

Collecting Baseline Information for National Morbidity Alleviation Programs: Different Methods to Estimate Lymphatic Filariasis Morbidity Prevalence

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Abstract. The lymphatic filariasis elimination program aims not only to stop transmission, but also to alleviate morbidity. Although geographically limited morbidity projects exist, few have been implemented nationally. For advocacy and planning, the program coordinators need prevalence estimates that are currently rarely available. This article compares several approaches to estimate morbidity prevalence: (1) data routinely collected during mapping or sentinel site activities; (2) data collected during drug coverage surveys; and (3) alternative surveys. Data were collected in Plateau and Nasarawa States in Nigeria and in 6 districts in Togo. In both settings, we found that questionnaires seem to underestimate the morbidity prevalence compared with existing information collected through clinical examination. We suggest that program managers use the latter for advocacy and planning, but if not available, questionnaires to estimate morbidity prevalence can be added to existing surveys. Even though such data will most likely underestimate the real burden of disease, they can be useful in resource-limited settings.

INTRODUCTION

Lymphatic filariasis (LF) caused by the mosquito-transmitted parasites *Wuchereria bancrofti* and *Brugia malayi* is endemic in 83 countries.¹ While one fifth of the world's population is at risk, an estimated 120 million people are infected.^{2,3} Although most people suffer from subclinical destruction of the lymphatic system, the proportion of symptomatic persons depends on the infecting parasite and the region of the world.⁴ It is estimated that 40 million persons in the world have disfiguring symptoms.⁵ This results in a ranking for LF as the second-leading cause of permanent disability worldwide.⁶ The estimated disability adjusted life years (DALY) burden due to LF is 5.55 million; in Africa alone, LF causes almost US\$1 billion in yearly losses, of which more than 80% is due to disability in men with hydrocele.⁷ Due to the fact that the lag time between infection and clinical symptoms can be more than 10 years, new manifestations of LF will appear, even when transmission is eliminated.^{8,9}

The WHO resolution WHA 50.29(1997) calling for the elimination of LF as public health problem was the basis for the creation of the Global Alliance for the Elimination of LF (GAELF). The elimination program consists of two pillars: stopping transmission and alleviating the morbidity caused by the infection.¹⁰ Initially, most energy and resources went into organizing the mass drug administrations (MDA) to stop transmission, which targeted an estimated 610 million people in 42 countries by MDA during the first 5 years of GAELF.¹ In contrast, morbidity alleviation projects exist only in a few endemic countries and LF program managers are looking into scaling up the geographical coverage of those interventions.¹ For the planning, advocacy, and funding of nationwide morbidity alleviation programs, it is important for LF coordinators to have an estimate of country-specific morbidity prevalence estimates—data that are currently rarely available. A laboratory test to determine the prevalence of clinical filari-

asis is not available and clinical examinations seem to be the most used and only validated methodology.¹¹ In the literature, morbidity prevalence estimates are frequently determined using clinical examination of the entire study population, but this is mainly done to answer specific research questions such as to define age-specific and gender-specific morbidity prevalence, to determine the impact of MDA on morbidity, or to describe the morbidity situation in specific villages.^{4,12–14} Most published population-based prevalence surveys involve a thorough clinical examination, such as described by Ngwira and colleagues who did a full body clinical examination among 3,000 persons in Malawi.¹⁵ Dunyo and colleagues set up a clinic in each of 9 surveyed communities in Ghana to examine more than 6,000 study participants to determine morbidity prevalence.¹⁶ Such methods are cumbersome and not convenient for determining the magnitude of LF morbidity for the purpose of countrywide project design. Besides the work done by Gyapong in Ghana, little research has been done to find a cost-effective way of collecting morbidity prevalence figures.¹⁴ To determine if there is a feasible way for country managers to estimate the burden of disease for advocacy purpose and program planning, we compared several ways morbidity prevalence data can be collected in study sites in Nigeria and Togo.

METHODS

Survey areas. The data from Nigeria were collected in Plateau and Nasarawa State, where The Carter Center assists the states.¹⁷ Depending on the parasite prevalence, areas in these two states are targeted by activities focusing on both onchocerciasis and filariasis (“oncho and LF” area) or on lymphatic filariasis alone (“LF-only” area) (Figure 1). In 2004, 3.5 million persons were reached by MDAs to interrupt LF transmission and control oncho. In Togo, data were collected in 6 districts co-endemic for LF and oncho with an estimated population of 0.8 million persons (Figure 2). Since 2000, the National Program to Eliminate Lymphatic Filariasis (NPELF) has organized yearly MDAs in the endemic districts.

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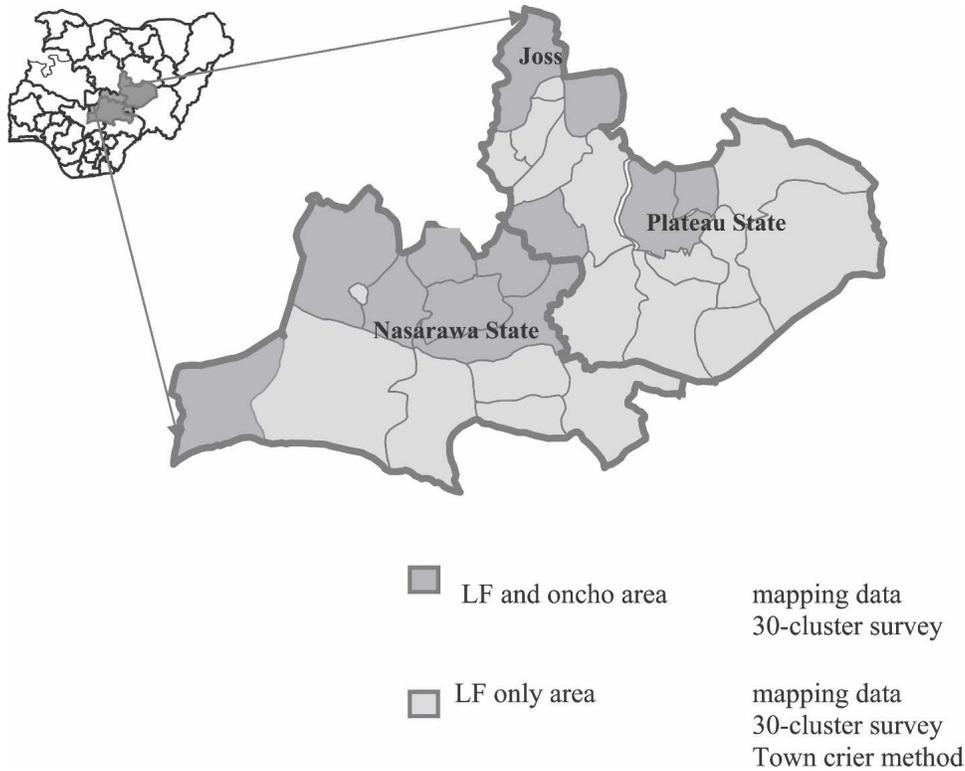


FIGURE 1. Map of Nigeria, identifying Nasarawa and Plateau State and LF and Oncho and “LF-only” area.

Data routinely available. Two sources of morbidity data are often available to the program coordinator. The first source is the information routinely collected during LF mapping. In the selected villages in West Africa, data are generally collected from 50 to 100 adults, defined as persons of 15 years or older. A second source is the data collected in sentinel site villages, which are used to follow up the impact of MDA on the trans-

mission of LF.¹⁸ In those villages, information is collected from a convenience sample of 500 persons volunteering for the microfilaria night bleeding. In both cases, the population undergoes a more or less rigorous clinical examination by medical staff.

Morbidity surveys. A second way to collect information is to conduct morbidity prevalence surveys during the WHO-

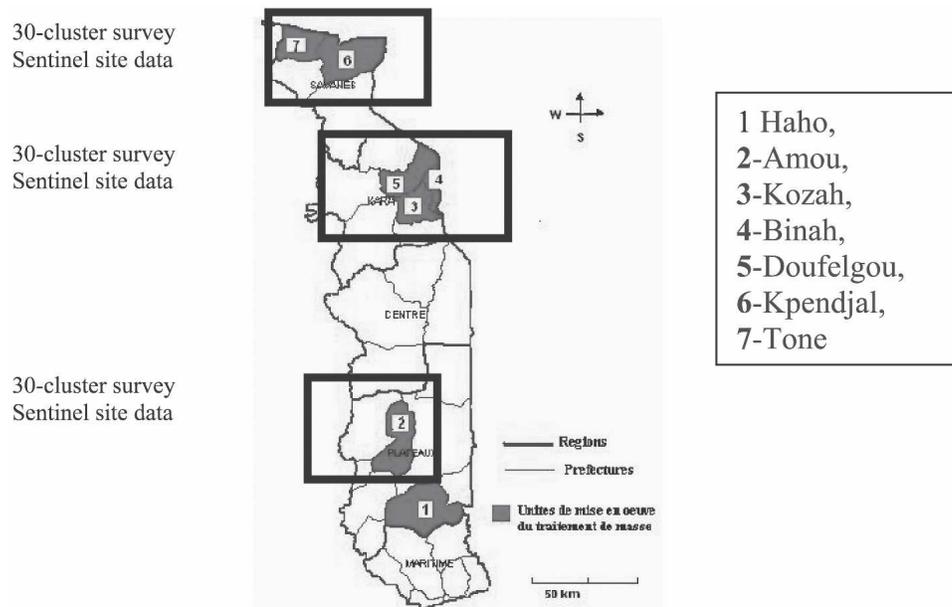


FIGURE 2. Map from Togo identifying the endemic district and the surveys conducted.

recommended drug coverage surveys after MDAs.¹⁸ We conducted several 30-cluster surveys in Nigeria and Togo, using a probability sample of all persons living in the surveyed area. The sample frame for each survey included all the villages in which a MDA was organized during the previous year. Thirty communities were chosen as primary sampling units (PSUs) with probability proportional to estimated population size. Population estimates were obtained from the census conducted by the village distributors prior to or during MDA.

From a central point or from the house of the head of the village, a random direction was selected by spinning a bottle. All the houses between the central point and the end of the village were counted and with the number on a bank note as a random number provider, a starting house was selected. A “next-nearest-house” path was used to select the next 9 houses. Local field staff was trained to select the households and to administer the questionnaire. In addition to the drug coverage questionnaire administered to all household members living in the selected houses (data not reported), morbidity information was collected from persons 18 years or older. A morbidity questionnaire was administered to one randomly selected adult household member. To estimate the morbidity prevalence, the following question was asked: “Does anyone in your household of the age of 18 years or older have lymphedema or hydrocele?” To estimate lymphedema stage, persons answering affirmative to this question had to indicate the stage on a drawing representing the different stages (Figure 3). The denominators for the hydrocele and lymphedema prevalence estimates were respectively, the male population and total population of the age of 18 years or above living in the surveyed households. Additional questions were asked to measure the impact of the LF morbidity on daily life, to determine the place where treatment was obtained, and other topics relevant for the national program manager (data not reported).

In Plateau and Nasarawa state (Nigeria), two 30-cluster surveys were conducted in 2003: one in the “oncho and LF” area and one in the “LF-only” area, covering a population of about 3 million persons (Figure 1). Sentinel site villages and urban areas were excluded from the sample frame. In Togo, three 30-cluster surveys were conducted in 6 endemic districts in 2004 with a population of approximately 820,000 (Figure 2).

Alternative survey. In an effort to find an easier way to conduct morbidity prevalence surveys, we tried a “town crier” method in Nigeria. Twenty villages were randomly selected from the sample frame used for the “LF-only” cluster survey.

After informing the local authorities, people suffering from lymphedema and hydrocele were asked by the town crier to gather in a central location. The interviewer counted the patients and recorded the staging of lymphedema, using the same drawing as was used for the cluster survey (Figure 3). To calculate the denominator for hydrocele prevalence, we used the data collected in the coverage survey to find out what proportion of the total population was represented by men 18 years of age or older. Based on this proportion and the village population, we determined the denominator. For the denominator for lymphedema, we used the same approach, using the proportion of the total population ≥ 18 years.

Data management. All data entry was done locally in Epi Info 6 (CDC) and analyzed using SAS (SAS Institute, Cary, NC) and SUDAAN (Research Triangle Institute, Research Triangle Park, NC) at CDC, Atlanta, GA. To be able to compare different methods, it was necessary that the collected data represent the same geographical area. For this reason some data were aggregated. To compare the prevalence estimates, we calculated 95% confidence interval with SUDAAN taking into account the cluster effect in all described methods. To compare the methods based on probability samples, we used the z-test.

RESULTS

Data routinely available. The two states in Nigeria collected morbidity prevalence data during the initial mapping. In each village selected for mapping, 30 to 100 adults (≥ 15 years) were randomly selected. From the 4,110 persons examined, 46 were diagnosed with Lymphedema, resulting in a prevalence of 1.09% (95% confidence interval [CI] CI 0.7–1.5). From the 2,185 men clinically examined, 168 or 7.7% (95% CI 5.6–9.7) were diagnosed with hydrocele (Table 1). In “LF-only area,” 25 persons (1.0%, 95% CI 0.4–1.6) were diagnosed with lymphedema and 113 men (8.4%, 95% CI 5.4–11.29) were diagnosed with hydrocele. Morbidity data were not collected in the sentinel sites. In Togo, data collected during mapping were not available, but prevalence data were collected in the sentinel sites through clinical examination by the district nurse (2001). Hydrocele prevalence among adult men (≥ 18 years) was 0.6% (95% CI 0.0–1.4) (Table 2). Lymphedema prevalence among adults (≥ 18 years) was 0.8% (95% CI 0.0–2.0).

Morbidity surveys. In Nasarawa and Plateau State, we interviewed 585 persons (median age: 39.6 years), giving us morbidity information from 2,899 adults (> 18 years) living in the selected households, including 1423 male. The reported

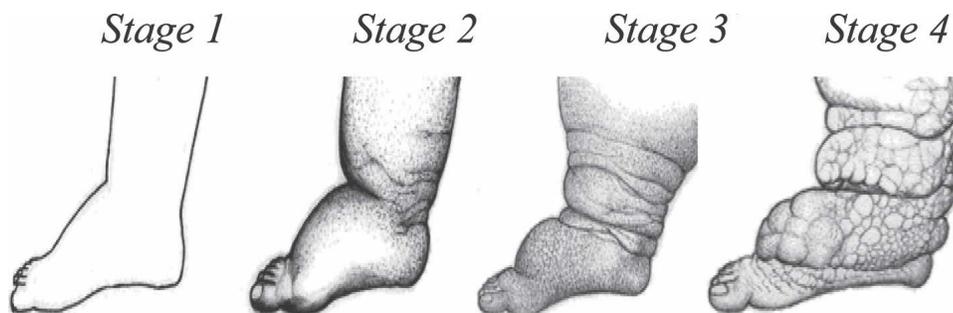


FIGURE 3. Staging of lymphedema.

TABLE 1

Hydrocele and lymphedema prevalence data collected during LF mapping, cluster survey and town crier method, Nasarawa and Plateau States, Nigeria

	Hydrocele			Lymphedema		
	N	Prevalence (%)	95% CI	N	Prevalence (%)	95% CI
Nasarawa and Plateau state						
Clinical examination (mapping)	2,185	7.69	5.65–9.73	4,110	1.09	0.66–1.52
Questionnaire (Cluster survey)	1,423	1.90	0.78–3.02	2,899	0.45	0.14–0.76
“LF-only” area in Nasarawa and Plateau state						
Clinical examination (mapping)	1350	8.37	5.45–11.29	2550	0.98	0.39–1.57
Town crier method	2,918	1.64	0.86–2.42	6,045	0.25	0.07–0.43
Questionnaire (Cluster survey)	768	2.34	0.97–3.71	1,562	0.58	0.01–1.15

hydrocele prevalence was 1.9% (95% CI 0.8–3.0) and the lymphedema prevalence was 0.4% (95% CI 0.1–0.8) (Table 1). In “LF-only area,” hydrocele and lymphedema prevalence was 2.3% (95% CI 1.0–3.7) and 0.6% (95% CI 0.0–1.2) respectively. For the 13 cases of lymphedema for which we collected staging information, 4 (30.8%) were stage 1, 5 (38.5%) were stage 2, 2 (15.4%) were stage 3, and 2 (15.4%) were stage 4.

For the cluster surveys in Togo, we interviewed 836 persons (median age: 38 years), giving us morbidity information from 2,891 adults (≥ 18 years) living in the selected households, including 1,266 men (Table 2). The prevalence of hydrocele was 0.6% (95% CI 0.2–1.1) among adult men and the prevalence of lymphedema was 0.2% (95% CI 0.0–0.3) among adults. Of the 5 cases of lymphedema from which we collected information, 2 (40.0%) were stage 1 and 3 (60.0%) were stage 2.

Alternative surveys. Based on the data collected in the morbidity survey mentioned previously, we calculated that 54.9% of the surveyed population in Nigeria was above the age of 18 and that 26.5% were male above the age of 18. From the estimated 6,045 adult persons living in the 20 surveyed villages using the “town crier” method, 15 or 0.2% (95% CI 0.1–0.4%) had lymphedema. This was not statistically significantly different from the morbidity cluster survey ($z = 0.33$, $P = 0.72$). We collected staging information from 12 lymphedema cases: 10 (83.3%) were stage 1, one (8.3%) was stage 2 and one (8.3%) was stage 3. From the estimated 2,918 adult men, 48 cases or 1.6% (95% CI 0.9–2.4%) of hydrocele were identified, which was also not statistically significantly different from the morbidity cluster survey ($z = 0.68$, $P = 0.48$).

DISCUSSION

The main purpose of this article was to find an acceptable way to estimate LF morbidity prevalence that would not re-

quire too many resources. It is important that these estimates are available for advocacy (e.g., adding morbidity alleviation programs to national action plans), fundraising, and allocation of resources in preparation for national morbidity programs. We describe different approaches LF country managers can use to collect nationwide morbidity prevalence figures: (1) data routinely collected during mapping or sentinel site activities; (2) morbidity surveys combined with coverage surveys; or (3) alternative surveys designed to identify persons with morbidity. None can be considered to represent a “gold standard,” which only could be obtained by expensive and time-consuming population surveys that include a thorough physical examination.

The data from Nigeria indicate that methods including physical examination find higher hydrocele prevalence rates than methods using questionnaires. The hydrocele rate was more than 4 times higher during a survey based on clinical examination compared with surveys based on reporting of pathology (7.6% compared with 1.9%). Due to the use of convenience sampling, statistical testing could not be applied and although confidence intervals can be compared, this also must be done with caution due to the used sampling frame. These data seem to confirm the findings from Eigege and colleagues who found that only 68% of men diagnosed with hydrocele by clinical examination had mentioned this symptom in the history taken prior to the examination.¹⁹ It also reiterates findings by Gyapong who stated that hydrocele estimates obtained from interviews are underestimates, but that this cheaper method can give a good estimate of lymphedema prevalence.²⁰ A possible explanation for the discrepancy between clinical examination and questionnaires may be that men are embarrassed about their hydrocele, so that they will not admit it to the interviewer or to family members. This limits the reliability of questionnaires for obtaining information from both patients and family members. The difference in sampling could also explain some of the discrepancy. The

TABLE 2

Hydrocele and lymphedema prevalence collected during sentinel site activities and cluster survey, Togo

	Entire endemic area			North (Kjendpal/Tone)			Central (Kozah, Binah, Doufelgou)			South (Amou)		
	N	Prevalence (%)	95% CI	N	Prevalence (%)	95% CI	N	Prevalence (%)	95% CI	N	Prevalence (%)	95% CI
Hydrocele prevalence												
In sentinel sites	655	0.61	0.00–1.41	177	0	–	340	0.89	0.00–2.36	138	0.73	0.00–2.14
Cluster survey	1,266	0.63	0.20–1.06	522	1.23	0.03–2.42	338	0.59	0.00–1.39	406	0.19	0.00–0.56
Lymphedema prevalence												
In sentinel sites	1,518	0.80	0.00–1.98	458	0.22	0.00–0.65	794	1.28	0.00–3.49	266	0.38	0.00–1.07
Cluster survey	2,891	0.17	0.00–0.34	1,108	0.2	0.00–0.59	793	0.13	0.00–0.38	990	0.18	0.00–0.43

mapping data were collected from a convenience sample of men who presented themselves for testing. Men with symptoms are probably more likely to volunteer. Another possible explanation could be that hydrocele is so common in certain communities that—if limited in size—it is not considered to be abnormal and for this reason not reported. Though the same trend was noticed in 2 of the 3 regions (Central and South) in Togo, national estimates were similar independent of the method used (both 0.6%).

We expected the underreporting in surveys to be less of an issue for lymphedema, which is visible to the interviewer and the family members; however, this was not the case. The prevalence estimates based on clinical examination were more than twice the estimates obtained by surveys in Nigeria (1.1% and 0.45%, respectively) and in Togo, the lymphedema prevalence obtained by clinical examination was more than 4 times the estimate obtained during the survey (0.8% and 0.2%, respectively). As mentioned previously, those data must be compared with caution. A possible explanation for this finding could be that in our surveys, almost 70% (Nigeria) to 100% (Togo) were mild cases of lymphedema with reversible (stage 1) or irreversible (stage 2) swelling without further symptoms. It is possible that the symptoms were not considered a problem for the patients and for that reason not reported. This could be due to the sample because in a survey conducted by Richard and colleagues in the same districts in Togo, only 28% of the LF patients were stage 1 or 2.²¹

Our data found that there was no statistically significant difference between prevalence estimates obtained by the town crier method and the estimates found in surveys using questionnaires and complicated sampling. The first method used fewer resources and could be used at the same time to educate patients. The feasibility of this method will of course depend on cultural aspects, which can be different depending on the country.²²

The pathology of lymphatic filariasis is complex and variable in clinical presentation, which makes it difficult to define prevalence accurately. We only collected information on hydrocele and lymphedema prevalence figures because these are the most common clinical manifestations and most morbidity programs are focused on those two pathologies. With the exception of the mapping data from Nigeria where the cut-off age was 15 years, we excluded persons younger than 18 years of age from the denominator because lymphedema and hydrocele prevalence increases with age and are rarely seen in children.^{4,12,16,23} This approach was also used by Bockarie and colleagues, who excluded males under 16 for determining hydrocele prevalence and persons under 21 for lymphedema because the likelihood of advanced pathology below those ages was low.¹³

An important shortcoming of using morbidity prevalence data from sentinel sites or coverage surveys is that they are limited to areas with active transmission, while it is well known that morbidity cases are also present where there is currently no transmission.²⁴ This indicates that prevalence surveys for morbidity must be further reaching than the MDA program. The lag time between infection and clinical symptoms can also take more than 10 years, so even when transmission is interrupted and MDAs are no longer necessary, the chronic manifestations of the disease will continue to appear.⁸ This is the case at the border area with Togo and Benin. There is no more transmission in this area due to an active

malaria vector control program in the 1970s, but there is a relatively high prevalence of LF morbidity (Sodahlon, personal communication). A possible solution could be to add morbidity questions to national surveys conducted for other programs. In Togo, similar morbidity prevalence questions used in this article were added to a nationwide household-based bed net coverage survey. Results indicated that 0.6% (95% CI 0.3–0.9) of the households had at least one person with lymphedema and 2.6% (95% CI 1.8–3.4) of the households had at least one male with hydrocele (Vanden Eng, personal communication). Taking into account that a household consists of an average of 6.6 persons (Mathieu, unpublished data), the prevalence data are similar to those found in our surveys.

Our data indicate that prevalence data collected by questionnaires tend to underestimate the morbidity prevalence compared with data collected by clinical examination. Due to the limited resources currently available for lymphatic filariasis in general and for morbidity programs in particular, it will be difficult for program managers to spend large amounts of resources on population-based clinical surveys. For this reason, we suggest the use of existing clinical data from mapping or sentinel site activities to estimate LF morbidity prevalence. If those data are not available or are too limited in geographical coverage, questions aimed at reporting of LF morbidity can be added to existing surveys conducted by the LF program or other programs with national scope. These data can be used for planning, advocacy, and fund raising although they must be used with caution because they seem to underestimate the real burden of disease. Our research seems to suggest that alternative surveys such as the town crier method can be used instead of surveys with a more complex design, but further research is necessary.

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