

SOCIOECONOMIC IMPLICATIONS OF BURULI ULCER IN GHANA: A THREE-YEAR REVIEW

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Abstract. This study examines some of the socioeconomic cost of treating 102 cases of Buruli ulcer between 1994 and 1996 at the St. Martin's Catholic Hospital in Agroyesum in the Amansie West district of the Ashanti region of Ghana. Seventy percent of the cases were children (up to 15 years of age). There was no sex difference in the distribution of cases. Hospitalization was prolonged (average = 186 days in 1994, 103 days in 1995, and 102 days in 1996) with no significant age and sex differences. There were 10 limb amputations, 12 patients were left with contracture deformities, one patient lost sight in one eye, and two died of sepsis and tetanus. The average total treatment cost per patient was \$966.85 (62% indirect) in 1994, \$706.08 (75% indirect) in 1995, and \$658.74 (79% indirect) in 1996. With increasing number of cases, high treatment costs, and serious complications, urgent attention should be given to the disease in terms of control and research efforts aimed at early detection and treatment.

Buruli ulcer is the third most common mycobacterial disease of nonimmunocompromised humans after tuberculosis and leprosy. It is caused by *Mycobacterium ulcerans* and often leads to massive destruction of the skin, often followed by debilitating deformities.¹ The disease typically affects impoverished inhabitants, primarily children, of remote areas where medical services are unavailable or too expensive.² It is commonly found around swampy and riverine areas in the tropics. Buruli ulcer is associated with nonspecific clinical manifestations and has an indolent course, with a resultant delay by those affected in seeking care until there is massive skin necrosis requiring extensive surgery and prolonged hospitalization.

The mode of transmission of the disease is not known, and this makes primary prevention strategies currently not possible. However, early diagnosis can simplify the surgical management and reduce the likelihood of deformities or the need for amputations. The increasing number of cases in recent years in West Africa has raised serious health concerns. Cases have been reported in Benin,^{2,3} Ghana,⁴⁻⁶ Togo,¹ Cote d'Ivoire,^{7,8} and Liberia.⁹

The first suspected case of Buruli ulcer in Ghana was reported at the Korle-Bu Teaching Hospital in Accra in 1971.¹⁰ Since then, other cases have been described: 96 cases in Ashanti Akim north,⁴ 90 cases in the Amansie West district,⁵ and recently another 105 cases in the Amansie West district.⁶ Buruli ulcer has been known in endemic communities for years. In the Greater Accra region, there are local names for the disease such as Odontihela, describing the cotton wool appearance associated with the fatty necrosis, Aboa gbonyo (dreadful disease), and Ashanti Asane, meaning the disease might have originated from Ashanti region. In Ghana, the number of reported cases has been increasing since 1993, and these cases have come from five of the 10 regions of this country (Figure 1). The disease is found predominantly in the Ashanti region. The management of Buruli ulcer is difficult, treatment costs are high, and complications of the disease are frequently severe. Buruli ulcer has a very low mortality rate, but the associated morbidity and treatment costs could pose a serious challenge to a struggling rural economy and its health system given the growing number of cases and current control strategies. There are practically no data on the cost of the disease to individuals af-

ected and society at large. Lack of such information, in addition to the associated low mortality, could be responsible for the low priority given to the disease by health authorities. The purpose of this study was to estimate the short-term treatment costs of Buruli ulcer in the Amansie West district, which may be important to both patients and health planners in decision making.

MATERIALS AND METHODS

We present a three-year retrospective study of the socioeconomic cost of treating 102 cases of Buruli ulcer between 1994 and 1996 at St. Martin's Catholic Hospital in Agroyesum in the Amansie West district of Ghana.

Study setting. The Amansie West district is one of the 18 districts in the Ashanti region. It has the highest number of reported cases of Buruli ulcer in Ghana. The district is approximately 60 km southwest of Kumasi, the Ashanti regional capital. It is one of the least developed districts in the country. All the social infrastructures are poorly developed. Roads are poor and access to health services is difficult. The population of the district is about 130,000 people; the majority are poor subsistence farmers. Annual per capita income is about US\$200.00 and the daily income for a farm laborer is about US\$1.00. There are five health facilities, a mission hospital, and four government health centers. There is poor distribution of the health facilities relative to the communities endemic for this disease. Most of the endemic communities are found in the southern half of the district along the Offin River. Tontokrom is the worst affected community with estimated prevalence of 22%.⁵

St. Martin's Catholic Hospital in Agroyesum is a 100-bed hospital that serves as the referral district hospital for the four health centers. All hospitalized patients require attendant relatives to provide indirect care and they must provide their own food. Since 1993, the hospital has become the focus for the treatment of Buruli ulcer cases from within and outside the district. This reputation has gained national and international recognition. A U.S.-based foundation, the Humanitarian Aid Relief Team (Provo, UT), has since 1995 been visiting this hospital annually with a team of medical staff to help in the management of patients and also to train local doctors on plastic surgical techniques.

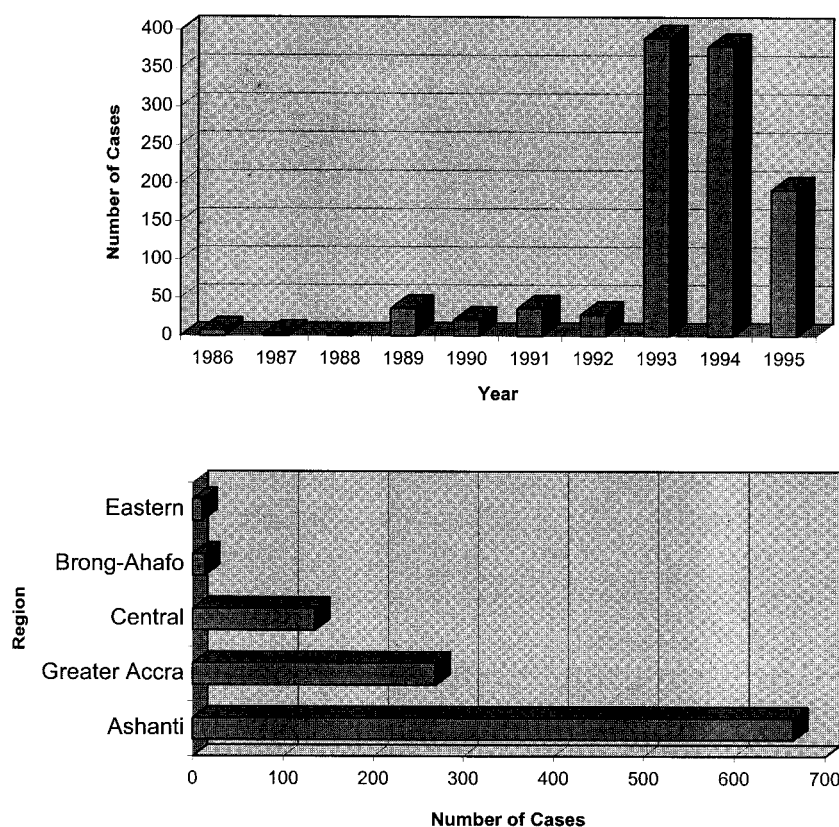


FIGURE 1. **Top**, number of reported cases of Buruli ulcer in Ghana (1986–1995). Source: Communicable Diseases 2 Returns and Special Survey Reports, Ministry of Health (Accra, Ghana). The 1995 figure is a projection of actual figures from January to April. **Bottom**, number of reported case of Buruli ulcer in Ghana by region (1986–1995).

Data. This was a retrospective study in which no personal identifiers were used. The study was reviewed and approved by the Regional Health Administration (Ashanti, Ghana). We initially identified 120 cases of Buruli ulcer through the hospital admission records between 1994 and 1996. There were complete medical records for 111 patients meeting the purpose of our study. These cases were then followed-up to the operating theater records and 109 cases were then identified. One hundred two of the 109 patients stayed in the hospital until their wounds healed. They were then discharged, which was our outcome of interest. The discharge remarks by the attending doctor in the patient's records were critical to the inclusion of a patient in the study. Of the seven patients who left before their wounds healed, three were referred to specialist care at the Komfo Anokye Teaching Hospital in Kumasi, and four requested discharge against medical advice. Additional data were extracted from medical records, pharmacy, laboratory, and medical stores. By these inclusion criteria, this number represents 85% of the total number of cases treated at St. Martin's Hospital within the three-year period. All cases were clinically diagnosed by the attending physicians with many years experience of diagnosing and treating Buruli ulcers. No microbiologic or histopathologic confirmation was possible given the limited resources at this hospital, but in a highly endemic community where most patients present late, clinical diagnosis is reliable.

The following assumptions were made. 1) All treatment costs were assumed to have been incurred in each year; thus,

this study was a prevalence-based rather than incidence-based.¹¹ 2) Charges instead of cost data were used in the estimation of direct cost. 3) It was assumed that all the costs incurred were related to the treatment of Buruli ulcer. 4) All costs were converted into the U.S. dollars based on the average exchange rate for that year. 5) Long-term economic loss due to disability and death was not included. 6) Children do not work. 7) Finally, we also assumed that the duration of hospitalization depended on the severity of the disease at the time of presentation and the available technology for treatment.

Estimation of direct cost. Inpatient. This is the inpatient charge per day multiplied by the number of days of hospitalization.

Surgery. This is the fee for a procedure multiplied by the number of times and the type of surgery.

Laboratory. This is based on the charge for each test and the number of tests performed during the period of hospitalization.

Dressing. This is the charge per daily dressing multiplied by the number of days of hospitalization.

Drugs. This is the total cost of drugs used by the patient during the period of hospitalization. It excludes drugs bought outside the hospital.

Miscellaneous. This includes consultation fees, stationery, and other charges levied on all patients.

Unmeasured cost. Because of the difficulty in measuring the staff time used on patients with Buruli ulcer, labor costs

were excluded. However, the time and energy spent on surgery (skin grafting) and wound dressing could be considerable and well acknowledged.

Estimation of indirect cost. *Loss of productivity.* This is the daily loss of productivity by the patient and the attending relative multiplied by number of days of hospitalization.

Feeding cost. This is the cost of feeding the patient and the attending relative per day multiplied by the period of hospitalization.

Miscellaneous. These were expenditures incurred outside the feeding cost and productivity losses. Miscellaneous cost was calculated based on 25% of the sum of feeding cost and productivity losses. This was to cover other costs such as firewood for cooking, kerosene for lanterns, drugs purchased outside the hospital, and laboratory tests done outside the hospital.

Unmeasured cost. We were unable to measure costs such as traveling to the hospital, previous costs of seeking care for the disease, productivity losses before and after hospitalization, and the cost of visiting the patient by friends and relatives.

We calculated the loss of productivity based on the average of two options as follows: Option 1: it was assumed that 30% of the patients who are adults work in addition to the attending relatives for seven days a week; Option 2: it was assumed that 30% of the patients who are adults work in addition to the attending relatives for five days a week.

RESULTS

Patient characteristics and duration of hospitalization.

Of the 102 patients with Buruli ulcer studied, 36 were treated in 1994, 34 in 1995, and 32 in 1996. There were 46 males (45%), 56 females (55%), 71 (70%) children (up to 15 years of age), and 31 (30%) adults. Figure 2 shows a typical Buruli ulcer. The average duration of hospitalization was 186 days in 1994, 103 days in 1995, and 102 days in 1996. There was no substantial difference by age or sex in the duration of hospitalization.

There were 10 amputations of limbs (Figure 3), 12 patients had contracture of a joint (elbow, wrist, and knee), one person lost sight in one eye, and there were two deaths due to tetanus and sepsis, respectively (complication rate = 24.5%). The average number of operations per patient was 1.45. Apart from the 10 amputations, 65 cases had wound excision(s) with a skin graft, 25 had only wound excisions, and two patients had bone involvement as a result of Buruli ulcer requiring wound excision and sequestrectomy. Forty-one patients were tested for human immunodeficiency virus (HIV) and three adults (one male and two females) tested positive. Forty-five (44%) patients received at least one pint of blood (average = 1.3 pints per person).

Seventy-four of the 102 cases (73%) came from our district, and 28 cases (27%) came from outside the district. The cases came from 51 communities, of which 30 (59%) were from the Amansie West district and 21 (41%) came from other districts. Tontokrom had the highest number of cases (14 of 102, 13.7%) in our district. Most of the cases came from Upper Denkyira district in the central region, which is adjacent to the Amansie West district. Denkyira Dominase

had the highest number of cases (4 of 102, 4%) in that district.

Treatment costs. The direct treatment cost shows a downward trend over the three-year period as shown in Table 1. The total direct treatment cost \$13,377.18 in 1994, \$6,000.23, in 1995, and \$4,468.15 in 1996. The average direct treatment cost was \$371.59 in 1994, \$176.48 in 1995, and \$139.63 in 1996. The total direct cost for the three years was \$23,845.56 and the average per patient cost was \$233.78. The distribution shows that drug cost was the single largest component of the direct cost, followed by dressings and inpatient costs (Figure 4a).

The total indirect cost in 1994 was estimated at \$21,429.59 and the average per patient was \$595.27. Similar estimates were \$18,006.53 and \$529.60 for 1995 and \$16,611.43 and \$519.11 for 1996. The total indirect cost for the three years was \$56,047.55 and the average per patient cost was \$549.49. The distribution of the indirect costs are shown in Figure 4b; feeding cost was the largest component followed by productivity losses and miscellaneous costs.

The total treatment costs were estimated to be \$34,806.77 in 1994, \$24,006.76 in 1995, and \$21,079.58 in 1996. The average total treatment cost per patient was \$966.85 in 1994, \$706.08 in 1995, and \$658.74 in 1996. The total treatment cost for the three years was estimated to be \$79,893.11 and the average cost per patient was \$783.27. The distribution of the total costs showed that indirect costs constituted 70% and direct costs constituted 30% of the total cost (Figure 4c).

The district health budget excluding salaries for the three-year period is also shown in Table 1. The budget was \$16,115.00 in 1994, \$15,271.00 in 1995, and \$27,449 in 1996. The total for the three-year period was \$58,835.00. The percentage of the total direct cost relative to the district budget was 83% in 1994, 39% in 1995, and 16% in 1996, and the overall total was 40%.

DISCUSSION

The age and sex distribution of the cases was consistent with other studies.^{4,5,8} Children were predominantly affected. Hospitalization was prolonged, reflecting the severity of cases at the time of presentation. There were no other concurrent illnesses that prolonged the period of hospitalization. The duration of hospitalization decreased considerably between 1994 and 1995, and we believe this was mainly due to the attention the disease received in 1992 and 1993. Other factors that might have contributed to the decrease were the incentives, such as food supplements and clothing, given to the patients. These may have led to early presentation of cases. Such incentives and the attention given to this disease have nearly vanished and this could explain the slight change between 1995 and 1996.

Surgery was the treatment of choice. All the patients were treated by wide excision with or without skin grafting. Sixty-five (64%) of the 102 patients had the excision and grafting done during the same surgical procedure; this resulted in an average number of surgical procedures of 1.45. Amputations were performed for extremely severe cases. Bone involvement in Buruli ulcer is uncommon but with the report of two cases, there is the need to consider Buruli ulcer-osteomy-



FIGURES 2 and 3. **2 (top)**, typical *Mycobacterium ulcerans* infections with massive destruction of the skin. **3 (bottom)**, amputation of the left upper arm as a result of infection with *Mycobacterium ulcerans*.

TABLE 1
Treatment costs and the district health budget (1994–1996); baseline figures used to estimate indirect cost*

	Year			Total
	1994	1995	1996	
Feeding cost per person per day	\$0.84	\$1.26	\$1.20	
Cost of hiring a farm labor per day	\$0.79	\$1.43	\$1.50	
Average exchange rate (USD1.00)	954.14†	1,192.49†	1,661.25‡	
Number of patients	36	34	32	102
Direct cost				
Total	\$13,377.18	\$6,000.23	\$4,468.15	\$23,845.46
Average	\$371.59	\$176.48	\$139.63	\$233.78
Indirect cost				
Total	\$21,429.59	\$18,006.53	\$16,611.43	\$56,047.55
Average	\$595.27	\$529.60	\$488.57	\$549.49
Total cost				
Total	\$34,806.77	\$24,006.76	\$21,079.58	\$79,893.11
Average	\$966.85	\$706.08	\$658.74	\$783.27
District health budgets§				
Total	\$16,115.00	\$15,271.00	\$27,271.00	\$58,657.00
% as a direct cost	83	39	16	41

* USD = U.S. dollars.

† Source: Bank of Ghana.

‡ Standard Chartered Bank, Cape Coast, Ghana.

§ Source: District Health Administration, Manso-Nkwanta.

elitis in the differential diagnosis of osteomyelitis in endemic communities. Increasing number of patients with Buruli ulcer–osteomyelitis could complicate the management of the disease. We believe that infection with HIV in three cases was just coincidental, given the fact that HIV infection is common in this district and other parts of the country. The clinical status of these three patients was good and their treatment outcomes were uneventful. Other studies have shown that HIV-infected persons are not at increased risk for Buruli ulcer,^{7,8} and HIV infection did not change the treatment outcome for this disease.¹² The rate of blood transfusions was high and this was the result of severity of the cases, with most of them requiring extensive surgery. Also, approximately 80% of the patients who visited the hospital had hemoglobin levels < 10 mg/dL (Annual Reports, St. Martin's Catholic Hospital, Agroyesum, Ghana, 1994–1996). Twenty-eight percent of the cases were from outside our district. This was due mainly to the reputation of the hospital in treating patients with Buruli ulcer. We also believe that the incentives provided to patients in 1992 and 1993 in this district also served to attract patients to our facility.

The treatment costs over the three-year period decreased. This reflected the decrease in the duration of hospitalization. There was a considerable decrease in treatment costs between 1994 and 1995 and this again reflected the substantial decrease in the duration of hospitalization over the same period. The change between 1995 and 1996 was slight, a reflection of the one-day decrease in the duration of hospitalization. We fear that the publicity, support, and incentives, which are almost nonexistent now, must have contributed to the slight change between 1995 and 1996. The minimum direct cost in 1996 was \$34.00 and the maximum direct cost was \$526.71. The minimum cost compared with to an average cost of \$139.63 in the

same year shows the potential savings that could be achieved if cases are detected and treated early. The costs of treatment relative to earnings in a community whose annual per capita income is about \$200.00 and the average income for a farm laborer (\$1.00 per day) indicate that only a few patients could afford treatment. The total treatment cost for an episode of malaria in the Amansie East and Kwaebibirem districts of Ghana was estimated to be \$8.67 (direct cost = \$1.80, indirect cost = \$6.87, productivity loss of 3.7 days for males and 4.7 days for females).¹³ The treatment cost for hydrocele, a complication of lymphatic filariasis, was estimated to be \$30.00 in the northern part of Ghana.¹⁴ Irrespective of methodologic differences, this comparison highlights the cost implications of Buruli ulcer. The average direct cost relative to wages implies that it would require working for 470 days in 1994, 123 days in 1995, and 93 days in 1996 to pay for the treatment cost assuming no other expenditures were incurred. The average total treatment cost was estimated to be \$966.85 for 1994, \$706.08 for 1995, and \$658.74 for 1996. The average total treatment cost compared with the Gross National Product per capita of \$410.00¹⁵ raises serious concerns of affordability for even the average Ghanaian. We suspect that the reported cases may be the tip of the iceberg for cost reasons.

Another factor that is responsible for the high indirect cost is the associated sick attendant relative phenomena. However, this is inevitable given our circumstances. The distribution of the total cost shows that indirect cost constituted 70% and the direct cost 30%. This demonstrates that even if the direct medical costs were free, the indirect cost formed the larger portion of the total costs. This shows the importance of indirect cost in the design of intervention programs especially for chronic diseases such Buruli ulcer, tuberculosis, and leprosy. Failure to do so may result in low com-

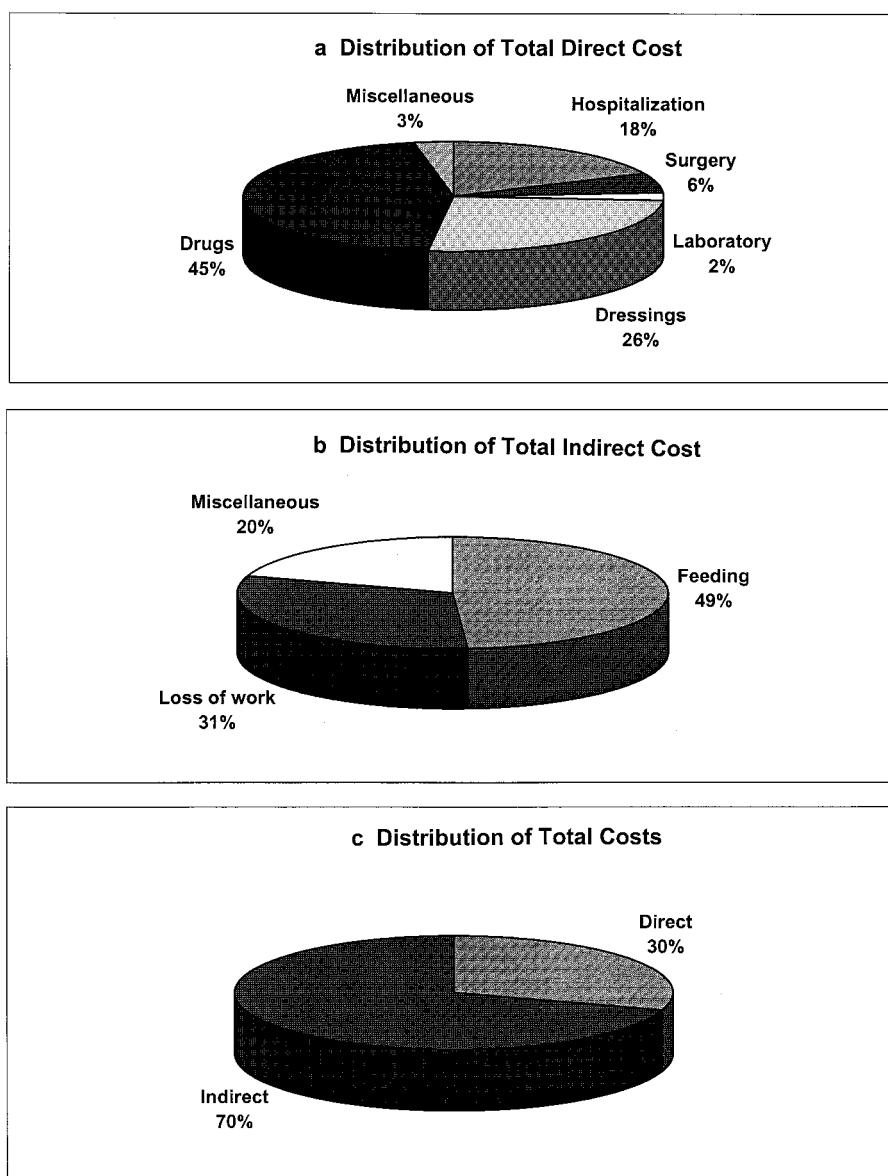


FIGURE 4. Distribution of **a**, the total direct cost, **b**, the total indirect cost, and **c**, total costs of Buruli ulcer over a three-year period (1994–1996) in Ghana.

pliance or treatment failures. It must be noted, however, that these estimates in our study should be interpreted with caution since cost estimates will vary from community to community and from hospital to hospital.

The distribution of the direct cost shows that most of the cost was incurred in drugs, dressing, and hospitalization. Drugs, especially antibiotics and dressing materials, are very expensive and patients paid full price for these. The charges for dressing might have been underestimated because of the majority of the patients required dressings at least twice a day. The low proportion of the laboratory cost reflects the low technology available at this hospital. The three heavy use areas are all a function of the duration of hospitalization. Despite the availability of the technology to treat Buruli ulcer since 1993, our results show that hospitalization is prolonged and complications are high. This implies that in hos-

pitals where this technology is not available and the only means of treatment is wound dressing, patients would be hospitalized much longer than those in our study for the same severity, and there would be more complications of the disease.

In poor remote areas, hospitals may not be able to recover cost of services to these patients because of their inability to pay. Also, the district health budget for the three-year period shows that even if the health services were to reimburse the hospital for the direct treatment costs, it would mean that 83% of the district's health budget in 1994 was used by 36 patients in a population of 130,000 people. Similarly, in 1995 and 1996, the treatment costs would account for 39% for 34 patients and 16% for 32 patients, respectively, of the district health budget. This could have a negative impact on the delivery of other health programs such as

immunizations, health education, and other disease control activities.

Several factors may be responsible for the late presentation of cases in the district. These include geographic access, lack of funds, attribution of the illness to superstition, stigmatization, and failure to make a diagnosis at the early stage.

In our health system, in which there is no social welfare program to take care of the disabled and the handicapped population, the burden of long-term care falls on the poor family members. Some of these patients who become handicapped will heavily depend on immediate family members to carry out activities of daily living. This may imply that in addition to the patient not being able to work, an immediate family member is equally handicapped.

The immediate and long-term impact of Buruli ulcer on children is two-fold. First, the prolonged morbidity and hospitalization could lead to serious disruption of school. Second, complications such as amputations and contracture deformities are frequent and children disabled by this disease will not be able to work (on farms). These children will grow into adulthood and may become a burden on society.

The disease has the potential of disrupting the family life of affected women. Studies on the social consequences of tuberculosis in Pakistan indicate that this disease may lead to stigmatization, social isolation, diminished marriage prospects, and divorce, particularly in women.¹⁶ Buruli ulcer could have similar repercussions.

The role of women in rural communities is enormous. They are involved in farming and other income-generating activities. These could be reduced considerably if they are left with a permanent disability. Such disability could potentially limit their ability to carry out activities such as trading, food preparation, obtaining water from streams or rivers, farming, and breast-feeding. The inability to perform these tasks could negatively affect their income-generating potential, and the health and welfare of their children.

Cost studies are not exact science and therefore tend to vary depending on the methodology, assumptions used, sources of data, the purpose and the perspective of the study, and whether charges or costs were used in the direct cost estimation. Inclusion of the feeding cost of both the patient and the relative, as well as productivity losses of the attending relative, are not in line with other cost studies,¹⁷ but we believe that these were important considerations in this study given the prolonged period of hospitalization and the social circumstances of the community where this study was conducted.

The results of this study show that most of the cases present late and therefore require prolonged periods in the hospital. Complications are devastating and treatment costs are high. Although cost-of-illness studies do not tell us anything about the efficacy, effectiveness, and cost-effectiveness of an intervention program, we have identified areas of heavy resource use (drugs, inpatient costs, and wound dressing). These areas need to be focused on in future research and treatment plans aimed at reducing the high treatment cost.

This study also shows that indirect treatment cost formed the larger portion of the total treatment cost, constituting as much as 70%. We have also shown that feeding cost is an

important consideration for hospitalized patients, especially when the period of hospitalization is prolonged.

We recommend that 1) Buruli ulcer should be given urgent attention in terms of research and control efforts aimed at early detection and treatment, 2) endemic countries should consider greater access to free treatment for those with this disease given the high cost, 3) Ministry of Health budgetary allocations to endemic districts should be increased, and 4) international health and development organizations should assist affected countries in containing the increasing number of cases and rehabilitating those already disabled by this disease.

Acknowledgments: We thank Professor F Portaels (Institute of Tropical Medicine, Antwerp, Belgium), Dr. T. S. van der Werf (Department of Pulmonary Medicine, University of Groningen, Groningen, The Netherlands), Dr. W. M. Meyers (Armed Forces Institute of Pathology, Washington, DC), Dr. Robert Horsburgh, Jr. (Schools of Public Health and Medicine, Emory University, Atlanta, GA), and Dr. Albert Berko (Voorhees College, Denmark, SC) for assistance and suggestions. We also thank Joe Forkuoh (Head of Medical Records, St. Martin's Catholic Hospital) and Joe Adomako (District Epidemiology Officer) for helping in the collection of data. Finally, we thank our typist, Andrew Barnie (alias Opponent), for assistance and the rest of the staff of the St. Martin's Catholic Hospital for support.

Financial support: We appreciate the financial support provided by individuals and institutions for Buruli ulcer patients at St. Martin's Catholic Hospital in 1994 and 1995. These included the EcoBank Ghana Limited, but there were others who preferred to remain anonymous. The Humanitarian Aid Relief Aid Team (Provo, UT) funded this paper.

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