# Newborn Screening Programs and Sickle Cell Disease

## A Public Health Services and Systems Approach

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**Introduction:** Despite universal newborn screening (NBS), children in the U.S. continue to experience morbidity and mortality from sickle cell disease and related causes. Recognizing that assessments of public health services and systems can improve public health system performance and ultimately health outcomes, this study examined variations in NBS program activities for sickle cell disease.

**Methods:** A mixed methods study included (1) a 2009 survey of NBS programs based on ten essential public health services (N=39 states with ten or more sickle cell births over a 3-year period) and (2) key informant interviews in 2011 with 13 states that had sufficient Phase 1 survey scores, black births, and variability in state legislation and geography. Key informants were from 13 NBS programs, 22 sickle cell treatment centers, and ten advocacy organizations. Analyses were conducted in 2009–2014.

**Results:** Considerable variability exists across states in program activities and roles. More programs reported activities oriented to care of individuals—ensuring access to services, coordination, and provider education; fewer reported planning and analysis activities oriented to statewide policy development and system change. Numbers of activities were not related to the number of affected births. In-depth interviews identified opportunities to enhance activities that support statewide comprehensive systems of care.

**Conclusions:** NBS programs perform important public health roles that complement and enhance clinical services. Nationwide efforts are needed to enable NBS programs to strengthen population-based functions that are essential to ensuring quality of care for the entire population of children and families affected by sickle cell disease.

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## Introduction

New York was the first state to implement newborn screening (NBS) for sickle cell disease in 1975; by 2006, universal NBS for sickle cell disease was in place in all U.S. states, Puerto Rico, and the U.S. Virgin Islands.[1] NBS permits timely diagnosis, initiation of penicillin prophylaxis, and establishment of comprehensive care.[2] NBS has contributed to decreased morbidity and mortality among those affected by sickle cell disease and increased receipt of specialized medical care necessary to manage the disease and its sequelae.[3] However, although notable improvements in survival for children with sickle cell disease have been realized, these children continue to experience disabilities, higher mortality, and greater difficulty accessing healthcare services relative to children without sickle cell disease.[4]

*Healthy People 2020* calls for increasing appropriate bloodspot NBS and follow-up testing.[5] Although variability in NBS and follow-up has been reported[6,7] and the need to consider NBS in light of public health functions recognized,[3] there has been no systematic reporting about the extent of variability in NBS specific for sickle cell disease across states

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or consideration of whether variability is a function of the size of the affected population. The gap is notable despite sickle cell disease being one of the first conditions recommended for NBS.[1,8] The lack of information is also concerning given that sickle cell disease disproportionately impacts populations of African, Mediterranean, Middle Eastern, and Indian descent,[8] many of whom are historically underserved in terms of healthcare services.

The objective of this study was to assess variability in NBS program structure and activities for sickle cell disease in the U.S. A two-phase study was conducted to

1. characterize organizational structures and functions of state NBS programs for sickle cell disease, from initial screening through longer-term follow-up and system-level roles; and
2. identify facilitators and barriers for developing a comprehensive public health approach to NBS and follow-up for sickle cell disease.

For Phase 1, the authors hypothesized that states with a larger number of affected births would have more-robust NBS activities specific to sickle cell disease.

## Methods

This mixed methods approach involved quantitative data gathered through surveys of NBS program directors (Phase 1) and qualitative data obtained from key informant interviews with stakeholders (Phase 2). This approach incorporated diverse perspectives to develop a more complete understanding of NBS activities.[9]

### Study Sample

**Phase 1: survey of newborn screening programs.** Study eligibility criteria for states were based on the number of confirmed or presumed newborns with sickle cell disease as reported in the 2001–2006 National Newborn Screening Information System database and with input from the study's Advisory Committee. A total of 38 states and the District of Columbia were eligible to participate based on the following:

1. twenty-eight states and the District of Columbia—ten or more confirmed cases of sickle cell disease in newborns over all 3-year periods between 2001 and 2006; and
2. ten states—ten or more confirmed cases ($n$=7) or presumed cases ($n$=3) of sickle cell disease over at least one 3-year period between 2001 and 2006.

All 39 eligible NBS programs participated in Phase 1.

**Phase 2: key informant interviews.** Eligibility for inclusion in Phase 2 was informed by input from the study's Advisory Committee to achieve purposive sampling and, empirically, reach saturation of relevant themes. Criteria for participation in Phase 2 included participation in the Phase 1 survey (N=39); total public health function domain summary score from Phase 1 survey of ≥11 (range, 0–38 out of possible score of 50 among the 39 states); and ≥3,000 live black births in 2007. The cutoff of 11 was selected based on a natural break in the data and the desire to include states with sufficient scores to examine NBS activities. Of the 24 states that met Phase 2 eligibility criteria, 13 were selected. They represented variability in geographic location, state legislation related to sickle cell disease, and federal funding for sickle cell–related activities. Attempts were made to minimize overlap with the seven states participating in the Registry and Surveillance System pilot being conducted by the National Heart, Lung, and Blood Institute (NHLBI) and the Centers for Disease Control and Prevention (CDC).

Within the 13 states, key informants included the NBS Program Director, director(s) of major sickle cell center(s), and the director of the state or regional Sickle Cell Disease Association affiliate(s) or major advocacy organization(s).

Key informant interviews were conducted with 17 of 17 representatives from the 13 NBS programs (ten states provided one key informant, two states provided two key informants, and one state provided three key informants); 22 of 26 sickle cell treatment center directors (one to three centers per state); and 10 of 16 advocacy organization representatives (zero to two organizations per state) for a total of 49 key informant interviews.

### Data Sources and Measures

**Phase 1: survey of newborn screening programs.** Surveys of state NBS programs were developed based on prior literature[10] and seven of the ten essential public health services[11] derived from the core public health functions of assessment, policy development, and assurance.[12] Services included data assessment and analysis, community partnerships and mobilization, policy planning and development, quality assurance, service linkages and coordination, workforce capacity, and program evaluation. The three essential public health services not relevant to this project were excluded: diagnosis and investigation (which is implicit to all NBS programs); research demonstrations (not relevant at state-level NBS); and information and education (because of focus on population-based systems and activities rather than the promotion of individual behavior change). The survey was piloted with NBS program directors in two states not eligible to participate and revised accordingly. The survey items comprising each of six composite domains were adapted from the essential public health services (Table 1).

**Abstraction of state legislation related to sickle cell disease and review of federal funding.** A systematic review of state legislation related to sickle cell disease was conducted to categorize states with regard to policy actions and guide key informant sampling from among the 39 states that completed the Phase 1 survey. This review included statutes and regulations and excluded executive orders. Data sources included websites of the National Conference of State Legislatures—Sickle Cell Legislation; U.S. Laws, Codes, Statutes & Cases—Justia (search terms: *sickle*, *sickle cell*, and *hemoglobinopathy*); and individual state legislature websites with searches for statutes and regulations with key terms.

Four categories of legislation were identified to establish

1. a commission, committee, or board specific to sickle cell disease;
2. a program specific to sickle cell disease;

**Table 1.** Newborn Screening for Sickle Cell Disease Survey Items by Public Health Domain (*n*=39 States)

| Survey items by public health domain | *n* (%) |
|---|---|
| Data collection and analysis | |
| • Collected population-level data in past 2 years[a] | 20 (51.3) |
| • Analyzed population-level data in past 2 years[a] | 13 (33.3) |
| • Conducted focus groups and/or needs assessment | 18 (46.2) |
| Policy development | |
| • Presented information to state political leaders at least annually | 4 (10.3) |
| • Developed fact sheets for community organizations | 18 (46.2) |
| • Participated in local advocacy coalition | 15 (38.5) |
| • Worked with state medical societies or other organizations to advocate | 16 (41.0) |
| • Produced a plan to address health needs of children with sickle cell disease | 7 (17.9) |
| • Undertook a formal process to develop strategies to improve health of children with sickle cell disease | 13 (33.3) |
| • Produced report on progress toward meeting state health goals | 17 (43.6) |
| • Provided expertise on sickle cell disease to state legislative or regulatory bodies for development of policy or programs | 13 (33.3) |
| • Provided expertise on sickle cell disease to other organizations in state | 20 (51.3) |
| Quality assurance | |
| • Developed standards of care for children with sickle cell disease | 11 (28.2) |
| • Participated in quality improvement of facilities/providers of care | 12 (30.8) |
| • Monitored providers of care/services[b] | 14 (35.9) |
| • Evaluated newborn screening services available to families | 10 (25.6) |
| • Worked with other organizations to collect/analyze data on consumer satisfaction, access issues, quality of care, etc. | 10 (25.6) |
| • Used data for quality improvement | 18 (46.2) |
| Coordination | |
| • Met with stakeholders to coordinate activities to meet needs of children with sickle cell disease[c] | 33 (84.6) |
| Workforce development | |
| • Disseminated comprehensive, written education plan for any audience (providers, consumers, policy makers, etc.) | 10 (25.6) |
| • Participated in developing strategies to address workforce shortages for care providers | 7 (17.9) |
| • Targeted care providers for education about sickle cell disease[d] | 24 (61.5) |
| Access to services | |
| • Tracked and followed up on receipt of care[e] | 32 (82.1) |
| • Met with service providers to enhance specific activities/services for families[f] | 18 (46.2) |

*Note*: Time frame for activities was past 4 years, unless otherwise noted. All activities are specific to children with sickle cell disease and their families.
[a]Includes one or more of the following: demographic characteristics, SES, health status, receipt of medical services, follow-up case management, and health outcomes.
[b]If responded yes to global question regarding monitoring. Follow-up questions assessed monitoring of managed care health plans, private providers and facilities, public health programs or services, and local hospital services.
[c]Includes one or more of the following groups: university medical facility/school, community organizations, local hospitals, primary care associations, state medical associations, community health centers, school-based health clinics, hematologists, WIC, Head Start, and other.
[d]Includes one or more of the following groups: doctors, nurse midwives, nurse practitioners, nurses, schools, and social workers.
[e]Includes one or more of the following: referrals of high-risk infants to specialty care, visit with hematologist, well baby visit in first month of life, appropriate penicillin prophylaxis, influenza and pneumococcal vaccines, transcranial Doppler ultrasound, and parental instruction in spleen palpation.
[f]Includes one or more of the following: identification of high-risk infants, treatment options, and other.

3. a commission or committee that addresses NBS and specifically identifies sickle cell disease; and
4. a program for NBS that specifically identifies sickle cell disease.

Of the 39 states, 18 states had no legislation, 14 states had one category of legislation, six states had two categories, and one state had three categories.

Additionally, states receiving Federal funding from NHLBI, Health Resources and Services Administration, and CDC initiatives were identified. These included NHLBI Comprehensive Sickle Cell Centers, Health Resources and Services Administration Sickle Cell Treatment Demonstration Grants, Health Resources and Services Administration Sickle Cell Disease and NBS Program grants, and CDC/NHLBI Registry and Surveillance System for Hemoglobinopathies.

**Phase 2: key informant interviews.** In-depth, structured telephone interview guides for each stakeholder type were developed. Interviews explored factors affecting implementation of public health activities and services for children with sickle cell disease. Interviews also elaborated on themes derived from the quantitative survey and explored issues amenable to qualitative data collection.

### Data Collection and Analysis

**Phase 1: surveys of newborn screening programs.** Two trained research assistants collected surveys by phone from May 26, 2009, to September 22, 2009, transcribing responses during the calls. Exploratory analyses of each of the variables were conducted, and missed skip patterns, outliers, and missing data were assessed.

The state was the unit of analysis. Descriptive tables and statistics were used to characterize public health activities.

Using survey data, composite measures were developed to describe public health services in each state related to NBS for sickle cell disease (Table 1). Summary scores (and the corresponding domain of essential public health services) related to data functions, policy functions, quality assurance, coordination and partnerships, workforce development, and access to services. In addition, a total cross-domain score was calculated (possible range, 0–50) and activities most and least commonly reported were identified for each domain (Table 2). Nonparametric tests (Mann–Whitney, Kruskal–Wallis) were used to examine associations between numbers of sickle cell births by state in 2006 and public health functions and capacities expressed as public health services.

**Phase 2: key informant interviews.** Data were collected from March 17, 2011, to June 15, 2011, in phone interviews conducted by two trained research assistants and a study investigator. Extensive notes were taken during the calls. Content analysis was conducted using an adapted version of Framework Analysis[13] to facilitate the systematic analysis of data through key stages. First, familiarization or the review of the data took place. Second, a thematic framework, or an initial coding framework for each respondent type and for each domain, was identified; they were refined during subsequent stages. These frameworks were developed from a priori issues (e.g., learned from Phase 1 analysis and discussions with the study's Advisory Committee) and from issues emerging during familiarization. Third, charting was used to create summaries of data based on headings emerging from the thematic framework. Charts that were thematic across all respondents rather than sorted by each respondent across all themes were constructed. All interview data were double coded and consensus reached between the two reviewers for any discrepancies. Fourth, patterns, associations, concepts, and explanations in the data were identified, in order to create typologies and develop strategies/recommendations.

Several aspects of program implementation were examined, consistent with the Donabedian[14] framework for evaluation using the key concepts of structure and process; outcomes were not assessed in these analyses. Also, contextual factors were explored with respect to the historic and political environment within which the sickle cell disease system of care evolved and currently operates. Study variables within the structure, process, and context domains derived from the theoretic literature on implementation, as well as from published studies of local and state systems interventions.[15–21] Themes from states scoring high in relevant domains from the Phase 1 survey and related challenges or issues were reported. Analyses were conducted in 2009–2014.

The Johns Hopkins Bloomberg School of Public Health IRB determined this study to not be human subjects research.

## Results

Among the 39 respondents who completed Phase 1 surveys (100% participation), 59.0% were NBS directors, 35.9% were NBS coordinators, and 5.1% held other job titles. On average, they had worked 13.0 years in the health department (range, 0.5–30 years) and 6.8 years in their current roles (range, 0.25–30 years). Most respondents ($n$=23, 59.0%) indicated their programs had staff designated for hemoglobinopathies. Participants reported that long-term follow-up for sickle cell disease is performed by state agency staff ($n$=3, 7.7%); by external entities ($n$=12, 30.8%); and a mix of state agency staff and external entities ($n$=12, 30.8%) or not performed ($n$=12, 30.8%).

More states collected than analyzed population-level data on child and family characteristics, such as SES, health status, receipt of medical services, or health outcomes ($n$=20, 51.3% vs $n$=13, 33.3%, respectively, Table 1). Only ten states (25.6%) collected data on three or more of these characteristics. Nineteen states reported no population-level data collection. Eighteen states (46.2%) conducted focus groups or compiled data for a needs assessment to learn about children with sickle cell disease.

Fewer than half of NBS programs engaged in most policy development activities. Activities related to serving as a resource for other entities were reported more frequently than formal policy development and planning processes. Although 20 (51.3%) provided expertise on sickle cell disease to other organizations in the state,

**Table 2.** Public Health Domain Scores for Newborn Screening for Sickle Cell Disease Survey (*n*=39 States)

| Public health domain | Potential range | Actual range | M | Median | SD | Most commonly reported activities | Least commonly reported activities |
|---|---|---|---|---|---|---|---|
| Data collection and analysis | 0–3 | 0–3 | 1.3 | 1 | 1.1 | • Collected population-level data on demographic characteristics (*n*=16)<br>• Conducted needs assessment (*n*=13) | • Analyzed population-level data on health status at birth or socioeconomic characteristics (both *n*=4) |
| Policy development | 0–9 | 0–8 | 3.1 | 3 | 2.3 | • Provided expertise on sickle cell disease to other organizations in the state (*n*=20)<br>• Developed/ disseminated fact sheets for community organizations (*n*=18) | • Presented information to state political leaders at least annually (*n*=4)<br>• Produced a state plan to address needs (*n*=7) |
| Quality assurance | 0–9 | 0–8 | 2.2 | 2 | 2.0 | • Used data to improve quality of services for families (*n*=18)<br>• Monitored private providers or facilities (*n*=13) | • Monitored managed care organizations (*n*=2) or local hospitals (*n*=5) |
| Coordination | 0–11 | 0–8 | 2.9 | 2 | 2.5 | • Coordinated with university-based medical facilities/ medical schools (*n*=28) or community organizations (*n*=20) | • Coordinated with WIC or Head Start (both *n*=3) |
| Workforce development | 0–8 | 0–7 | 2.1 | 1 | 2.1 | • Targeted education to physicians (*n*=21) or nurses (*n*=16) | • Developed education plan for policy makers or birthing facility staff (both *n*=3) |
| Access to services | 0–10 | 0–9 | 3.9 | 4 | 2.4 | • Tracked if children received penicillin prophylaxis (*n*=27) or kept a visit with hematologist (*n*=26) | • Tracked if children received transcranial Doppler ultrasound (*n*=5) or parents received education about spleen palpation (*n*=6) |
| Total | 0–50 | 0–38 | 15.6 | 14 | 9.4 | | |

WIC, Special Supplemental Nutrition Program for Women, Infants and Children.

only four (10.3%) presented information annually to state political leaders and seven (17.9%) produced a plan to address the health needs of children with sickle cell disease.

Eleven (28.2%) programs participated in developing standards of care for children with sickle cell disease. Twelve (30.8%) participated in quality improvement activities of facilities or providers of care. Only ten (25.6%) reported that they evaluated the NBS services available to families. However, 18 (46.2%) programs reported using data for quality improvement.

Workforce development activities included providing education about sickle cell disease for healthcare professionals (*n*=24, 61.5%) and, less frequently, disseminating comprehensive written education plans (*n*=10, 25.6%) or developing strategies to address workforce shortages (*n*=7, 17.9%).

Most respondents reported that they met with stakeholders to coordinate services for children with sickle cell disease and track receipt of care (*n*=33, 84.6% and *n*=32, 82.1%, respectively). Twenty-seven states (69.2%) reported having processes to track receipt of penicillin prophylaxis, two thirds of which do so until at least age 18 years. Twenty-six states (67%) tracked if hematology visits were kept, and 11 states (28.2%) tracked receipt of influenza and pneumococcal vaccines.

Numbers of activities were not related to the numbers of affected births (all $p > 0.05$).

**Table 3.** Themes From Key Informant Interviews With Newborn Screening Stakeholders, by Public Health Domain ($n$=13 States)

| Public health domain | Selected activities among high scoring states | Related challenges or issues |
|---|---|---|
| Data collection and analysis | • Disseminated questionnaires to individual providers or contracted treatment centers on quarterly or annual basis to ensure children were in care<br>• Used secondary data (e.g., hospital data, Medicaid claims) for comprehensive needs assessments and activity planning | • Limited capacity to analyze data from providers or secondary sources<br>• Most data collection/analysis activities are not truly population level given population mobility, loss to follow-up |
| Policy development | • Convened newborn screening advisory committees/ hemoglobinopathies working groups<br>• Created state plans/consortia with key stakeholders<br>• Used a regular planning cycle that coincided with grant funding for treatment centers to assess needs and inform programs<br>• Collaborated with state Medicaid or children with special health care needs programs<br>• Held annual family day at state capitol to increase awareness of sickle cell disease among policy makers | • Demographic changes in states with increasing Hispanic and African populations<br>• Lack of funding for planning or fully implementing plans and programs<br>• Reliance on dedicated individuals (advocates, specialists) who drive policy and programming and are difficult to replace |
| Quality assurance | • Developed standards of care (e.g., care pathways for emergency department visits, pain management, medical home) in consultation with stakeholders at the state or regional level<br>• Leveraged contracts (funded or unfunded) with designated treatment centers to set standards, and to monitor and evaluate activities<br>• Conducted annual newborn screening program evaluations that included parent satisfaction surveys<br>• Utilized newborn screening program–funded hemoglobin follow-up coordinator at treatment centers to track and reduce time to specialist referral<br>• Reported to state legislature on meeting goals for penicillin prophylaxis | • Reliance on treatment centers/specialists to develop standards of care; perception of limited role for newborn screening program<br>• Most quality improvement activities conducted at individual treatment centers; not statewide and not coordinated with or led by newborn screening program |
| Coordination | • Held annual conferences that included professional organizations, churches, community-based organizations, etc.<br>• Convened contracted treatment centers annually to increase coordination and communication<br>• Created memoranda of understanding or other agreements with community-based organizations so information on positive screens/trait could be sent to them and they could do outreach<br>• Automatically referred affected children to children with special health care needs case management, early intervention; collaborated with school nurses | • Difficulty engaging primary care physicians across the state; many individuals each with very few affected patients<br>• Collaborations with community-based organizations vary greatly depending on their size, number, history of leadership, and involvement with newborn screening program and other organizations<br>• Lack of mechanisms to encourage coordination and communication across treatment centers |
| Workforce development | • Provided updated information on sickle cell disease for clinicians and families in print and online<br>• Held annual symposia or meetings to provide education to a range of providers and consumers<br>• Created a written education plan for clinicians; comprehensive treatment handbook<br>• Funded local newborn screening projects to provide outreach and education to providers (example: survey of school nurses to inform education efforts)<br>• Trained counselors at community-based organizations to provide trait counseling to families | • Need for increased knowledge among care providers in emergency departments, urgent care centers in treating sickle cell crises<br>• Lack of providers for adolescents transitioning out of pediatric specialty care |

*(continued on next page)*

**Table 3.** Themes From Key Informant Interviews With Newborn Screening Stakeholders, by Public Health Domain ($n$=13 States) *(continued)*

| Public health domain | Selected activities among high scoring states | Related challenges or issues |
|---|---|---|
| Access to services | • Increased access to care through mobile clinics in rural areas or collaborations with community health centers<br>• Used regional social workers or newborn screening program–funded coordinators at treatment centers to ensure follow-up<br>• Developed computer systems that allow newborn screening program and treatment centers to share information about screening results, treatment plans, etc.<br>• Conducted focus groups with parents to evaluate access to care<br>• Tracked receipt of services (when number of treatment centers is small) | • Most states track children only until confirmatory diagnosis or initiation of specialty care<br>• Size/structure of state and location/number of treatment centers may have a significant impact on follow-up and tracking activities<br>• Few states reported transition services for adolescents moving on to adult care |

The mean total cross-domain score was 15.6 (SD=9.4) out of a possible score of 50 and actual range of 0–38 (Table 2). For each public health domain, mean and median scores were less than the midpoint of the range. Common themes from the key informant interviews were identified for each domain (Table 3). The activities reported by higher-scoring states reflected strategic use of public agency tools (e.g., contracts, memoranda of understanding, planning and reporting cycles) as levers to build system capacity and promote a comprehensive approach to quality care. These higher-scoring programs also often served as conveners, creating focal points for planning, policy development, and capacity-building activities. Challenges comprised a mix of issues, including those mutable in the short term with additional resources (e.g., limited capacity for data analysis or implementing programs); those amenable to quality improvement activities (e.g., need to engage primary care physicians, need for coordination mechanisms across treatment centers); and those that may be more intractable or require substantial resource commitments (e.g., reliance on individual leaders to shoulder efforts, variability in capacity of community organizations).

## Discussion

Using a public health services and systems approach, considerable variability was observed across states in program roles and activities related to NBS and follow-up for sickle cell disease. More programs report activities oriented to care of individuals—ensuring access to services, coordination, and education for providers; fewer report policy development and data analysis activities oriented to statewide policy and systems change. Variability in program activities is not related to number of sickle cell disease–affected births. This may be due to limited funding, lack of political will, low priority on the public health agenda, ineffective advocacy, or challenges of collaborating or changing policies/practices in some states with more affected births.

The findings suggest a need for enhanced efforts to strengthen population-based functions that support statewide comprehensive systems of care for children affected by sickle cell disease. These efforts are made even more necessary by demographic shifts in many states, growing accountability by the healthcare sector for population-level outcomes, and a heightened recognition of health disparities. Moreover, recent advances in health information technology[22,23] and administrative data linkages[24] may provide opportunities to generate population-level data and encourage pooled resources to enhance analytic capacity. The performance evaluation and assessment scheme,[25] in combination with quality indicators specific to sickle cell disease (including measures for genetic counseling and penicillin prophylaxis),[26] provide opportunities for states to use quality improvement processes to promote population health.

To date, federal investments in demonstration programs and private sector advocacy efforts[27] have not enabled NBS programs to reach their full potential or maintain gains already achieved. In the current economic environment, substantially more resources are unlikely to be available to bolster systems of care for children affected by sickle cell disease. However, promising practices do exist in many states. Working together, federal public health leaders and national professional and consumer/advocacy organizations could draw on those practices to formulate model program elements and provide and guide progress nationwide.

### Limitations

Several limitations are noted. First, data were collected from 2009 to 2011, and NBS programs have evolved with expansion of the Recommended Uniform Screening Panel, recommendations for implementing health information technology, and implementation of quality improvement initiatives in selected states.[28] However, despite demonstration projects, fundamental gaps persist regarding public health functions. Second, these assessments depend on respondent knowledge and experience; to facilitate accuracy, surveys were shared in advance and additional participants joined calls as appropriate. Third, social desirability bias may have led some to overestimate achievements; however, varied responses with regard to public health services and triangulated qualitative responses within and across states suggests a willingness to report accurately. Fourth, there were small sample sizes with states as the unit of analysis; however, no other states had sufficient number of births to be included. Also, for key informant interviews, saturation was reached for most themes, suggesting adequate numbers of study participants. Finally, the impact of alternative organization and delivery strategies on health outcomes that would strengthen public health services and systems approach was not examined.[29] However, strategies were noted that study participants across stakeholder groups identified in states with favorable public health services and systems domain scores.

This examination of variability in NBS programs also benefited from several strengths. The mixed methods design enabled the authors to validate and elaborate on survey responses. Relevant themes were identified for state agencies seeking to assess their functioning relative to other states and federal agencies charged with supporting the development of state public health capacity. The findings also reflect contextual factors regarding states' organization of services and development of collaborative relationships. In addition, stakeholders outside the public health sector were included to encompass community organizations as well as public health agencies and healthcare providers, recognizing each of their contributions to population health.[30]

## Conclusions

Current NBS programs are situated to perform important public health roles that complement and enhance clinical services, fulfilling statewide planning, quality assurance, and policy functions that are beyond the purview of individual clinical centers but are essential to ensuring quality of care for the entire population of affected children and families. The findings of variability across NBS programs with regard to public health functions for sickle cell disease suggest the need to pursue strategies that enable public health NBS programs nationwide to achieve their full potential.

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