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Eileen Stillwaggon  
*Gettysburg College*

Larry Sawers  
*American University*

Jonathan Rout  
*Church's Auxiliary for Social Action*

*See next page for additional authors*

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## Abstract

Lymphatic filariasis afflicts 68 million people in 73 countries, including 17 million persons living with chronic lymphedema. The Global Program to Eliminate Lymphatic Filariasis aims to stop new infections and to provide care for persons already affected, but morbidity management programs have been initiated in only 24 endemic countries. We examine the economic costs and benefits of alleviating chronic lymphedema and its effects through a simple limb-care program. For Khurda District, Odisha State, India, we estimated lifetime medical costs and earnings losses due to chronic lymphedema and acute dermatolymphangioadenitis (ADLA) with and without a community-based limb-care program. The program would reduce economic costs of lymphedema and ADLA over 60 years by 55%. Savings of US\$1,648 for each affected person in the workforce are equivalent to 1,258 days of labor. Per-person savings are more than 130 times the per-person cost of the program. Chronic lymphedema and ADLA impose a substantial physical and economic burden on the population in filariasis-endemic areas. Low-cost programs for lymphedema management based on limb washing and topical medication for infection are effective in reducing the number of ADLA episodes and stopping progression of disabling and disfiguring lymphedema. With reduced disability, people are able to work longer hours, more days per year, and in more strenuous, higher-paying jobs, resulting in an important economic benefit to themselves, their families, and their communities. Mitigating the severity of lymphedema and ADLA also reduces out-of-pocket medical expense.

Disclaimer: The findings and conclusions in this report are those of the authors and do not necessarily represent the views of CDC.

## Keywords

lymphatic filariasis, odisha state, community-based limb-care program, lymphedema and acute dermatolymphangioadenitis

## Disciplines

Community Health and Preventive Medicine | Health Economics

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## Authors

Eileen Stillwaggon, Larry Sawers, Jonathan Rout, David Addiss, and LeAnne Fox

## Economic Costs and Benefits of a Community-Based Lymphedema Management Program for Lymphatic Filariasis in Odisha State, India

Eileen Stillwaggon,<sup>1\*</sup> Larry Sawers,<sup>2</sup> Jonathan Rout,<sup>3</sup> David Addiss,<sup>4</sup> and LeAnne Fox<sup>5</sup>

<sup>1</sup>Department of Economics, Gettysburg College, Gettysburg, Pennsylvania; <sup>2</sup>Department of Economics, American University, Washington, District of Columbia; <sup>3</sup>Church's Auxiliary for Social Action (CASA), Bhubaneswar, India; <sup>4</sup>Children Without Worms, Task Force for Global Health, Decatur, Georgia; <sup>5</sup>Division of Parasitic Diseases and Malaria, Centers for Disease Control and Prevention, Atlanta, Georgia

**Abstract.** Lymphatic filariasis afflicts 68 million people in 73 countries, including 17 million persons living with chronic lymphedema. The Global Programme to Eliminate Lymphatic Filariasis aims to stop new infections and to provide care for persons already affected, but morbidity management programs have been initiated in only 24 endemic countries. We examine the economic costs and benefits of alleviating chronic lymphedema and its effects through a simple limb-care program. For Khurda District, Odisha State, India, we estimated lifetime medical costs and earnings losses due to chronic lymphedema and acute dermatolymphangioadenitis (ADLA) with and without a community-based limb-care program. The program would reduce economic costs of lymphedema and ADLA over 60 years by 55%. Savings of US\$1,648 for each affected person in the workforce are equivalent to 1,258 days of labor. Per-person savings are more than 130 times the per-person cost of the program. Chronic lymphedema and ADLA impose a substantial physical and economic burden on the population in filariasis-endemic areas. Low-cost programs for lymphedema management based on limb washing and topical medication for infection are effective in reducing the number of ADLA episodes and stopping progression of disabling and disfiguring lymphedema. With reduced disability, people are able to work longer hours, more days per year, and in more strenuous, higher-paying jobs, resulting in an important economic benefit to themselves, their families, and their communities. Mitigating the severity of lymphedema and ADLA also reduces out-of-pocket medical expense.

### INTRODUCTION

Lymphatic filariasis (LF) afflicts an estimated 68 million people in 73 countries of Africa, Asia, Oceania, and the Americas<sup>1</sup> and is one of the diseases targeted for elimination by the World Health Assembly (World Health Assembly Resolution WHA 50.29: Elimination of lymphatic filariasis as a public health problem. Fiftieth World Health Assembly, 5–14 May 1997, Resolutions and Decisions). The Global Programme to Eliminate Lymphatic Filariasis (GPELF) embodies two “pillars”: stopping new infections by the year 2020 and managing morbidity and preventing disability for persons already infected.<sup>2</sup> Of the 73 endemic countries, 62 had initiated mass drug administration (MDA) for elimination of new infections as of 2014.<sup>3,4</sup> As of 2015, 45 countries were considered “on track” to achieve elimination targets by 2020.<sup>5</sup>

An estimated 36 million people live with the disabling effects of LF, including about 17 million persons with chronic lymphedema, primarily of the legs, and also of the arms, breasts, and scrotum, and 19 million men with hydrocele.<sup>1</sup> The remaining LF-infected persons are at risk of developing lymphedema or hydrocele. Programs to manage morbidity and prevent disability among infected persons, the second pillar of the GPELF, had been initiated in only 24 of the 73 endemic countries by 2014.<sup>4</sup> This article examines the economic costs and benefits of one such program of morbidity management and disability prevention (MMDP) for alleviating the causes and effects of chronic lymphedema. Interventions for hydrocele differ from those for lymphedema and are not included in this study.

**Nature of the disease.** Larval forms of *Wuchereria bancrofti*, *Brugia malayi*, and *Brugia timori* are transmitted to humans by different species of mosquitoes, depending on world region. Lymphatic vessels are damaged by the presence of adult

worms, causing lymphedema that worsens with age. Lymphedema is generally considered an adult condition, but damage to lymph vessels from filarial infection can begin in childhood.<sup>6,7</sup> The progressive worsening of lymphedema is not inevitable; rather it is accelerated by recurrent episodes of acute dermatolymphangioadenitis (ADLA), disabling bouts of fever and intense pain lasting several days that are caused by bacterial infections.<sup>8,9</sup> These infections generally enter the lower limbs where the skin is damaged by wounds or interdigital fungal infections.<sup>8,10</sup>

Each episode of ADLA further damages the lymph system and contributes to progression of chronic lymphedema, the severity of which has been classified into seven stages by Dreyer and others.<sup>8,10</sup> In stage 1, lymphedema is generally relieved by limb elevation overnight. By stage 7, lymphedema is characterized by deep skin folds, knobs or protrusions, mossy lesions, interdigital lesions, and bad odor; the lymphedema usually extends above the knee and prevents activities of daily living.<sup>8,10</sup> Worsening lymphedema increases vulnerability to entry lesions that lead to ADLA, which in turn worsens lymphedema. Several studies confirm the higher incidence of ADLA at higher stages of lymphedema,<sup>11–18</sup> but others do not.<sup>19–21</sup> Nevertheless, “[t]he epidemiologic association between ADLA frequency and stage, as well as extensive clinical experience from both filariasis-endemic and non-endemic areas, strongly suggest that ADLA episodes are a major—likely the most important—factor in lymphedema progression, particularly in filariasis-endemic areas.”<sup>11</sup>

**Prevention of increasing disability.** With recognition of the causes of ADLA and their role in progressive worsening of lymphedema came the realization that very simple and low-cost methods could prevent recurrent ADLA episodes and thus lymphedema progression. Washing the legs and feet with soap and clean water, drying the limbs with clean towels, applying antifungal creams or antibiotic ointments to interdigital lesions, elevating affected limbs, exercising to improve lymphatic and venous drainage, and wearing shoes have been shown to be effective in reducing the number of episodes

\*Address correspondence to Eileen Stillwaggon, Department of Economics, Gettysburg College, 300 North Washington Street, Gettysburg, PA 17325. E-mail: stillwaggon@gettysburg.edu

of ADLA<sup>12,15,17,22–34</sup> (see Supplemental Information for additional discussion). Several studies have also found that limb hygiene was associated with reduced leg volume and regression in lymphedema stage.<sup>25,31,35</sup>

Very simple interventions can have a substantial impact on quality of life. Each episode of ADLA can mean several days of excruciating pain. Lymphedema limits mobility and daily activities; it can be disfiguring and lead to stigma. Reducing ADLA and lymphedema allows LF-affected persons to engage more easily in family and community life as well as employment.

**Previous studies of the economic cost of LF.** Numerous studies have described the economic cost imposed by ADLA or lymphedema. When people seek medical attention for their chronic lymphedema or during an ADLA episode they incur out-of-pocket expenses for consultations, tests, medications, and transportation costs. The quantitatively more important components of the lifetime economic cost of LF are the lost earnings in paid employment and loss of unpaid household labor. Those with lymphedema or ADLA may be forced to work fewer days per year or fewer hours per day and may earn a lower wage because they cannot engage in strenuous labor.

Chu and others<sup>36</sup> estimated the benefits of MDA from the first 8 years of the GPELF. They included patient out-of-pocket costs of medical consultations, medications and travel, lost wages due to reduced hours of work or days lost to ADLA, and medical costs to the public sector. They found that the economic cost of LF could be reduced by at least US\$20 billion by preventing transmission of filarial infection.

Two studies in Odisha State (formerly Orissa), India, found that the average annual out-of-pocket cost for medical care for lymphedema and ADLA was about US\$14,<sup>37,38</sup> more than 10 times the average daily wage of unskilled rural workers in the state.<sup>39</sup> Studies elsewhere in India and in other countries also found substantial out-of-pocket costs for medical care for lymphedema and ADLA.<sup>40–44</sup>

Each ADLA episode leads to a loss of 3–12 days of work, with an average reported in India of more than 4 days; annual incidence of ADLA varies widely among different studies in India, ranging from 1.6 to 7.6 episodes.<sup>11,15,40,45–50</sup> Chronic lymphedema at advanced stages can be completely disabling and prevent wage employment or the performance of household tasks. Those at intermediate stages of lymphedema may have partial disability with substantial earnings loss. For example, daily output measured in yards of cloth was found to be 27% lower for weavers with lymphedema than for those without.<sup>51</sup> Estimates of reduced time spent in paid or unpaid employment (measured in either hours per day or days per year) for those with lymphedema range from 13.0% to 19.5%.<sup>38,42,44,49,51,52</sup> Together with out-of-pocket medical costs, those earnings losses are an extraordinary economic burden on some of the poorest people in India.

#### A COMMUNITY-BASED PROGRAM IN KHURDA DISTRICT, ODISHA STATE, INDIA

The present work examines the economic costs and benefits of the lymphedema management program implemented by the Church's Auxiliary for Social Action (CASA), an Indian non-governmental organization (NGO), with technical assistance provided by the U.S. Centers for Disease Control and Prevention. In 2005, 40 local NGOs conducted a house-to-house census of 1.3 million persons in the rural and peri-urban areas

of Khurda District in Odisha State, an LF-endemic area, and identified all residents with lower-limb lymphedema, recording age, gender, lymphedema stage, and number of ADLA episodes in the previous year.<sup>53–55</sup> From 2007 to 2010, CASA provided services to more than 21,000 persons identified in 1,447 villages in a community-based program utilizing community health workers to train LF patients in leg washing and use of topical antibiotic and antifungal treatments.<sup>54</sup> In 2009 to 2011, 370 patients from villages not yet enrolled in the CASA program in Khurda were recruited in a prospective cohort study examining the effectiveness of the lymphedema management program. Participants reported a significant decrease in perceived disability after 2 years in the program, with greater improvements in patients with moderate or advanced lymphedema. Patients also reported losing 2.5 fewer work days per month after 1 year in the program.<sup>56</sup>

In another study of the 370 patients in the limb-care program, ADLA episodes decreased 34% over 24 months. The percentage of persons whose lymphedema progressed (worsened) decreased and the percentage of those whose lymphedema regressed (improved) increased. Use of soap was associated with decreased incidence of ADLA among persons without entry lesions.<sup>35</sup> Per-person program costs were US\$10.00 to US\$12.50 for the 24 months. Based on 29 days of lost productivity per year recovered as a result of the limb-hygiene program, it was estimated that 1,600 person-years of labor were saved in the first year of the CASA program covering more than 21,000 people.<sup>54</sup> Clinical data for this economic analysis are based on the 2005 census and the pilot studies of 2009–2011 mentioned above.

#### METHOD

Although the individual experience of persons with lymphedema due to LF varies, there is a general tendency, in the absence of intervention, toward increasing stage of chronic lymphedema and increasing frequency and severity of ADLA episodes with age.<sup>57–59</sup> The purpose of MMDP for people with lymphedema is to prevent ADLA and stop the progression of chronic lymphedema. We began by calculating the age distribution of chronic lymphedema and number of ADLA episodes per year for the population in the 2005 census of households in Khurda District mentioned above. We grouped people from 8 to 72 years of age into 5-year age cohorts and calculated the number of people at each stage of lymphedema and the number of ADLA episodes in the previous year for each age cohort. Using Microsoft Excel (Microsoft Corporation, Redmond, WA), we estimated the economic cost of morbidity and disability over the working lives of affected persons without lymphedema management and the projected reduction in those costs that would result from implementation of a community-based lymphedema management program.

**Age distribution of lymphedema stage and ADLA without and with lymphedema management.** We postulated two scenarios. In both scenarios, MDA has ended transmission of the LF parasite, but it has not reduced lymphedema or ADLA. Every 5 years, the oldest age cohort is retired from the population and younger cohorts move forward. Below age 8 years, no newly infected persons enter the treatment population because of the effects of five rounds of MDA in stopping transmission of filariasis.<sup>1</sup>

In the no-treatment scenario, there was no intervention to improve limb care, prevent ADLA, or slow progression of lymphedema. As each cohort ages, its average lymphedema stage and frequency of ADLA increase so that, 5 years from now, each group will have the morbidity distribution that its next older cohort has at present.

In the treatment scenario, we assumed that the community lymphedema management program on average halts the progression of lymphedema. In every age cohort, we assumed that progression of lymphedema stage for some is offset by regression for others, and thus each age cohort maintains the same distribution of lymphedema that it had at the beginning of the lymphedema management program. Based on the results of the limb-care program in Khurda, we assumed that the number of ADLA episodes for each age cohort will be one-third less than that in the no-treatment scenario.<sup>35</sup>

**Costs.** Using the two sets of morbidity distributions—ADLA and lymphedema stage with and without lymphedema management—we calculated the economic cost for each scenario. The difference between the two (the cost saving) is the economic benefit of lymphedema management.

Costs were calculated from the societal perspective, but we included only out-of-pocket costs to patients and their caregivers for clinic visits, medical tests, travel, and medications, and lost earnings for patients due to chronic lymphedema and ADLA. The earnings loss due to lymphedema and ADLA episodes arises from fewer work days, fewer hours per day, and/or lower daily wages. Based on costs reported in the literature, we calculated the total out-of-pocket costs and lost earnings with and without lymphedema management for each age cohort during the initial 5-year period, and then for every 5 years until the cohort ages out of the analysis at the age of 72 years. The economic benefit from the lymphedema management program is the difference between the two estimates of the total cost. We then compared the direct costs of

implementing the program to the economic benefits of lymphedema management.

All costs were estimated in US dollars for 2008, discounting future costs at 3% per year. To determine what economists call the present discounted value, future costs and benefits are assumed to be worth less than current ones and are weighted less than those in the near term.<sup>60</sup> Real wages (adjusted for inflation) and real expenditure on medical care were projected to rise 4% per year. Total cost was estimated over the working lives of all persons up to age 72. Table 1 lists the parameter values estimated for the calculation of lifetime out-of-pocket cost and earnings loss. (See Supplemental Information for explanation of data sources and derivations of parameter values for out-of-pocket medical costs, average number of days worked per year, lost work days due to chronic lymphedema and ADLA, wage rate, and the rate of increase in real wages and in the real cost of medical care.)

We used conservative estimates for improvement in ADLA, stage progression, and lost work days and hours. Predictions for real wage growth and the cost of medical care over the next 60 years are subject to considerable uncertainty. Thus, we performed sensitivity analysis using higher estimates of lost work days and lower and higher estimates of rate of growth of real wages and costs, the results of which are reported in the Supplemental Information.

## RESULTS

We found progression of lymphedema with age in Khurda District as found in other studies.<sup>57–59</sup> Table 2 shows the distribution of lymphedema stage by age, grouped in 5-year cohorts. From the youngest to the oldest, there is a steady decrease in the proportion of people in stage 1. For higher stages, we found the opposite trend. The average lymphedema stage rises monotonically with age (from 1.20 in the youngest cohort to 2.35 for people in their 70s.). Table 3 shows the percentage of persons in each lymphedema stage experiencing 0, 1, 2, and 3 ADLA episodes in the previous year. Our analysis of the Khurda data confirmed, as some studies have found<sup>11,13–18,48,57</sup> but others have not,<sup>19–21,58</sup> that those in higher stages of lymphedema are likely to have more ADLA episodes.

### Economic cost with and without lymphedema management.

Days of work lost due to chronic lymphedema and to ADLA episodes for each age cohort without an intervention are shown in the second and third columns of Table 4. We calculated the lost earnings from partial or total disability as the total number of work days lost times the average wage for rural households in Odisha State. Derivation of work days lost and the wage is described in the Supplemental Information.

Current out-of-pocket spending for medical attention for lymphedema and for ADLA episodes for each age cohort is shown in the fifth and sixth columns of Table 4.

We calculated the economic cost of lymphedema and ADLA in two scenarios, with and without a community-based lymphedema management program. Without the program, each cohort would progress through lymphedema stages as had older cohorts and more people would experience episodes of ADLA, replicating the experience of older cohorts. The total economic cost of lymphedema and ADLA is calculated as the present discounted value of the sum of out-of-pocket costs and lost earnings over the working lives of all persons

TABLE 1

Parameter values for medical costs and earnings loss due to lymphedema and ADLA\*

Parameter	Baseline estimate 2008–2009	Source
Annual per-person out-of-pocket medical costs for chronic lymphedema	US\$10.96	38
Per-episode out-of-pocket medical costs for ADLA	US\$2.04	37
Annual increase in real cost of medical care for chronic lymphedema and ADLA	4%	61–66
Annual discount rate	3%	60
Average daily wage rate	US\$1.31	39
Annual increase in real wages	4%	61–66
Lost work days per episode due to ADLA	4	37
Average number of days worked per year	289	67
Percentage of work days lost annually due to chronic lymphedema		38,42,44,49,51,52
Stages 1–2	0	
Stage 3	20	
Stage 4	50	
Stages 5–7	100	

ADLA = acute dermatolymphangioadenitis.

\*Derivation of values is explained in Supplemental Information.

TABLE 2  
Stage of lymphedema by age cohort in Khurda census, 2005

Age cohort (years)	Number of respondents	Percentage of age cohort at each stage of lymphedema							Total	Average stage
		Stage of lymphedema								
		1	2	3	4	5	6	7		
8–12	74	86.5	6.8	6.8	0.0	0.0	0.0	0.0	100.0	1.203
13–17	137	78.8	15.3	2.9	2.2	0.0	0.7	0.0	100.0	1.314
18–22	267	70.4	18.0	8.6	2.6	0.0	0.0	0.4	100.0	1.453
23–27	443	61.9	24.6	9.5	2.9	0.2	0.9	0.0	100.0	1.578
28–32	866	56.8	24.0	15.1	2.2	0.7	0.9	0.2	100.0	1.696
33–37	1,158	47.8	30.3	16.4	3.7	0.7	0.5	0.6	100.0	1.832
38–42	1,845	43.0	29.7	19.1	5.0	1.2	1.0	1.0	100.0	1.987
43–47	1,789	40.9	29.2	21.0	5.5	1.7	1.2	0.6	100.0	2.037
48–52	2,257	38.0	29.2	23.4	6.0	1.7	1.0	0.8	100.0	2.104
53–57	1,723	34.5	28.1	25.2	8.9	1.7	0.8	0.8	100.0	2.208
58–62	2,441	31.0	30.1	25.8	8.6	2.5	1.4	0.6	100.0	2.280
63–67	1,400	29.3	31.2	25.8	9.1	2.0	1.9	0.7	100.0	2.318
68–72	1,453	29.9	28.4	26.8	10.1	2.3	1.7	0.8	100.0	2.352
Total	15,853	39.5	28.6	21.9	6.6	1.6	1.1	0.7	100.0	2.084

with morbidity identified in Khurda. For this population, the total lifetime economic cost without lymphedema management is US\$47.4 million.

We then calculated the economic cost for this population in a scenario with community-based lymphedema management; people on average remain in the same stage of lymphedema over time and experience on average one-third fewer ADLA episodes per year than they would have without the limb-care program. This scenario, based on the results of the Khurda limb-care program, represents a substantial gain in quality of life for more than 17,000 people who can expect a reduction in number of episodes of ADLA and stabilization of lymphedema stage or possible improvement. The present value of the total economic cost for this population after lymphedema management is US\$21.3 million.

The present value of the benefit of lymphedema management (the reduction in economic cost) for this population is US\$26.1 million, or US\$1,648 per participant of working age. When the community-based lymphedema management program was implemented in Odisha, the average daily wage for low-skilled agricultural workers in the state was US\$1.31.<sup>39</sup> Thus, the present value of per-person economic benefit from the limb-care program was equivalent to 1,258 days of earnings. To implement and operate the Khurda community-based lymphedema management program for 2 years cost between US\$10.00 and US\$12.50 per person.<sup>35</sup> The average participant in the program can expect lifetime economic benefits that are between 132 and 165 times the

per-person cost of the program. The results are robust to changes in parameters for wage and price increases and work days lost (see Supplemental Information for sensitivity tests of our assumptions about parameter values).

## DISCUSSION

Lymphedema and episodes of ADLA in filariasis-endemic areas diminish the quality of life of affected persons due to pain, stigma, numerous days of illness each year, restricted mobility, and reduced participation in family and community life. They also impose a substantial economic cost on affected persons and their families and diminish the potential economic strength of communities. Programs to provide care for persons with lymphedema and ADLA (as well as hydrocele) in filariasis-endemic areas are mandated by the GPELF. Beyond the ethical mandate to improve quality of life for affected persons, there are strong economic arguments for investing in the care of persons affected by filariasis, which the results of this research confirm. With adequate limb care, patients are better able to support themselves and provide for their families. Children and other dependents of affected persons could have greater access to better nutrition and the

TABLE 4

Work days lost annually, annual earnings lost, and annual out-of-pocket medical costs due to lymphedema and ADLA for each age cohort at program start, 2008–2009\*

5-year cohort	Work days lost annually		Annual earnings lost due to lymphedema and ADLA	Annual out-of-pocket medical costs	
	Due to lymphedema	Due to ADLA		Due to lymphedema	Due to ADLA
18–22	2,630	1,084	US\$4,865	US\$2,926	US\$553
23–27	5,751	1,796	US\$9,887	US\$4,855	US\$916
28–32	14,941	3,568	US\$24,247	US\$9,491	US\$1,820
33–37	23,265	4,784	US\$36,744	US\$12,692	US\$2,440
38–42	50,835	7,568	US\$76,508	US\$20,221	US\$3,860
43–47	53,754	7,460	US\$80,190	US\$19,607	US\$3,805
48–52	72,626	9,204	US\$107,197	US\$24,737	US\$4,694
53–57	63,725	7,096	US\$92,775	US\$18,884	US\$3,619
58–62	98,405	10,224	US\$142,303	US\$26,753	US\$5,214
63–67	57,858	6,196	US\$83,910	US\$15,344	US\$3,160
68–72	64,245	6,192	US\$92,272	US\$15,925	US\$3,158

ADLA = acute dermatolymphangioadenitis.

\*See Supplemental Information for derivation of values.

TABLE 3

ADLA episodes in previous year experienced by persons in each lymphedema stage in Khurda census, 2005

Stage of lymphedema	Percentage of persons in each stage with ADLA episodes				
	0 episode	1 episode	2 episodes	3 episodes	Total
1	17.1	68.7	7.8	6.4	100.0
2	16.1	71.4	7.2	5.2	100.0
3	15.2	69.4	9.2	6.3	100.0
4	14.2	68.5	9.6	7.7	100.0
5	15.7	58.7	12.3	13.3	100.0
6	10.8	62.1	12.8	14.3	100.0
7	12.6	57.1	15.1	15.1	100.0
Average	16.1	69.3	8.2	6.4	100.0

ADLA = acute dermatolymphangioadenitis.

opportunity to attend school if the wage earner is healthier. Family members are relieved of the burden of caring for persons who are bedridden due to ADLA or advanced lymphedema and can contribute better to household income and domestic tasks. The community's economy is strengthened with fewer of its members disabled by lymphedema and ADLA and fewer of its families in poverty.

**Extent of the problem.** Our dataset was based on a morbidity census that was taken by visiting every household in the rural and peri-urban areas of Khurda District and found more than 17,000 persons with some degree of lower-limb lymphedema, 1.3% of the regional population.<sup>55</sup> Two-thirds of persons with lymphedema, however, were in stage 1 or 2. These results suggest the possible invisibility of persons in other locations who have subclinical lymphatic damage due to LF infection, early-stage lymphedema, or infrequent episodes of ADLA and who remain at risk for worsening ADLA, advanced lymphedema, and disability. Where morbidity estimates are based not on a census, but on the number of people who seek treatment of chronic lymphedema, ADLA, or hydrocele, prevalence could be greatly underestimated.

Another issue highlighted by the Khurda census data is the long time horizon of lymphedema and ADLA and their ongoing economic cost. Even if new infections are stopped by 2020, some people whose lymph vessels are already damaged will experience ADLA episodes and lymphedema for the rest of their lives. Lymphedema and ADLA can necessitate out-of-pocket medical costs and cause a loss of earnings from reduced hours, absenteeism, and reduced intensity of work for 60 years or more. Indeed, the younger the cohort, the greater are the economic losses that accrue over their working lives. Thus, it is of critical importance to begin lymphedema management as soon as possible and to include young people and others who may have subclinical lymphatic damage and few or no ADLA episodes. Very low-cost interventions initiated now can save a lifetime of suffering and lost earnings.

**Potential benefits nationally and internationally.** Implementation of lymphedema management throughout India would reap benefits many times greater than in Odisha alone, one of the poorest Indian states. In ranking 20 Indian states by the daily wage rate, Odisha is in the bottom quartile in nine of 10 unskilled rural occupations (Table 3a in Labour Bureau<sup>39</sup>). Even though LF generally affects the poorest people, in most other states, rural wages at all levels are higher than in Odisha. Consequently, the earnings loss of lymphedema and ADLA would be greater and the benefits of a lymphedema management program would also be greater in other Indian states than in Odisha. In other countries, community- and clinic-based limb-care programs have demonstrated the efficacy of low-cost interventions in reducing the number of ADLA episodes and stabilizing or improving lymphedema stage. It is reasonable to conclude that those improvements in quality of life would also yield economic benefits. Since the largest component of the cost of lymphedema and ADLA is the loss of wages—and the largest benefit is regained productivity—it is likely that gains elsewhere would be greater than in Odisha because it has lower wages than in most other LF-affected areas.

**A public health approach: integration with other programs.** Every filariasis-endemic country has numerous other serious health problems competing for scarce resources, whether from government sources or community NGOs. While some aspects of elimination programs may require a vertical, or disease-

specific, approach, policymakers are finding that integration of control programs for multiple diseases can have logistical and economic advantages.

With morbidity management as well, there could be important advantages to integrated programs. Limb care in particular might be integrated across several diseases common in India and in other countries. India has the world's highest burden of Hansen's disease (leprosy),<sup>68</sup> also present in several other LF-endemic countries, which can necessitate lifelong limb care. There are an estimated 4 million people globally with podocoinosis, for whom limb treatment is similar to that for LF.<sup>69</sup> Diabetes, now common in affluent countries, is an increasing problem in low- and middle-income populations. Foot protection, wound care, and limb hygiene are all important for diabetes care as well. Providing education and support for people with limb-care needs can be carried out in the public sector or in NGO-run programs, whether at the health facility or community level, with the potential for substantial cost economies as well as social benefits. Integrated programs can help reduce the social isolation of disfiguring and debilitating diseases. The emphasis on rehabilitating people in traditionally marginalized groups, such as people with Hansen's disease and LF, and helping them maintain their work performance or return to participation in community life, carries an important message of inclusion.<sup>70</sup>

Programs to educate people in limb washing require access to clean water. Water, sanitation, and hygiene (WASH) programs are essential for limb care, as well as to reduce breeding grounds for species of mosquito vectors of LF that flourish in open sewers. Reduced costs for limb-care programs, as well as reduced disability for LF patients, are important externalities that should be included in estimations of the benefits of WASH programs.

**Limitations.** To model the economic impact over the lifetimes of those with lymphedema and ADLA, we have made conservative assumptions about labor markets, the impact of disability on productivity, length of working life, and other parameters. We assumed flexible labor markets that could absorb workers who are rehabilitated through lymphedema management programs without exerting downward pressure on wages. This is reasonable because, although LF morbidity is a serious problem in endemic areas, the number of people affected is still a small proportion of the available labor force. Increases in labor supply from reduced morbidity would have little effect on the labor market because the expected gains from lymphedema management programs extend over the working lives of cohorts, rather than acting as a shock to labor markets at a moment in time. Moreover, any increase in earnings in the wake of a lymphedema management program is likely to be spent in the local economy, which could stimulate job growth and offset any downward pressure on wages from increased labor supply. We have not included a local multiplier effect and therefore the economic benefit of the intervention over time is substantially underestimated.

While the present analysis shows substantial gains from a community-based lymphedema management program for the Khurda population—US\$26.1 million or US\$1,648 per person enrolled—we think that those figures underestimate the economic costs of untreated LF morbidity and the benefits of lymphedema management. Our baseline estimate for earnings loss due to chronic lymphedema and ADLA was below the range found in several other studies. We did not include lost

work time for youths until they reach the 20-year cohort, although young people in poor rural areas are generally employed, even below the age (15 years) included in government employment statistics. In addition, we chose only low-wage occupations in rural areas (omitting semi-skilled trades with higher wages) to set the daily wage rate in our modeling. Finally, we assumed that a successful lymphedema management program would freeze the age structure of lymphedema. Recent studies, however, find that lymphedema management leads to net regression of lymphedema stage as well as reduction in the number of ADLA episodes.<sup>12,15,17,23–35,56</sup>

This study underestimates the costs of LF morbidity and the benefits of lymphedema management in other ways. We have attempted to measure only the economic costs that fall directly on persons with chronic lymphedema and ADLA in a filariasis-endemic area. We exclude costs to others, including society as a whole or government. Subsidized care in government-run clinics, for example, is ultimately financed by the taxpayer. Reducing disease progression and disability reduces the need for subsidized care in the future, a benefit to taxpayers that is not included in our analysis.

We do not include any accounting of other externalities of chronic lymphedema and ADLA. For example, we do not include the lost work time of family caregivers for those disabled from ADLA and lymphedema, nor the impact on child nutrition and schooling, which would affect the child's future earnings. Since we have not measured these second-order costs of morbidity and benefits of lymphedema management programs, our calculations substantially understate the reduction in the economic cost of lymphedema and ADLA that a lymphedema management program would generate.

Chronic lymphedema and episodes of ADLA impose a substantial physical burden on the population of Khurda District, a filariasis-endemic area, and that disease burden increases with age. The economic burden of lymphedema and ADLA is also substantial. A low-cost program of lymphedema management based on limb washing and topical medication for infection can reduce the economic burden on poor populations affected by filariasis morbidity by 55%. The net benefit per person over the lifetime is more than 130 times the per-person cost of the program and equivalent to more than 1,250 days of earnings for the average person affected by filariasis.

Programs for MMDP are mandated by the twin pillars of the GPELF. Low-cost interventions have been shown to be effective in reducing the frequency of episodes of ADLA and slowing progression of lymphedema. This study demonstrates that the economic benefits of such interventions far exceed the costs and result in very significant benefits to filariasis-affected people and their communities.

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Authors' addresses: Eileen Stillwagon, Department of Economics, Gettysburg College, Gettysburg, PA, E-mail: [stillwagon@gettysburg.edu](mailto:stillwagon@gettysburg.edu). Larry Sawers, Department of Economics, American University, Washington, DC, E-mail: [lsawers@american.edu](mailto:lsawers@american.edu). Jonathan Rout, Church's Auxiliary for Social Action (CASA), Bhubaneswar, India, E-mail: [jroutcasa@gmail.com](mailto:jroutcasa@gmail.com). David Addiss, Children Without Worms, Task Force for Global Health, Decatur, GA, E-mail: [daddiss@taskforce.org](mailto:daddiss@taskforce.org). LeAnne Fox, Division of Parasitic Diseases and Malaria, Centers for Disease Control and Prevention, Atlanta, GA, E-mail: [lff4@cdc.gov](mailto:lff4@cdc.gov).

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## SUPPLEMENTAL INFORMATION

### DETAILED METHODS AND SOURCES

This supplement provides detailed information on the sources used to determine parameter values and the method of calculating the results for our modeling of the economic cost of lymphedema and acute dermatolymphangioadenitis (ADLA) in an area with endemic lymphatic filariasis (LF).

**Efficacy of lymphedema management.** Numerous studies have provided evidence of the efficacy of simple programs of limb care in stopping or reversing progression of lymphedema and reducing the number of episodes of ADLA. Many successful lymphedema management programs have been community-based educational campaigns. The World Health Organization explained the rationale for community-based efforts, saying “the social model views disability as a matter of an individual’s full integration into society and prescribes social action to make the environmental modifications necessary for the full participation of people with disabilities in all areas of social life.”<sup>1</sup>

In Egypt, 2 years after instruction in limb washing and application of antifungals and antibacterial ointments to small wounds, participants reported a 57% reduction in ADLA episodes per year.<sup>2</sup> In Nigeria, under community-based care, 90% fewer patients experienced two episodes of ADLA per month after 12 months of treatment.<sup>3</sup> In Burkina Faso, after 4½ months of a leg-hygiene program, the percentage of patients experiencing an ADLA episode in the previous month was reduced by half.<sup>4</sup> A community program of home-based care in Sri Lanka found that the number of persons experiencing one or more ADLA episodes decreased by 64% (Table 3)<sup>5</sup> and 66% perceived reduced lymphedema after 1 year.<sup>5</sup>

Most community-based lymphedema management programs for filariasis reported in the literature have been in India. In Kerala, 1-day health camps to teach leg washing and care of bacterial entry points were followed up after 6 months by questionnaires on disease symptoms (redness, swelling, odor, wound, and fever) and quality of life. Almost all participants (96%) reported reduced symptoms after following the hygiene regimen.<sup>6</sup> Also in Kerala, placebo-controlled trials examined the efficacy of different treatments for reducing ADLA that included diethylcarbamazine (DEC) and either ivermectin or penicillin. Participants in all arms were enrolled in a comprehensive foot-care program.<sup>7,8</sup> Improved foot care was found to be an effective treatment even for those who received no medication. A follow-up study in which visits were carried out a year or more later, with no additional interventions, found a 72.5% reduction in the frequency of ADLA, confirming the lasting efficacy of education efforts to reduce morbidity.<sup>9</sup>

In Tamil Nadu, in a trial of three drugs to prevent ADLA, all arms of the trial practiced limb hygiene. Even the control group, practicing only leg washing, had reduced incidence of ADLA in the treatment year and beyond.<sup>10</sup> In two LF-endemic districts in Kerala and Karnataka States, patients with grade II and III lymphedema (of grades I–IV) were trained in limb hygiene in LF camps and evaluated 3 months later. In Kerala, 25% fewer patients had episodes of ADLA, and in Karnataka, 73% fewer patients had ADLA episodes<sup>11</sup> (see also Ref. 12).

In addition to community-based educational programs, a few lymphedema management programs have been based in clinics. In Haiti, one such program emphasizing self-care, especially washing of the legs, was associated with a 69% reduction in ADLA episodes for all participants and reduction in leg volume for participants with stage 2 lymphedema.<sup>13</sup> In another study in Haiti, it was found that washing affected legs with soap was associated with a reduction of ADLA episodes from 1.1 to 0.4 per person-year, a 64% reduction, over the 12-month study.<sup>14</sup> In contrast, a clinic-based program in Togo led to a small, but statistically insignificant, decline in the number of ADLA episodes and a statistically significant increase in the number of patients whose lymphedema prevented them from washing or getting out of bed.<sup>15</sup>

A recent systematic review and meta-analysis concluded that the “available evidence strongly supports the effectiveness of hygiene-based lymphedema management in LF-endemic areas” for the prevention of ADLA.<sup>16</sup> Their meta-analysis found that participation in such programs was associated with decreased percentage of patients reporting at least one episode of ADLA (odds ratio [OR] = 0.29, 95% confidence interval [CI] = 0.12–0.47) and lower incidence of ADLA (OR = 0.32, 95% CI = 0.25–0.40).<sup>16</sup>

Some studies suggest that mass drug administration (MDA) can reduce lymphedema and frequency of ADLA in persons already infected,<sup>17–26</sup> but other studies do not.<sup>10,27–29</sup> Consequently, available evidence does not warrant incorporating in the model any effect of MDA on lymphedema and ADLA.

The results of the Khurda community-based limb-care program are in line with the results discussed above. We based our parameters—halting progression of lymphedema and one-third reduction in ADLA episodes—on the literature above and the outcomes of the Khurda interventions.<sup>30,31</sup>

**Clinical data.** The objective of the study is to assess the economic effect of a morbidity management and disability prevention program that changes the age distribution of lymphedema and ADLA in a population over time. We derive the age distribution of morbidity from a census of households conducted in 2005 in Khurda District, Odisha State, India, by 40 Indian nongovernmental organizations. The census recorded age, gender, lymphedema stage, and number of ADLA episodes in the previous year. The data we used were stripped of personal identifiers and were thus anonymous. Consequently, no ethical clearance was warranted. We used Microsoft Excel (Microsoft Corporation, Redmond, WA).

The census generated a list of 21,496 persons. We eliminated cases with no data and those with no lower-limb lymphedema, and 17,036 respondents remained in the dataset.

**Gender and age distribution of morbidity.** From the Khurda morbidity census data we generated the gender and age distribution of persons with lymphedema. As shown in Supplemental Table 1, there is only a small gender difference in the number of ADLA episodes, as others have found<sup>32,33</sup> (for contrary findings, see Refs. 34, 35).

As shown in Supplemental Table 2, there were only small differences in lymphedema stage between females and males. The “average stage” was slightly higher for females than males (2.14 versus 2.06). (Average stage is in quotation marks in the previous sentence since stage is an ordinal, not a cardinal, variable.) Men were more likely to be in stage 1 lymphedema than women, and women were more likely to be in stages 2 and 3 than men. There was more gender balance at higher stages.

SUPPLEMENTAL TABLE 1  
Number of ADLA episodes in previous year by gender in Khurda census, 2005

Gender	Number of respondents	Percentage of females and males having ADLA episodes in 1 year					Average number of ADLA episodes
		0 episode	1 episode	2 episodes	3 episodes	Total	
Female	8,746	16.8	68.8	8.1	6.2	100.0	1.04
Male	8,290	15.3	69.8	8.4	6.6	100.0	1.06
Total	17,036	16.1	69.3	8.2	6.4	100.0	1.05

ADLA = acute dermatolymphangioadenitis.

**Start date.** CASA scale-up of the lymphedema management program was carried out between mid-2007 and mid-2010, with the largest number of people enrolled in 2008–2009.<sup>36</sup> We took mid-2008 to mid-2009 as our start date and calculated real wages and out-of-pocket medical costs from that time.

**Age cohorts.** Age was reported as ending in 0 or 5 for 72% of respondents, rather than the approximately 20% that one would expect. Accordingly, we grouped people in 5-year cohorts: 18- to 22-year-olds belong to the 20-year cohort, 23- to 27-year-olds belong to the 25-year cohort, and so on.

**Age limits.** The Khurda dataset included persons aged 3 to 99 years. We omitted the 18 children from 3 to 7 years of age because of small numbers, uncertainty of the cause of their reported lymphedema and ADLA, and the expected effect of cleared infection from their having experienced multiple rounds of MDA at a very young age.<sup>18</sup> Ultrasonographic and histological evidence shows lymphatic vessel damage in infected children long before the typical age at which lymphedema may become evident, generally around the age of 20 years.<sup>37,38</sup> In India and other endemic countries, there are reported cases of children in filaria-endemic zones with lymphedema thought to be of filarial origin.<sup>39</sup> Among respondents in the dataset, 1.3% were under the age of 18 years.

We assumed that those in the 10- and 15-year cohorts would incur out-of-pocket costs for lymphedema and ADLA from the beginning of the period calculated. The Indian Bureau of Labour does not include children under 15 years in the labor force, and we do not include these two cohorts in the lost earnings calculations until they reach the 20-year cohort (minimum age of 18 years). There is no maximum age listed for labor force participation in the Indian government labor statistics, but we calculated only up to the 70-year cohort (with 72 years as the maximum age) because of the sharp decline in respondents older than 72 years, Indian life expectancy of 66 years (<http://data.worldbank.org/indicator/SP.DYN.LE00.IN>), and decreased labor force participation. We calculated economic cost for the 15,853 people aged 8–72 years.

**Women's labor force participation.** We assigned economic loss to the disability of patients whether or not they were in the paid labor force. Specifically, women whose only or primary occupation is to prepare food, care for children or the sick and elderly, and other domestic activities are necessary for the household and the community to function as economic entities. We assign the same number of work days lost to men and women and the same average wage to each work day lost by either gender.

**Out-of-pocket costs of medical treatment of ADLA per episode.** We calibrated our model based on a study in Khurda District in 2000–2001 by Babu and Nayak (Table 1)<sup>40</sup> who reported that ADLA patients who sought medical care paid ₹ (Indian rupees) 92.3 on average (arithmetic mean) per episode of ADLA, which was equal to US\$2.04. (For this amount and all other costs reported in rupees, we convert to U.S. dollars for the appropriate dates using the OANDA online currency converter at <http://www.oanda.com/currency/historical-rates/>).

Babu and Nayak's estimated cost of US\$2.04 per episode in Khurda is somewhat higher than reported in earlier studies in other regions of India. Nanda and Krishnamoorthy (p. 57)<sup>41</sup> reported per-episode treatment costs averaging US\$0.46. From the data given in Table 2 in the work of Ramaiah and others,<sup>42</sup> one can calculate the arithmetic mean of per-episode spending on ADLA, which is US\$0.61. Other studies provide estimates of annual spending on ADLA, not per-episode costs. Krishnamoorthy<sup>43</sup> reported spending of US\$0.69 to US\$3.00. In Tamil Nadu, Ramaiah and others (Table 2)<sup>44</sup> estimated the number of ADLA episodes and total spending by patients on ADLA in India. From those estimates, one can calculate the annual per-person (but not per-episode) cost of ADLA: US\$1.01 for men and US\$0.77 for women. We do not consider reported costs of treating ADLA in other countries.<sup>45–47</sup>

**Annual out-of-pocket costs of medical treatment of lymphedema patients.** We calibrated our model based on a study in Khurda District by Babu and others (p. 34)<sup>48</sup> who found that the average male lymphedema patient spent ₹ 576 annually and the average female spent ₹ 425, the weighted

SUPPLEMENTAL TABLE 2  
Stage of lymphedema by gender in Khurda census, 2005

Gender	Number of respondents	Percentage of females and males at each stage of lymphedema							Total	Average stage
		Stage of lymphedema								
		1	2	3	4	5	6	7		
Female	8,746	34.6	31.3	24.8	6.3	1.7	0.9	0.5	100.0	2.14
Male	8,290	43.4	25.7	19.6	7.2	1.8	1.5	0.9	100.0	2.06
Total	17,036	38.9	28.6	22.3	6.7	1.7	1.2	0.7	100.0	2.10

average of which was ₹ 478, or US\$10.96. The reported averages are geometric means, which are lower than arithmetic means. Even though the geometric mean for some purposes may offer a better sense of the central tendency of a highly skewed distribution, the appropriate mean for our modeling is the arithmetic mean since we estimated spending on medical services by all lymphedema patients. The estimate based on geometric mean thus understates the cost of treatment and generates a more conservative estimate of the benefits of the intervention.

Estimates of treatment costs for chronic lymphedema in other locales and at other times are lower. Nanda and Krishnamoorthy (p. 57)<sup>41</sup> reported per-visit treatment costs in south India averaging US\$0.56 but did not report annual number of visits. Ramaiah and others (p. 21)<sup>49</sup> found that the geometric mean of annual spending on medical care for lymphedema patients in south India in 1993–1994 was ₹ 76 or US\$2.42. Ramaiah and others (Table 2)<sup>44</sup> found annual treatment costs of chronic lymphedema (and hydrocele) to be US 2.25 and US\$1.88 for men and women, respectively, in India as a whole.

**Annual real increase in out-of-pocket treatment costs.** We assumed that the demand for medical services will grow as household income rises (demand is income elastic). Moreover, since labor is a chief input into medical care provision, the cost of medical services will also rise as real wages rise. We assumed that the rate of growth in real (adjusted for inflation) expenditure on medical services will be the same as the rate of growth in real wages.

**Average daily wage.** The wage is useful for estimating the impact on individuals and their families, as well as the impact of their reduced spending in the community. It would also be desirable to measure the lost productivity for society that morbidity and disability entail. For that purpose, the wage is an imperfect proxy since it may not accurately measure the contribution of workers to output, nor the loss of their contribution to society. We estimated the average daily wage of unskilled, mostly agricultural workers, who comprise almost all (98%)<sup>36</sup> of the people in our dataset and are also representative of the population across India affected by lymphedema and ADLA. We calculated the average daily wage in rural occupations in Odisha State during the crop year July 2008 through June 2009. To determine the average daily wage, we used data from *Wage Rates in Rural India 2008–2009* (Tables 3a–14b),<sup>50</sup> which gives average wages for men and women for every month reported separately for ploughing, sowing, weeding, transplanting, harvesting, winnowing, threshing, picking, herding, and unskilled nonagricultural labor. We took the unweighted (the number of workers in each occupation is not given) average of wages in those 10 occupations averaged over 12 months of the crop year. Finally, we averaged men's and women's wages, weighted by their relative presence in the Khurda census, which was 48.7% male.

**Annual increase in real daily wage.** Between 2006–2007 and 2011–2012, real GDP in India grew 7.9% annually and real rural wages grew 6.8% (p. 12)<sup>51</sup> (see also Refs. 52–54). Since 2003, real GDP growth exceeded 9% in 4 years and 7% in 8 years. The OECD projects 6.1% growth of real GDP in India in 2016, gradually trending downward to about 4.6% annually by 2040.<sup>55</sup> Between 2007 and 2011, Odisha had the most rapid growth in rural wages in India (p. 189).<sup>54</sup> Indian economists report the following forces pushing up rural wages: rapid growth in GDP, a shrinking share of agriculture

in GDP, growth of nonagricultural economic activity in rural areas, and rapid urbanization due to rapid growth in urban employment, causing a growing gap between urban and rural wages and fostering rural labor shortages, growing mechanization, and growing demand for skilled agricultural workers. Other factors include increasing integration of labor markets across states and national and state government policies that promote growth in rural wage rates, especially the Mahatma Gandhi National Rural Employment Guarantee Act, which has boosted rural wages since 2006 by guaranteeing employment of at least 100 days/year for low-income workers.<sup>51–54</sup> All of that suggests that rural wage growth could easily reach 4% annually over the coming decades. Accordingly, we set our baseline prediction of real rural wage growth at 4%. The recent rapid growth in real rural wages may be a harbinger of even more rapid growth in wages over the coming decades. Our sensitivity analysis, therefore, explores the possibility that real rural wages in India could grow as rapidly as 5% annually over the coming decades.

On the other hand, real farm wages in India grew at an average annual rate of only 2.9% from 1990 to 2012.<sup>51</sup> Furthermore, between 2000 and 2006, real farm wages fell by nearly 2% annually.<sup>51</sup> Despite rapid wage growth in 2006–2011, both Indian GDP growth and real rural wage growth slowed sharply in 2012.<sup>56</sup> Persistently weak economic performance in Europe and the United States is currently a drag on the Indian economy that could continue long into the future. All of this could suggest that the baseline assumption of 4% annual growth in real wages is too optimistic, so the sensitivity analysis shows the effect on the economic cost of lymphedema and ADLA if real rural wages continue the 2.9% pace set between 1990 and 2012.

**Days of work lost per year by people with chronic lymphedema.** Lymphedema can reduce one's ability to engage in productive work, and we measured that as days of work lost. Several studies record reduced hours of labor per day, reduced physical output, absenteeism, and coping by performing less strenuous and lower paid jobs.<sup>44,48,49,57–59</sup> We estimated a composite figure to represent all of the ways that morbidity reduces productivity and earnings. Lower productivity that reduces the wage rate is treated as a partial reduction in days worked and is subsumed into our measure of days of work lost. To determine the days lost, first we found the number of days a usually occupied rural laborer in Odisha could expect to work in a year. We relied on data for Odisha State from Table 4.1.1.1.1, Average Annual Number of Days Not Worked by Usually Occupied Men Belonging to All Classes of Rural Labour Households, from the Rural Labour Enquiry (61st Round of N.S.S.) 2004–05, Report on Employment & Unemployment of Rural Labour Households (Main Report), published by the Labour Bureau of the Government of India.<sup>60</sup> The report indicates that men in rural areas of Odisha State did not work an average of 76 days during the crop year 2005–2006 because no work was available or they were not available for work (since they were, for example, students, pensioners, or disabled). Consequently, usually occupied men in rural Odisha worked an average of 289 days per year. Many women are engaged fully or partially in unpaid work. As indicated above, we assigned women the same number of potential work days as men since their work in the household is essential to the household and community economy.

SUPPLEMENTAL TABLE 3  
Sensitivity testing: variations in wages, prices, and work time loss

	Assumptions subject to sensitivity testing				
	Annual increase in real wages and real out-of-pocket spending on medical services			Loss of work time due to lymphedema	
	Baseline 4.0%	2.9%	5.0%	Baseline* work time loss (11.2%)	Higher† work time loss (15.2%)
Economic cost without lymphedema management (in millions of US\$)	47.4	39.2	57.0	47.4	58.8
Economic cost with lymphedema management (in millions of US\$)	21.3	19.2	23.5	21.3	27.1
Reduction in economic cost with lymphedema management (in millions of US\$)	26.1	20.0	33.6	26.1	31.7
Reduction in economic cost per person with lymphedema management (in US\$)	1,648	1,263	2,119	1,648	1,998

\*Stages 1–2 = 0%; stage 3 = 20%; stage 4 = 50%; stages 5–7 = 100%.

†Stage 1 = 0%; stage 2 = 10%; stage 3 = 25%; stage 4 = 50%; stages 5–7 = 100%.

Although there are numerous estimates of lost work time due to chronic lymphedema, none reports the degree of disability by stage of lymphedema, measured either as hours or days of work (and thus lost wages) or reduced wage rates. For the baseline calculations, we assumed that lymphedema patients in stages 1 and 2 do not lose any work time due to symptoms of lymphedema. We assume that lymphedema patients in stages 5–7 cannot work, losing 289 days of productive activity annually because of chronic lymphedema. We further assumed that patients with stage 3 lymphedema experience a 20% reduction in work time and patients with stage 4 lymphedema experience a 50% reduction in work time as a composite estimate of reduced hours, reduced intensity of work, lower pay grade, and absenteeism. The Khurda census allows us to compute the number of respondents in each age cohort at each stage of lymphedema. We multiplied that number by the average daily wage and summed over all cohorts and all stages to determine the economic cost of chronic lymphedema from lost earnings.

Our assumptions about degree of disability at each stage of lymphedema were quite conservative. They produce an average of 32 lost work days annually for persons with chronic lymphedema in stages 1–7. That is much lower than the loss reported in the work of Ramaiah and others, who found that in India as a whole those with lymphedema or hydrocele lost 51 days of work annually (Table 3).<sup>44</sup> Several studies show that lost work days for men with lymphedema or hydrocele are about the same, so Ramaiah and others' estimate is an appropriate indicator of lost work time for lymphedema.<sup>48,49,57,58</sup>

Our assumptions about lost work time at different stages of lymphedema result in an 11.2% reduction in work time, which is substantially lower than other studies have found. Ramaiah and others (Table 4)<sup>49</sup> indicate that lymphedema patients worked 15.2% less than controls. Ramaiah and others (Tables 3 and 4)<sup>58</sup> reported that those with chronic lymphedema worked 13.7% less than controls in paid work and 13.0% less in unpaid domestic work. Babu and others (Table 3)<sup>48</sup> found males with lymphedema worked 15.4% less than controls and women worked 23.5% less. Babu and others (p. 714)<sup>57</sup> measured a 20.2% drop in earnings for weavers with lymphedema (8.0% from lower wages and 12.2% from fewer hours worked). Similarly, Ramu and others (p. 670)<sup>59</sup> found that male weavers with lymphedema produced 27.4% less cloth than those with no lymphedema. Our baseline assumption about productivity loss from lymphedema is thus lower than any other published estimate.

Accordingly, the sensitivity analysis determines the effect of assuming greater disability among those with lymphedema than in the baseline analysis, with work time loss as follows: stage 1 = 0%, stage 2 = 10%, stage 3 = 25%, stage 4 = 50%, stages 5–7 = 100%. These assumptions produce a 15.2% average work time loss for all persons in the Khurda census dataset (or 44 lost work days annually), which is just below the mean of what other studies have found.

**Average length of ADLA episodes/days of work lost from ADLA.** We calibrated our model based on the work of Babu and Nayak (p. 1104)<sup>40</sup> and Babu and others (Table 1)<sup>61</sup> who reported on a study in Khurda District that finds a mean duration of ADLA episodes of approximately 4 days (3.93 days). The length of ADLA episodes in adults has been reported in numerous other studies.<sup>34,35,42,44,58,62–68</sup> The range of reported average duration of an ADLA episode is 3–8.6 days with a mean duration of 4.86 days.

**Discount rate.** We used an annual discount rate of 3%, which is the standard rate used for analyzing health interventions.<sup>69</sup>

**Results of sensitivity analysis.** We addressed uncertainty about appropriate values for some of the parameters in our modeling with sensitivity analysis. We projected a 4% annual growth in real wages and a commensurate growth in out-of-pocket spending for medical services associated with chronic lymphedema and ADLA. As shown in Supplemental Table 3, reducing predicted growth in the real wage to 2.9% (the average rate between 1990 and 2012) reduced the per-person reduction in the economic cost of lymphedema and ADLA by 23%, from US\$1,648 to US\$1,263. In the last decade, rural real wages in India and in Odisha have grown far faster than 2.9%, suggesting that a 5% annual growth in real wages is within the realm of possibility. Modeling a 5% real wage growth raised the per-person payoff of implementing a lymphedema management program by 29%, to US\$2,119.

Our assumptions about the degree of disability caused by lymphedema at different stages produced a loss in work time of just over 11%. Other studies on the burdens of lymphedema have found a loss of work time between 13.0% and 23.5% with a mean of 15.6%<sup>48,49,58</sup> (see also Refs. 57,59). Our 11.2% worktime loss may be appropriate, given that our data were based on a census that sought to find every person in the target districts with lower-limb lymphedema. Studies that recruit participants who come to clinics as patients, for example, may overrepresent individuals at higher stages of lymphedema since those with early or mild lymphedema may not seek health services. In case our estimates were too

conservative, we reestimated our model to produce a 15.2% total loss in work time, just below the mean work time loss found in other research. Doing so raised the per-person reduction in economic burden of lymphedema and ADLA by 21%, from US\$1,648 to US\$1,998, at 4% rate of growth of real wages and costs and 3% discount rate. The comparison of results with baseline parameters and sensitivity analysis is shown in Supplemental Table 3.

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