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Multimethod evaluation of health services integration for neglected tropical diseases requiring case management in Liberia

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ABSTRACT

Introduction The WHO neglected tropical disease (NTD) roadmap stresses the importance of integrating NTDs requiring case management (CM) within the health system. The NTDs programme of Liberia is among the first to implement an integrated approach and evaluate its impact.

Methods A retrospective study of three of five CM-NTD-endemic counties that implemented the integrated approach was compared with cluster-matched counties with non-integrated CM-NTD. We compared trends in CM-NTD integrated versus non-integrated county clusters. We conducted a pre-post comparison of WHO high-level outcomes using data collected during intervention years compared with baseline in control counties. Changes in health outcomes, effect sizes for different diseases and rate ratios with statistically significant differences were determined. Complementary qualitative research explored CM-NTD stakeholders' perceptions, analysed through the framework approach, which is a transparent, multistage approach for qualitative thematic interdisciplinary data analysis.

Results The detection rates for all diseases combined improved significantly in the intervention compared with the control clusters. Besides leprosy, detection rates improved with large effects, over fourfold increase with statistically significant effects for individual diseases (p<0.000; 95% CI 3.5 to 5.4). Access to CM-NTD services increased in integrated counties by 71 facilities, compared with three facilities in non-integrated counties. Qualitative findings highlight training and supervision as inputs underpinning increases in case detection, but challenges with refresher training, medicine supply and incentives negatively impact quality, equity and access.

Conclusions Integrating CM-NTDs improves case detection, accessibility and availability of CM-NTD services, promoting universal health coverage. Early case detection and the quality of care need further strengthening.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ There is increasing advocacy and calls for the integration of case mangement-neglected tropical diseases (CM-NTDs) through health system strengthening; while evidence exists on the integration of leprosy into the health system, evidence on the impact of integrating the case management of multiple NTDs in practice is sparse.

WHAT THIS STUDY ADDS

⇒ The CM-NTDSs integration model was found to be feasible and impactful in Liberia, strengthening the health system, improving case detection and enhancing the accessibility and availability of CM-NTD services. There was, however, variation in the quality of care and some challenges with respect to the sustainability of the integrated approach, such as the continual provision of medicines and supplies for CM-NTDs.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ We suggest a stepwise approach to CM-NTDS integration, beginning with the prioritisation of integration in national health policies through a participatory and consultative approach to ensure stakeholders' input and ownership at all levels. This should be followed by the evidence-based development and implementation of integrated strategies that reflect the country's context and are evaluated through a multimethod approach.

INTRODUCTION

Neglected tropical diseases (NTDs) are a group of 20 communicable diseases affecting more than one billion people living in poverty, which can lead to long-term disability, economic hardship and stigmatisation.[1] Many of these diseases can be prevented, controlled or potentially eliminated or eradicated. Global policymakers, NTD community activists and academic experts are increasingly calling for health service integration[2–4];

however, there needs to be more evidence of the impact of practically integrating national case management (CM) NTD services within health systems. A resolution adopted in 2013 by the WHO (WHA66.12) member states urged all member countries to increase their integration efforts to prevent, control and eliminate priority NTDs.[5]

It is useful to define the integration of health services, as different interpretations exist or have evolved. The WHO defines the integration of health services as the 'management and delivery of health services so that clients receive a continuum of preventive and curative services, according to their needs over time and across various levels of the health system'.[6] However, within the WHO's new NTD roadmap, such processes are referred to as mainstreaming, with integration referring to shared programme delivery across multiple disease conditions.[3] As this study was conducted under the previous roadmap framework, the former definition of integration is used.

Armitage *et al* point to the limited available evidence on successful public health programme integration and the need for more quality evidence to show that health system integration improves the delivery of health services and population health status.[7] Despite limited evidence on NTDs and integration, Director Tedros Ghebreyesus, Director-General of the WHO, in his message during the launch of the WHO NTDs roadmap (2021–2030), proposed that:

> 'Over the next decade, we will move from a disease-specific to an integrated approach that cuts across all 20 diseases and disease groups, to ensure country ownership and leadership, to work even more closely with countries and partners, and to promote the development of new tools for prevention, diagnosis and treatment'.[2]

Alongside the WHO NTDs roadmap (2021–2030), the WHO has also launched the complementary integrated skin NTD strategy with the aim of enhancing people-centred care for those affected by skin NTDs in proximity to their communities.[3] In Lalo, Benin, Barogui *et al* conducted a 7-month study to assess the optimal use of scarce resources in the context of NTDs. After months of intervention covering Buruli ulcers, leprosy and yaws, they concluded that an integrated approach to skin NTDs was essential to maintaining optimal use of resources at the community level.[8] Similarly, over a period of 11 months, Koffi *et al* carried out a cross-sectional study in Côte d'Ivoire on skin NTD (Buruli ulcer, leprosy and yaws) integration, providing evidence to promote this approach, particularly where co-endemicity is present within communities.[9] However, current research on the importance and benefits of health integration is not underpinned by practical, routine national programmatic evidence.[10]

Liberia NTD integration

Liberia is co-endemic for several NTDs that require disease-specific treatment and/or morbidity management, including Buruli ulcer, yaws, leprosy, hydrocele and lymphoedema related to lymphatic filariasis (LF).[9] Since 2017, the Liberia Ministry of Health (MoH) NTD programme has been implementing the first integrated CM-NTD approach with national-level activities and county-level activities in 5 of the 15 counties of Liberia: Lofa, Nimba, Bong, Maryland and Bomi counties (figure 1). The rationale for the selection of the five counties includes diversity in disease burden and geographical characteristics. The goal of the integrated strategy was '*to strengthen the health system through integration so that it is able to provide early identification and successful management of all NTDs requiring intensive disease management (IDM)*'.[9]

Prior to the integration approach, CM-NTDs were managed in a fragmented manner, with some conditions, such as lymphoedema, not being a specific programme priority. The implementation of the integration plan introduced training modules covering all priority CM-NTDs. This was followed by integrated planning, monitoring, budgeting and human resource strengthening through integrated cascaded training, which progressed from the national to the community level. Community awareness, mobilisation and data management for CM-NTDs were strengthened through the integrated national health information system, specifically the District Health Information System 2 (DHIS2).[11]

This study aims to evaluate the measurable and perceived impacts of CM-NTD integration across diseases and on the larger Liberian health system after 3 years of intervention. We used case detection, access to improved quality of service delivery, health outcomes and health system stakeholders' views and perceptions as benchmarks for the evaluation. The primary research question that this manuscript aims to answer is: what is the impact of integrating services for NTDs requiring CM across diseases and through the existing health system of Liberia? The specific research objectives are to: (1) evaluate the effect of integration on case detection rates for a group of NTDs over the first 3 years of implementation, compared with a control sample of non-intervention counties; (2) compare profiles of case detection rates for different NTDs over the implementation period; (3) evaluate the accessibility of CM-NTD services and quality of care in an integration approach and (4) explore stakeholder views of the effectiveness of the integration strategy.

METHODS

Study setting and design

Current context

We selected six counties for the research: three intervention counties (Lofa, Bomi and Maryland) from among the five counties where the integrated approach has been in place since 2017 by the Ministry of Health NTD programme. We could not use the entire five integrated counties due to resource constraints, which is a limitation of the research.

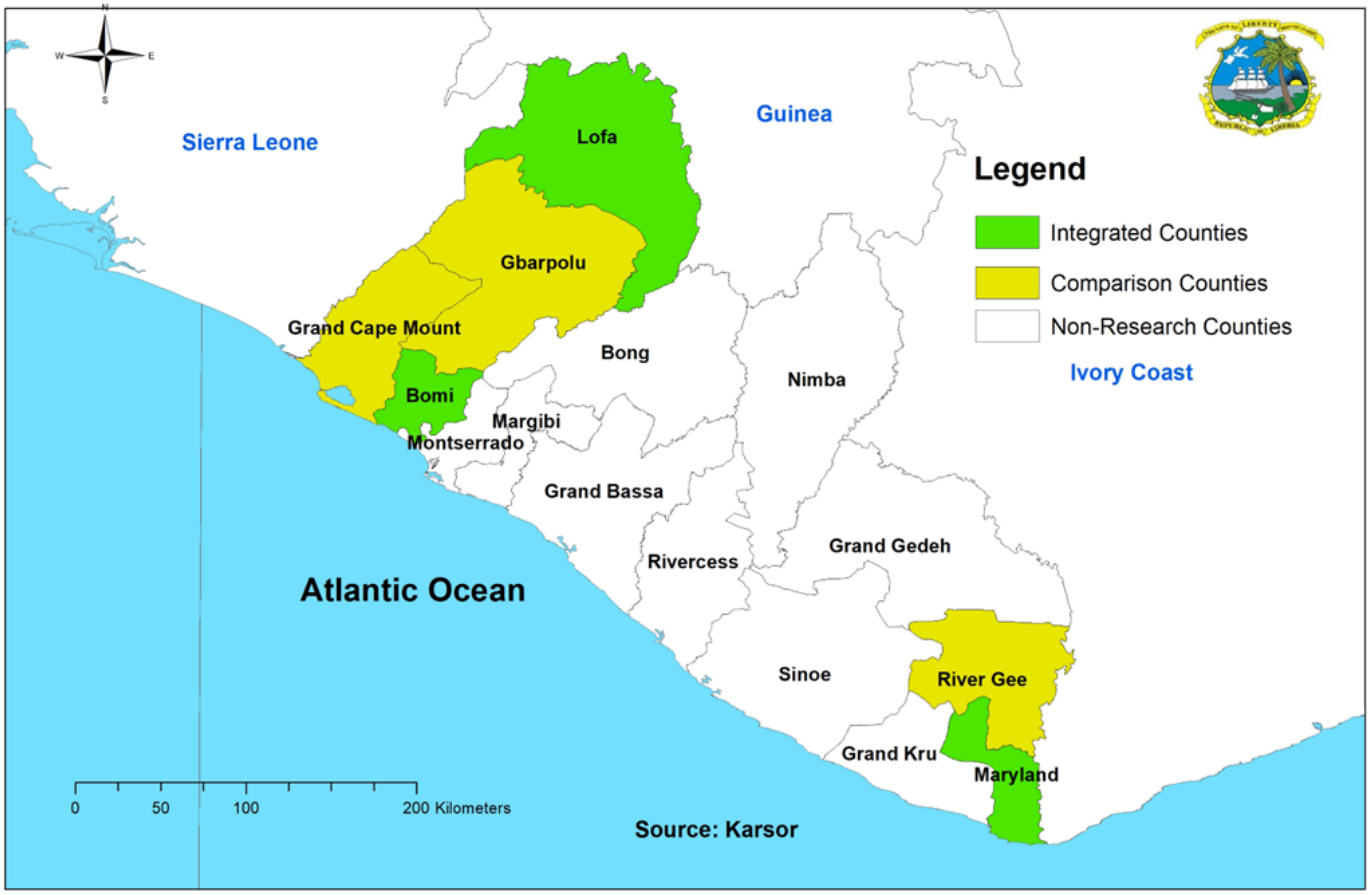

Figure 1 Sites of multimethods research in Liberia.

However, the three integrated counties were selected among the five counties due to their diversity in geography and culture; they are NTD co-endemic but with differences in disease burden (figure 1 and table 1).

Three comparison counties (counties where integration was not happening) with similar characteristics in disease co-endemicity and diversity in disease burden were selected. These counties include River Gee, which is a good match for Maryland, Cape Mount County, which is a good match for Bomi County and Gbarpolu County, which is a good match to Lofa County (table 1).

We conducted a retrospective trend study in the three selected NTD high-burden counties in which NTD programme integration was implemented. For controls, we cluster-matched the three selected counties with non-integrated NTD (or standard care).

We examined and compared the levels and trends of our outcomes of interest in integrated versus non-integrated county clusters. We did a pre-post comparison of WHO high-level outcomes in three domains in the same clusters. To achieve this, we adapted the WHO guide for the Monitoring and Evaluation of Case Management NTD Control and Elimination Programmes Performance. The guide outlines three high-level groups of indicators (sensitivity of case detection, access to care and quality of care) (see table 5 for a detailed impact domain).[3 12] The evaluation depicts the performance of the programme based on routine programme indicators triangulated with the qualitative findings.

Comparing counties implementing the integrated approach and those that are not, for the first objective, we report detection rates, rate ratios, 95% CI and p values using appropriate tests (χ^2 or Fisher's exact tests). To achieve the second objective, we compare trends in detection rates for the different selected NTDs. Finally, for the third and fourth objectives, we present an evaluation of the high-level performance indicators alongside qualitative evidence against each of the key domains of the CM-NTDs programme using proxy indicators from the routine NTD data.

To further broaden the evaluation of the effectiveness of the integration strategy over the first 3 years of its implementation, we collected and analysed qualitative interviews with key stakeholders to understand the wider perceptions of the impact of NTD integration. This combined approach is helpful when making sense of complex data, such as those gathered within health systems, and provides a more complete and coherent understanding of the effectiveness of the integrated approach.[13 14] Complex real-world data such as the CM-NTD health system data requires an understanding of context as there will be gaps in data

Table 1 Study site contextual information about three interventions and three non-intervention counties prior to integration

Key contextual information about the three intervention counties and non-intervention counties prior to integration	
Intervention counties	**Non-intervention counties (matching or control counties)**
Maryland County is a southeastern county bordering Côte d'Ivoire, with an estimated population of 174 441 based on the 2008 National Population Housing Census. The county is inhabited by the Gribo tribe. The county is co-endemic for some of the CM-NTDs, and among the first five counties implementing the integrated CM-NTDs approach. It is among the four counties (Grand Bassa, Sinoe, Grand Kru and Maryland) with the highest lymphatic filariasis prevalence of infection >10% by ICT (MoH 2016). The county has one government hospital. Buruli ulcer and hydrocele cases were detected in <1/10 000 population prior to the integration approach, leprosy in 2.9/10 000 population and lymphoedema in 11.3/10 000 population.	River Gee County was chosen because it is a better match to Maryland. Like Maryland County, it is a southeastern county bordering Côte d'Ivoire, with an estimated population of 85 707. The county is inhabited by the Gribo tribe and their culture. It is co-endemic for some of the CM-NTDs. Buruli ulcer detected in 0.23/10 000 population, leprosy in 3.38/10 000 population, hydrocele in <1/10 000 population, lymphoedema in 20/10 000 population and microfilaria in 0.99%.
Lofa County has an estimated population of 355 283 (National Population Housing Census 2008) and the county is in the northwest of Liberia, bordering Guinea in the northeast and Sierra Leone in the northwest. It consists of six major tribal groupings, including the Lorma, Kissi, Gbandi, Madingo, Mende and Kpelleh. It was among the first three counties that implemented the Buruli ulcer project* and is now among the first five counties implementing the integrated CM-NTDs approach. As a strength, there are four hospitals in Lofa County led by a medical doctor. This county is also co-endemic for CM-NTDs. It is moderately prevalent (1%–10% ICT) for lymphatic filariasis. Leprosy prevalence in Lofa County at <1/10 000 population, Buruli ulcer <1/10 000 population (WHO 2016), leprosy <1/10 000 population, hydrocele <1/10 000 population and lymphoedema <1/10 000 population.	Gbarpolu County was chosen because it is a good match for Lofa County. It has an estimated population of 107 007 according to the 2008 National Population Housing Census and the county is in the northeast bordering Sierra Leone. Gbarpolu was established out of Lofa County and therefore has all the major characteristics of Lofa, including tribes and cultural practices. Leprosy prevalence in Lofa County was <1/10 000 population prior to the integration, Buruli ulcer <1/10 000 population, hydrocele <1/10000 and lymphoedema 0/10 000 population (WHO 2016).
Bomi County is estimated to have a population of 107 945 (National Population Housing Census 2008), and it is one of the smallest counties located in the southwest of the country. The major tribal groupings in Bomi are Vai, Mede and Gola, and it is the only western county among the five counties implementing an integrated CM-NTDs approach in Liberia. It is bordered by Montserrado, Cape Mount and Gbarpolu counties. The burden of CM-NTDs is relatively low based on annual reports. Bomi County is one of the two counties that is not endemic for lymphatic filariasis (MoH 2016). Buruli ulcer in 1/10 000 population, leprosy in 18/10 000 population, hydrocele in 4/10 000 population and lymphoedema in <1/10 000 population.	Grand Cape Mount County was chosen because it has a better match with Bomi County. It is located in the southwest of Liberia, bordering Sierra Leone and Bomi County. Like Bomi, the major tribal groupings in Cape Mount County are Vai, Mede and Gola. It has an estimated population of about 163 059 according to the 2008 Liberia National Census. Prior to the CM-NTD integration, Buruli ulcer detection rates for Buruli ulcer, leprosy and lymphoedema were all <1/10 000 population, except for hydrocele 12/10 000 population.

*The Buruli ulcer project was the first package of interventions instituted for the control of Buruli ulcers after the first assessment in Lofa, Nimba and Bong counties that established the existence of Buruli ulcers in Liberia in 2012. Details of the results of that assessment can be found here: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3944850/.
CM-NTD, case management-neglected tropical disease; ICT, immunochromatographic test.

and offset timelines for phases of data collection that require consideration. Bringing in broader sources of information provides a more rounded understanding to help explain and interpret patterns and their real and perceived impacts.

Intervention descriptions

The differences between the programmatic input in integrated and non-integrated counties are outlined in table 2. These inputs are categorised by levels based on international partners, national, county, district, health facility and community levels. In these non-integration counties, there are still specialised treatment centres, for example, for leprosy, but no integrated training for NTDs, no use of an integrated ledger for data collection and other integration elements are also lacking. In the integrated counties, combined interventions and non-interventions are carried out through the health system (see in detail in table 2).

Table 2 Operational inputs that differentiate integration and non-integration counties

Inputs for integrations	Inputs for non-integration
International partner level	
Flexible and coordinated resources for priority NTDs among international partners. The multiyear strategic partnership between the Ministry of Health and international partners. Partnership approach dynamic to emerging health system challenges and encourages learning. Multiyear funding and programmatic commitment to enable planning.	Restricted funding is intended for specific approaches and diseases. Lack of resource integration and coordination among international partners.
National levels	
Integrated planning for NTDs. Integrated training materials developed on all priority CM-NTDs interventions. Integrated supervisory tools developed. Intra-NTDs supportive supervision. Integrated human resource structure operating on one platform. Integrated data management system with HMIS (DHIS2). Inclusion of diseases in the IDSR guidelines. Training lab staff.	Standalone supply chain for individual disease. Vertical data management and not embedded in DHIS2. Individual disease planning. Specific disease training materials and guidelines are developed separately. Individual disease supervision.
County levels	
Intra-NTDs supportive supervision. Supplies and medical products are channelled through county supply chain structures. Joint integrated supportive supervision. Coordinated and collaborative human resources. Integrated training approach for county level.	Specialised health workers for specified NTDs. Disease-specific supply chain. Disease-specific supervision.
District level	
Integrated case detection (IDSR) and referral. Integrated case reporting through IDSR and the HMIS. Intra-NTDs supportive supervision. Joint integrated supportive supervision. Integrated training approach for district level.	Individual NTD surveillance activities. Individual NTD supervision.
Health facility level	
Integrated training approach for training all health workers and community health workers (CHA and CHVs). NTD services are provided at the point of service delivery across all health facilities. Integrated case detection and referral.	Specialised health workers for specified NTDs. Individual NTD supervision. Specialised treatment facility for individual NTDs. Reporting outside the integrated health facility ledger.
Community level	
The integrated training approach is used for training all community health workers (CHA and CHVs). Performance-based incentive. Integrated case detection and referral.	Individual NTD surveillance activities. Ad hoc incentives.

CHA, community health assistant; CHV, community health volunteer; CM, case management; DHIS2, District Health Information System 2; HMIS, Health Management Information System; IDSR, Integrated Disease Surveillance and Response; NTD, neglected tropical disease.

Data sources

Quantitative measurement

Trained research assistants extracted the routine CM-NTDs programme data from the health facility ledger with unidentifiable participant codes into Microsoft Excel V.13 with all indicators included in the health facility ledger. We visualised and inspected the data to identify duplicates and have them removed. The rationale for using the health facility ledger was to ensure that we were using quality data without duplication. We used health facility data because we wanted case-based data that accounts for individual patient detail data elements that would give us access to different options for analysis that cannot be obtained at the central level due to the aggregated nature of central-level Health Management Information System (HMIS) data. In the CM-NTDs health facility ledger, there is a unique identification number for each NTD-affected person. There is also enough additional space for recording during the revisit in the course of treatment that helps reduce the chances of double counting errors.

Table 3 Qualitative research participants by sites

Stakeholder category	Stakeholder roles	No. of participants			Total
International partners	Partners KII	7 (5 females and 2 males)			7
National	National managers and supervisors KII	17 (5 females and 12 males)			17
County and district levels		Lofa	Bomi	Maryland	
	County health team KII	17 (4 females and 13 males)	17 (4 females and 13 males)	17 (6 females and 11 males)	51
Community level	Community health assistants and volunteers (focus group discussion)	2 sessions (3 females and 15 males)	2 sessions (4 females and 14 males)	2 sessions (12 females and 8 males)	6 (56 participants)
	Patient group IDI	4 males	4 (3 females 1 male)	4 (3 females and 1 male)	12
Total		47	23	23	93 (143 participants)

Qualitative data sources

Qualitative research was undertaken in the three intervention counties included in this study (table 3) to explore health system actors' perceptions of the impact resulting from the integrated CM-NTDs approach. We purposively selected our respondents based on their expertise or understanding of CM-NTDs. Participants with insight were recruited from various levels of the health system and included international partners, national-level programme managers and supervisors, county health team members, clinicians at health facilities, community health assistants (CHAs), community health volunteers (CHVs) and people affected by CM-NTDs. The research assistants supported the recruitment with support from the lead author. The initial step was to contact participants via phone calls, face-to-face or by email to explore interest and initiate verbal consent. The next step was to send the participants an information sheet so that they could read it in advance of the interview. On arrival or before the interview, there was a step-by-step discussion of the participant information sheet to discuss any concerns and the consent of the participants. A topic guide was developed and used to interview the informants. Interviews were conducted between July 2019 and May 2020, led by the lead author (KK) and trained and experienced research assistants (LJ, DB and OK). We also took diary notes on observations during meetings and field activities that involved discussion on integration activities.

Data analysis

Quantitative analysis

Routine programme data were collated, coded and uploaded to SPSS V.24 for descriptive analysis. The data were collected across a 3-year period, starting with baseline data from 2015 and 2016. Changes in health and patient outcomes across comparison counties and integrated counties were recorded across the integration implementation years of 2017, 2018 and 2019. To clean the data, we filtered the data, removed repetitions and retained the completed version. We observed that there was less than 3% missing data (this was mainly for treatment outcomes for leprosy, which has treatment duration beyond the study period), so we used the pairwise method to address missing data.

Differences in detection rates between the integrated and non-integrated counties were evaluated for statistical significance using χ^2 tests, having met test assumptions.

Qualitative analysis

All interviews and focus group discussions (FGDs) were conducted in English, transcribed verbatim and analysed with diary notes using the framework approach.[15] The lead author (KK) led the analysis with support from the local research assistants using the computer-assisted software QSR NVivo V.11. During the familiarisation of the data, we took note of emerging issues, developed codes, charted the data based on categories, summarised and interpreted the data. This enabled triangulation across different stakeholder groups and the interrogation of gaps and patterns identified in the quantitative data.

Quality assurance and positionality

KK, as the first author and Liberian NTD programme manager, was critically reflexive about his dual role of researcher and lead practitioner (head of the Liberian NTD programme). This was recognised from the outset and is integral to research design and data collection decisions. KK's insider positionality meant he had access to strategic meetings, was engaged in supervision and field visits and took diary notes throughout on issues influencing the integration of the CM-NTDs. KK undertook some interviews, but the research assistants conducted interviews where it was felt that interviewees may be

Table 4 Comparison of case detection rates between integrated and non-integrated counties

	Integrated		Non-integrated		Total	Rate ratio	95% CI	P value
All case management-neglected tropical diseases		Detection rate		Detection rate				
All diseases combined	723	0.116%	92	0.0264 %	815	4.385	3.5 to 5.4	<0.00001
No cases	623 810		348 413		972 223			
Lymphoedema cases	246	0.039%.	1	0.00029%	247	137.258	19.3 to 978.3	<0.00001
No lymphoedema cases	624 287		348 464		972 751			
Yaws cases	21	0.0034%	0	??	21	23.992	1.5 to 396.1	<0.0263
No yaws case	624 512		348 465		972 977			
Hydrocele cases	123	0.0197%	7	0.002%	130	9.804	4.6 to 20.9	<0.00001
No hydrocele case	624 410		348 458		972 868			
Buruli ulcer cases	180	0.029%.	14	0.004%	194	7.174	4.2 to 12.4	<0.00001
No Buruli ulcer cases	624 353		348 451		972 804			
Leprosy cases	151	0.0242%	70	0.02%	221	1.203	0.90 to 1.6	0.199282
No leprosy cases	624 382		348 395		972 777			

less likely to be critical if led by KK.[16] The experienced research assistants were supported by KK and supervisors to apply the principles of rigour in qualitative research. The analysis process included others, ensured openness to critical views that may not necessarily support the integrated approach and enhanced the trustworthiness of the process. For example, results were regularly shared with peers (county NTD focal points, data managers and NTD programme supervisors in the MoH) who were familiar with the data for transparency.

Reflexivity statement

In order to promote equitable authorship in the publication of this manuscript, we have adopted the 'consensus statement on measures to promote equitable authorship of research publications from international partnerships',[17] along with the BMJ Global Health editorial on using scientific authorship criteria.[18] In line with these guidelines, we have included junior researchers and data collectors in line with the authorship criteria as discussed in our reflexivity statement (online supplemental appendix 1).

Ethical approval

LSTM Research Ethics Committee and Liberia UL-PIRE Internal Review Board (IRB) both issued approval (#19-05-163 UL-PIRE and #18-035 LSTM Research Ethics Committee).

Patient and public involvement

Following a detailed explanation and understanding of the research information sheet, all informed consent forms were signed and completed before the interview was conducted.

RESULTS

A comparison of case detection rates between integrated and non-integrated counties is provided in table 4. Reporting numbers of cases and non-numbers for each disease, with detection rates, rate ratio, 95% CI and p value from the significance test (χ^2 or Fisher's exact test).

Comparing case detection rates between the integrated and non-integrated counties, rates were improved across all diseases, with statistically significant effects observed for all individual diseases except for leprosy. When considering all diseases combined, the all-case detection rates differed significantly between the intervention and control clusters (0.116% vs 0.0246%). This resulted in a statistically significant rate ratio of 4.385% (p<0.000; 95% CI 3.5 to 5.4). The rate of detection of lymphoedema cases differed significantly between the intervention and control clusters (0.039% vs 0.00029%). This resulted in a statistically significant rate ratio of 137.3% (p<0.00001; 95% CI 19.3 to 978.3). As reported in table 4, in terms of greater effect, four of the five diseases, including LF-related lymphoedema and hydrocele, yaws and Buruli ulcer, have large effects with over a fourfold increase in case detection rate. Cases of leprosy were just 1.2 times more likely to be detected in an integrated county than in a non-integrated county (rate ratio 1.2036, 95% CI 0.9066 to 1.5979, p=199 282). See table 4 for the full statistical details.

Changes in patterns of detection in individual diseases, when compared with different approaches (integration and non-integration), are shown in figure 2. The cumulative detection rate showed slightly different trajectories over the study period for different diseases. Lymphoedema and hydrocele have a similar pattern (as both are features of LF); they are characterised by the rapid detection of new cases in the first 12 months and a lower

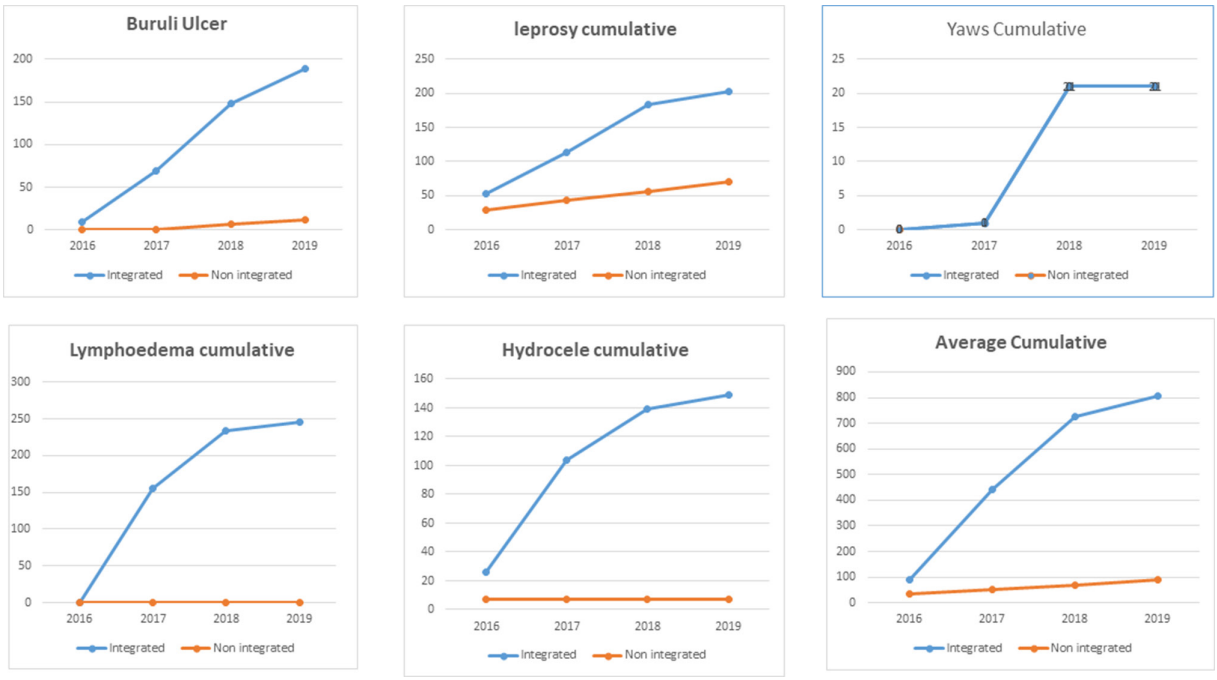

Figure 2 Cumulative frequency of individual diseases between integrated and non-integrated counties.

rate of new cases detected over the next 2 years. This differs from Buruli ulcer and leprosy, which accumulate less rapidly over a longer period. This may relate to their lower frequency in the community or being less easy to detect. The yaws curve, which was rapidly detected in a specific period of year two, is attributed to a snapshot survey done in one of the research counties (Maryland)[19] using different case detection strategies outside of the normal CM-NTDs routine programme.

The period from Q1 of 2015 to Q4 of 2016 is included to observe and compare changes in the trend before the intervention and trend after the intervention. Q4 of 2016 to Q2 of 2017 saw the staggered introduction of integration processes and inputs, including the hiring of county

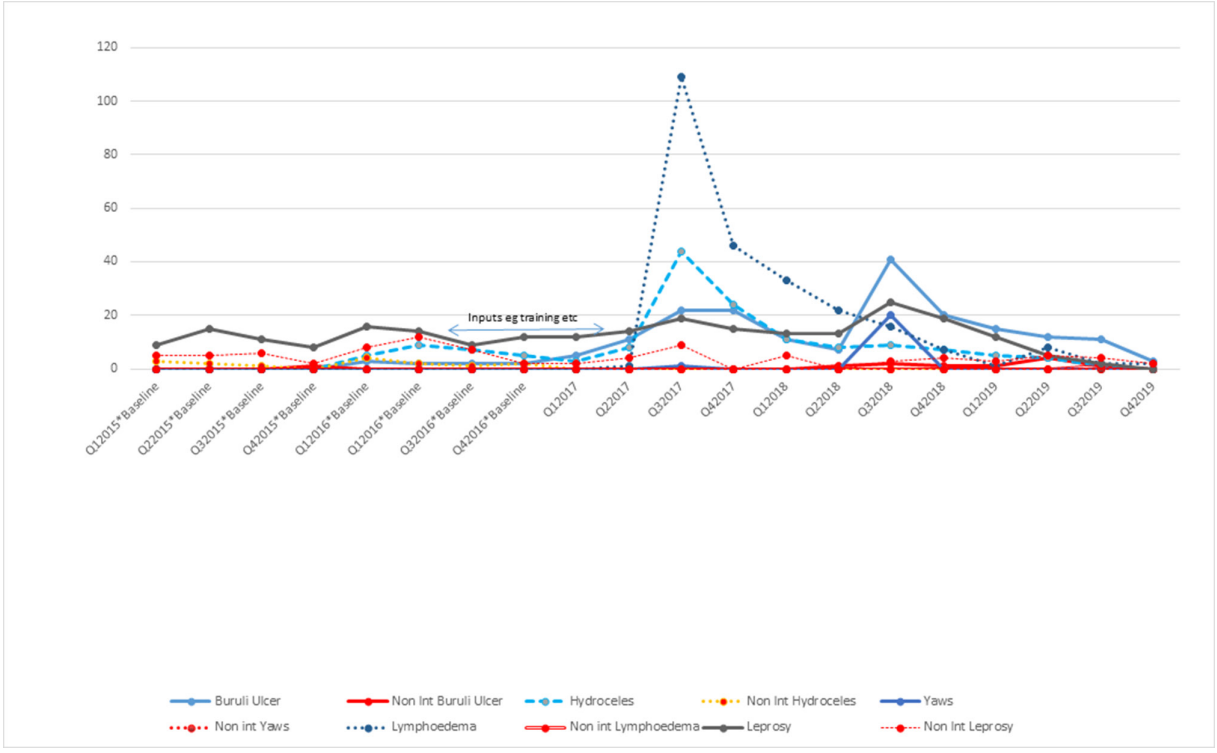

Figure 3 Number of case management-neglected tropical diseases per quarter (Q) in integrated and non-integrated countries.

NTD focal points, training of health workers, CHAs CHV, supportive supervision, provision of CM-NTDs medical commodities and community awareness (figure 3).

Following the completion of these processes, we observed a marked increase in cases of detection in Q3 of 2017, which then declined over 2018 until reaching baseline levels in 2019. An isolated intervention (an integrated skin NTD survey conducted in one of the research counties) increased the number of new cases in Q3 of 2018.[19]

Qualitative evidence supports patterns documented within the quantitative data in relation to case detection. Most FGD participants described how there was a noticeable increase in cases after the integrated training. Even with the noticeable reduction, most key informants explained that more CM-NTD cases were identified and reported during the integration period compared with the preintegration period (Maryland FGD 1 and Lofa County Key Informant) (see table 5, qualitative evidence).

Domains of impact: cross-domain summary of results

Adapted from the WHO Regional Office for Africa guide for the Monitoring and Evaluation of CM NTD Control Programmes Performance (WHO, 2015), table 5 outlines

Table 5 Measurable indicators of performance against baseline data and qualitative evidence

Measurable indicators of performance against baseline data and qualitative evidence. The data in the table represent the percentage of cases as a proportion of the total N in brackets representing total absolute numbers or denominators.

Value of indicator	Measures of sensitivity in case detection	Non-integration year as the baseline	Integration intervention years			
	Indicators	2016 Baseline	2017	2018	2019	Qualitative evidence: illustrative quotes
Sensitivity of case detection	Percentage of children diagnosed with leprosy and visible disability	9% (n=23)	15% (n=60)	19% (n=70)	11% (n=9)	(1) 'The first time (2017) we were trained here and then we did the training (cascaded training) for the CHA along with the CHVs. We used to find many cases of the big foot, leprosy and Buruli ulcer, but since last year (2019) to now, we do not find many cases; maybe there are not many new infections now, or some of our friends have forgotten the signs and symptoms of the diseases' (Maryland County, FGD-1).
	Percentage of newly diagnosed patients with Buruli ulcer with category III lesions	0	73% (n=59)	44% (n=41)	38% (n=79)	
The measure of access to services	Percentage of health facilities provided CM-NTD services per year	0	58% (n=71)	59 59% (n=71)	39.4% (n=71)	(2) "Even though the number of cases is reducing mainly in this year, we still see more cases than before the integration; this is because more health workers and community volunteers were trained. And more health facilities are providing CM-NTDs services (Lofa County, Key Informant). (3) 'The reasons we are not seeing more CM-NTDs compared with the past 2 years might be because the CHVs are not getting the 5 dollars promised them when they find cases. And some CHV or CHA are leaving the job because they are not motivated' (Maryland Key Informant-14). (4) 'This time we can be scared to send people to the clinic because when we referred the people with the big bag (hydrocele) and the one with the big sore (Buruli ulcer) to the clinic, they always tell them no medicine and no materials to dress the sore. So, when the patients come back from the clinic, we are the ones they can talk to (angry at) and they take us to be liar (FGD-2, Maryland county).
	Number of health workers trained in integrated NTDs	0	222	0	0	
	Number of CHA trained in integrated NTDs	0	175	0	0	
	Number of CHV trained in integrated NTDs	0	355	0	0	
	Percentage of NTD cases referred by CHAs	–	4% (n=358)	6.3% (n=285)	6% (n=172)	
	Percentage of NTD cases referred by CHVs	–	58% (n=357)	56% (n=282)	72% (n=83)	
Quality of care	Percentage of patients with hydrocele that underwent hydrocelectomy	–	12% (n=78)	22% (n=22)	60% (n=10)	(5) 'Hydrocele surgeries are specialised interventions that require special training of medical doctors. We have not had any funding within the integrated approach that supports hydrocele surgeries. The number of hydrocele surgeries reported are those coming to the hospital as emergency or elective surgeries with direct out-of-pocket payment' (National Programme Informant). (6) 'Our people can respond well to the treatment advice we ask them to take. They want to get well; they do not feel fine to be in the condition they find themselves. Our challenge is that we do not always have the supplies for home-based care and medicines to continue to give to them (Bomi County Key Informant). (7) 'At the time I was using the medicine…my foot was not hurting, when I rubbed the medicine on my foot…it was not hurting until after 2 months when my medicine finished, my foot started hurting again' (Bomi County IDI-03).
	Percentage of new clinically diagnosed Buruli ulcer cases confirmed by PCR	–	27% (n=41)	0	0	
	Percentage of patients with Buruli ulcer having completed 56 doses of antibiotics	100% (n=9)	82% (n=55)	84% (n=77)	44% (n=41)	
	Number of confirmed (DPP and RDT) patients with yaws	0	1	20	0	
	The percentage of patients with Buruli ulcer healed with 'limitation of movement'	–	2% (n=56)	8% (n=79)	0	
	Percentage of patients with lymphoedema practicing self-care	–	96.1% (n=152)	85.1% (n=74)	77% (n=13)	

CHA, community health assistant; CHV, community health volunteer; DPP, Dual Path Platform ; FGD, focus group discussion; NTD, neglected tropical disease; RDT, Rapid Diagnostic Test.

high-level indicators that the NTDs programme used as proxy indicators (sensitivity to threshold of case detection, access to care and quality of care) for measuring health system performance in the context of integration. These indicators are discussed in turn in the subsequent results sections, which further include qualitative evidence (objective 4).

Domain 1: sensitivity to case detection

When we evaluate the sensitivity of case detection, the integrated surveillance system has incrementally detected cases of both category III Buruli ulcer at 72%, 43% and 37% and children with leprosy disability at 55%, 19% and 10% in 2017, 2018 and 2019, respectively. There was, however, a gradual reduction in new cases across the two diseases in the third year of intervention of the integrated approach. Insights from interviews with CHVs and CHAs suggest that the decrease in the number of cases in the third year of the intervention may have resulted from several factors, including the poor morale among CHVs and CHAs to continue identifying cases amid medicines and supplies being out of stock at the health facility and the absence of incentives for them for case finding (see, eg, quotes 3 and 4 in table 5). A few key informants were also of the view that the reduction in cases in the subsequent years could be attributed to a lack of motivation to retain the community and professional health workers (Maryland Key Informant-14, table 5). The CM-NTD measurable data demonstrate improvements following the implementation of an integrated approach in the capacity of the surveillance system to rapidly detect cases at the start of the intervention. However, the data do not indicate a significant improvement in the system's sensitivity to detect cases early in the progression of the disease.

Domain 2: measure of access to CM-NTD services

There has been an increase in access to NTD services in the integrated counties, as evidenced by an increase in the identification and referral of suspected cases at the community level. We observed that the health system now reports cases from over 71 health facilities, bringing CM-NTD services closer to the communities. Over 222 professional health workers were trained to provide CM-NTDs health services in addition to other health services in the study settings compared with the preintegration approach (see, eg, quote 2 in table 5). Additionally, qualitative analysis showed that most facility-level informants identified treatment adherence by people affected by CM-NTDs to have improved (see table 5, qualitative evidence), with fewer complications following treatment.

The first and second years of implementation of the integrated approach saw about 59% (41) of health facilities reporting and managing CM-NTD cases in the integration counties; however, by the third year, the number of health facilities reporting new cases of CM-NTDs had reduced to 39% (28). Qualitative evidence illustrates that the reduction in health facility, reporting and managing cases could be attributed to health system challenges, including limited financial support during that period, staff turnover and a lack of refresher training. It was also likely that fewer new cases were identified in this period as most of the existing burden of infection had already been identified (see, eg, quotes 1 and 3 in table 5).

Domain 3: quality of care for NTD cases

There was an apparent variation in the quality of care; we observed strengths in treatment and lymphoedema self-care adherence. There was also an improved quality of care relating to the limitation of movement after healing occurs. These strengths indicate about 80% treatment adherence for Buruli ulcers and 90% self-care adherence for lymphoedema in the first 2 years of implementing the integrated approach. Of all Buruli ulcer cases treated and healed, less than 10% experienced limitation of movement, which was far less than the 25% set as a target by the WHO for 2020 (see, eg, quote 6 in table 5).

Most lymphoedema-affected participants appreciate the importance of the home-based self-care package but stress the need to supply this on an ongoing basis (see, eg, quote 7 in table 5). There are potential weaknesses in clinical diagnosis, especially for Buruli ulcer, which may have led to over-reporting and unnecessary treatment with specific antibiotics. We found that of the only batch of 44 Buruli ulcer specimens shipped and tested during the intervention years using PCR, only 26% were positive in the three integrated counties. Poor clinical diagnosis (limited clinical diagnostic experience, including limited refresher training or mentoring for clinicians) and test sensitivity may impact over-reporting or under-reporting. We also observed fewer surgeries for hydroceles compared with the number of cases detected over the period, as these may have had less priority compared with other NTDs during resource allocation. National-level participants disclosed that hydrocele surgeries reported to the DHIS2 are emergencies or elective surgery cases requiring patient fees. A national-level informant stressed the challenges in funding and training for hydrocele surgeries (see quote 5 in table 5). Diary notes from a discussion on integrated training detail how one international partner explained: '*Karsor! Sorry, unfortunately, we cannot include all CM-NTDs in this training because we cannot justify it to the donor…in as much I would love to, but there is no justification under our contract with the donor*' (International partner). Negotiating donor priorities was critical to effective and efficient health services integration and country ownership of the overall approach.

Challenges in quality of care were also identified by study participants as impacting the level of trust between people affected by CM-NTDs and the health system, including CHAs and CHVs. For example, the lack of medicine to treat cases when they are referred to health facilities by CHA and CHV was one perceived factor that may have impeded trust and led to a plateauing of case detection in some areas. Despite some challenges to

intervention quality, largely impacted by broader systems factors (eg, supply chain management; see Kollie *et al* forthcoming), most national and international key informants were of the view that the integration of CM-NTDs into the health system was feasible. They described key successes in the embedment of the CM-NTDs data system into the national DHIS2, embedding CM-NTDs medical commodities into the national supply chain system and national joint integrated supervision within the national community health assistant programme.

> 'So yes, the programme succeeded in embedding CM-NTDs indicators in the National MOH DHIS2, the CM-NTDs training materials were also integrated into the training module of the community health training modules, the inclusion of NTDs in the supply chain and the national health promotion division of the MOH and the joint integrated supportive supervisions tools' (International Key Informant-7)

DISCUSSION

To our knowledge, this paper is the first to evaluate the impact of integrating CM-NTDs into an existing health system. Our quantitative and qualitative results demonstrate the positive impact of integration on the health system's response to CM-NTDs. This approach has improved the overall case detection rate more than fourfold and enhanced accessibility through the provision of coordinated inputs, including training activities, awareness of CM-NTD prevention and control, medical supplies and scaling up of CM-NTD services into primary health facilities. Qualitative evidence supports disease patterns documented within the quantitative data related to noticeable increases and subsequent decreases in case detection. Challenges emerged with respect to poor morale linked to a lack of refresher training, medicines and supplies being out of stock at the health facility and the absence of incentives impacting quality, equity and access. We now highlight learning about strengthening integrated case detection, access and quality of care.

Learnings to strengthen integrated case detection, access and quality of care

Case detection

Except for leprosy, there was a significant fourfold improvement in detecting cases in integrated counties compared with non-integrated counties. Our findings corroborate the assertions of many theoretical papers that an integrated approach to disease surveillance would improve case detection.[2 20–22] This was greatly enhanced by the community-level case detection approach led by CHVs, who identified more than 62% of all cases compared with other cadres of community health workers. The excellence of CHV performance in the first and second years of integration might be partly attributed to factors, including motivation, presented within our qualitative findings. Additionally, the nature of the intensive training received by the CHVs compared with less intensive modules for community health services training that had less emphasis on CM-NTDs might be a factor. Despite increases in case detection, there was a variation in decline in new cases across diseases in the third intervention year, potentially explained by limited refresher training and the difference in modes of transmission of the CM-NTDs.[21 23] This points to the need for further investigation into the sustainability of the approach. There was an additional increase in case detection resulting from an extra survey in one of the integrated counties. This shows that the integration approach was effective but suboptimal for detecting and reporting all existing cases. This demonstrates the importance of strengthening ongoing epidemiological surveillance for early detection of cases, tracking ongoing changes through time and identifying and responding to health system weaknesses. Our analysis revealed that the impact of integrating leprosy with the other CM-NTDs was not very powerful compared with others, which requires further investigation.

Access to CM-NTD services

In the integrated counties, access to CM-NTD services has increased and improved, supporting the position of WHO that integrated health services have the potential to increase access to services,[24] through the training of professional health workers, CHVs and CHAs.[25] Integrated training of the health workforce has led to the scaling up of CM-NTD services to all government health facilities in the integrated counties and consequently enhanced the progress of Liberia towards the goal of Universal Health Coverage for health services for those who are in need.[3] Several other factors, including supportive supervision, mentoring and a sufficient supply of medicine and supplies, have enhanced access and scaled up the process. However, key barriers remain, such as weakness in the availability of medicine, uncertainty about the sustainability of medical supplies through the national supply chain system[26] and the need for continuous in-service training. While our findings confirmed the importance of training for the success of the integrated approach as suggested by Barogui *et al* and Means *et al*.,[20 22] training as an isolated intervention, as recommended by Barogui *et al* cannot achieve the level of success without other inputs and can be counterproductive to strengthening health systems if increased demand is not met with increased supply.[22] For other countries that are contemplating adopting an integrated approach to CM-NTDs, and indeed for their scale-up in Liberia. Investment in strengthening the supply chain and training, support and supervision are important for success and are needed to underpin increased access to CM-NTD services for all to achieve Universal Health Coverage. There is a need to monitor and further investigate equity in access to these services, including by gender, disability and mental health conditions, to ensure these integrated services are accessible to all of those in need.

Quality of services

Our results demonstrate variations in the quality of care. One of the benefits has been that integration has promoted lymphoedema self-care and adherence to medication for those affected by CM-NTDs. Integration of health services has also led to a lower proportion of cases with limitations of movement after management. However, challenges remain that impact the quality of services, including weaknesses in the supply chain and limited training to strengthen the capacity of the health workforce, which are needed to provide the necessary quality of services. Mitjà *et al* also recognise the importance of capacity strengthening at all levels of the health system and emphasise that for a CM-NTD approach to be successful, training of the health workers and CHA or CHV must be a priority.[21] Our data contest the recommendation from Means *et al* that CM of individuals with advanced NTD complications should remain a vertical programme because of the less extensive overlap of CM-NTDs in the population.[20]

Recommendations

Our recommendations are directed at three levels: (1) Liberia's national NTD programme, (2) policymakers and programme managers in other contexts and (3) the WHO. Within Liberia, we hope to scale up CM-NTDs' integrated approaches within all counties. The national programme requires overarching health system strengthening that will fill the gaps and weaknesses identified, especially in the provision of quality CM-NTD care. There is a need to strengthen existing policies and develop a strong national policy on integration, training and retraining in integrated approaches of health facility staff and CHVs or CHAs in CM-NTD awareness (including correct identification of Buruli ulcer, hydrocele and other NTDs), detection and management. Investing in the health workforce and supply chain in a sustained manner is critical to the sustainability of this approach in the context of high levels of donor dependency.[27] For other countries and programme managers interested in integrating CM-NTDs, the experience in Liberia highlights the importance and potential benefits of mainstreaming into the health system; however, the process was complex and needs to consider elements of the health system that may not have been so critical when implementing vertical disease control programmes, such as general health worker training, robust supportive supervision and flexible donor partners who are committed to integration. Internationally, we stress the importance of WHO's focus on developing or adopting cross-cutting measurement indicators for integration at a country level and standard integrated training modules and tools related to the health systems building blocks for the integration strategy of CM-NTDs. Currently, there are no cross-cutting measurable indicators for integration except for individual diseases used as proxy indicators. We recommend a multimethod evaluation of CM-NTD integration as a way of complementing and further illuminating the findings of the integration process.

Limitations and strengths

The study design was limited in terms of verification of the accuracy of clinical diagnosis of diseases and conditions, in particular Buruli ulcer and hydrocele. These results need to be reviewed with a critical lens and strategies to support improved diagnosis are required. The lack of standardised, measurable indicators for integration prompted the use of proxy indicators. The study used routine programme data rather than being specifically designed as a highly controlled empirical study. The study was limited to three of the five intervention counties due to resource constraints. As the data analysed were health facility-based, the study could not identify discrepancies that may have occurred between health facility to district and from district to national-level HMIS. However, in terms of strength, the study was situated in real-world programme data collection and has benefited from the use of routine data for decision-making. A holistic and multimethod analysis was undertaken, and data were generated from the perspectives of different health system actors. Our approach highlights the importance of ongoing multimethod evaluation and learning within national programmes when integration is taking place. Using quantitative indicators in isolation would not have uncovered the quality and equity challenges identified through qualitative findings.

CONCLUSION

Integration of the CM-NTDs into the Liberia health system is impactful; it has led to improved case detection, accessibility and availability of CM-NTD services, thereby promoting universal health coverage. Addressing in a sustained manner the issues of capacity strengthening, staff motivation, tools to measure the integration approach, early and equitable case detection, access and quality of care are critical. Additionally, addressing the gaps in the health management information system and supply chain will significantly impact the likelihood of successful and sustainable integration.

In the evaluation process, qualitative methods complemented and further illuminated quantitative evidence of the impact of integration. Thus, measuring the impact of CM-NTD integration relies on the consideration of multiple domains (case detection, access and quality) simultaneously and requires both qualitative and quantitative data to create a holistic understanding of the complex impacts of CM-NTD integration.

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