that solely target the sickle cell trait community to provide education and resources but there is an over-reliance on messages that are often inconsistent, incorrect, outdated due to lack of research, or focus too heavily on one outcome (such as reproduction) and not enough on the variety of possible outcomes. For example, results of a thorough environmental scan of these organizations found that some organizations incorrectly report that sickle cell trait is benign and will not cause any issues for the carrier, while other organizations correctly report sickle cell trait can cause debilitating complications and even death [3]. Thus, the need for consistent, accurate, and holistic information around sickle cell trait is essential for providers to be properly trained to deliver education to their patients so patients are correctly informed about sickle cell trait and can make educated decisions to promote positive health outcomes.

Typologies resulting from comprehensive scoping reviews seek to understand human behavior, leading to more successful health promotion efforts, by bridg-

ing the gap between cross-case and within-case approaches to data analysis and focus on exploring similarities and differences in participants' whole accounts of their experiences [27]. An overview of sickle cell trait interventions, the awareness efforts surrounding them, the organizations supporting the interventions, and the impact of the efforts is not well documented in the literature. To address our research questions around what educational and awareness interventions exist around sickle cell trait, we aimed to uncover the landscape of existing sickle cell trait intervention. Understanding the landscape allowed us to uncover the gaps in education and awareness and make recommendations to move toward improving the low awareness in the United States. To do so, we conducted a scoping review of United States-based sickle cell trait interventions and the messaging within those interventions and performed a content analysis to identify the categories and characteristics of different efforts in an effort to inform future efforts. The typology, which is the first to focus on sickle cell trait interventions, provides sickle cell trait organizations and other implementers comprehensive findings to directly educate patients and guide recommendations about targeted interventions to raise awareness through consistent and scientifically-accurate messaging, to educate people and providers on sickle cell trait and the related health complications, and to support the design and implementation of comprehensive sickle cell trait awareness initiatives across the field. The result of this comprehensive education and messaging is to allow people responsible decision-making to promote their individual health and wellbeing.

2. Methods

2.1. Scoping Review

The development of the typology started with a scoping review on a topic that has not been comprehensively reviewed to date [28]. Specifically, the research questions included the following: 1) "What awareness and educational interventions targeting those at risk of having a sickle cell trait exist in the United States?"; and 2) "What educational interventions exist that target those who have a sickle cell trait but do not understand the related health complications?" Literature searches were performed using the following databases: Pub Med, JSTOR, CINAHL, and Science Direct for papers published between 2000 and the present day. Other sources included searches in Google Scholar as well as a snowballing approach to locate additional manuscripts by scanning the manuscript reference lists. Additionally, stakeholder interviews and focus groups with leadership from sickle cell trait organizations (n = 23) resulted in further recommendations for exploration. The following search terms were used in isolation or in combination: "Sickle cell trait interventions", "Sickle cell trait programs", "Sickle cell trait education", "Sickle cell trait screening program", and "Sickle cell trait support".

2.2. Typology Components

The typology is organized into two components: 1) Intervention Component

Categories and 2) Typology Categories. Intervention Component Categories consist of how the intervention is described to provide a holistic picture of the intervention, as in the case of this typology, the strategies or activities, setting, target population, intervention outcomes, and current operating status of the program. Typology Categories are how the interventions are grouped based on similarities identified in the Intervention Component Categories, so in the case of this typology, the interventions are grouped into *Educational Interventions, Combined Educational and Screening Interventions, National Screening Policy or Guideline-Based Interventions*, and *Sickle Cell Trait Organization-Led Interventions.*

2.3. Intervention Inclusion Criteria

The sickle cell trait community is defined as those who have a sickle cell trait, those who are at high risk of having a sickle cell trait but do not know their status, or caregivers of those with a sickle cell trait, such as parents of newborns. Inclusion criteria included interventions after 2000 (unless they were a major intervention in the field or currently active); interventions that target the sickle cell trait community as defined above; and, interventions that targeted those with sickle cell trait even if it also targeted people with SCD. If an intervention did not have evaluation outcomes, they were still included if evaluation was ongoing and/or they fit the other inclusion criteria in order to access the most available information possible. This is especially relevant for the *Sickle Cell Trait Organization-Led Interventions* category, as these organizations often did not report outcomes or effectiveness of their interventions, most often because the programs are ongoing or were implemented in the last 3 years [29] [30] [31] [32]. Interventions were excluded if they occurred outside of the United States, were clinical interventions, or interventions aimed at healthcare providers given the focus of this typology is awareness and education efforts that target the sickle cell trait community in the United States. Websites were excluded if they did not contain information about a particular intervention or its evaluation. Only interventions in the United States were included in order to capture and subsequently inform targeted awareness efforts [21].

2.4. Content Analysis

According to previously established typology methods [33], two reviewers, SC and JG, reviewed each identified manuscript to perform a content analysis and determine the intervention component categories for each manuscript. Reviewers completed comparison of each intervention component category until each category had a distinct set of characteristics with no overlap. Discrepancies were discussed until consensus was reached. After all manuscripts were reviewed for inclusion eligibility, the following five Intervention Component Categories were determined: *Strategies or Activities of the Program, Setting, Target Population, Outcomes,* and *Status of the Program*. We then analyzed program components and created the typology. We iteratively compared and contrasted different pro-

gram components to identify patterns and groupings that were often observed together or appeared to influence other program elements. We also compared any overlapping components or patterns found in multiple categories to further explore these classification definitions, identifying distinguishing characteristics. We continued this process through five iterations until each category had a distinct set of characteristics, resulting in a draft typology. Finally, we validated the draft typology against the research articles to ensure accuracy, with each article fitting distinctly into each category.

2.5. Typology Grouping

Once all articles were collected and reviewed and data was extracted, the constant comparative method [34] was used to create a typology. The articles were grouped into the following typology based on the type of intervention with the following Typology Categories: 1) *Educational Interventions*, delivered in a variety of formats and settings, for people who have a sickle cell trait or people who are at increased risk of having a sickle cell trait; 2) *Combined Education and Screening-Based Interventions* that take place in medical settings or the community; 3) *National Screening Policy or Guideline-Based Interventions* that are implemented to mandate sickle cell trait screening for specific populations, and 4) *Sickle Cell Trait Organization-Led Interventions* that are implemented to raise awareness of sickle cell trait or increase testing rates across the United States or in specific populations (**Figure 1**). These four categories emerged as the most common types of education and awareness interventions for sickle cell trait.

3. Results

3.1. Content Analysis: Informing the Intervention Component Categories for the Typology

Among 164 interventions, twenty-five (15%) met the typology inclusion criteria described above and were further grouped into five Intervention Component Categories. The main reason for exclusion was a website hosting information but not a specific intervention. The five Intervention Component Categories included:

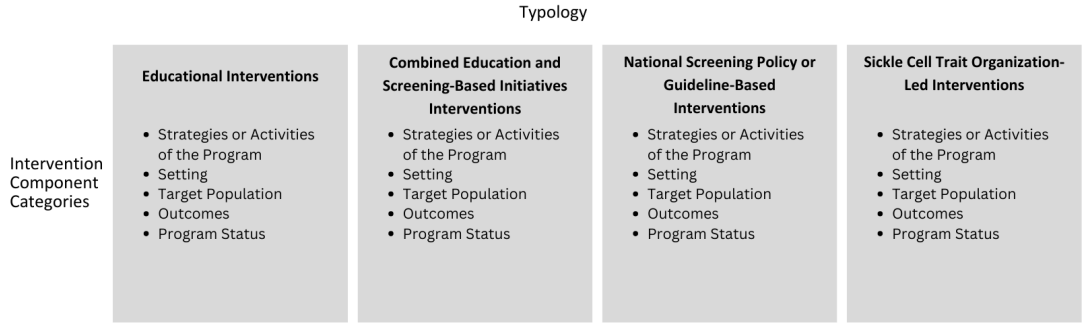

Figure 1. Sickle cell trait typology and intervention component categories structure, results from scoping review of sickle cell trait interventions from 2000-2024 United States.

Strategies or Activities of the Program, *Setting*, *Target Population*, *Outcomes*, and *Program Status*. *Strategies or Activities of the Program* include the methods that the intervention utilized to deliver the educational or awareness to the sickle cell trait community. *Setting* describes the type of institution of intervention delivery (e.g., in a hospital, online, etc.), and the state in which the intervention took place, if available. The *Target Population* specifies who within the sickle cell trait community was targeted. For example, a program targeting parents of newborns with sickle cell trait compared to adults with sickle cell trait. *Outcomes* are the results and effectiveness of the intervention, including any reported increases in sickle cell trait knowledge and awareness. *Program Status* is whether the intervention is active as of 2024. These components informed the creation of the Typology and provided the organizing structure to group information and evidence for future efforts.

Strategies or Activities of the Program: All of the articles discussed the strategies and activities of each program in depth. Of the twenty-five programs uncovered in this component, eleven (44%) [21] [29] [35]-[44] provided education about having a sickle cell trait, whether to people who already know they have a sickle cell trait or those who have just been provided a positive test. Eleven of twenty-five (44%) programs [28] [31] [39] [40] [41] [43]-[48] included screening participants for sickle cell trait. Eight of twenty-five programs (32%) [42], [44]-[50] included introducing policies or guidelines for screening various populations for sickle cell trait to increase awareness of one's status. Ten of these twenty-five interventions (40%) [21]] [31] [35]-[43] used individual-level education with a provider or trained educator providing education to the participant one-on-one.

Setting: Six out of twenty-five programs (24%) took place in healthcare settings [21] [35] [36] [37] [39] [41] [42], three of twenty-five programs (12%) were implemented in specific communities [31] [40] [43], and thirteen (52%) were nationwide [29] [30] [32] [42] [44]-[52]. Four of twenty-five interventions (16%) were delivered virtually such as through video modules or providing education over the telephone [35] [38] [41] [44].

Target population: Of the twenty-five interventions, seven (28%) targeted the parents of newborns who were diagnosed with sickle cell trait [20] [31] [36]-[41], and twelve (48%) targeted people who already know their sickle cell trait status [30] [32] [35]-[42] [51] [52]. Nine of twenty-five (36%) targeted specific populations of people, such as newborns, athletes or those in the military [40] [42] [44]-[51].

Outcomes: Sixteen of twenty-five interventions (64%) had outcomes that reported increased participant knowledge or awareness of sickle cell trait [35] [36] [37] [38] [40] [42] [44]-[51]. Nine programs (36%) have not yet undergone evaluation or did not increase participant knowledge or awareness [21] [29] [30] [31] [32] [39] [41] [43] [52]. Fifteen programs (60%) were overall effective as reported by the authors of the studies [21] [35]-[50].

Program Status: Eleven of twenty-five interventions (44%) are still active [29] [31] [32] [35] [37] [38] [41] [42] [47] [51] [52], three of which (27%) are na-

tionwide policies or guidelines [42] [44] [46]. Five programs (20%) designed to increase knowledge and perceptions within a designated period of intervention time ended by the time the typology was conducted [21] [30] [39] [40] [42], one intervention (4%) resulted in determination of feasibility of future community-based interventions [43], one (4%) was a policy implemented to provide short term funding [50], and one intervention (4%) informed an awareness and education program that is still active today [37].

3.2. Typology Results

The Intervention Component Categories inform the Typology. The articles were grouped into four categories representing the most common type of educational and awareness interventions for sickle cell trait: 1) *Educational Interventions*, 2) *Combined Education and Screening-based Interventions*, 3) *National Screening Policy or Guideline-Based Interventions*, and 4) *Sickle Cell Trait Organization-Led Interventions*.

Educational Interventions: Seven of twenty-five interventions were Educational Interventions (28%) and utilized various methods of providing knowledge, including video, audio, and text-based, mass media, and one-on-one virtual and in-person counseling [21] [35]-[40] (**Table 1**). While some interventions focused on educating people who already know their sickle cell trait status on the impact having the trait means for their life, many of these interventions aimed to educate parents of newborns who recently received a positive screening

Table 1. Sickle cell trait typology of United States interventions from 2000-2024 educational interventions (n = 7).

Intervention Name	Strategies or Activities	Setting	Target Population	Outcomes	Program Status
1. The CHOICES Intervention: An internet -based, tailored, multimedia education program about reproductive options and consequences [35] (2015)	• Participants completed a computer-based questionnaire (SCKnowIQ) before the intervention to assess baseline knowledge. The development of this intervention was based on the Kolb Experiential Learning Theory. A video, audio, or text-based case study or simulation was used so the participant could experience a scenario that included reproductive issues due to SCD or sickle cell trait. • Participants reflected on the simulated experience from different perspectives, then the participants were given information about SCD and sickle cell trait and reproductive health issues that are related. • The participants were shown videos of other couples who experienced decision making, and consequences related to reproduction with SCD and sickle cell trait.	University of Illinois Sickle Cell Clinic and University of Illinois Pediatric Sickle Cell Clinic.	Participants who had SCD or a sickle cell trait, were able to have children, and were 18 - 35 years old.	Study findings provide important insights for designing a national trial of the CHOICES intervention focusing on subjects whose partner status puts them at risk for having a child with SCD. At baseline, 114 (48.7 %) participants reported having partners who would not put the couple at risk for their children inheriting SCD. Of the 116 (49.6 %) at-risk participants, a higher proportion of those who were in the CHOICES group chose partners that reduced their risk by the last visit than the text-based group ($p = 0.04$).	This intervention is active, to complete in 2025.

Continued

2. Using qualitative and quantitative strategies to evaluate knowledge and perceptions about sickle cell disease and sickle cell trait [21] (2006)	• Targeted education was provided based on pre-intervention assessments in order to evaluate knowledge, perceptions and the effectiveness of different sources of information about sickle cell trait and sickle cell disease (SCD) and to determine individual knowledge of sickle cell trait status. • A series of focus groups were conducted with the goals of: 1) identifying barriers to sickle cell trait follow-up; 2) gathering perceptions of the general awareness of SCD; 3) generating potential solutions to the problem of low rate of trait follow-up. • Neighborhood surveys were conducted with the goals of: 1) determining the extent of community members' exposure to different sources of information about SCD and sickle cell trait in the past year; 2) evaluating community knowledge about SCD and sickle cell trait; 3) determining if individuals actually knew their own trait status and 4) evaluating the effectiveness of different sources of information about SCD and sickle cell trait in improving knowledge.	Northern California and the Northern California Comprehensive Sickle Cell Center in Oakland, CA, in 2006.	28 individuals participated in three focus groups (healthcare providers, people affected by SCD or have a sickle cell trait and community members). Surveyors interviewed 282 respondents within their neighborhoods.	Common themes across the focus groups included the limited general awareness of SCD and the sickle cell trait, the emphasis on the benign nature of sickle cell trait rather than on future implications, and the need for public health education campaigns about SCD and sickle cell trait involving media strategies. The majority of community survey respondents (n = 243, 86.2%) had correct general knowledge about the genetic basis and severity of SCD, but only 16% (n = 45) knew their own trait status. When respondents had received information about SCD from friends and acquaintances, they were three times more likely to know their sickle cell trait status, compared with respondents who had not received information from a personal source ($p < 0.01$).	This intervention is no longer active.
3. Sickle cell trait knowledge and health literacy in caregivers who receive in-person sickle cell trait education [36] (2017)	• A trained educator provided in-person sickle cell trait education to caregivers of referred infants with a sickle cell trait. • Participants were recruited and completed a health literacy assessment and a sickle cell trait knowledge assessment (SCTKA) before and after receiving education. • Caregivers repeated the SCTKA again after ≥ 6 months if they could be contacted.	In-person training at Nationwide Children's Hospital in Ohio from August 2015 to July 2016.	113 primarily English-speaking caregivers of infants with hemoglobin S-trait.	• (38.1%) percent of 113 caregivers had high SCTKA scores (≥75%) before education but 90.3% achieved high scores after education. • Caregivers with low SCTKA scores after education had significantly lower health literacy and baseline SCTKA scores compared to those with higher scores after education. • At ≥6 months, caregivers' scores were significantly higher ($p = 0.014$) than baseline, but only 73.3% scored ≥ 75%. • Results suggest that caregivers' baseline sickle cell trait knowledge is low, improves with in-person education but may decline with time. Caregivers who do not achieve high sickle cell trait knowledge after education had lower health literacy and baseline knowledge.	This intervention informed the creation of SCT aware, which is active: See Intervention #4.

Continued

4. A health literate approach to address health disparities: a virtual program for parents of children with sickle cell trait (2022) [37]	• Central Ohio's Sickle Cell Trait Education Program was delivered in-person. • The education program included having a parent of a child with SCT meet with an educator who provided verbal education using supporting visual materials. • The program did not have a formal curriculum but aimed to provide comprehensive SCT education to enable parents to learn about SCT and explain it to others. • Evaluators determined if health literacy affected understanding of concepts in order to create a health literacy-informed virtual training.	Education delivered in person in Central Ohio in 2019.	Seven English -speaking parents, of which three had limited health literacy.	• During the evaluation, parents asked few questions; noted undefined technical terms, closed questions, key concept omission, and limited explanation of visuals scoring low for understandability, actionability, and clarity. • The output of this study was a virtual sickle cell trait education program (SCT aware) for individual video conference delivery (knowledge objectives; plain language guide; health literacy-informed communication strategies; new visuals scoring highly for understandability, actionability, and clarity; narrated post-education version; standardized educator training).	SCTaware is ongoing in Central Ohio.
5. Closing Knowledge Gaps Among Parents of Children with Sickle Cell Trait: SCTaware program [38] (2021)	• SCTaware, a video conference-administered education program was delivered by a trained educator after being developed by a multidisciplinary team. • Parents reported their and their child's sickle cell trait status, completed a demographic survey, a health literacy assessment, and the published Sickle Cell Trait Knowledge Assessment (SCTKA) consisting of eight questions. • The video conference included sickle cell trait knowledge objectives, health literacy-informed communication strategies (e.g. teach-back), visuals scoring highly for understandability, and access to narrated post-education review materials. • After completing the program, parents repeated the SCTKA and report of their and their child's sickle cell trait status immediately and one month later.	Education delivered via video conference in Central Ohio between March 2020 and October 2021.	• Through electronic medical record review, English -speaking biological parents of infants with sickle cell trait who were informed of their child's sickle cell trait by telephone were identified. • 63 parents enrolled, 54 completed baseline surveys, 44 completed SCTaware, and 35 completed the one-month follow-up.	• Following telephone education, 44% of participants had high sickle cell trait knowledge. Participants' mean SCTKA scores post-telephone education did not significantly differ whether they had received telephone education ≤ 3 months or > 3 months before enrolling in the study (64% correct vs. 71% correct, p = 0.30). • Of those who had completed SCTaware, 42 (95%) achieved high sickle cell trait knowledge immediately after. Of those who have completed the one-month follow-up, 94% continue to have high knowledge. Participants with low HL had significantly lower SCTKA scores post-telephone education but those with high and low HL achieved and maintained high sustained knowledge one month after receiving SCTaware.	At the time of the article (2021), the intervention was ongoing.

Continued

6. A Pilot Study to Explore Knowledge, Attitudes, and Beliefs about Sickle Cell Trait and Disease [39] (2009)	In-person questionnaires were administered to parents with a sickle cell trait and parents of a child with either SCD or a sickle cell trait to examine the knowledge, attitudes, beliefs, and disclosure patterns about sickle cell trait of parents. These participants receive care and education from three different sickle cell disease clinics.	Chicago, IL in 2009.	Parents were recruited from (1) the University of Chicago Sickle Cell Disease Clinic, where their children are patients; (2) the Sickle Cell Disease Association of Illinois (SCDAI); (3) a federally qualified health clinic affiliated with the University of Chicago; and (4) the University of Chicago postpartum inpatient unit.	• The study found that there is significant misinformation about what it means to be a carrier and its health and reproductive implications. Formal professional counseling is rare, especially for those families without an affected proband. Strategies to increase the utilization of counseling and improve genetic literacy are necessary. • Fifty-three adults were interviewed, half (27) of whom had a child with SCD. There was significant misunderstanding about sickle cell inheritance (mean score, 68%), but parents who have a child with SCD have better knowledge compared to those without a child with SCD (78% vs 58%, p = 0.002). • Respondents perceive minimal stigma associated with sickle cell trait. • Individuals with a sickle cell trait rarely receive counseling or education outside of the family. • Parents report receiving their child's sickle cell trait diagnosis in the early newborn period from their child's doctor but indicate they received incomplete information. • Opportunities exist in primary care pediatrics to better align sickle cell trait disclosure timing and counseling content with parent desires.	This intervention ended in 2009 and is no longer active.
7. Parents' Experiences and Needs Regarding Infant Sickle Cell Trait Results [40] (2022)	• Parents were delivered their infant's positive newborn screening results. • This study explored parents' experiences with and desires for sickle cell trait disclosure and counseling for their infants with a sickle cell trait identified via newborn screening. • Parents of infants 2 to 12 months old with a sickle cell trait were recruited through a state newborn screening program for semi structured interviews to explore their experiences with and desires for sickle cell trait disclosure and counseling. • Inductive thematic analysis was conducted.	Virginia, from January to August 2020.	Parents of infants 2 to 12 months were recruited through the Virginia newborn screening program.	• Sixteen interviews were completed from January to August 2020. • Five themes were identified: parent knowledge before child's sickle cell trait disclosure, family planning, the dynamics of sickle cell trait disclosure and counseling, emotions and actions after sickle cell trait disclosure, and parent desires for the sickle cell trait disclosure and counseling process. • Two primary parent desires were revealed. Parents want more information about sickle cell trait, particularly rare symptomatology, and they want sickle cell trait counseling repeated once the child approaches adolescence.	This intervention ended in 2020 and is no longer active.

for their child. Mixed methods assessments were used to evaluate these interventions, including a survey [35] [36] [37] [38] [39], focus group [21], or interview [37] [40] to assess baseline knowledge pre-educational intervention and post-intervention.

Combined Screening and Educational-Based Interventions: Three of twenty-five interventions (12%) were categorized as *Combined Screening and Educational-Based Interventions* [29] [41] [42] [43] (**Table 2**). These interventions either encourage people to know their sickle cell trait status by getting tested and then provide education to those who screen positive, or include combined newborn screening and follow up education. Educational efforts were delivered in a variety of formats. For example, Red Cross Sickle Cell Trait Screening provided SCT testing for blood donors who opted in; if the donor receives a positive test result, they are directed to the Red Cross website for brief education on sickle cell trait but also encouraged to follow up with their healthcare provider [29].

Table 2. Sickle cell trait typology of United States interventions from 2000-2024 combined education and screening-based Initiatives (n = 3).

Intervention Name	Strategies or Activities	Setting	Target Population	Effectiveness	Program Status
1. Factors that influence parents' experiences with results disclosure after newborn screening identifies genetic carrier status for cystic fibrosis or sickle cell hemoglobinopathy [41] (2013)	• Parents were given their newborn's sickle cell trait screening results by their infant's PCP in various settings. • This study aimed to identify factors during initial newborn screening carrier results disclosure by primary care providers (PCPs) that influenced parents' experiences and reactions. • Open-ended responses from telephone interviews with 270 parents of carriers were analyzed using mixed-methods.	Wisconsin, between March 2008 and August 2010.	270 parents of children with a sickle cell trait.	• Parents identified aspects of PCP communication which influenced their reactions and results disclosure experiences. • Parents reported positive (35%) or negative (31%) reactions to results disclosure. • Parents' experiences were influenced by specific factors: content messages (72%), PCP traits (47%), and aspects of the setting (30%). Including at least one of five specific content messages was associated ($p < 0.05$) with positive parental reactions; omitting at least one of four specific content messages was associated ($p < 0.05$) with negative parental reactions. • Parents reported positive reactions when PCPs avoided jargon or were perceived as calm. Parents reported negative reactions to jargon usage and results disclosure by voicemail. • Findings suggest ways PCPs may improve communication of carrier results. PCPs should provide specific content messages and consider how their actions, characteristics, and setting can influence parental reactions.	This intervention is active.

Continued

2. Prenatal sickle cell screening education effect on the follow-up rates of infants with sickle cell trait (2000). [42]	• Mothers of infants with sickle cell trait were shown a 10-minute education videotape specifically produced for this target population during pregnancy and were given the opportunity for in-person notification of screening results and follow-up counseling/education. • This study assessed the effect of prenatal education about newborn sickle cell screening on parents' compliance with the follow-up for infants with sickle cell trait. • Follow-up rates, anxiety and retention of information were assessed for the case-control study.	Alabama, from May 1993 through October 1995.	Mothers in prenatal clinics.	• There were a total of 15,670 infants born in the region, and 647 infants were identified with sickle cell trait. The follow-up rate for parents of infants with sickle cell trait was significantly higher (76%) for study group than the control group (49%) ($p = 0.0006$). • Parents whose prenatal education included sickle cell hemoglobinopathy information retained significantly more of the information given during the post-natal education than did controls. • Data suggests that prenatal education for expectant mothers which includes information about newborn sickle cell screening significantly increases the follow-up rate for infants with sickle cell trait and contributes to a greater retention of information.	This intervention ended in 1995 and is no longer active.
3. Feasibility of a Community-Based Sickle Cell Trait Testing and Counseling Program. [43] (2016)	• Community intervention tested in eight community sites in which participants were invited to complete a community-based educational program and hemoglobin analysis. The analysis was followed by individual meetings with a genetic counselor to discuss test results and receive further information about their diagnosis, as well as resources for care and support moving forward. • As part of the formative work for this study, a multidisciplinary team developed educational materials to promote sickle cell trait testing among African American families. Using the recommended program development stages of formative research, message development, pretesting and message refinement, the multidisciplinary team created their messaging strategy. • This 15-minute educational program explained traits, genetic inheritance, and general information about sickle cell trait and SCD.	St. Louis, Missouri, between July 14, 2010 and May 31, 2012.	Participants were recruited from various branches of the St. Louis Federally Qualified Health Centers. 637 people completed the educational program.	• The study found that the program would likely be feasible in many communities, due to its success in the experimental eight communities. • The program also increased the number of individuals who know their sickle cell trait status. • Between July 14, 2010, and May 31, 2012, of the 637 participant who completed the educational program, five hundred seventy (89.5%) provided a blood sample, and 61 (10.9%) had a sickle cell trait or other hemoglobinopathies. The genetic counselor met with 321 (56.3%) participants.	This intervention ended in 2012 and is no longer active.

In two interventions, the follow-up education for people who screened positive was provided as part of the intervention by a primary care provider [41], or a genetic counselor [43]. In one intervention, the education began during pregnancy with a 10-minute education videotape specifically produced for this target population, and then post-natal sickle cell trait education was provided. The evaluation of this intervention showed that when parents of children infants with sickle cell trait were given education during and after pregnancy versus just after pregnancy, their knowledge retention was greater and follow up rates for post-natal education were greater [42].

Policy and Guideline-Based Interventions: Eight of twenty-five interventions (32%) were categorized as *Policy and Guideline-Based Interventions* [42] [44]-[50] (**Table 3**).These interventions mainly focus on introducing mandated screening for sickle cell trait in order to increase awareness of status in the

Table 3. Sickle Cell Trait Typology of United States Interventions from 2000-2024: Policy or Guideline-Based Interventions (n = 8).

Intervention Name	Strategies or Activities	Setting	Target Population	Outcomes	Program Status
1. Policy: Universal newborn blood screening (NBS) program [20] (2006)	• Newborn sickle cell trait screening programs have been adopted by all US states since 2006. • All newborns undergo heel-prick blood collection and trained medical professionals fill out a newborn screening card which is sent to a state lab for analysis. • Follow-up education or counseling is not mandated but some hospitals provide this service, although there is unclear evidence of how many. Many sickle cell organizations have received grants from HRSA to provide follow up counseling to those who ask for it. (More details on HRSA grant here). • Prior to 1993, CDC Hemoglobinopathy Reference Laboratory provided national standardized testing protocols; distributed manuals and training courses (required for clinics to receive federal funding). • CDC maintains hemoglobinopathy proficiency testing program to ensure reliability of state testing. • Commercially available tests now widely used, and genetic testing may someday replace dried blood drop as standard.	The United States, 2006 through present day.	All newborns in the United States and their parents.	• Despite 1987 federal recommendation, there was high variability in state newborn screening program adoption prior to 2006. • Universal screening identifies almost all cases of a sickle cell trait, but follow-up conversations about sickle cell trait status/implications are not standard.	This intervention is active, but there have been cuts to funding.

Continued

2. Mitigating sickle cell trait Associated Risks for War fighters and Athletes [45] (2012)	• Uniformed Services University convened an expert panel to: 1) Provide physical training that further mitigates the risk of fatal and nonfatal sickle cell collapse from intense exercise with a sickle cell trait and 2) Create clinical guidelines for frontline responders to recognize and treat sickle cell collapse, enabling warfighters and athletes to return to duty/their sport.	The summit was held on September 26 and 27, 2011 in Bethesda, Maryland.	Members of the military and athletes in the United States who have a sickle cell trait.	• This initiative was effective in introducing new terminology, exploring areas of controversy surrounding having a sickle cell trait, exploring methods for mitigating risk and adverse outcomes for members of the military and athletes, and identifying gaps in the research on sickle cell trait.	The clinical guidelines have evolved but are still in place.
3. Screening for Sickle-Cell Trait at Accession to the United States Military [46] (2014)	• All military recruits were screened for sickle cell trait over a 10 year period. • This review explores the benefits and harms associated with sickle cell trait screening on military accession. • An analytical framework was developed to address five key questions (KQs) and address benefits and harms of sickle cell trait screening: KQ1: Does Sickle Cell Trait Screening of Healthy Adults Directly Decrease Morbidity and Mortality? KQ2: Does Sickle Cell Trait Screening of Healthy Adults Lead to Improved Knowledge of the Condition? KQ3: Does Improved Knowledge of the Sickle Cell Trait Condition Lead to Behavior Change? KQ4: Does Behavior Change Lead to Decreased Morbidity and Mortality? KQ5: What Are the Harms Associated With Sickle Cell Trait Screening of Healthy Adults?	United States military over 10 years.	General US military recruits.	• KQ1: Despite the increased risk of rhabdomyolysis and sudden exertional death among military recruits with a sickle cell trait, there is no evidence in the medical literature that screening directly reduces morbidity and mortality. • The only large-scale intervention study (N = 2.8 million) that showed a reduction in exercise-related mortality among military recruits with a sickle cell trait also showed a reduction among recruits without a sickle cell trait. • Over the course of the 10-year study period, the investigators estimated that nearly 15 deaths were averted among recruits with a sickle cell trait and nearly eight deaths were averted among recruits without a sickle cell trait. • KQ2: There are no published data regarding whether sickle cell trait screening of healthy adults leads to improved knowledge of the condition. • KQ3: Although it is presumed that knowledge of sickle cell trait positivity would cause individuals to be more cautious during training, there is no evidence to substantiate this hypothesis. • KQ4: There is strong evidence to confirm that behavior change leads to decreased morbidity and mortality in both recruits with and without a sickle cell trait. • KQ5: Potential harms of sickle cell trait screening include fiscal costs, opportunity costs, genetic labeling and discrimination, and a false sense of security among individuals without a sickle cell trait, including approximately 1% of trainees who will falsely screen negative. • The current evidence is insufficient to support a conclusive recommendation for or against universal sickle cell trait screening on military accession.	All military recruits are screened for a sickle cell trait before joining the military.

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4. Policy: Public Law 92-294 (1972) Title XI-Sickle Cell Anemia Program [50] (1972)	• Nixon administration Sickle Cell Anemia Control Act (1972) provided funding for sickle cell trait/SCD screening and follow up counseling programs through the Department of Health, Education, and Welfare under the National Heart, Lung and Blood Institute (NHLBI). • The act gave the health secretary the power to grant research/training funding, development of public education programs on inheritance of a sickle cell trait and sickle cell anemia. • National Genetic Disease Act (1978) provided additional federal funding for years 1976-1978. • Nixon inaccurately stated SCD "strikes only blacks and no one else".	People with a sickle cell trait and SCD across the United States.	Voluntary participation available to "any person requesting screening, counseling, or treatment."	• Significant and highly visible financial investment in sickle cell disease; acknowledgement of lack of prior funding. • Funded 41 sickle cell centers/clinics, 250 + general screening programs, 69 research grants/contracts to support screening, education, and counseling. • Made note of counseling/education for people with a sickle cell trait and SCD. • Voluntary nature of program hindered universal adoption for decades. • Lack of information about having a sickle cell trait led to confusion, stigmatization, and distrust.	This intervention ended in 1978 and is no longer active.
5. Screening Student Athletes for Sickle Cell Trait—A Social and Clinical Experiment [47] (2010)	• The National Collegiate Athletic Association (NCAA) implemented mandatory testing for sickle cell trait status among all student athletes participating in Division One sports in 2010. Prior to the beginning of the athletic season, all students must submit documentation, but the study does not explain how institutions reinforce the mandate. • Sickle Cell Solubility Tests (SST) are provided by the NCAA Committee on Competitive Safeguards and Medical Aspects of Sports, and results are shared directly with students and coaches.	Colleges and Universities in the United States from 2010-2011.	All Division One College Athletes.	• This initiative was effective in screening around 167,000 student athletes between 2010 and 2011 alone. However, this screening intervention was not paired with any treatment or resources. • Students can avoid the testing if they provide a signed waiver releasing their university and the NCAA from liability. • This intervention has created a model for expanding testing to other groups. Still, only Division One athletes receive testing at this time.	Screening for a sickle cell trait in Division One athletes is active.
6. Implementation of the NCAA Sickle Cell Trait Screening Policy: A Survey of Athletic Staff and Student-athletes [44] (2018)	• The National Collegiate Athletic Association (NCAA) implemented mandatory testing for sickle cell trait status among all student athletes participating in Division One sports in 2010. • Two online surveys were used to assess knowledge, perspectives, and experiences with the NCAA Division One Sickle Cell Trait Screening Policy.	10 NCAA D1 Colleges and universities in North Carolina from January-June 2014.	228 student-athletes, 32 athletic trainers, and 43 coaches.	• 63% of student athletes supported the idea of required screening. Many felt strongly that the screening should be mandatory regardless of race/ethnicity (66%) or sport (71%). • 13% of student-athletes felt that non-black student-athletes did not have to be concerned with testing. • Some student-athletes did not know whether they had been tested (22%) or indicated they had not been (10%) offered sickle cell trait screening by their athletic departments.	Screening for a sickle cell trait in Division One athletes is active.

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				• After screening, 35% of student-athletes wanted to know more about the reasons for screening and 23% of student-athletes who tested negative for a sickle cell trait wanted to know more about sickle cell trait. • Twenty-eight percent (28%) of coaches, but no ATs, reported that at least one student-athlete at their institution had been removed from activity because of concern of developing a dangerous sickle cell trait-related condition. • Almost all student-athletes with a sickle cell trait said that more education and information about sickle cell trait would be helpful to them.	
6. Implementation of the NCAA Sickle Cell Trait Screening Policy: A Survey of Athletic Staff and Student-athletes [44] (2018)	• The National Collegiate Athletic Association (NCAA) implemented mandatory testing for sickle cell trait status among all student athletes participating in Division One sports in 2010. • Two online surveys were used to assess knowledge, perspectives, and experiences with the NCAA Division One Sickle Cell Trait Screening Policy.	10 NCAA D1 Colleges and universities in North Carolina from January-June 2014.	228 student-athletes, 32 athletic trainers, and 43 coaches.	• Four students learned their status through the NCAA policy and were notified in person (two), by email (one), and by mail (one). All four had questions about sickle cell trait and the implications of their positive status, but only one received genetic counseling. • The primary benefits of the screening policy identified by student-athletes with a sickle cell trait was that it created awareness among athletic staff and student-athletes that could help avoid adverse outcomes and could offer information for future family planning. • More education about sickle cell trait is needed for student-athletes and athletic staff in order to help make the implementation more successful. All parties need to be in agreement regarding the importance of knowing which student-athletes have a sickle cell trait and how that information will be utilized.	Screening for a sickle cell trait in Division One athletes is active.
7. Athletes' Perceptions of National Collegiate Athletic Association -Mandated Sickle Cell Trait Screening: Insight for Academic Institutions and College Health Professionals [48] (2010)	• The National Collegiate Athletic Association (NCAA) implemented mandatory testing for sickle cell trait status among all student athletes participating in Division One sports in 2010. • The study objective was to explore athletes' perspectives of National Collegiate Athletic Association (NCAA)-mandated sickle cell trait-screening policy by examining race-and gender-related differences in athletes' perceptions regarding risk of having a sickle cell trait and concern about loss of playing time.	Southeastern college campus in the US during April-August of 2010.	259 college athletes.	• The majority of athletes (81.7%) perceived that they would have a high level of concern if found to carry a sickle cell trait. • African Americans were 9.07 times more likely than Caucasians to perceive risk of having a sickle cell trait. • The majority of athletes disagreed (38.4%) or did not know (50.8%) if they would lose playing time related to carrying a sickle cell trait.	Screening for a sickle cell trait in Division One athletes is active.

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8. A Policy Impact Analysis of the Mandatory NCAA Sickle Cell Trait Screening Program [49] (2012)	• The NCAA implemented mandatory sickle cell trait screening for all Division One athletes, proof of previous test, or signed waiver releasing the school from liability.	Colleges and Universities in the US with Division One athletes.	All Division One College Athletes.	• Screening efforts identified over 2,000 sickle cell trait carriers over a four-year period. • Screening efforts could prevent 7 student athlete deaths over the course of a decade (1 in every 144,181 students screened). • Annual estimates of identifying 530 new sickle cell trait carriers. • ⅓ of students report wanting to learn more information behind the reasoning of NCAA screening and 1/4 of students report wanting to learn more about having a sickle cell trait.	Screening for a sickle cell trait in Division One athletes is active.

United States. These policies and guidelines either target newborns [42], athletes [44] [47] [48] [49] or those in the military [45] [46] and therefore are not inclusive of all populations of people who potentially have a sickle cell trait. Given the inherent tailored approach, these interventions were successful in increasing testing and/or awareness of sickle cell trait in individuals. Specifically, universal newborn screening was implemented in the United States in 2006 [20], screening for all NCAA Division One athletes was implemented in 2010 [49], and screening of all people entering the military was implemented in 2020 [46]. The Universal Newborn Screening Policy is reported to identify all cases of sickle cell trait [20], but there have been gaps in whether these test results are accurately communicated to carriers or recorded in medical records [53]. The Uniformed Services University Expert Panel was successful in introducing new terminology, exploring areas of controversy surrounding having a sickle cell trait, and exploring methods for mitigating risk and adverse outcomes for members of the military and athletes [45]. One study that examined the policy that all military recruits be tested for sickle cell trait upon entering the military reported that over a ten year study period, around fifteen deaths were averted among military recruits with sickle cell trait [46]. While the 1972 Sickle Cell Anemia Program does not report increase in knowledge or awareness on an individual level, the program funded 41 sickle cell centers/clinics, more than 250 general screening programs, and 69 research grants/contracts to support screening, education, and counseling [50]. Finally, mandated sickle cell trait screening for NCAA Division One athletes identified over 2,000 sickle cell trait carriers over a four year period and estimates identifying 530 new sickle cell trait carriers per year [44] [47] [48] [49].

Sickle Cell Trait Organization-Led Interventions: Six of twenty-five interventions (24%) were Sickle Cell Trait Organization-Led Interventions [29] [30] [31] [32] [51] [52] (**Table 4**). Half of these (n = 3) focused on providing sickle cell trait testing across the United States [29] [32] [52], while the other three interventions aimed to provide sickle cell trait education [30] [31] [51]. The three interventions that provided sickle cell education provided education to people who already knew they had a sickle cell trait, and each intervention targeted a

different community: young athletes (high school and younger) [51], parents of newborns with a sickle cell trait in California [31], and people with a sickle cell trait across the United States [30]. Only one of these interventions was not nationwide [31]. Two of these six interventions had effectiveness or outcomes reported, the first being the As One Foundation Operation Hydration Program, which provided an 80% increase in awareness among participants [51]. The second is the 23 and Me Sickle Cell Carrier Status Awareness Program, and while it does not report how many people total they have tested for a sickle cell trait, 19,000 people with a sickle cell trait in their database consented to be part of a study that determined people with sickle cell trait are at higher risk of experiencing pulmonary embolism [52] [54]. All of the interventions are still active, except one that ended in 2022 but continues to make educational materials accessible on their website [30].

Table 4. Sickle cell trait typology of United States interventions from 2000-2024: sickle cell trait organization-led interventions (n = 6).

Intervention Name	Strategies or Activities	Setting	Target Population	Outcomes	Program Status
1. Red Cross Blood Services Free Sickle Cell Trait Screening [29] (2023)	• A recent intervention by the Red Cross has expanded its testing to include sickle cell trait screening on all donations from self-identified African American donors. • This additional screening will help the Red Cross identify compatible blood types more quickly to help sickle cell patients and provide Black donors this important health insight. • African American donors can opt out of the screening if they wish. • Upon receiving a positive screening result, donors are directed to the Red Cross website to learn more about sickle cell trait and are encouraged to follow up with their healthcare provider.	Red Cross Centers across the United States from 2023 to present day.	All self-identified African American blood donors.	This intervention has not been evaluated and outcomes have not been reported.	This intervention is active.
2. As One Foundation Operation Hydration [51]	• A signature training program that teaches student athletes, coaches, training staff and parents about sickle cell trait awareness, hydration, and prevention of sports-related deaths. • Seeks to create earlier awareness of the correlation between sickle cell and lack of hydration—preventing sports related deaths due to dehydration and exhaustion. • Provides an annual training program for high school coaches, other athletic related staff as well as student athletes.	The As One Foundation is located in Florida and many Operation Hydration events happen in Florida, but it is a nationwide program.	Youth across the US, specific high school athletes, as only Division One college athletes undergo mandatory testing for sickle cell trait before participating in sports.	• Trainees are pre- and post-tested to measure the effectiveness of the training—outcomes include an 80% increase in awareness.	This intervention is active.

Continued

3. 23andMe Sickle Cell Carrier Status Awareness Program [52] (2022)	• Partnership between 23andMe and various sickle cell trait organizations to provide free testing and awareness. • 23andMe is collaborating with Breaking the Sickle Cell Cycle on a Sickle Cell Carrier Status Awareness Program that offers free 23andMe Health + Ancestry kits to participants and provides education and counseling services.	Nationwide, limited to people in the United States. The intervention began in 2022, and is active today.	People who do not know their sickle cell trait status who are over the age of 18.	• 23andMe does not report how many people in total they have tested for sickle cell trait, but 19,000 people in their database with sickle cell trait were part of the largest study of sickle cell trait to date that determined that people with sickle cell trait are at higher risk of experiencing pulmonary embolism [54].	This intervention is active.
4. Kidney Cancer Association and Sickle Cell Disease Association of America (SCDAAA) KNOW & TELL [30] (2021-2022)	• KNOW & TELL aims to raise awareness of sickle cell trait and its link to RMC. • The year-long initiative seeks to promote the early identification of RMC by encouraging people to know their sickle cell trait status and inform their family and health care providers about the connection between sickle cell trait and RMC.	Nationwide, 2021-2023.	People who are at risk of having a sickle cell trait.	No outcomes were reported.	This intervention is no longer active—the initiative took place from 2021-2022, however the educational materials are still present on both the Kidney Cancer Association and SCDAAA websites.
5. Sickle Cell Disease Foundation Hemoglobin Trait Follow-Up Program [31]	• The Sickle Cell Disease Foundation (SCDF) is a certified sickle cell trait counseling center. In partnership with the California Department of Public Health, Division of Genetic Disease Screening-Newborn Screening Program the SCDF provides parents of infants identified with sickle cell trait, hemoglobin C trait, and hemoglobin D trait free trait counseling and family testing.	California.	Parents of infants who screen positive for sickle cell trait, hemoglobin C trait and hemoglobin D trait.	No outcomes have been reported.	This intervention is active.
6. Sickle Cell Disease Association of America Free sickle cell trait testing through local sickle cell organizations that are members of SCDAAA [32]	• SCDAAA partners with local sickle cell trait organizations such as SCDAAA chapters and state organizations (e.g., the Massachusetts Sickle Cell Association) to provide funding for tests.	Nationwide.	People at risk for having a sickle cell trait.	No outcomes have been reported.	This intervention is active.

4. Discussions

The results of this typology lend insight to the strategies, activities, and messages of existing interventions that have either enhanced the awareness of sickle cell trait or that have contributed to the confusion around it. Grouping interventions into categories trough a typology summarizes current efforts in order to inform future ones through identification of successful intervention components and exposing inconsistencies in messaging. The successes and lessons learned that result from this typology will contribute to scientifically correct and consistent interventions to arm people with a sickle cell trait with the education to make informed healthcare decisions, thus collectively promoting positive health behaviors. Sickle cell trait interventions have historically been overshadowed by comprehensive SCD interventions; people with a sickle cell trait are in need of this investigation and subsequent further focus in this area and for organizations that serve people with sickle cell trait to be more effective in their collective approach to educating not only patients, but providers and health systems for a more holistic approach to education and awareness.

Our findings show that the current interventions that aim to increase education and awareness of sickle cell trait in the United States fall into four categories: *Educational, Combined Screening and Educational, Policy and Guideline-Focused,* and *Sickle Cell Trait Organization-Led*. Each of these efforts focuses on either encouraging people who do not know their sickle cell trait status to get tested, or to educate people who do know their status about the potential complications of having a sickle cell trait. The interventions most commonly focused on reproduction and newborn screening [20] [31] [35] [41] [42], especially the possibility of having a child with SCD and safety around athletics [44]-[49] [51]. Following, we summarize the findings and explore the connection to existing context and application in practice.

The *Educational Interventions* provides insight into the methods that currently exist to close knowledge gaps in sickle cell trait, which is important given the lack of knowledge in the sickle cell trait community. The interventions in this category suggest further sickle cell trait awareness and educational efforts could be delivered effectively in many modalities, including virtual or in-person formats and in individual or group settings. While most of these interventions happened pre-COVID-19 pandemic, the shift to virtual education strategies has increased and the effectiveness of virtual education to change behavior and practice is important to consider when designing interventions in a post-COVID-19 context [55]. Additionally, these interventions offer various ways to evaluate the effectiveness of educational interventions, specifically by using surveys, focus groups, or interviews to assess baseline knowledge pre-intervention and educational attainment post-intervention though focus less on how knowledge will be translated to practice behaviors. The drawback of these Educational Interventions is that they only target those who already know they have a sickle cell trait, leaving out those who do not know their status, further warranting more efforts

in this area.

The *Combined Screening and Educational Interventions* allow a deeper understanding of the importance of providing education to a patient once they screen positive for a sickle cell trait to ensure they know how this result impacts their health. When this type of intervention is implemented, awareness increases in two ways: more people in general will know their sickle cell trait status, and more people will be aware of how having a sickle cell trait impacts their life. Combining screening and educational interventions allows for larger intervention impact [56]. By alerting more people to know their sickle cell trait status, not only will more individual deaths and health complications be avoided, but more people will be able to make informed decisions about reproduction, as SCD can occur if one or both parents have a sickle cell trait [57]. In many of these interventions, brief education was provided and then the patient was referred to further counseling, either through their primary care or a genetic counselor. It is important for interventions to consider partnerships with these types of providers, as connecting these patients with long-term care after a positive test is important for ongoing monitoring and education [58]. The outcomes of these interventions provide insight into patients' motivations to pursue follow-up education and preferences for receiving this education once they receive a positive screening, which is an essential consideration for future interventions for maximum impact and sustainability.

While the *Policy and Guidelines* show that these types of interventions are effective in increasing the number of people who know their status, without proper follow-up counseling or education, there is a disconnect how people apply that information. This is exemplified by universal newborn screening for a sickle cell trait in the United States. While all newborns born in the United States are screened for a sickle cell trait at birth, only 16% of people know their sickle cell trait status [21]. Without investment in ensuring test results are accurately charted in a newborn's medical chart (which was shown to not be done consistently despite universal testing [20]) and ensuring follow-up education and counseling, testing-based interventions often do not improve awareness of sickle cell trait nearly as much as they could [59]. Focusing on the outcome of identifying more people who have a sickle cell trait, the Policy and Guideline category was found to be most successful, likely because policies are the most expansive in reach. For example, screening all Division One college athletes identified over 2000 sickle cell trait carriers over a four-year period, and identifies around 530 new sickle cell trait carriers per year [49]. Identifying sickle cell trait among athletes is especially important given the most serious complications of sickle cell trait (e.g. rhabdomyolysis and exercise-related sudden death) are occur by strenuous exercise and dehydration [14]. This exemplifies the necessity of education following a positive test; without education, athletes with a sickle cell trait will not know how to decrease their risk of these serious complications (proper hydration, building exercise intensity slowly, etc. [14]), or the symptoms to watch for when exercising.

The interventions led by Sickle Cell Trait Organizations provide insight into what the organizations that are closest to the problem are doing to address the lack of awareness in the space, however, also highlight the gaps in what they can accomplish given less resources. These interventions sometimes addressed certain groups within the target population, for example, athletes or parents of newborns, but many interventions aimed to increase the amount of people who do not know their status across the United States. Unlike the other intervention categories in this typology, the interventions led by Sickle Cell Trait Organizations aim to remove barriers to testing by making testing free and convenient with consistent education or referral to education after a positive test results. This suggests the organizations addressing sickle cell trait understand the importance of education to increase awareness. The most prominent theme that emerged in this research was that messaging about sickle cell trait and the related health complications is inconsistent and even conflicting across interventions and organizations. For example, messaging on one website states "A trait IS NOT A DISEASE and will not cause your baby to become ill" [32]. Meanwhile, messaging on another website "[Sickle cell trait] coupled with physical rigor and lack of hydration, could be fatal [···] Over 20 high-school and collegiate football players' deaths have been related to sickle cell trait since 2000, making exertional sickling the leading cause of death in football players in recent history" [51]. The two statements drastically conflict and suggest that interventions for sickle cell trait in general, and the messaging that form their foundation, need to be more consistent in the education they provide.

There are limitations to this typology process. First, is that we do not include information on length of intervention or sustainability efforts for the intervention. Second, our focus was on awareness and education efforts and not on medical interventions, which may provide more evidence of effective approaches. Since our focus was to enhance messaging and awareness efforts these other medical interventions fell outside the scope of review. This is an important feature to understand effective approaches but was not available for many of the studies we found. Finally, this scoping review and typology only included interventions from the peer-reviewed literature or from existing websites in the sickle cell trait space. There is likely institutional knowledge and other efforts that the field would benefit from learning for example communities of practice, tailored forums, and other information sharing may be other areas to explore to enhance the findings of this typology.

The main lessons from this typology, as summarized by the above descriptions, are that many people do not know their sickle cell trait status in the United States, and of those who do know their status, many do not know the possible complications of having a sickle cell trait or how to decrease their risk [21] [39]. There is a lack of consistency in messaging across interventions whether delivered by credible healthcare institutions or national organizations, which can result in lack of education and awareness and confusion around sickle cell trait. Further, for an intervention to be comprehensive and effective a variety of strat-

egies should be employed. The four categories in this typology largely remain siloed; interventions do not often employ the methods of the other categories to create a holistic approach to sickle cell trait. For example, mandated testing policies provide the recipient with a positive test, but without follow-up education, the person will not understand what precautions they will need to take to avoid potential complications. These lessons learned are important to inform future interventions in sickle cell trait. Specifically, interventions must have scientifically accurate and consistent messaging in order to ensure all people know whether they have a sickle cell trait, and what that means for their life. As such, the research approach and typology allowed us to compile what exists in the field and distill it into recommendations for practice. These recommendations should be applied to both existing and future interventions to increase awareness and education of sickle cell trait in the United States:

1) Facilitation of meaningful partnerships between research and practice to disseminate scientifically accurate information. Many of the messages that exist within interventions online are scientifically inaccurate or conflicting from source to source. By strengthening partnerships between research and practice, interventions will be infused with more scientifically accurate and consistent information [60].

2) Implementation of consistent and accurate messaging around sickle cell trait among organizations to avoid confusion and lead to health promoting behaviors. Once interventions in the sickle cell trait field are infused with scientifically sound messages, organizations implementing these interventions will be able to provide messaging that is consistent and not conflicting so all people with sickle cell trait clearly understand what this means for their life and the behavior changes to make to avoid health complications [61].

3) Utilization of multi-pronged strategies for interventions to ensure a comprehensive approach. Interventions that utilize a multi-pronged approach are more successful in promoting awareness, education, and behavior change [56]. When organizations are creating interventions, they should consider utilizing interventions that use multiple approaches, such as combined screening and educational interventions.

4) Expansion of education efforts for medical students, providers, health systems, and patients to ensure bidirectional education and communication around sickle cell trait. This research shows that while one way to increase awareness and education is by reaching people who have a sickle cell trait, another important way is to reach medical students, healthcare providers, and health systems, who have been identified as having a lack of knowledge in this space [16] [17]. By educating these populations, we create a more holistic way of ensuring the public is more informed about having a sickle cell trait by introducing education and awareness from multiple angles. In order to improve education, awareness, and health outcomes of people with a sickle cell trait, there needs to be improvement in provider knowledge and the information that they provide their patients, as patients often rely heavily on their providers for infor-

mation about their health [62].

5) Implementation of flexible communication strategies to ensure that messages reach those who need it and are tested by the target population. All interventions created to increase awareness and education about sickle cell trait should be accompanied by flexible communication strategies for dissemination of the interventions. Organizations implementing interventions should focus on refining the messages within those interventions and testing them with the target population [63] through various methods such as focus groups and interviews. Finally, organizations should create communication and dissemination strategies assisted by a media planning guide to get the messages to those who need to hear them through communication avenues such as websites, multi-media advertising strategies, and communication campaigns.

Having historically been overshadowed by interventions aimed at SCD, interventions aimed at increasing education and knowledge of sickle cell trait deserve focus and recommendations to move the field forward. This typology, which is the first to focus on sickle cell trait interventions, will provide sickle cell trait organizations and other implementers recommendations for scientifically accurate and consistent messages to holistically educate patients and providers. Additionally, this typology will provide organizations the support to design and implement comprehensive sickle cell trait awareness initiatives across the field. This education and intervention guidance will ultimately encourage health promoting behaviors and health behavior change to an overlooked population.

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There are no conflicts of interest to disclose.

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