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# Enablers and barriers to newborn screening for sickle cell disease in Africa: results from a qualitative study involving programmes in six countries

Natasha M Archer ,[1] Baba Inusa,[2] Julie Makani,[3] Siana Nkya,[3] Léon Tshilolo,[4] Venee N Tubman,[5] Patrick T McGann ,[6] Emmanuela Eusebio Ambrose,[7] Natalie Henrich,[8] Jonathan Spector,[9] Kwaku Ohene-Frempong[10]

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NMA and BI are joint first authors.

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For numbered affiliations see end of article.

**Correspondence to**
Dr Natasha M Archer;
natasha.archer@childrens.harvard.edu

## ABSTRACT

**Objectives** Given the fundamental role of newborn bloodspot screening (NBS) to enable prompt diagnosis and optimal clinical management of individuals with sickle cell disease (SCD), we sought to systematically assess enablers and barriers to implementation of NBS programmes for SCD in Africa using established qualitative research methods.

**Setting** Childbirth centres and NBS laboratories from six countries in East, West and Southern Africa.

**Participants** Eight programme leaders involved with establishing and operating NBS programmes for SCD in Angola, Democratic Republic of Congo, Ghana, Liberia, Nigeria and Tanzania.

**Primary and secondary outcome measures** Data obtained through a structured, phased interview approach were analysed using a combination of inductive and deductive codes and used to determine primary themes related to the implementation and sustainability of SCD NBS programmes.

**Results** Four primary themes emerged from the analysis relating to governance (eg, pragmatic considerations when deploying overcommitted clinical staff to perform NBS), technical (eg, design and execution of operational processes), cultural (eg, variability of knowledge and perceptions of community-based staff) and financial (eg, issues that can arise when external funding may effectively preclude government inputs) aspects. Key learnings included perceived factors that contribute to long-term NBS programme sustainability.

**Conclusions** The establishment of enduring NBS programmes is a proven approach to improving the health of populations with SCD. Organising such programmes in Africa is feasible, but initial implementation does not assure sustainability. Our analysis suggests that future programmes should prioritise government partner participation and funding from the earliest stages of programme development.

## Strengths and limitations of this study

- This is one of the largest studies of enablers and barriers to successful implementation and sustainability of sickle cell disease (SCD) newborn screening programmes in Africa, where no national-level programmes currently exist.
- Applying established qualitative research methods, this study investigated the first-hand experiences of clinical and coordinating leaders involved in establishing and operating programmes in six African countries: Angola, Democratic Republic of Congo, Ghana, Liberia, Nigeria and Tanzania.
- Six programmes were included in the analysis, which is a sample of the total number of newborn screening programmes for SCD that have been implemented in Africa.
- By design, a single or small number of participants were surveyed from each programme.
- The lessons learnt from one country may not always be immediately transferable to other countries due to various local factors.

## INTRODUCTION

Sickle cell disease (SCD) is one of the world's most common haemoglobinopathies, estimated to affect in excess of 400 000 newborns annually with 80% of patients born into populations living in low-income and middle-income countries.[1 2] The disease is caused by a single point mutation in the beta-globin gene that results in the formation of sickle haemoglobin (HbS).[3] Under certain conditions, including hypoxia, HbS polymerises and creates distorted (ie, 'sickle' shaped), adherent and less deformable red blood cells (RBCs).[4] The result is easily haemolysed RBCs with a shortened lifespan, endothelial damage, vessel obstruction and other pathophysiological effects that collectively contribute to the development of a vast constellation of acute and chronic clinical manifestations and, often, premature mortality.

Fetal haemoglobin (HbF), the predominant haemoglobin during gestation and in neonates, is the most potent known inhibitor

of HbS polymerisation. As such, infants with SCD are asymptomatic until HbF levels decline to low levels, typically within the first 6–24 months of life. Early diagnosis prior to the predominance of HbS is critical to allow for provision of early lifesaving interventions. Since SCD cannot be diagnosed by clinical signs at birth, newborn bloodspot screening (NBS) materialised decades ago to be a standard approach in many high-resource countries for identifying babies with SCD before complications develop.[5 6] Early detection enables the prompt initiation of parental education and evidence-based preventative care practices that include penicillin prophylaxis and pneumococcal vaccination.[7 8]

In the 1980s, a randomised, placebo-controlled trial in the USA confirmed the efficacy of penicillin prophylaxis in significantly reducing incidence of and mortality due to *Streptococcus pneumoniae*, the leading cause of death in young children with SCD.[5] Evidence from that study provided the impetus for the US National Institutes of Health Consensus Development Conference on Newborn Screening for SCD and Other Hemoglobinopathies to recommend that all babies born in the USA be screened for SCD.[9] In the USA, where universal NBS for SCD (ie, testing newborn babies within the first few weeks after birth) has existed in all 50 states since 2006, NBS is largely acknowledged to be among the most important factors leading to high rates (well over 90%) of survival into adulthood.[5 10 11] Universal screening for SCD now constitutes national policy in the USA, Brazil, UK, Germany, Spain, the Netherlands and Malta;[12–15] longstanding NBS programmes have also been in place in other parts of Europe, Jamaica, Ghana and Canada.[13 16 17] Targeted screening of newborns (eg, according to ancestry) is implemented in some regions but has been shown to be less effective compared with universal screening at identifying infants with disease and preventing deaths.[18]

The vast majority of people with SCD globally are born in Africa, where up to 2% or more of births are reported to be affected in some regions, contributing silently but significantly (8%–16%) to under 5 years of age mortality in high burden countries.[19–21] While no country in Africa has yet implemented policies for universal screening, various national NBS programmes for SCD have been organised, and with heightened awareness about the impact of the disease, there is optimism for increased progress in the future.[19 20 22–26] In this context, we sought to characterise the enablers and challenges to conducting NBS for SCD based on the experiences of previous and ongoing programmes. Specifically, we assessed programmes in Angola, Democratic Republic of Congo (DRC), Ghana, Liberia, Nigeria and Tanzania.[19 20 23–25 27] Using established qualitative research methods,[28–30] we conducted semi-structured interviews with clinical and coordinating leaders involved in each programme and extracted key messages to codify main lessons learnt. This analysis is envisioned to be a resource for patients, clinicians, policy-makers and other stakeholders seeking to improve health systems relating to NBS for SCD in Africa and other limited resource settings globally where SCD occurs in high prevalence.

## METHODS

### Study design

We conducted a qualitative descriptive study that incorporated data from semi-structured interviews with individuals who were responsible for, or significantly involved in, the design and implementation of NBS programmes for SCD in an African country (hereafter referred to as 'participants').[31] The purpose of the interviews was to describe the process for designing and implementing the programmes, identify enablers and challenges, and elicit lessons learnt in order to facilitate a concise summary of learnings that could be used to inform future SCD NBS programmes. Additionally, participants provided background information about their programme by email in advance of their interview. If a participant did not provide the information prior to their interview, then these questions were asked at the start of the interview. See online supplemental materials for the background questions and interview guide.

Interviews were conducted in two phases. The first phase included four participants (representing programmes in Ghana, Angola, DRC and Liberia), who answered a comprehensive set of questions about their programmes. Interviews were transcribed, coded and analysed after the first phase of data collection. From this analysis, the study team identified aspects of SCD NBS programme that warranted deeper exploration either because they emerged as critical to the success of the programme or because they were characterised by variability that prompted deeper investigation across programmes. The latter included aspects of the programme that were subjective (eg, cultural attitudes toward SCD) as opposed to mechanistic (eg, the type of test used to screen for SCD). The second phase included two participants (representing programmes in Nigeria and Tanzania), who answered questions on the topics determined in phase 1 that required further discussion. By limiting the number of questions asked in the second phase, the study team was able to conduct deeper exploration of each of the topics. The findings from phase 2 supplemented the results from the corresponding topics in phase 1. The results from the two phases were analysed together to identify key learnings for the establishment and maintenance of SCD NBS programmes in Africa.

### Patient and participant involvement

Patients were not involved in this study. Participants were identified by study members as programme leaders after reviewing publications related to SCD NBS in African countries. Participants were recruited by email. During the recruitment, all participants confirmed that they were programme leaders and they reported various levels of public engagement in their respective countries. All participants were invited to review the results and to

contribute to identifying key messages and implications of the results, clarify or correct any information from their interviews, and co-author the resulting manuscript (ie, in alignment with a form of 'member checking' described in the literature).[32] One participant was also a study member (KO-F). This study member was not involved in the coding, analysis or preliminary interpretations of the data to minimise the risk that this study member's own experiences would bias the results.

### Interview guide

We designed the interview guide to gain insight into how participants developed, implemented and, when applicable, sustained their programme. The team's qualitative researcher (NH) led the creation of the interview guide with input from a study team member with extensive knowledge about SCD newborn screening programmes in Africa (KO-F) and from study team members with general expertise about SCD (JS and NMA). Collectively, the study team identified the key steps of establishing and implementing a screening programme as well as other factors that were likely to impact the success of the programme. These high-level topics included: programme partners, planning the programme, launching the programme, logistics of day-to-day operations, establishing and running the laboratory, patient notification and follow-up, funding and costs, programme disposition and perceptions of the programme by families of newborns. The interview guide was piloted with a member of the study team (KO-F) for clarity, flow and duration. Minor revisions to the interview guide were made based on his feedback and his responses were included in the dataset.

### Data collection and analysis

Participants were interviewed one time for approximately 1 hour. Phase 1 interviews took place between October 2017 and December 2017. Phase 2 interviews took place between July 2019 and September 2019. All interviews were conducted by phone, audio recorded and transcribed verbatim. Phase 1 interviews were conducted by the qualitative specialist on the team (NH), who received training on SCD-specific content from the other team members and studied relevant literature to become additionally familiar with the topic. Phase 2 interviews were conducted by a team member with content expertise who had prior interviewing experience (JS).

We performed a thematic analysis of the interviews using a coding scheme developed with a combination of inductive and deductive codes. In phase 1, coding was performed in NVivo (QSR) and the content from each code was summarised in a table, including key quotes and identification of key findings. Key findings were used to identify areas that required more in-depth exploration during the second phase of data collection. Phase 2 interviews were analysed by directly adding key findings into the summary tables from phase 1. Results were shared with the participants for feedback and, if needed, corrections, clarifications and the addition of missing information.

## RESULTS

### Study sample

The study involved data collection relating to NBS programmes in six countries in Africa (figure 1) with representation from West Africa (Ghana, Liberia and Nigeria), Central Africa (Angola and DRC) and East Africa (Tanzania). Participants were based at academic institutions and professional societies; many had worked in conjunction with government agencies and external collaborators. The planning period before the initiation of screening ranged from approximately 9 months to 4 years, and the duration of screening ranged from 21 months to 25 years. The number of birth centres involved in the NBS programmes ranged from 1 to approximately 250. Most programmes are ongoing in some capacity, although several with reported periods of inactivity due to various operational challenges as described below.

### Qualitative findings

Four primary themes emerged in the analysis relating to (a) structure and governance; (b) technical aspects; (c) culture and (d) finances. Within these four main themes, we identified 12 subthemes that are summarised in table 1 and described below. A summary of major lessons learnt/recommendations is provided in table 2.

#### Primary theme I: structural and governance aspects

The role of national health authorities was universally felt to be a critical determinant of success. Government entities, including Ministries of Health and/or other national health service delivery units, were involved in each of the programmes with a level of engagement that ranged along a continuum from passive (eg, conceptual 'support' of the programme and allowance to proceed without allocating new resources) to active (eg, recognising the NBS programme as a core part of the health system and providing clinical staff and other resources to maintain its continuity). While in several countries the government was involved from the early stages of NBS programme design, in no country was the government, the initial actor, involved in establishing the NBS programme. Programmes that continued beyond a 'pilot' phase ascribed government involvement as a key enabler; likewise, programmes that met with challenges in achieving long-term sustainability pointed to a lack of government ownership as a main reason.

All participants reported the topic of programme structure and governance to be an essential consideration. Programmes were each championed by clinician-led teams with specialised expertise in caring for patients with SCD. All programmes focused mainly on births taking place in public health facilities (ie, government operated); private sector birth centres were less commonly included. Clinical and ancillary staff (eg, midwives and

**Country (approximate population size and total births):** Liberia (population 5 million; 165,000 annual births)

**Province or city where the program took place (approximate population size and total births):** Greater Monrovia (population 1 million; 33,000 annual births)

**Approximate planning period and duration of screening:** 2 years planning beginning 2010; 21 months screening

**Number of birth centers involved at any stage in the duration of the program:** 1

**Timing of screening:** In the days following birth

**Approximate numbers of babies screened:** 3,986

**Location of laboratory and laboratory screening method:** Noguchi Memorial Institute for Medical Research, University of Ghana, Legon; initial screening method: testing by IEF

**Main partners involved:** Thrasher Research Fund; Boston Children's Hospital; John F. Kennedy Hospital, Monrovia

**Status (2021):** Screening paused due to Ebola epidemic and limited funding; planning to resume screening with support from ASH CONSA

**Country (approximate population size and total births):** Ghana (population 30 million; 870,000 annual births)

**Province or city where the program took place (approximate population size and total births):** Mainly Kumasi and surrounding districts (population 3.3 million; 96,000 annual births) and one site in Accra (population 2.5 million; 73,000 annual births)

**Approximate planning period and duration of screening:** 4 years planning beginning 1991; 25 years screening

**Number of birth centers involved at any stage in the duration of the program:** 39

**Timing of screening:** In the days following birth; if missed, then at the first well-baby visit (approximately 2-4 weeks of age)

**Approximate numbers of babies screened:** 523,159 as of June 30th 2020

**Location of laboratory and laboratory screening method:** Noguchi Memorial Institute for Medical Research, University of Ghana, Legon Accra; IEF for screening and HPLC for confirmatory testing (however, unaffordability of HPLC reagents led to testing by IEF only)

**Main partners involved:** Sickle Cell Foundation of Ghana; US National Institutes of Health; Ghana and Brazilian government; Pfizer (supporting NBS at Korle Bu Teaching Hospital, Accra, since 2017); ASH CONSA (supporting 37 Military and Greater Accra Regional Hospitals since Dec 2020)

**Status (2021):** Active; reduced funding has forced reduction in screening sites (to 6 in 2021)

**Country (approximate population size and total births):** Angola (population 32 million; 1.3 million annual births)

**Province or city where the program took place (approximate population size and total births):** Luanda Province (population 7 million; 287 annual births) and Cabinda Province (population 800,000; 33,000 annual births)

**Approximate planning period and duration of screening:** 1-2 years planning beginning 2011; 10 years screening

**Number of birth centers involved at any stage in the duration of the program:** Initially 2 large maternity hospitals in Luanda province with expansion to 22 health centers with maternity wards in Luanda and Cabinda province

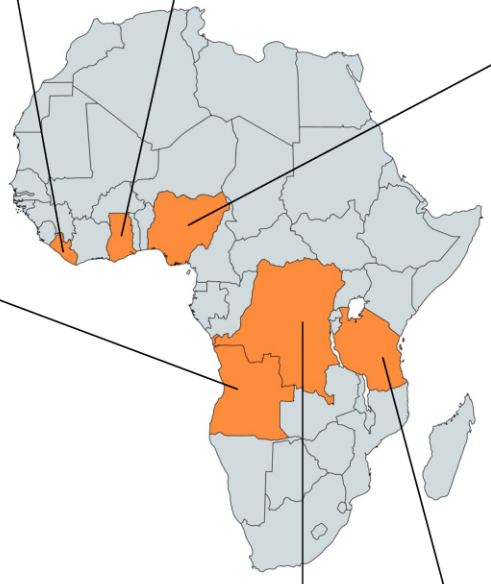
**Timing of screening:** In the days following birth

**Approximate numbers of babies screened:** 485,955

**Location of laboratory and laboratory screening method:** Centralized laboratory within the public pediatric hospital in Luanda utilizing IEF

**Main partners involved:** Texas Children's Hospital, Angola MoH, Chevron corporation

**Status (2021):** Paused; Chevron and Texas Children's funding/support completed in June 2020; MoH working to transition to public ownership



**Country (approximate population size and total births):** Nigeria (population 201 million; 7.6 million annual births)

**Province or city where the program took place (approximate population size and total births):** Kaduna (population 1.1. million; 42,000 annual births), Katsina (population 505,000; 19,000 annual births), and Abuja (population 1.2 million; 46,000 annual births)

**Approximate planning period and duration of screening:** 9 months planning beginning 2010; 18 months screening

**Number of birth centers involved at any stage in the duration of the program:** 4

**Timing of screening:** Ranged from the days following birth to 6 months of age

**Approximate numbers of babies screened:** 660

**Location of laboratory and laboratory screening method:** Abuja-Zankli Medical Centre (private hospital); HPLC (Classic model)

**Main partners involved:** Kafanchan and Zankli Medical Centre (Abuja), Guy's and St Thomas NHS Trust, UK; Michigan State University, US; NGO Fantsuam Foundation

**Status (2021):** Re-starting with EU funded project (African Research and Innovative Initiative for Sickle cell Education and ASH CONSA)

**Country (approximate population size and total births):** Democratic Republic of Congo (population 87 million; 3.6 million annual births)

**Province or city where the program took place (approximate population size and total births):** Mainly Kinshasa (population 17 million; 697,000 annual births) and also involving 3 additional provinces: Bas Congo, Kasai, Katanga (total population 14.3 million; 586,000 annual births)

**Approximate planning period and duration of screening:** 2 years planning beginning 2005; 14 years screening

**Number of birth centers involved at any stage in the duration of the program:** 262

**Timing of screening:** In the days following birth, in children under age 5 in tandem with an immunization program, or when newly diagnosed patients required transfusion

**Approximate numbers of babies screened:** Greater than 180,000 newborns and a total of more than 230.000 young children

**Location of laboratory and laboratory screening method:** Centre Hospitalier Monkole/Centre de Formation et d'Appui Sanitaire (CEFA) in Kinshasa and an antenna laboratory in Lubumbashi/ Katanga; IEF for screening and capillary electrophoresis for confirmatory testing

**Main partners involved:** Centre Hospitalier Monkole/Centre de Formation et d'Appui Sanitaire (CEFA); European Union; Agence Française de Développement (AFD), DGD Coppération Belge; Pierre Fabre Foundation; Association for Cultural, Technical, and Educational Cooperation/Belgium (ACTEC) ; Institut Européen de Coopération et de Développement/France (IECD) ; Instituto per la Cooperazione Universitaria, Italy (ICU)

**Status (2021):** Reduction of screening due to lack of funding

**Country (approximate population size and total births):** Tanzania (population 58 million; 2.1 million annual births

**Province or city where the program took place (approximate population size and total births):** Dar-es-Salaam (population 4.4 million; 163,000 annual births) and Mwanza (population 2.8 million; 104,000 annual births)

**Approximate planning period and duration of screening:** 1 year planning beginning 2015; 24 months of screening

**Number of birth centers involved at any stage in the duration of the program:** 3

**Timing of screening:** In the days following birth

**Approximate numbers of babies screened:** 6,000

**Location of laboratory and laboratory screening method:** Muhimbili University of Health and Allied Sciences, Dar es Salaam, Tanzania; Isoelectric focusing and HPLC

**Main partners involved:** Muhimbili University of Health and Allied Sciences

**Status (2021):** Active through research activities (Fogarty K43 Emerging Global Leader Award and the Sickle Pan-African Research Consortium) and health projects (ASH CONSA)

**Figure 1** Location and characteristics of included programmes. Programme data provided by country participant(s) who were interviewed. Reference for demographic data: World Bank. Map design credit: Mapchart.net. ASH, American Society of Hematology; CONSA, Consortium on Newborn Screening in Africa; HPLC, high-performance liquid chromatography; IEF, isoelectric focusing; MoH, Ministry of Health; NGO, non-governmental organisation; NHS, National Health Services.

**Table 1** Summary of main results

| Subtheme | Core concept | Principal stakeholders | Enablers | Challenges | Examples |
|---|---|---|---|---|---|
| **Theme: programme structure and governance** | | | | | |
| Health authority endorsement | ▶ Endorsement by government and incorporation into core health systems is fundamental to operational success and sustainability | ▶ Governments, ministries of health and other local health authorities | ▶ Government involvement from the start, in particular with plans for financial investment by national health authorities, facilitates national 'ownership' of NBS programmes and rational integration with routine healthcare delivery processes | ▶ Non-clear or unclear involvement of government risks prioritisation uncertainties, ineffective communication and implementation challenges<br>▶ Small-scale 'pilot' programmes can be useful for establishing proof-of-concept but may risk sustainability challenges if they do not involve buy-in from national government authorities from the outset | ▶ In Ghana, support from Ashanti local government is recognised to be a main factor in the programme's 25+ year duration<br>▶ In Angola, while the MoH was involved in the programme design from the start and supported the programme conceptually, financial investment to launch the programme was received from a private sector partner and the motivation of MoH to fund the programme long term was unclear |
| **Theme: technical** | | | | | |
| Workflow mapping | ▶ Optimal workflows (eg, that involve sample collection, sample transfer to laboratories, testing and patient follow-up) must be fully integrated with local health systems | ▶ Programme leaders, coordinators, health workers, laboratory staff and families | ▶ Programme design conducted in collaboration with all local stakeholders<br>▶ Recognition that workflows will need to be tailored to local settings and may require iterative refinement after initial implementation | ▶ Follow-up with patients for results notification and to enrol in comprehensive care programmes is recognised as a common challenge across programmes | ▶ In Ghana, the Ghana Health Service (GHS) staff conducts most activities along the spectrum of sample collection to counselling families on results and referral for medical care; activities are integrated with the laboratory and coordinated by the dedicated staff at the Sickle Cell Foundation of Ghana |
| **Theme: cultural** | | | | | |
| Community engagement | ▶ Family participation is fundamental to screening and follow-up | ▶ Programmes leaders, coordinators, families, patient organisations and support groups | ▶ Providing education about SCD can help families to understand the importance of NBS and following up in the event of positive screening results | ▶ Families may not believe positive test results or fail to follow-up for routine healthcare visits since babies are asymptomatic in early infancy<br>▶ SCD is stigmatised in many communities | ▶ Newborn screening, similar to immunisation was described as a 'silent' public health activity that, when successful, works in the background to help keep the population healthy<br>▶ Some programmes described community engagement to be helpful at initiation, but specific ongoing engagement was often not necessary as long as the structures are in place for programme implementation. |
| **Theme: funding** | | | | | |
| Role of government | ▶ NBS must be prioritised by government in order to assure long-term sustainability | ▶ Governments, Ministries of Health and other local health authorities | ▶ Government involvement from the start facilitates national 'ownership' of NBS programmes and financial planning | ▶ Government agencies in Africa have many competing interests for spending on health | ▶ Typically, NBS is provided free of charge to families and may be funded through a national health insurance programme<br>▶ In private systems, the cost of NBS is often either paid by private insurance or families<br>▶ In Africa, unlike early childhood immunisation, no country's government fully funds NBS programmes |

Table 1 summarises the main results of the study. It is organised by the four primary themes that emerged from the analysis, including governance (eg, considerations in deploying already overcommitted clinical staff to perform NBS), technical (eg, design and execution of operational processes), cultural (eg, variability of knowledge and perceptions of community-based staff) and financial (eg, issues when relying on external funding to the exclusion of government contribution). Subthemes are also highlighted as well as corresponding core concepts, stakeholders, enablers and challenges. Examples from various country programmes are also included for validity.
MoH, Ministry of Health; NBS, newborn bloodspot screening; SCD, sickle cell disease.

**Table 2** Major lessons learnt/recommendations

| Subtheme | Lessons learnt/ recommendations | Participant quotes |
|---|---|---|
| **Theme: programme structure and governance** | | |
| Health authority endorsement | Receive endorsement by government at start of programming | ► It was designed as a pilot project within the Public Health Service so that it would be incorporated. That was the plan right from the start. That it would end with government takeover was our goal<br>► The deputy minister of health was always a huge supporter. I would have the opportunity to meet with her whenever I wanted to, and she was always a huge supporter of the programme. The Ministry wasn't able to financially support the programme, but they made sure that I was able to get around stumbling blocks. And continued to do so after the study ended<br>► With our Ministry of Health, we have an official partnership because all the different hospitals need to have relation with the health minister<br>► There was some interest by the First Female at the time, but ultimately their involvement or—especially from the Ministry of Health side was quite low |
| **Theme: technical** | | |
| Workflow mapping | Integrate NBS into the local health system | ► We would rely on public health nurses and doctors working in that system<br>► The hospital director Helped to facilitate things primarily. So, we had a laboratory that we allocated within the hospital, so he helped allocate space for us to renovate a laboratory area. [This country] is one of the probably more difficult places to get either personally in and out of as a human being or to get materials in and out of. So, they helped to barter some of the supply chain stuff a little bit so that things weren't stuck in customs and people couldn't come into the country<br>► Whereas initially we thought once we get the funding, we thought we're going to go straight to screening. And when we went, we realised we actually had to have initial engagement with the traditional leaders and also to do some counselling work before we actually did the screening<br>► [One of our learnings was to] start in a place where some resources already exist (nurses, labs, etc) having a good lab in particular is crucial |
| **Theme: cultural** | | |
| Community engagement | Maintain interest at the MoH and hospital administration level | ► There are a huge number of competing interests and everybody is overburdened and overworked and very dedicated. So, it's really easy for people to lose sight of what—of the long-term goal of all the different projects that are going on. So, it was important to keep people's attention … at the ministry level and at the hospital administration level<br>► The Ministry of Health was always there to snap a photo. Unfortunately, not always there to do anything else |
| **Theme: funding** | | |
| Role of government | Obtain financial commitment from government prior to the start of programming | ► But we have not financial support from the government. That's the real problem in most of the African countries. It's the reason why we have foreigner partners for the financial support … . It's the reason why we can say most of our partners are foreigners<br>► [A recurrent challenge was engagement on the Ministry of Health side.] So, for example, the people who we hired, these laboratory technicians, were supposed to be Ministry of Health employees which … being a government employee is a complicated thing. And they—I don't even think still since—from when we started the programme until now, have had official quote unquote openings for jobs. So, they haven't hired anyone new into the system in five or 6 years<br>► There was severe engagement by the community leaders, but somehow, we could not follow that through with making the government—so I think one of the major challenges that I would think is really the government not only engaged by accepting that is their work, but actually to get funded. So, government funding is limited. And government implementation or what they have agreed to do is significantly limited |

Table 2 summarises the most consistent lessons learnt/recommendations highlighted across country programmes for each of the primary themes. Select quotes from different respondents are included to support our recommendations. Quotes have been anonymised.
MoH, Ministry of Health; NBS, newborn bloodspot screening.

nurses) that worked at birth centres and were responsible for the hands-on aspects of screening (ie, conducting heel sticks, communicating with families, etc) were generally government-employed workers who had been on staff prior to the initiation of the NBS programme. In most cases, therefore, the work associated with NBS constituted a new task they were asked to perform in addition to other duties. Across the programmes, coordinating staff played a fundamental role in organising and overseeing a vast array of logistics and managing the relationships with multiple stakeholders that variably included families, birth centre staff, SCD clinical experts, government representatives and external collaborators, including clinician colleagues and funding partners.

An important subtheme relating to staffing concerned the availability of specialised clinical 'centres of excellence'

that would be capable of providing holistic preventative and treatment services for individuals that were diagnosed with SCD through the NBS programmes. Participants recognised that the existence of such centres, and their accessibility to patients, was a pre-requisite to the initiation of NBS programmes such that families could be immediately offered a clinical service for follow-up on notification of positive test results.

### Primary theme II: technical aspects

While the general workflows involved in NBS programmes are conceptually straightforward (eg, sample acquisition, laboratory testing and notification of results), the design and execution of consistent operational processes were reported by several programmes to be an intensive and challenging exercise in practice. This was felt in part to be due to the very high level of coordination that was required between practitioners at birthing sites (who were responsible for collecting specimens, organising specimen transport to the laboratory, receiving laboratory results and notifying families), technicians in laboratories (who were responsible for receiving and testing specimens, and reporting laboratory results) and coordinators that oversaw NBS programmes (responsible for ensuring adequate training of staff, reliable availability of equipment and supplies, reporting to national authorities and other activities). In one programme, the laboratory was located in a different city from the birth centres, requiring the specimens to be transported by an approximately 7-hour car ride from the birthing sites to the laboratory. Another programme shipped specimens in a sealed container at 4°C by plane to the NBS programme laboratory in another country. The ambition of most programmes was to fully integrate the NBS workflows into routine health system processes; ultimately, this was achieved to a variable degree by different programmes. All programmes had a common aim to keep the cycle duration (ie, from the time of specimen acquisition to the time when families were notified of results) as short as possible. One commonly cited reason for delays in the NBS workflow was tracking down families to share laboratory results—some families were not able to be contacted by phone, which necessitated in-person visits that were time consuming for NBS staff and not always successful.

Robust data collection and management systems were important to support workflows (ie, registering babies that underwent testing, storing laboratory results and keeping record of when families were notified of results), facilitate quality improvement of NBS programmes (ie, as a means to identify when the workflows were operating suboptimally) and generate evidence that could be used for advocacy, research or to inform health policy (eg, incidence data, cost effectiveness or impact on health outcomes). Most programmes used a hybrid model that involved some paper-based record keeping and some digital components. One of the programmes (Ghana) converted entirely to a digital 'app'-based system beginning in 2018 accessible on the phones of birth attendants, laboratory technicians and programme coordinators.

All programmes, except Nigeria (where high-performance liquid chromatography (HPLC) was used), used isoelectric focusing (IEF) as the primary technique for screening or diagnosis, and some programmes used HPLC or capillary electrophoresis for confirmatory testing after screening. While none of the programmes surveyed reported that NBS laboratory equipment was a main barrier, virtually all of the programmes reported challenges with maintaining regular maintenance of equipment or reliable access to reagents. In some cases, periodic unavailability of reagents led to delays in testing.

### Primary theme III: cultural aspects

Some NBS programmes reported quick adoption of new technical practices by staff (eg, conducting heel sticks and managing bloodspot specimens), whereas other programmes met with some challenges in fully integrating this practice due to the perception of increased workload. Some programmes described clinical staff 'champions' who became highly dedicated to the programme (in the same way that many of the participants were), helped to advocate for the programme and trained other staff members. Ultimately, most programmes reported achieving a state of cultural adaptation resulting in a sense of pride among the programme staff for being involved in a novel programme with profound implications for the health of individuals with SCD.

Community engagement was highlighted by several programmes as an important determinant of success. It was reported that knowledge about SCD among community members varied widely and was occasionally confounded by false perceptions about the disease or stigmatisation. In some cases, the cultural aspects of community engagement were noted to be a determinant in the ability of NBS programme staff to follow-up with families to provide notification of test results (ie, if families were fearful of receiving results). Participants noted that families could also be dubious of positive results in the face of a baby who is healthy appearing (since babies with SCD are universally asymptomatic in early infancy).

### Primary theme IV: financial aspects

In all programmes, NBS services were provided free of charge to families. Participants reported an idealised scenario, where NBS programmes were entirely funded by local or national governments such that programmes were fully integrated as part of routine public health services.

Several programme leaders raised the idea of cost sharing between NBS programmes as a potential approach for reducing the costs borne by each individual programme. One example that was implemented was the shipping of laboratory specimens from one country to another for testing. Another example that was raised as a concept but not yet implemented was purchasing materials such as reagents for laboratory equipment in bulk.

All programmes received some form of external funding, defined as funding from out-of-country entities. Sources of external funding included foundations, non-governmental organisations, private sector companies and governments of other countries. Many participants reported external funding to have been an important enabler in helping to establish and/or maintain operations, and in some cases the cessation of external funding resulted in the need to scale down or halt the programme. External funding was, therefore, generally perceived to be a 'double-edge sword', whereby it had been necessary for some programmes to manifest but at the same time it complicated the attainment of long-term sustainability since permanent funding from outside sources was not feasible.

## DISCUSSION

Newborn screening programmes constitute a standard approach for diagnosing SCD in several countries and are urgently needed in Africa to assure that affected individuals promptly receive essential counselling as well as preventative and therapeutic care.[2 33] The reality, however, is that the establishment and sustained operation of NBS programmes in Africa is complex due to many factors. In an effort to better understand experience-based and pragmatic determinants of success, this study sought to harness lessons learnt from participants involved in establishing and operating NBS programmes that took place across West, Central and East Africa. While there are numerous published reports of progress achieved with subnational NBS programmes for SCD in individual countries,[19 20 23–25] we had identified only a single previous report that analysed cross-country experiences; that study described pilot programmes in DRC and Burkina Faso and presented an excellent review of the rationale for SCD NBS programmes along with high-level guidance for selected aspects of their implementation.[34] Thus, to the best of our knowledge, the current study involving programmes in six countries constitutes the first attempt to integrate learnings from a 'critical mass' of NBS programmes for SCD in Africa. Through standard qualitative methods, four main themes encompassing 12 subthemes emerged that highlighted enablers and barriers to implementation.

A main and crucial finding of this study was confirmation that NBS programmes for SCD are feasible to successfully implement in Africa, as evidenced by the large numbers of babies screened (eg, tens of thousands) and the long duration of screening (eg, more than 25 years) that was demonstrated in some programmes. Nevertheless, a consistent narrative emerged that feasibility did not ensure sustainability. Many of the programmes reported periodic setbacks in their capabilities to maintain their planned level of operations or to expand, and some programmes were forced to cease operations. Technical or workflow issues were never the primary challenge; rather, there was general consensus that the greatest barrier to the long-term success of NBS programmes resulted from their incomplete adoption into routine health systems. This was attributed mostly to inter-related aspects of governance (in particular, government involvement) and funding.

Government commitment was recognised by all interviewees as an essential element of success, and government entities routinely played important roles in the design and implementation of programmes. Even so, in none of the programmes was the government the primary driver behind programme inception and, as a result, several programmes innovatively sought and applied external resources (eg, grants or philanthropy) in order to initiate NBS with the hope that demonstrated success would provide evidence that governments could use to rationalise investing in NBS programmes. While that logic stands to reason, unfortunately, none of the programmes have been fully integrated widely into public health systems despite all six of the programmes having achieved operational success in different ways. Furthermore, it is possible that external funding received from some programmes complicated the 'handover' to government agencies, even while that funding was foundational to establishing the NBS programmes in the first place, a paradox that perhaps could only be avoided by confirming full government support from the outset (ie, NBS designated as a core service and budgeted accordingly). Indeed, the longest running NBS programme in Africa (Ghana) appears to have had the most substantial commitment from local government.

Another finding was the high degree of effort and dedication on the part of teams of SCD clinicians and advocates that was required to establish NBS programmes. Planning routinely took a year or longer before screening started, during which time many team members worked without extra compensation and in addition to an already full workload. Therefore, progress in each of the NBS programmes was all the more remarkable given the natural barriers that existed to establish them. At the same time, the achievements of each programme also served to highlight how much more work is needed given the coverage gaps resulting from high numbers of unscreened babies in each country (figure 1). Other learnings from this study related to operational considerations (eg, data collection and management systems) and cultural aspects (eg, strengthening the education of community members about SCD and the rationale for screening).

Limitations of this study include the sample of programmes assessed, which is less than the total number of NBS programmes for SCD that have been implemented in Africa and, therefore, is associated with an inherent bias based on the selection of included programmes. For practical reasons, we surveyed a single or small number of participants from each programme, and it is possible that by involving a larger cohort then additional perspectives may have been captured. Finally, it is recognised that local factors between countries, and even within countries, can influence health programmes and so the lessons learnt in one region will not always be immediately transferable to another. The above notwithstanding, the methodology was designed to involve a sufficiently large number of programmes across different parts of the continent in order that lessons learnt would be as applicable as possible across countries.

## CONCLUSION

This study codified learnings that may be useful to help inform the design and conduct of future NBS programmes for SCD in Africa. A key finding was that the capability of establishing a new programme was not a guarantee that the programme would endure; on the contrary, some aspects of programmes that were recognised enablers of their establishment (eg, funding from external sources) may have ultimately confounded sustainability (ie, by complicating ownership from government entities). Put another way, simply demonstrating that a programme is feasible, and gathering evidence to show it is associated with positive outputs and health outcomes, may not be sufficient to garner the support needed to sustain the programme in the long term. Being aware of this scenario at the outset may help stakeholders to emphasise certain aspects of programme design, including the role of government, with an aim to incorporate NBS programmes into routine public health services. As such, continuing to increase awareness of the burden of SCD and the critical importance of NBS among policymakers in Africa may be a priority in order to improve the timely detection of patients and promote optimal health outcomes.

**Author affiliations**
[1]Division of Hematology/Oncology, Dana-Farber/Boston Children's Cancer and Blood Disorders Center, Boston, Massachusetts, USA
[2]Evelina London Children's Hospital, London, UK
[3]Muhimbili University of Health and Allied Sciences, Dar es Salaam, Tanzania
[4]Institut de Recherche Biomédicale/CEFA and Centre Hospitalier Mère–Enfant Monkole, Kinshasa, Congo
[5]Texas Children's Cancer and Hematology Centers, Houston, Texas, USA
[6]Division of Hematology, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio, USA
[7]Department of Paediatrics and Child Health, Bugando Medical Centre, Mwanza, Tanzania
[8]Ariadne Labs, Harvard T.H. Chan School of Public Health, Boston, Massachusetts, USA
[9]Department of Global Health, Novartis Institutes for Biomedical Research, Cambridge, Massachusetts, USA
[10]Sickle Cell Foundation of Ghana, Accra, Ghana

**Twitter** Natasha M Archer @NatashaArcherMD

**Contributors** NMA, BI, NH, JS and KO-F planned the study, developed the interview guides and related questions, recruited participants and reviewed revisions, including final revision. NMA, BI, NH and JS reviewed the transcripts and data and wrote the initial draft of the paper. JM, SN, LT, VNT, PTM and EEA shared their work with the newborn bloodspot screening programmes in respective countries and reviewed revisions, including final revision. NMA is responsible for the overall content as the guarantor.

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**Ethics approval** This study involves human participants, and was approved by the institutional review board (IRB) of Boston Children's Hospital (IRB-P00025918). The IRB reviewed the above referenced protocol and determined that it qualifies as exempt from the requirements of 45 CFR 46. This protocol was determined to be exempt because it is limited to research activities in which the only involvement of human subjects will be in the following category/ies described in 45 CFR 46.101. (b): research conducted in established or commonly accepted educational settings, involving normal educational practices, such as (a) research on regular and special education instructional strategies or (b) research on the effectiveness of or the comparison among instructional techniques, curricula or classroom management methods. Participants gave informed consent to participate in the study before taking part.

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**ORCID iDs**
Natasha M Archer http://orcid.org/0000-0002-6460-5872
Patrick T McGann http://orcid.org/0000-0001-6198-4785

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**Supplemental Materials**

## I. Background questions

Questions sent by email ahead of interview and discussed at the start of each interview

a. In what city or geographic region was/is the program?

b. What is the approximate population size of the catchment area(s) covered?

c. About how long was the program planning process before screening started?

d. When did screening start?

e. Did the program end or is it ongoing?

f. If it ended, how long did it run for?

g. How babies were, or have been, screened in total?

h. How many birth centers were/are involved in the program?

**II. Interview guide: Phase one**

**INTRODUCTION**

Thank you for speaking with me today. My name is [name here]. As I mentioned in our email exchange, we are doing a study to inform success of newborn screening programs in Africa by assessing enablers and barriers to these programs by learning from the experiences of programs that had been established in the past and programs that are ongoing.

Over the next few months we aim to speak with representatives from various programs. Our plan is to distill the learnings into a format that can be used practically by various stakeholders including health workers, policy makers, NGOs, and others. We anticipate a publication, which we would invite you to review and take part in.

Note that this project received Institutional Review Board (IRB) exemption from the Boston Children's Hospital. We won't be asking for any patient information.

Today, I'd like to learn about your experience with the SCD newborn screening program in [country]. By agreeing to this interview, it is understood that you are in a position to comment on the newborn screening program that took place there and have the necessary authorization to speak on behalf of the program.

Would it be ok for me to audio-record the interview? That will help be sure we don't miss anything when we do the analyses. In the write-up, we won't attribute any specific statements to you unless we get your permission for that.

Any questions or comments?

Thank you so much again. Ok—let's get started with the interview, which will take about 45 minutes.

**INTERVIEW**

**1. Email survey questions**

*If any email survey questions not answered or need clarification—ask those first. If all have been answered, then move on to next section.*

**2. Partners**

- Who were all the partners involved in the program?
  *[Govt, MOH, University, teaching hospital, NGO, professional societies, consultants, other]*
  *[Categorize: local partner vs international partner]*

- Which partner or partners would you say had the biggest role in planning the program? Can you describe their role?

- Which partner or partners would you say had the biggest role in running the program? Can you describe their role?

- What were the main roles of the other partners?
  *[Ask specifically about role of government/MOH]*

- What was it like to get buy-in from the other partners? What was your approach? Could you tell me more?

**3. Planning**

- How did the idea for the program come about in the first place?

- What was helped the program most in the planning phase?

- What was the biggest challenge you faced in the planning phase?

- Was it envisioned at the start as a "pilot" program with a defined endpoint? Could you tell me more about that?

**4. Launch**

- Was there some sort of launch event when screening started?

- If so, was that important? What did the launch event consist of? Could you tell me more about that?

**5. Logistics**

- Who managed the day-to-day operation of the program?
  *[Profile of managers (nurse, doctor, etc), team composition (how many), full-time/part-time]*

- Was there a "headquarters" for the newborn screening program? If so, where was it located?

- Could you describe the birth centers where newborn screening took place?
  *[Clinics, hospital, urban, rural]*

- Were babies screened before leaving facility, or did they return for screening at a later date? How do you think this affected the success of the program?

- Who did most of the heelsticks? About how many participated in the program?
  *[Want to learn how many nurses and/or other health workers were trained/participated in the program in the various birth centers where screening took place]*

- Was there a consent process for families before obtaining heelstick? If so, could you please describe it?

- Could you briefly describe the sample collection and transport process from the point of heelstick to the screening laboratory? Were there any major problems in handling the samples?

- *How did patient information get to the screening lab? How did results get back to patients?* Did you use a specific computer program to manage information—if so, which one? Were there any major problems in collecting or managing data/information?

- What in your opinion were the most important factors that led to success in day-to-day operation of the program?

- What were the biggest challenges in day-to-day operation of the program?

- Were modifications to the way the program ran made over time?

**6. Laboratory**

- Was a SCD screening lab newly set up in conjunction with the screening program, or was an already established SCD screening lab used? Was the lab located in the same facility where screening occurred? How did that affect success?

- Who worked in the laboratory to analyze the samples?
  *[Profile of staff (techs, etc), team composition (how many), full-time/part-time]*

- Did the lab have equipment problems? Staffing problems? Could you tell me more? [*How did this affect how the lab ran?]*

- What method was used to conduct the screening test?
  *[For example, isoelectric focusing]*

- Do you happen to know what specific equipment was used in the lab?
  *[E.g., brand name of isoelectric focusing machine]*

- What was the most important factor in the successful running of the lab?

- What was the biggest barrier to running the lab?

**7. Notification and follow-up**

- If a baby screened positive, how were the parents notified? Who did that communication? What messages were delivered?

- What was the process for babies that screened positive—for example, did they get enrolled in a clinical management program? Could you tell me more about that? *[Seeking details of the sickle cell management program, if there was one]*

**8. Funding**

- How was the program funded? Were the costs shared by different parties

- What were/are the parts of the program that are most expensive?

- Would you be comfortable sharing the approximate cost of the program?
  *[Start-up costs, annual running costs]*

- How did costs affect the program? *[were activities, services, scale, sustainability etc. affected for financial reasons?]*

**9. Program disposition**

- **If the program has ended**—did it end when planned, go for longer than planned, or end prematurely? What do you think were the main reasons for this?

- **If the program is ongoing**—has it remained stable size, grown, or diminished in size? What do you think have been the main reasons for this?

**10, Perceptions**

- How did you and the other leaders of this program define [and measure] success?

- Could you comment on how families viewed the program? Could you tell me more about that? [*if viewed negatively, how did the program deal with that?]*

- What was your own biggest learning in doing this program?

**11. Closing**

- Are there any reports or publications about the program that could be shared with me?

- Is there anything else that you think I should know that we didn't talk about?

- Based on the interviews, we'll be writing a report summarizing the findings and we would like to acknowledge your contribution. Is that ok? We will share the report with you when it's ready and it would be great to get your feedback.

- In addition to you, we have also spoken with Dr. [name] from [country, Dr [name] from [country], etc. Are you aware of other newborn screening programs in Africa and contacts that we haven't yet connected with?

Thank you very much for speaking with me.

*Bye!*

**III. Main topic categories for phase two interviews**

For each, discussing how it impacted success, challenges, enablers, and other lessons learned.

- Cultural issues (among providers and community)
- Sustainability
- Balance of involvement between external and local partners
- Notification and follow up

## IV. Interview guide: Phase two

### INTRODUCTION

Thank you for speaking with me today. My name is Natalie. As I mentioned in our email exchange, we are doing a study to inform success of NBS programs in Africa by assessing enablers and barriers to these programs by learning from the experiences of programs that had been established in the past and programs that are ongoing.

Over the next few months we aim to speak with representatives from various programs. Our plan is to distill the learnings into a format that can be used practically by various stakeholders including health workers, policy makers, NGOs, and others. We anticipate a publication, which we would invite you to review.

Note that this project received Institutional Review Board (IRB) exemption from the Boston Children's Hospital. We won't be asking for any patient information.

Today, I'd like to learn about your experience with the SCD newborn screening program in [country]. By agreeing to this interview, it is understood that you are in a position to comment on the NBS program that took place there and have the necessary authorisation to speak on behalf of the program.

Would it be ok for me to audio-record the interview? That will help be sure we don't miss anything when we do the analyses. In the write-up, we won't attribute any specific statements to you unless we get your permission for that.

Any questions or comments?

Thank you so much again. Ok—let's get started with the interview, which will take about 45 minutes.

### INTERVIEW

#### 1. Email survey questions

*If any email survey questions not answered or need clarification—ask those first. If all have been answered, then move on to next section.*

#### 2. Partners

Who were the partners involved in the program?

- What was the role of local leaders and champions in the program?
- What was the role of external partners?
- What was the role of the government?

- How did they affect the success of the program?

- What lessons learned or recommendations do you have about working with partners?

#### 3. Logistics

- Can you please walk me through the entire screening process for one baby starting with how the baby is identified through how the parents are notified?
    - Probes: data management systems, equipment and supplies needed, getting results back to patients

- What in your opinion were the most important factors that led to success in day-to-day operation of the program?

- What were the biggest challenges in day-to-day operation of the program?

- What lessons learned or recommendations do you have about running the day to day operations of the program?

- Probe: challenges and facilitators for running the lab, recommendations

**4. Program disposition**

- **If the program has ended**—did it end when planned, go for longer than planned, or end prematurely? What do you think were the main reasons for this?
    - What would be needed in order to have a sustainable program?

- **If the program is ongoing**—has it remained stable size, grown, or diminished in size? What do you think have been the main reasons for this?

- Who pays for it?

- What recommendations do you have for other programs in the planning and implementation phase that can set them up to be sustainable?

**5. Perceptions**

- Could you comment on how families and the community viewed the program? Could you tell me more about that? [*if viewed negatively, how did the program deal with that?]*
    - *Probes: stigma, need for education*

- How did this impact the success of the program?

- What was your own biggest learning from the program?

**6. Closing**

- Are there any reports or publications about the program that could be shared with me?

- Is there anything else that you think I should know that we didn't talk about?

- Based on the interviews, we'll be writing a report summarizing the findings and we would like to acknowledge your contribution. Is that ok? We will share the report with you when it's ready and it would be great to get your feedback.

Thank you very much for speaking with me.

*Bye!*