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Original Research Article

Developing and testing sickle cell trait awareness messages to strive towards consistent and accurate education

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ABSTRACT

Background: Sickle cell trait is often misrepresented as benign, leading to misinformation and a lack of awareness about potential health complications. In the United States, 1 in 13 Black individuals are affected, though people of any race or ethnicity can have the condition. Misconceptions, systemic racism, and underfunded research contribute to inconsistent communication and education. To that end, this initiative aimed to identify gaps in awareness and education and develop consistent, comprehensive, scientifically informed messages to improve public understanding. The goal was for these messages to be adopted by sickle cell trait organizations nationwide to promote consistency in education and communication.
Methods: Using a mixed-methods approach, 19 key stakeholders including sickle cell organizations, medical experts, and individuals with sickle cell trait were engaged to identify communication gaps. The consolidated framework for implementation research (CFIR) guided survey and interview analysis, ensuring a structured approach to understanding stakeholder perspectives. The nominal group technique (NGT) validated findings during expert meetings, helping achieve consensus on messaging before testing with the sickle cell trait community.
Results: Findings highlighted inconsistent messaging, limited public understanding, insufficient research funding, and inadequate provider knowledge. Stakeholders and community members stressed the need for accurate, culturally sensitive, consistent, and accessible messaging to improve awareness and patient-provider communication.
Conclusions: Through collaboration and community validation, evidence-based messages were developed to promote informed decision-making, encourage self-advocacy, and enhance provider-patient communication. Standardized messaging is essential to combat misinformation, raise awareness, and improve health outcomes for individuals with sickle cell trait.

Keywords: Consolidated framework for implementation research, Health communication, Nominal group technique, Stakeholder Sickle cell trait awareness

INTRODUCTION

In the US, people who are Black carry the burden of sickle cell trait with 3.6 million, or 1 in 13 people affected.[1] However, anyone of any race or ethnicity can have a sickle cell trait including 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 with potential associated health outcomes treated on an individual basis.

Sickle cell disease (SCD), a different health condition than sickle cell trait, overshadows awareness efforts. Sickle cell trait is often misrepresented as benign despite mounting evidence that proves otherwise and many remain unaware of their status until they experience a

debilitating or life-threatening complication or have a child with SCD.[1-13] The incorrect belief that having a sickle cell trait is benign compounded by systemic racism and underfunding of research leads to poor understanding and inconsistent communication.[14,15] Limited investment in medical education also contributes to underdiagnosis.[14,15] Demonstrating the limited awareness, despite the high prevalence of the condition in the US, only 14% of the US population knows their status.[16]

Access to consistent and accurate information is essential, from providers and more broadly in the field, as correct information is needed to facilitate behavior change that leads to improved health outcomes. An environmental scan and typology found messages from organizations are often inconsistent, incorrect, outdated due to lack of research, and do not report the full spectrum of scientifically-proven related health outcomes.[17]

The Boston University School of Public Health Sickle Cell Trait Awareness Initiative aimed to enhance existing efforts by creating a set of consistent, comprehensive, and scientifically-informed messages. We assessed existing messaging, engaged with national stakeholders to determine communication gaps, designed messaging with these expert stakeholders for providers to use with patients and for patients to directly access, and finally, tested the messages with the sickle cell trait community in order to create consistent messages that can be used across organizations. Stakeholder engagement was essential for adoption across organizations to achieve consistency in communication across the field.

The consolidated framework for implementation research (CFIR), an implementation science framework, was used to distill qualitative findings and apply themes to create actionable messages.[18] The Nominal Group Technique (NGT) guided all phases of the project due to its success in guiding group discussions and feedback.[19] We engaged multiple perspectives from 19 stakeholders across the country including providers, organization leaders, experts, and people who have or are at risk of having a sickle cell trait to produce and test messages that have an impact on behavior change to ultimately improve health outcomes.

Project phases

A formative evaluation was conducted to guide project goals and design interventions for people who have a sickle cell trait. Additionally, we conducted scientific research into sickle cell trait related-health outcomes and formative research into existing awareness efforts. The formative evaluation aimed to 1) assess the messaging and intervention landscape, including challenges and facilitators to inform subsequent message and intervention design; 2) engage appropriate stakeholders to develop and refine messaging to test with the sickle cell trait community; and, 3) achieve stakeholder buy-in for ongoing collaboration.[20] Both qualitative and quantitative methods were used to collect data from these stakeholder groups (Table 1).[20]

Table 1: Project phases and activities.

Project phases	Activities
Phase 1: landscape and stakeholder analysis through environmental organization scan and prioritization matrix	The environmental organization scan identified organizations and experts and gathered data on their awareness efforts to inform creation of messages and interventions.
	The prioritization matrix scored several factors to decide which organizations or stakeholders were most important to collaborate with and engage based on factors such as connection to the sickle cell community and other sickle cell organizations, and alignment of their mission with this project.
Phase 2: sickle cell trait-focused organization interviews and surveys	Formative research questions were generated and refined through the research methods described below.[20]
	These research questions were the basis for the organization interview guide, which was used during these interviews to understand the landscape of sickle cell trait messages, initiatives, and motivations to collaboration and engagement.
	The follow-up survey gathered quantitative data on a subset of the questions asked during the interview for data visualization purposes.
Phase 3: drafting project messages based on interviews and surveys	The organization interviews and surveys were analyzed and informed creation of salient themes and draft messages to be presented to the sickle cell trait experts in the next step of the process: meetings convening these experts.
Phase 4: sickle cell trait expert meetings – message development and refinement	Sickle cell trait experts identified through the prioritization matrix and organization interviews were convened to develop and refine awareness messages to be tested with the community.
	This process used the nominal group technique (NGT) to structure each meeting and ensure actionable messages.

Continued.

Project phases	Activities
Phase 5: community message testing focus groups	Messages that were refined alongside the experts were tested with the community through focus groups to produce final messages.
Phase 6: finalized messages: presentation to sickle cell trait-focused organizations and experts	The messages were presented to the organizations and experts during a final meeting. Participants reached a consensus that they supported adopting these messages.
Phase 7: development of website	A website was created to house these messages and additional information about having a sickle cell trait, including testing information for those who do not know whether they have a sickle cell trait, information for loved ones on supporting someone who has a sickle cell trait, and providers who may care for people who have a sickle cell trait.

METHODS

This mixed-methods study was guided by two scientific frameworks to formulate innovative and informed messages – CFIR to consolidate qualitative findings into themes for application and NGT to provide structure to engagement of the stakeholders representing a variety of disciplines and perspectives.

The consolidated framework for implementation research (CFIR) framework

CFIR was used to: 1) understand the present landscape of sickle cell trait messaging; 2) guide the message development and formative evaluation; and, 3) organize data collection, analysis, results, and recommendations.[18] This framework is used for systematically analyzing and organizing qualitative findings and allows for generalizing findings to allow for more immediate application to message and intervention development.[18] CFIR constructs are organized into five domains and are detailed in results.

Nominal group technique

The nominal group technique (NGT) has been used extensively in public health and is a decision-making technique that we employed to validate information gathered in the sickle cell trait expert group meetings ("Expert Meetings") to reach consensus on messaging.[21,22] NGT is primarily useful at the preliminary phase of an initiative as it offers a structured way to generate ideas and to quantify the group's assessment of the ideas.[22] NGT provides an orderly procedure to meetings for obtaining qualitative information from target groups who are most closely associated with the problem area.[23]

Sickle cell trait-focused organizations

Recruitment and data collection methods

Sickle cell trait-focused organizations ("Organizations") were identified for interview through an environmental organization scan, which assessed the contextual landscape in the US, and a prioritization matrix, which allowed follow-up with relevant organizations according to a pre-determined set of factors The research team contacted 50 organizations outlined in the Prioritization Matrix, and 13 interviews were conducted. After interviews were completed, a follow-up survey was sent to collect quantitative data on similar questions from the interview. A follow-up email was sent one week later to those who did not complete. The survey was administered online and took no more than fifteen minutes to complete. Nine surveys were completed (69.2% of the completed interviews).

Data analysis methods

Stakeholders were recruited for semi-structured interviews, which were conducted over Zoom and recorded and transcribed for analysis. The interview lasted around one hour and was tailored based on information in the Environmental Organization Scan, such as the mission of the organization and their partnerships in the field. The interview, guided by questions aligning with CFIR domains, elicited information about the Organization's awareness and attitudes on efforts surrounding sickle cell trait education and their organization's history with and willingness to collaborate with similar organizations.

The stakeholder interviews and surveys were analyzed, and themes and draft messages were presented to the Experts in subsequent meetings. The draft messages emerged from the results of the interviews and the surveys reflecting prominent themes, barriers, gaps, and facilitators to sickle cell trait awareness in the community. Qualitative data was analyzed in NVivo, and quantitative data was analyzed in Qualtrics.[24,25]

Sickle cell trait expert group meetings

Recruitment and data collection methods

Expert meeting recruitment entailed emailing each of the organizations identified in the environmental organization scan as being focused on sickle cell trait awareness.

Twenty-one experts from these organizations were contacted to participate in a meeting to develop sickle cell trait awareness messages, and over the course of four meetings spanning four months, 19 experts participated. The meetings were held over Zoom and were recorded and transcribed for analysis.

Data analysis methods

Meeting notes were analyzed by reviewing the transcript and chat log from the meetings and annotating the messages presented to the group with each person's suggestions and revisions. The research team reviewed these annotations together and revised each message based on the consensus reached by the Experts, and with consideration of the supporting scientific evidence. This meeting process was guided by NGT.

Sickle cell trait community

Recruitment methods

Sickle cell trait community members ("Community") were recruited for the community message testing focus groups through connecting with groups, organizations, healthcare clinics, social media platforms, and stakeholder groups who met the inclusion criteria (over 18 years old and have or are at high risk of having a sickle cell trait based on race/ethnicity). This approach ensured that results would best represent the target audience of the messages. The University Institutional Review Board approved this study as exempt (H-43320).

Data analysis methods

Focus group notes were taken by one or two members of the research team while the third led the conversation, the chat log and poll results were downloaded, and all data was compiled into a master document. Analysis of the results was performed in a separate document and guided by CFIR. Individual participant feedback was examined across all focus groups and analyzed for themes with two members of the research team, and any inconsistencies or disagreements were discussed with a third member of the research team until resolution.

RESULTS

Sickle cell trait-focused organization interviews

All 13 organizations that responded were interviewed. Nine stakeholders completed the follow-up survey (69.2%). Interview participants included eight organizations solely focused on sickle cell trait, and five organizations focused on the target audience more broadly, such as those focused on both SCD and sickle cell trait, hematologists, or physicians who were experts on sickle cell trait health outcomes such as renal medullary carcinoma.

Results of the organization interviews are presented by CFIR domain with illustrative themes and quotes in Table 2. Organization interview and survey data collection results. These themes and quotes represent the variety of Organizations who responded within that construct. If a construct was not used in the analysis process, it is not represented in the below description or table.

Table 2: Sickle cell trait-focused organization interview and survey data collection results.

CFIR construct	Survey results	Interview results
Intervention characteristics	Respondents represented nonprofits focused on the sickle cell trait and/or disease community, national organizations, athletics-focused organizations, hospitals, private foundations, and research organizations and held roles including program-focused roles, research, physicians, and other (patient advocate, athletic trainer).	Participants often praised their own messages and interventions but expressed concern that other organizations in the field were producing inconsistent messages.
	Interventions were distributed mostly on social media channels, email campaigns and on the organization's website.	Quote: "I think [messaging] needs more consistency because I think the biggest messaging is wrong. I think the biggest messaging has been that it's benign, and you don't have to worry about it."
	Respondents measured success of their interventions mostly from patient feedback, but also through decreased sickle cell trait outcomes and increased engagement of other stakeholders in their field.	It is challenging to break into the community of people they want to target with their messages.
	Respondents' organizations were funded largely through private foundations and/or grants and state sources.	It is important to understand specific needs of the target audiences for their messages and interventions, as they must be crafted and delivered in appropriate and specific ways.

Continued.

CFIR construct	Survey results	Interview results
Outer setting	Respondents often collaborated with other local and/or community-based organizations, research institutions, and national organizations to develop of joint messages, programming, and awareness efforts.	Participants expressed that patients are not being given information about their sickle cell trait status and when they do have this information, education about the related health outcomes is severely lacking.
	The majority of respondents felt there was a strong need for sickle cell trait awareness in the community (88%) and 63% strongly disagreed, disagreed, or felt neutral that the major messages that exist in the field provide awareness in communities.	Quote: "Even in the sickle cell disease community they tend to think that sickle cell trait is benign, and it's not a big deal, but what they don't see is sickle cell trait is really the nucleus of all these problems or concerns or issues. Without sickle cell trait, you wouldn't have sickle cell disease, but they don't look at it that way, and I think the hardest job is educating people, helping them to understand sickle cell trait."
	Respondents largely felt that the messaging was inconsistent, there was a lack of funding, and lack of awareness of sickle cell trait status.	
Inner setting	Organizations mainly expressed that they work in silos; there is a desire to break down those silos to become more collaborative to benefit the community.	Quote: "Everybody's doing great things in silos. So, if there could be some cohesiveness to make [communication and messaging] more streamlined - and even if it wasn't connected - just making it have some of the same messaging. So, no matter where you go, when you learn about sickle cell trait, or sickle cell disease, the messaging will be the same."
		Common incorrect beliefs among the community, providers, and organizations: having a sickle cell trait is only a condition that people who are Black have, and that having a sickle cell trait is benign.
		Quote: "I know the key is education, and most of our education is […] through virtual seminars. But what's so heartbreaking is even when you have a virtual seminar, and it's free, the participation is low, because once again they believe there's nothing we need to know, because [sickle cell trait is] benign, right?"
		There was a desire for more quality education for all medical students to provide better quality healthcare for people who have a sickle cell trait, more testing for and education about having a sickle cell trait, and more public awareness of having a sickle cell trait in general.
Characteristics of individuals	N/A	Participants were extremely passionate and knowledgeable about sickle cell trait, with many having personal connections.
		There were conflicting opinions among the community about whether people who have a sickle cell trait should have children, given the risk of having a child with SCD.
		Most organizations had consistent messaging on making your own informed choices about having children when you have a sickle cell trait.

Continued.

CFIR construct	Survey results	Interview results
Process	Organizations mainly measured the success of their interventions through feedback from the community.	Participants expressed that engaging provider has been challenging and has been a barrier to implementation of their interventions.
		Quote: "We tried to reach physicians, and that was a dead end. Nobody would come to our conferences. We put them on at the beach. Nobody would come except the nurses who were really doing the right thing, anyway. So really, we reached out to make patients the expert, and we wanted them to understand their disease so they can advocate for themselves."
		Providers struggled to engage patients, especially when it comes to getting them to return for follow-up education after a positive sickle cell trait newborn screening.

*All quotes are presented as stated unless edited to remove unnecessary information as indicated by […] in the quote. The use of brackets is also used to insert clarifying information not part of the response and/or quote.

CFIR Domain #1: Intervention characteristics

This domain addresses factors that may influence whether an organizations intervention is implemented successfully. This domain was a main focus of the interviews as we aimed to learn more about the awareness efforts surrounding sickle cell trait that exist in the field already.

CFIR Domain #2: Outer setting

The outer setting domain addresses external factors impacting the intervention such as agency networks, communication, policies and incentives, barriers and facilitators to meeting needs of the population, and competitive pressure to implement an intervention. As stakeholders detailed interventions existing within their organizations, outer setting constructs apply to factors external to their organizations.[18] This domain was prevalent in these interviews, especially the patient needs and resources construct, as participants often spoke about community needs and how their organization attempts to fill those needs.

CFIR Domain #3: Inner setting

This domain considers internal factors of an organization that impact the implementation of an intervention including internal communication, norms and values, structure, capacity for change, receptivity of individuals, and agency commitment to the intervention. The inner setting domain gave insight into the inner workings of each organization, the challenges and successes they face when implementing interventions, and what the participants would like to change about the field.

CFIR Domain #4: Characteristics of individuals

Characteristics of Individuals domain include the thoughts, perceptions, knowledge, confidence, beliefs, and commitment to change of individuals within the organization. The domain revealed how the participants view the efforts of their organization and how they personally identify with the organization.

CFIR Domain #5: Process

This domain addresses not only the implementation of interventions but the initial and ongoing use of the interventions through consideration of engagement strategies (i.e., education, marketing, training), established plans for implementing the intervention, delivery of the intervention according to the established plan, and communication and feedback about the intervention and its implementation.[18] The domain allowed participants to reflect on the implementation process and outcomes of their interventions and consider how they engage their target audiences. Once again, participants expressed a desire for more consistent information among organizations. A benefit to having interviewed both organizations and providers were their reflections on engaging not only with patients, but with each other.

Sickle cell trait-focused organization surveys

Quantitative analyses organized by CFIR were conducted on the organization survey results to describe participant characteristics and summarize perceptions and knowledge of organizational efforts. Findings from the Organization interviews are also presented in Table 2: sickle cell trait-focused organization interview and survey data collection results.

Surveying the organizations who were interviewed provided deeper insight into their efforts and supported the qualitative data collected through interviews. For example, that 88% of organizations interviewed believed there is a strong need for sickle cell trait awareness in the community reflected the extensive qualitative data

collected through interviews that provided more contextual findings. The survey results also reinforced the qualitative interview findings that messaging is inconsistent, there is a lack of funding, and lack of awareness of sickle cell trait status. These responses highlighted the need for increased awareness and consistent messaging in the field.

Organization interviews and surveys resulted in major themes that we used to draft messages that achieved the following goals: 1) People with the potential to have a sickle cell trait will seek to know their status, 2) People with a sickle cell trait will be motivated to know and understand symptoms and conditions linked to having a sickle cell trait that can have an impact on their health, and, 3) People with a sickle cell trait will be knowledgeable enough to feel comfortable to self-advocate when they visit health care providers. The most prominent themes that emerged after quantitative and qualitative analysis revealed barriers to sickle cell trait awareness, and the resulting draft messages were created to address those themes. See Table 2: sickle cell trait-focused organization interview and survey data collection results.

Sickle cell trait expert group

Nineteen experts participated across four expert meetings. Some of these stakeholders attended all four meetings while others attended less than four due to time constraints. Experts spanned roles, such as directors, professors, medical directors, hematologists, a president/founder of a nonprofit, a medical director, a chief health officer, and more.

The NGT guided each of these meetings and allowed the experts to reach consensus on messaging to be tested with the community. As these messages evolved over the meetings, changes were made to reflect each stakeholder's expertise, scientific understanding, and lived experiences. During each meeting, all the steps of NGT were followed while discussing messages, including Step 1: Generating ideas, step 2: Recording ideas, Step 3: Discussing ideas, and Step 4: Voting on ideas to reach consensus. Table 3: meeting actions and outcomes guided by NGT shows how each meeting was structured by NGT.

Table 3: Meeting actions and outcomes guided by NGT.

NGT step	Meeting action	Action outcome
Step 1: generating ideas	Participants were shown a draft set of messages one by one that were developed by the project team based on analysed information resulting from the stakeholder interviews and surveys.	New pieces, parts, or wording of messages were generated.
	Participants were then asked to individually consider each message and generate new parts or pieces of the message if necessary.	
Step 2: recording ideas	The project team recorded each participant's ideas and feedback on each message.	A list of ideas and feedback on each message was created for further discussion and development.
Step 3: discussing ideas	Each idea or piece of feedback was discussed to work toward consensus on various phrasing, importance of proposed messages, and introduction of new messages.	Messages were tailored based on the ideas and pieces of feedback and these changes were discussed message by message.
Step 4: voting on ideas to reach consensus	If there were multiple suggestions or ideas for parts or entire messages, the participants voted on which part they preferred. The votes were tallied to decide which adjustments would be made to the messages.	Message by message, each change made to each message was voted on and a finalized list of messages to test with the community emerged.

Over four meetings, the experts developed and refined draft messages. Through discussion of the nuances of each message, an important need surfaced: consensus on the foundational parts of sickle cell trait. This became a focus of the meetings where the Experts received information surrounding foundational sickle cell trait science and engaged in discussion about the nuances of how to talk about sickle cell trait. Historically, the condition has generally been referred to as "sickle cell trait" and people who have a sickle cell trait have been called "sickle cell trait carriers". The Experts determined that calling people "sickle cell trait carriers" does not utilize person-first language, which is a recent standard for how to refer to people who have various conditions, and that "having a sickle cell trait" is the most appropriate, scientifically backed, and culturally sensitive way to refer to the condition.[26] Additionally, the team presented a comprehensive list of scientifically proven outcomes associated with having a sickle cell trait to

reach consensus on the outcomes that should be included in awareness efforts, which are included throughout the project messages.

Another important discussion that emerged was whether the sickle cell trait related complications should be described as occurring rarely, as this may have contributed to ineffectiveness of past awareness efforts. The research team presented the likelihood of select complications, such as that people with a sickle cell trait are two times more likely than the average population to have venous thromboembolism.[27] Experts (especially physicians) were able to weigh in on their perception of rarity when looking at the statistics, and the group discussed whether "rare" is inactivating and may make people less likely to be aware of the outcomes, or if "rare" makes people more interested in the outcome and more likely to examine the risk further. The group consensus was reached that we should generally not use "rare" in messaging.

Sickle cell trait community

Six focus groups (n=32) were held to test the Project Messages and two focus groups tested messages that currently exist in the field (n=33), for a total of 65 participants across eight focus groups. Approximately 70% (45/65) of participants had a sickle cell trait.

The messages were tested for overall impressions, tone, whether it was compelling and/or motivating, and in some cases, certain parts of the message were tested. In Table 4: message progression through project phases based on expert and community feedback, each original message is presented with the feedback from the experts and the community to show the evolution of each message, and the final project messages. There were no noticeable differences in the focus groups in the quality of participation among attendees who did and did not have a sickle cell trait.

Table 4: Message progression through project phases based on sickle cell trait expert and community feedback.

Original draft message	Summary of feedback: sickle cell trait expert group meetings	Revised message	Summary of feedback: community message testing focus groups	Final project messages
1. If you don't know if you are a sickle cell trait carrier, get tested.	Add statistics to grab the attention of the reader.	Anyone of any race or ethnicity can have a sickle cell trait; if you don't know [whether you have the gene], get tested. In a United States study, the gene was 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.	Add "know your risk"; use "risk" over "status". Couple message with information about sickle cell trait.	Anyone of any race or ethnicity can have a sickle cell trait. Know your risk; get tested. In a United States study, the trait was 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.
2. The sickle cell trait gene you carry puts you personally at risk of critical and even life-threatening health complications.	"Carry" could be stigmatizing, consensus on using "have a sickle cell trait"; abbreviating to sickle cell trait is confusing. Break into two messages: one about what the complications are, and one about understanding the complications.	Having a sickle cell trait can put you at increased risk for some serious health complications, including... (list of associated health complications). Understand the [possible/potential] health [conditions/complications] associated with having a sickle cell trait.	Prefer "potential" and "complications.	Having a sickle cell trait can put you at increased risk for some serious health complications, including: • Hematuria (blood in the urine) • Hyposthenuria (trouble concentrating urine) • Renal papillary necrosis • Chronic kidney disease • Renal medullary carcinoma • Venous thromboembolism

Continued.

Original draft message	Summary of feedback: sickle cell trait expert group meetings	Revised message	Summary of feedback: community message testing focus groups	Final project messages
				• Splenic infarction • Exertional • rhabdomyolysis • Exercise-related sudden death • Glaucoma and hemorrhage post-eye injury • Having a sickle cell trait also provides protection against severe malaria. • Understand the potential health complications associated with having a sickle cell trait.
3. Your doctors may not be as knowledgeable about sickle cell trait as you are; be prepared to advocate for yourself.	Don't create more medical distrust. Move language around providers toward the positive as to not inactivate the reader; encourage advocating for oneself.	Some healthcare providers may be unaware of emerging research about a sickle cell trait; be prepared to [self-advocate and collaborate with your healthcare provider].	Did not elicit a positive reaction, further created distrust of medical providers and had the opposite effect: "If my providers don't know, why would I go see them?". Additional message was added after participants expressed wanting tangible instructions on what to ask their provider for.	Our understanding of potential health complications associated with having a sickle cell trait is constantly improving. Be prepared to self-advocate and collaborate with your healthcare providers. Be sure to visit your healthcare provider for at least an annual checkup with urine testing that can catch signs of early complications.
4. Tell all of your doctors you have sickle cell trait and that additional testing could save your life.	Combine message 4 and 5.	Message was combined with the message below.		
5. Understand the risks of having sickle cell trait and what you and your doctors do could save your life.	Combine message 4 and 5. Test the wording in brackets with the community.	Tell all of your healthcare providers you have a sickle cell trait and additional testing [may be warranted/needed/helpful] [could save your life].	Use "may be needed and could save your life" as it is the most motivating and urgent.	Tell all of your healthcare providers you have a sickle cell trait and additional testing may be needed and could save your life.
6. Symptoms that would be	Test the wording in brackets with	Symptoms that would be considered trivial in others may	"Use critical" as it is the most	If you have a sickle cell trait, certain s

Continued.

Original draft message	Summary of feedback: sickle cell trait expert group meetings	Revised message	Summary of feedback: community message testing focus groups	Final project messages
considered trivial in others may be critical for your health.	the community. List the symptoms in the message.	be [important/critical] for your health, such as… (List of symptoms).	motivating and provide a call to action. The use of "trivial" is confusing. Specify that this message is for people who already know they have a sickle cell trait.	you experience could be critical to your health; talk to your health care provider if you experience: • Hematuria (blood in the urine), urinating more or less than usual, or dark urine • High blood pressure • Shortness of breath • Chest or flank pain • Muscle cramps or weakness when exercising • Blood in the eye
7. Some external conditions may make sickle cell trait-related health outcomes more likely. These include altitude changes, intense exercise and dehydration.	List the conditions. List the interventions that reduce the outcomes.	Certain extreme conditions, including altitude changes, intense exercise, and dehydration, may make outcomes associated with a sickle cell trait more likely. To reduce the potential for these outcomes… (Interventions that are presently believed to reduce those outcomes, such as hydration, breaks, etc.).	Well received.	Certain extreme conditions, including altitude changes, intense exercise, and dehydration, may make health outcomes associated with having a sickle cell trait more likely. To reduce the potential for these outcomes: • Stay hydrated • Pay close attention to symptoms when in high or low altitudes • Build exercise intensity slowly • Keep your body temperature cool • Seek medical care when experiencing any of the above symptoms.

DISCUSSION

This project, guided by the frameworks CFIR and NGT, provided deep understanding of the awareness and education efforts being delivered in the sickle cell trait field and recommendations for creating messages that address the gaps in education through consistency and scientific backing. By involving sickle cell trait stakeholders including organizations, experts, and the community, this project resulted in consensus among many organizations that they would adopt the messages in an effort to work toward collective consistency in information dissemination across organizations.

To ensure messages were not only consistent, but to ensure the process of creating and testing the messages was scientifically-backed, using CFIR and NGT was at the forefront of all efforts. CFIR allowed thorough understanding of the current landscape of messaging; guided the message development process and formative evaluation; and, provided the structure to organize data collection, analysis, results, and recommendations. Further, NGT provided structure to the decision-making process used in expert meetings to reach consensus on messaging.[21,22] As evidenced in this project and prior research, using these frameworks to guide the message development and testing process yields actionable public health messages that change behavior.[21,28]

Table 5: Semi-structured interview and survey recommendations based on results.

CFIR Domain	Recommendations
Intervention characteristics	Increased collaboration among organizations in order to align messages to provide clear and consistent information and education. Organizations should deeply understand the unique needs of the specific community they serve so they can deliver their messages in ways that are useful and relevant.
Outer setting	Further research is needed to ensure sickle cell trait test results are included in every newborn's medical record, and consistent follow-up education and counselling is implemented to ensure everyone knows whether they have a sickle cell trait. Consistency across organizations in their messaging about having a sickle cell trait so unified, scientifically backed education is provided to the community.
Inner setting	Organizations should expand collaborations with each other to break down silos, and efforts to unite the field and increase collaborations should be expanded. Increased collaboration is a way to work toward aligning messages across the field. Organizations should expand efforts to decrease stigma and correct assumptions about having a sickle cell trait so the general public, the community, and providers are more well-informed. More funding should be provided for sickle cell trait research and awareness efforts to better understand outcomes, increase quality of healthcare for the community, and increase awareness of having a sickle cell trait and its implications.
Characteristics of individuals	Organizations should consider their organizational stance on reproductive choices for people who have a sickle cell trait and make their stance very clear to avoid providing unclear recommendations. Organizations should also provide resources to support their stance. However, the field should work toward consistent messaging surrounding reproductive choices and healthcare for people with a sickle cell trait, and further research into this area could provide a foundation for more informed recommendations.
Process	Consistency across organizations in their messaging about having a sickle cell trait so consistent education and awareness is provided to the community. To reach providers and community members more effectively, organizations could consider their communication efforts that surround their programs and consider additional modes of communication, such as websites and social media engagement. Organizations should focus on expansion and sustainability of their efforts through perhaps identifying program champions or spokespeople.

Developing and testing the messages with the various stakeholders yielded actionable insights for not only message content, but message delivery. In both the organization survey and interviews, the CFIR intervention characteristics domain highlighted the lack of collaboration among organizations, hindering consistent messages. It was also found that organizations focused on chronic health conditions must work together to align education messages to improve health of communities.[29] It is important that organizations understand the unique needs of the community so they can deliver messages in the most effective ways to create behavior change.[30,31] The outer setting domain identified a root cause of a lack of sickle cell trait awareness in is likely that sickle cell trait test results at birth are often not included in a newborn's medical record. Additionally, follow-up education and counseling after a positive test is even less frequently offered. Provider communication and follow up education and counseling following a positive newborn screen is paramount in improving health outcomes and preventing future complications.[32–34]

The inner setting domain underscored the need for increased funding for sickle cell trait research and awareness efforts to enhance understanding of sickle cell trait among providers and the community, as expanded scientific understanding would form the basis for improved messages.[14,35] The characteristics of individuals domain detailed varying organizational stances on reproductive choices for people with sickle cell trait,

which introduced variability in information provided. Despite personal beliefs about reproduction with sickle cell trait, organizations should provide scientifically-backed resources to support their message so people with sickle cell trait can make informed healthcare choices.[36,37]

Finally, the Process domain, while reiterating the importance of message consistency, also provided context around how providers and organizations communicate their messages to their target audiences. While many organizations expressed desire to improve their message delivery methods, the community provided concrete ways that they want to receive information. Participants strongly favored receiving these messages from their healthcare provider, whether verbally or in written form. Participants also reported interest in receiving the messages on social media, but one participant, however, expressed concern that information on social media is not always accurate and that people could respond to the message with incorrect information.

Using NGT gave structure to the decision-making process during the Expert Meetings and provided a process by which messages were developed, tested, and finalized. during Step 1: Generating Ideas, discussion of the nuances of each message produced the need to strive toward consensus on the most basic parts of sickle cell trait. Therefore, in the subsequent meetings the research team presented scientific information surrounding various topics that form the foundation of sickle cell trait and opened up discussion for input from the experts, as it is important for the most basic parts of the condition to form the foundation of the messages.[38]

Step 2: Recording ideas allowed discussion around the importance of how to talk about sickle cell trait through the lens of stigma and scientific understanding of the term. Given patients with the condition are referred to in many ways, such as “sickle cell trait carrier”, “having a sickle cell trait”, etc., the experts and community agreed that consensus was needed on terminology as the foundation for all messaging. Using the terms “SCT” and “sickle cell trait carrier” were not preferred by the community, and therefore, the team agreed that “having a sickle cell trait” is the most clear, appropriate, scientifically backed, and culturally sensitive way to refer to someone who has the condition. This consensus is supported by research including that all chronic conditions should use person-first language, as in “person with a sickle cell trait” vs. “sickle cell trait carrier.”[26] Additionally, messages should use self-empowering language as this is more effective in motivating patients, such as in the project message “Some healthcare providers may be unaware of emerging research about a sickle cell trait; be prepared to (self-advocate and collaborate with your healthcare provider)” versus “Our understanding of potential health complications associated with having a sickle cell trait is constantly improving.[39,40] Be prepared to self-advocate and collaborate with your healthcare providers.” Further, Experts and the Community alike felt the abbreviation “SCT” was not different enough from “SCD”, which contributes to confusion between sickle cell trait and sickle cell disease. Given someone with a sickle cell trait has one copy of the sickle cell gene and sickle cell disease requires inheriting two copies of the sickle cell gene, experts and the community agreed that SCT should not be abbreviated, and “having a sickle cell trait” adequately differentiated sickle cell trait from SCD.[3] Using the “a” was vetted with a hematologist and a geneticist to ensure scientific accuracy.

As a result of the discussions sparked from Step 1 and Step 2, in Steps 3 and 4, messages were further developed and experts voted on a finalized list of messages to test with the community. Consensus among all members of the expert group was important to not only ensure the messages were the most effective they could be, but also to increase the likelihood of adoption among Organizations to improve consistency.[19,23]

The process this project followed to develop and test messages to be delivered to improve health outcomes is a model for the field going forward. Whether for sickle cell trait or other chronic health conditions, using this process and putting stakeholders and community members at the forefront of each step and guiding the process with scientific frameworks is of utmost importance for success

CONCLUSION

Sickle cell trait affects many people in the US, with 1 in 13 people who are Black having the condition. Despite the wide prevalence, the condition is not widely understood, and research and awareness efforts are lacking. Contributors to this lack of awareness include inconsistent messaging being promoted by organizations and healthcare providers. To address this lack of awareness and consistent information, engaging diverse stakeholders and the community was of paramount importance in this effort. Ensuring that organizations, Experts, and the Community were involved heavily in the development, testing, and finalizing of the messages ensured that the messages were holistic, scientifically backed, and would be understood and motivating to the audience. Not only did involving these stakeholders ensure quality messages, but this worked toward achieving the goal of fostering more collaboration among organizations and promoted adoption of a consistent set of messages. Overall, engaging these stakeholders and using CFIR and NGT helped us achieve consistent, clear, and evidence-based messages that are holistically informed and fostered stakeholder alignment.

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