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ORIGINAL ARTICLE

# Financial burden of health care for Buruli ulcer patients in Nigeria: the patients' perspective

**Joseph N. Chukwu[a], Anthony O. Meka[a], Charles C. Nwafor[a], Daniel C. Oshi[a,1], Nelson O. Madichie[a], Ngozi Ekeke[a], Moses C. Anyim[a], Alphonsus Chukwuka[a], Mbah Obinna[a], Julie Adegbesan[a], Martin Njoku[b], Festus O. Soyinka[c], Adebola O. Adelokiki[c], Isreal O. Enemuoh[d], Patrick I. Okolie[d], Joseph E. Edochie[d], Jonah B. Offor[e], Joseph Ushaka[e] and Kingsley N. Ukwaja[f,*]**

*[a]Medical Department, German Leprosy and TB Relief Association, Enugu State, Nigeria; [b]St Benedict's Tuberculosis & Leprosy Rehabilitation Hospital, Ogoja, Cross River State, Nigeria; [c]Ogun State Tuberculosis, Leprosy and Buruli Ulcer Control Programme, Ogun State, Nigeria; [d]Anambra State Tuberculosis, Leprosy and Buruli Ulcer Control Programme, Anambra State, Nigeria; [e]Cross River State Tuberculosis, Leprosy and Buruli Ulcer Control Programme, Cross River State, Nigeria; [f]Department of Medicine, Federal Teaching Hospital Abakaliki, Ebonyi State, Nigeria*

[1]Present address: Department of Community Health and Psychiatry, University of West Indies (UWI), Mona, Kingston 7, Jamaica.
*Corresponding author: Tel: +234 803 624 3196; E-mail: ukwajakingsley@yahoo.co.uk

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**Background:** The economic burden of Buruli ulcer for patients has not been well-documented. This study assessed the costs of Buruli ulcer care to patients from the onset of illness to diagnosis and to the end of treatment.

**Methods:** This was a cross-sectional cost of illness study conducted among patients with Buruli ulcer in four States in Nigeria between July and September 2015. A structured questionnaire was used to collect data on the patients' characteristics, household income and out-of-pocket costs of care.

**Results:** Of 92 patients surveyed, 54 (59%) were older than 15 years, 49 (53%) were males, and 86 (93%) resided in a rural area. The median (IQR) direct medical and non-medical cost per patient was US$124 (50–282) and US$3 (3–6); corresponding to 149% and 4% of the patients' median monthly household income, respectively. The overall direct costs per patient was US$135 (58–327), which corresponded to 162% of median monthly household income, with pre-diagnosis costs accounting for 94.8% of the total costs. The direct costs of Buruli ulcer care were catastrophic for 50% of all patients/households – the rates of catastrophic costs for Buruli ulcer care was 66% and 19% for patients belonging to the lowest and highest income quartiles, respectively.

**Conclusions:** Direct costs of Buruli ulcer diagnosis and treatment are catastrophic to a substantial proportion of patients and their families.

**Keywords:** Buruli ulcer, Cost of illness, Health economics, Illness costs, *Mycobacterium ulcerans*, Nigeria

## Introduction

Buruli ulcer (BU) is a neglected infectious disease caused by *Mycobacterium ulcerans*.[1] It is the third commonest mycobacterium after *M. tuberculosis* and *M. leprae*. BU disease manifests as skin and soft tissue infections and can lead to permanent disfigurement and disability.[1] Although the case fatality of BU is low, the disease is associated with substantial morbidity and disability.[1,2] BU has been reported in 33 countries predominantly in Africa and the Western Pacific.[1] Currently, 15 countries regularly report BU data to WHO with West and Central Africa reporting the majority of cases.[1,2] BU can affect people of all ages particularly children and individuals living in swampy, rural and remote settings.[2–4] *Mycobacterium ulcerans*, the causative agent, thrives at a temperature of 29–33 °C and requires low oxygen concentration (2.5%) to grow.[1–4] The exact mode of transmission of *M. ulcerans* is still unknown; however not wearing protective footwear, previous trauma and living in BU endemic settings has been identified as risk factors of acquiring the infection.[1–4] Two clinical forms of the disease have been

described: the active and inactive form. The inactive form is characterised by previous infection with depressed stellate scar with or without sequelae. The active form is an ongoing infection, which can occur as an ulcerative or non-ulcerative disease.[1–4] Treatment requires the use of antibiotic therapy with or without surgery and physiotherapy.[1–4]

The burden of BU notified from Nigeria is increasing and it is one of the few countries reporting increasing cases annually.[5] Whilst the endemicity of BU in Nigeria is increasingly being recognised, the burden of the disease to patients and their households in the country has not been adequately evaluated. Thus, while case finding strategies for BU in Nigeria are being scaled-up, most of the cases identified have been found with advanced lesions and ulcers.[6] If BU disease is notified early, the treatment is easier requiring mainly the use of combination antibiotics therapy (usually streptomycin and rifampicin).[1,2] The small lesion responds to this treatment reducing the need for additional surgical intervention and or physiotherapy. However, lesions that are diagnosed late—in the advanced stages—have extensive necrosis of the skin which requires wide surgical excision and skin grafting in addition to antibiotic therapy. Management of these lesions may sometimes involve amputation of affected limbs or require physiotherapy to correct contractures.[1,2] The treatment for BU disease is provided in selected hospitals in endemic regions in Nigeria which have specialised programmes funded by international development partners. These hospitals offer free medical treatment and financial support to patients with BU. These BU services include free in- and out-patient treatment, meals and complementary accommodation for in-patients and their caretakers during hospitalisation, funding for affected children for enrolment into school and the provision of a basic allowance (US$4) for other needs.

The cost of health-seeking for diseases is a key factor associated with poverty, particularly in developing countries. Illness diminishes health status, drains incomes and impoverishes households.[7] The burden of diseases, including BU, has been found to have two dimensions: economic and social.[7] Economic burden consists of the direct cost of illness which may include medical costs of treating a particular illness (such as cost of drugs, hospitalisation, laboratory tests, surgery) and non-medical costs (costs of transportation, feeding, amenities and others). Also, patients may incur indirect costs which includes the opportunity cost of time lost by the patient for seeking healthcare, and income loss due to the illness.[7] The economic costs of diseases are important for policy-makers in making decisions on the allocation of scarce resources. They are also important for the patients in making household decisions. The economic burden of BU has not been adequately studied.[7,8] Thus, while the costs of hospitalisation, surgery, laboratory tests, daily wound dressings, drugs and miscellaneous services provided by the health system and development partners to BU patients have been documented,[9–11] little has been studied regarding any additional financial costs incurred by patients and their household due to BU illness before diagnosis and even during treatment.[7–8,12,13] Costs incurred by patients for other free of charge treatment for diseases like TB have been found to be catastrophic to the patients and their households, discouraging such patients from initiating or continuing treatment.[14,15] The economic burden of BU illness for patients in Nigeria has not been documented. Also, no studies have estimated the rate of catastrophic out-of-pocket payments for patients receiving free-of charge BU treatment services. In this study, we sought to evaluate the financial burden of health care for patients with BU in a multisite survey. Specifically, we assessed the rate and type of health service use, direct costs of BU care, the rate of catastrophic payments for BU and evaluated associated factors.

## Materials and methods

### Study design

This was a cross-sectional cost of illness study from the patient's perspectives conducted among patients with BU between July and September 2015. The study aimed to assess the costs of BU care to patients from the onset of the BU illness to diagnosis to the end of treatment. For patients whose BU lesions were less than one year old, we collected all cost data due to the ulcer care-seeking from the onset of illness until the end of treatment; while patients who had lesions that have lasted over a year were asked to provide costs data in the last one year prior to diagnosis until they were evaluated and managed by the BU treatment programme. Direct (out-of-pocket) costs, defined as all medical costs (medication, laboratory tests, hospitalisation and others) and non-medical costs (transportation, food and others) incurred before diagnosis and during diagnosis and treatment of BU by patients and their households were considered. The BU direct cost of illness associated with care-seeking were broadly categorised as pre-diagnosis and diagnosis/treatment costs. These costs were further classified as medical costs (for drugs, laboratory tests, wound care, hospitalisation and others) and non-medical costs which included transportation and feeding costs.[15] Pre-diagnosis costs included all costs (for medications, drugs, wound care, hospitalisation, transportation, food, hospitalisation, tests and others) incurred by the patients during health care seeking for the ulcer lesion before a diagnosis of BU was made. Diagnosis/treatment costs included all costs (for medications, drugs, wound care, hospitalisation, transportation, food, hospitalisation, tests and others) incurred by the patients during BU diagnosis and treatment by the BU control programme. Patients' direct costs are considered catastrophic if they exceed 10% of their total annual household income.[14–17] Patients from households that earned less than the median monthly income reported during the study (US$83.3) were classified as poor households and those that earned the median reported income or more were classified as less-poor. The local cost in Nigerian currency (i.e., Nigeria Naira [N]) was converted to US$ equivalents using the average exchange rate during the study period.

### Study area

The study was carried out in four States (Cross River, Anambra, Imo and Ogun) in Southern Nigeria. The States belong to the tropical rain forest belts characterised by several rivers and swamps. In each of the selected State four local government areas (administrative districts) where case finding intervention for BU was ongoing were used as the study sites. Patients with BU diagnosed during the intervention were the study participants

and were invited to complete a survey on patients' perspectives regarding BU disease and its consequences to patients and their households.

### Study population and sampling

Patients with confirmed BU disease during the study period constituted the sampling frame. An initial 102 confirmed cases were diagnosed and commenced treatment at the time of the survey.[18,19] In addition, 10 persons who had completed BU treatment in one of the study communities, were conveniently sampled for pretesting of the questionnaire. We recruited all consenting laboratory-confirmed patients with BU during the study period.

### Data collection

The patients were recruited at the health facilities offering BU treatment services and a structured questionnaire was used to collect data on the patients' socio-demographic characteristics, household income and direct (out-of-pocket) costs associated with seeking BU-related treatment. Cost and household income data from paediatric patients and respondents 18 years old or lower was obtained from their parents or other accompanying adult relatives. The questionnaire captured other issues related to patient's perspectives on BU, like local illness concepts on the disease, stigma/discrimination and quality of BU services offered in the health facilities.

### Statistical and data analysis

Data was double-entered, cleaned and analysed using Epi Info 3.5.4 (CDC, Atlanta, GA, USA). The economic cost burden was estimated as a percentage of the cost of BU care (diagnosis and treatment) divided by the total monthly/annual household income. Continuous variables were reported as median (IQR) while categorical variables were reported as proportions (%). Categorical variables were compared using the $\chi^2$ test. Odds ratios and their 95% CIs were estimated using multivariable logistic regression analysis, with household catastrophic payments (i.e., costs >10% of annual household income) as an outcome variable. The likelihood ratio test was used to assess the association between explanatory variables and household catastrophic payments. We performed a stratified analysis to determine the occurrence of interaction and confounding between the outcome variable and explanatory variables. A multivariable logistic regression model was constructed using the fixed model fits. All p-values were bidirectional and significance was set at a p-value of <0.05.

## Results

### Socio-demographic characteristics

Overall, 92 laboratory-confirmed patients with BU completed the study. The socio-demographic characteristics are as shown in Table 1. In all, 54 (59%) were older than 15 years while 38 (41%) were children ≤15 years. The median age of the patients was 18.5 years (IQR 6–43.5; mean 26.6±18.6). There was an almost balanced sex distribution of the respondents: 49 (53%) were males and 43 (47%) were females. Among children (≤15 years), 23 (61%) were males and 15 (39%) were females; while among adults (>15 years) 28 (52%) were females and 26 (48%) were males. In addition, 85 (92%) of the respondents were either in primary school or had completed some levels of formal education, and 86 (93%) resided in a rural area. The majority of the patients were either farmers 25 (27%) or students 48 (52%); 85 (92%) were Christians, and 28 (30%) had regular and dependable income. All the patients either self-presented or presented to the various BU facilities following community

**Table 1.** Socio-demographic characteristics of the patients (n=92) according to household income groups

| Variables | All n (%) | Poor[a] n (%) | Less-poor[b] n (%) | $\chi^2$ (p-value) |
|---|---|---|---|---|
| Age groups (years) | | | | 0.60 (NS) |
| ≤15 | 38 (41) | 12 (58) | 16 (42) | |
| >15 | 54 (59) | 27 (50) | 27 (50) | |
| Gender | | | | 0.01 (NS) |
| Female | 43 (47) | 23 (54) | 20 (46) | |
| Male | 49 (53) | 26 (53) | 23 (47) | |
| Residence | | | | 1.02 (NS) |
| Rural | 86 (93) | 47 (55) | 39 (45) | |
| Urban | 6 (7) | 2 (33) | 4 (67) | |
| Education | | | | 3.70 (NS) |
| No formal education | 7 (8) | 2 (29) | 5 (71) | |
| Primary | 51 (55) | 31 (61) | 20 (39) | |
| Secondary | 26 (28) | 13 (50) | 13 (50) | |
| Tertiary | 8 (9) | 3 (38) | 5 (62) | |
| Occupation | | | | 8.10 (NS) |
| Civil servant | 6 (7) | 0 (0) | 6 (100) | |
| Farmer | 25 (27) | 15 (60) | 10 (40) | |
| Others | 7 (8) | 3 (43) | 4 (57) | |
| Student | 48 (52) | 28 (58) | 20 (42) | |
| Trader | 6 (6) | 3 (50) | 3 (50) | |
| Religion | | | | 1.10 (NS) |
| Christian | 85 (93) | 44 (52) | 41 (48) | |
| Islam | 3 (3) | 2 (67) | 1 (33) | |
| Traditional religion | 4 (4) | 3 (75) | 1 (25) | |
| Patient income | | | | 5.70 (NS) |
| Irregular | 28 (30) | 16 (57) | 12 (43) | |
| Not applicable | 44 (48) | 27 (61) | 17 (39) | |
| Regular and dependable | 20 (22) | 6 (30) | 14 (70) | |
| Ethnic group | | | | 0.09 (NS) |
| Igbo | 29 (32) | 16 (55) | 13 (45) | |
| Others | 35 (38) | 18 (51) | 17 (49) | |
| Yoruba | 28 (30) | 15 (54) | 13 (46) | |

NS: not significant (p>0.05).
[a] Poor household: monthly income <US$83.3.
[b] Less poor household: monthly income ≥US$83.3.

interventions notifying them of BU treatment services. Most of the patients 84 (91%) reported visiting several health providers before coming to the hospital closest to their community where BU was diagnosed. In addition, although BU services offered were completely free to patients, 81 (88%) admitted to spending some money in the hospital where BU was diagnosed. Also, following diagnosis, 70 (76%) admitted to receiving free BU treatment including reimbursements for feeding and transportation, 16 admitted receiving free BU treatment except for food and transportation costs, while 6 (7%) admitted receiving free treatment including reimbursements for food and transportation costs except costs for additional medicines.

Before BU diagnosis, the patients visited several places for healthcare (Table 2). The most commonly visited place was patent medicine vendors/dealers (75, 82%), followed by traditional medicine practitioners (66, 72%), and churches/prayer houses for faith-healing (31, 34%). Primary health centres, secondary-care public hospitals, or any hospital received less than 30% of the health-seeking visits before BU diagnosis (Table 2).

**Table 2.** Places visited for healthcare by Buruli ulcer patients before diagnosis (n=92)

| Place | n (%)[a] |
|---|---|
| Patent medicine dealer/vendor | 75 (82) |
| Traditional medicine practitioner | 66 (72) |
| Prayer house/faith-healing | 31 (34) |
| Primary health centre | 26 (28) |
| Public secondary-care hospital | 25 (27) |
| Private hospital | 17 (19) |
| Mission hospital | 10 (11) |

[a] Sum exceeds 100% because most patients visited multiple places.

## Direct costs of Buruli ulcer care

Table 3 shows the summary of the median direct costs incurred by patients before BU diagnosis and during diagnosis/treatment periods. The median (IQR) direct medical cost per patient was US $124 (50–282). The median non-medical direct cost per patient was US$3 (3–6). The median medical and non-medical direct costs corresponded to 149% and 4% of median monthly household income, respectively. Also, the median pre-diagnosis cost per patient was US$128 (50–319), and the median treatment cost per patient was US$6 (3–9). The median pre-diagnosis and diagnosis/treatment direct costs corresponded to 154% and 7% of median monthly household income, respectively. The overall sum of pre-diagnosis and diagnosis/treatment direct costs per patient was US$135 (58–327), which corresponded to 162% of median monthly household income (13.5% of annual income), with pre-diagnosis costs accounting for 94.8% of the total costs. Median direct costs incurred by individuals classified as poor were substantially lower than those who were classified as less-poor; persons classified as poor incurred total median direct costs of US$89 (33–161) compared with US$250 (84–622) incurred by persons classified as less-poor (p<0.001).

The median total direct costs incurred by persons classified as poor did not differ according to residence, occupation, educational status, religious affiliation, age or gender categories (for all p>0.05) (Table 4). Also, the median total direct costs of care for patients classified as less-poor did not differ according to residence, occupation, educational status, religious affiliation, age or gender categories (for all p>0.05).

## Burden and determinants of catastrophic costs for Buruli ulcer care

Table 5 shows the distribution of median direct costs and incidence of catastrophic payments for BU care according to income quartiles. The median total direct cost of care for patients in the four income quartiles varies. Patients belonging to the lowest income quartile spent a median US$100 for BU care compared with the median US$256 spent by those

**Table 3.** Median (IQR) patients' costs (US$) of Buruli ulcer care: costs summary

| Variables | All patients Median[a] (IQR) | Costs as % of household income | Poor[b] Median (IQR) | Less-poor[c] Median (IQR) | p-value |
|---|---|---|---|---|---|
| Medical costs | 124 (50–282) | 149 | 89 (22–144) | 222 (78–447) | <0.001 |
| Non-medical costs | 3 (3–6) | 4 | 3 (2–6) | 6 (3–6) | NS |
| Pre-diagnosis costs | 128 (50–319) | 154 | 83 (19–144) | 233 (83–589) | <0.001 |
| Diagnosis/treatment costs | 6 (3–9) | 7 | 3 (3–6) | 6 (3–13) | 0.030 |
| Total costs | 135 (58–327) | 162 | 89 (33–161) | 250 (84–622) | <0.001 |

NS: not significant (p>0.05).
[a] Median household income: US$83.3.
[b] Poor household: monthly income <US$83.3.
[c] Less poor household: monthly income ≥US$83.3.

**Table 4.** Median patients' costs (US$) of Buruli ulcer care according to demographic and economic profile

| Variables | Poor[a] Median (IQR) | p-value | Less-poor[b] Median (IQR) | p-value |
|---|---|---|---|---|
| Age groups (years) | | NS | | NS |
| ≤15 | 64 (19–144) | | 189 (96–309) | |
| >15 | 117 (42–283) | | 289 (84–756) | |
| Gender | | NS | | NS |
| Female | 106 (42–161) | | 214 (76–699) | |
| Male | 72 (20–167) | | 250 (173–506) | |
| Residence | | NS | | NS |
| Rural | 89 (33–161) | | 239 (81–622) | |
| Urban | 168 (17–319) | | 378 (262–1086) | |
| Education | | NS | | NS |
| No formal education | 150 (17–283) | | 123 (111–289) | |
| Primary | 94 (38–161) | | 309 (189–659) | |
| Secondary | 64 (19–111) | | 144 (70–307) | |
| Tertiary | 122 (117–1294) | | 261 (189–467) | |
| Occupation | | NS | | NS |
| Civil servant | 0 | | 276 (72–467) | |
| Farmer | 117 (44–283) | | 699 (288–833) | |
| Others | 17 (11–1294) | | 286 (146–420) | |
| Student | 65 (19–136) | | 181 (76–273) | |
| Trader | 111 (106–556) | | 250 (44–1180) | |
| Religion | | NS | | NS |
| Christian | 100 (36–176) | | 250 (84–506) | |
| Islam | 51 (17–86) | | 233 (233–233) | |
| Traditional religion | 44 (17–117) | | 1125 (1125–1125) | |

NS: not significant (p>0.05).
[a] Poor household: monthly income <US$83.3.
[b] Less poor household: monthly income ≥US$83.3.

**Table 5.** Distribution of direct costs and incidence of catastrophic costs[a] for Buruli ulcer care across income quartiles, Nigeria, 2015

| Indicator | Income quartile[b] | | | | |
|---|---|---|---|---|---|
| | Q1 | Q2 | Q3 | Q4 | All |
| Frequency | 32 | 17 | 27 | 16 | 92 |
| Mean direct costs of BU care (US$)[c] | 100 | 78 | 239 | 256 | 128 |
| Median annual household income (US$) | 667 | 1000 | 1333 | 4167 | 1000 |
| Direct costs share of household income (%) | 15 | 8 | 18 | 6 | 13 |
| Households with catastrophic costs n (%) | 21 (66) | 7 (41) | 15 (56) | 3 (19) | 46 (50) |

[a] Catastrophic costs: direct costs >10% of household income.
[b] Quartile 1 (Q1) is the poorest and Quartile 4 (Q4) is the wealthiest.
[c] Based on a currency exchange rate of 180 Nigeria Naira to US$1.

belonging to the highest income quartile. However, this direct costs for BU care was catastrophic to households of 66% of patients belonging to the lowest income quartile; while direct costs of care was catastrophic to households of 19% of patients belonging to the highest income quartile. Overall, for all patients surveyed, the direct costs of BU care were catastrophic to 50% for all patients and/or their households (Table 5).

Table 6 shows the proportion of patients who incurred catastrophic costs for BU care according to their demographic and clinical characteristics. In bivariate analysis, the rate of catastrophic payments for BU care did not differ according to residence, occupation, educational status or religion of the patients. Also, the rate of catastrophic payments for BU care did not differ according age or gender categories. After adjustments for potential confounders in multivariable logistic regression analysis, none of the factors evaluated was a significant determinant of catastrophic payment for BU care (adjusted p>0.05; Table 6).

## Discussion

In this study, we have shown that patients with BU visited several places during care-seeking before a diagnosis was made; the majority reportedly visited patent medicine vendors or dealers, traditional medicine practitioners' and churches or prayer houses for faith-healing. Also, we have shown the patients with BU incur substantial direct costs of US$135 which corresponded to 162% of median monthly household income, with pre-diagnosis costs accounting for 94.8% of the total direct costs. We have also demonstrated that an inverse relationship existed according to the patients income quartiles with patients belonging to the lowest income quartile who incurred the mean lowest costs for BU care having the highest rate of catastrophic payments for care and vice versa. We also showed that overall, the direct costs of BU care were catastrophic to the households of 50% of patients and their households. Furthermore, we demonstrate that socio-demographic characteristics did not alter the proportion of patients who incurred catastrophic payments for TB care.

A number of studies have evaluated costs of BU treatment.[7–11] These studies followed the health care perspective and assumed that costs incurred by patients may be negligible.[7,8] As a result, economic decisions on the delivery of BU treatment services mainly considered these costs. In this study, we found that median out-of-pocket payments for BU care were substantial (US$135). The median out-of-pocket costs reported in this study was higher than the median direct patient costs of US$71 reported among patients with BU in Cameroon, but lower than a mean direct cost of US$548.2 incurred by patients in Ghana.[12,13] The very high direct cost reported from Ghana may be because mean values were reported. Median values are preferable for cost burden research as they avoid outliers and, as such, give a more accurate estimation

**Table 6.** Determinants of catastrophic costs for Buruli ulcer care, Nigeria, 2015

| Variables | All n (%) | Catastrophic costs n (%) | Crude OR (95% CI) | Adjusted OR (95% CI) | Adjusted p-value |
|---|---|---|---|---|---|
| Age groups (years) | | | | | |
| ≤15 | 38 (41) | 17 (45) | 1 | 1 | |
| >15 | 54 (59) | 29 (54) | 1.4 (0.6–3.3) | 1.2 (0.3–4.4) | NS |
| Gender | | | | | |
| Female | 43 (47) | 20 (47) | 1 | 1 | |
| Male | 49 (53) | 26 (53) | 1.3 (0.6–3.0) | 1.5 (0.6–3.6) | NS |
| Residence | | | | | |
| Rural | 86 (94) | 43 (50) | 1 | 1 | |
| Urban | 6 (6) | 3 (50) | 1.0 (0.2–5.2) | 1.8 (0.3–12.0) | NS |
| Education | | | | | |
| No formal education | 7 (8) | 3 (43) | 1 | 1 | |
| Formal education | 85 (92) | 43 (51) | 1.4 (0.3–6.5) | 1.5 (2.8–8.2) | NS |
| Occupation | | | | | |
| Farmer | 25 (27) | 17 (68) | 1 | 1 | |
| Others | 19 (21) | 8 (42) | 2.9 (0.8–10) | 3.5 (0.9–13.9) | NS |
| Student | 48 (52) | 21 (44) | 2.7 (0.9–7.5) | 4.3 (0.9–20.0) | NS |
| Religion | | | | | |
| Christian | 85 (92) | 43 (51) | 1 | | |
| Others | 7 (8) | 3 (43) | 1.4 (0.3–6.5) | 2.6 (0.4–15.9) | NS |
| Patient income | | | | | |
| Poor | 49 (53) | 28 (57) | 1.9 (0.8–4.2) | 1.9 (0.8–4.7) | NS |
| Less poor | 43 (47) | 18 (42) | 1 | | |

NS: not significant (p>0.05).

of the economic burden than average values.[14,20] Furthermore, the median direct costs reported corresponded to 162% of median monthly household income (13.5% of annual income). This was similar to the study by Grietens et al., which showed that direct costs represented 8% of patients' and households' earnings.[13] However, it was lower than the economic burden of 45% of earnings reported from Ghana.[12] Also, we found that pre-diagnosis costs represented 94.8% of the total direct costs. This suggests that the most expenses are incurred by BU patients prior to diagnosis—these go towards paying informal providers—resulting in late presentation and advanced lesions. WHO has observed that these costs can be reduced in the case of BU by early detection and treatment (i.e., pre-ulcerative stages).[1,19] This suggests that there is a need to increase community education about BU in endemic settings in order to substantially lower inappropriate visits to informal providers and costs of care-seeking.

In this study, we have shown that an inverse relationship existed between the economic burden of BU illness and the patients' earnings—with patients belonging to the lowest income quartile who incurred the mean lowest costs for BU care having the highest rate of catastrophic payments for care and vice versa. Thus, although BU patients in the poorest group had the mean lowest directs costs, two-thirds incurred catastrophic payments compared to one-fifth observed for patients in the richest income group. This is consistent with previous observations that reporting costs data as averages and estimating economic impact using average incomes do not represent the poor because although the amount spent by the poor is low, this amount represented a higher proportion of their earnings.[15,21] Therefore, reports based on average income figures will most likely underestimate the economic burden of disease for impoverished BU patients.[21]

Furthermore, we have shown that direct costs of BU care were catastrophic to 50% of patients or their households surveyed. This suggests that despite receiving free BU treatment services, that the costs incurred by patients with BU along the care-seeking pathway is enough in more than half of the patients for their families to sacrifice other basic necessities, further impoverish them and make them adopt coping strategies. Although we did not evaluate for specific coping strategies adopted by patients or their families in this study, in Ghana and Cameroon, patients with BU have been found adopt coping strategies like 'selling assets', 'borrowing money', use of savings, making claims from social networks, reducing consumption of nonessentials and adopting social isolation measures.[12,13] Moreover, none of the socio-demographic factors evaluated was a significant predictor of incurring catastrophic payments for BU care. This suggests that all patients irrespective of their profile are prone to incurring catastrophic payments for BU care and calls for urgent strategies to reduce these costs. One key strategy beyond improved community awareness and education is the incorporation and training of

informal providers like traditional healers, faith healers and patent medicine vendors to urgently refer suspicious cases to appropriate facility for treatment.[6]

The study has a number of limitations. First, this was a hospital-based survey which utilised laboratory-confirmed patients with BU, thus may be prone to selection bias. However, we were able to interview all but 10 patients notified with BU during the period. We reported mainly on direct (out-of-pocket) costs of care; previous studies have shown that indirect costs from lost earnings and productivity as well as intangible costs are also substantial—in most cases more than twice the direct costs.[12,13] We focused on the direct costs because its effect specifically has been shown to encourage or discourage a family from initiating or continuing care.[15] Another crucial limitation is that we did not evaluate coping strategies, while many exist.[12]Future studies need to evaluate financial coping strategies that may lead to future impoverishment. Due to the long recall periods and the internal variation in BU costs due to stage of illness at presentation, varying places visited for care-seeking, socioeconomic status, medical complications, distance to the health facility, and other competing household needs,[12] we did not evaluate a breakdown of these costs according to these variations rather the paper emphasizes the importance of patient costs and costs burden in a free BU treatment programme. Finally, we did not evaluate the effect of co-morbidities on patients' costs of BU care. Co-morbid conditions like diabetes mellitus, HIV/AIDS, anaemia and others may have contributed to higher direct costs of BU care before diagnosis and during treatment of the patients. This needs to be investigated in further studies.

## Conclusions

In conclusion, patients and their households incur substantial expenditure for BU care corresponding to 162% of median monthly household income, with pre-diagnosis costs from informal provider visits accounting for about 95% of these costs. Also, we demonstrated an inverse relationship between patients' income and their burden of catastrophic payments for BU care, and showed that overall, the direct costs of BU care were catastrophic to 50% of patients or households. Improved community mobilisation and patients' education concerning BU, decentralisation and strengthening of rural BU services and further engagement of informal healthcare providers are strategies that could mitigate the tortuous inappropriate BU care-seeking, reduce diagnostic delays and the associated pre-diagnosis costs. Also, other interventions like financial protection specific for BU patients from the poorest households may be needed. Furthermore, future research should investigate the impact of these interventions on costs for patients with BU and their treatment outcomes within both the local context, and different BU endemic settings.

**Authors' contributions:** JNC, AOM, MCA, CCN, DCO, NOM, NE, JA and OM conceived the study; MCA, CN, and KNU designed the study protocol; all authors collected data, performed data entry and carried out the data analysis and interpretation; KNU, AOM, DCO, JNC, IE, CN and AHB, drafted the manuscript; All authors critically revised the manuscript for intellectual content. All authors read and approved the final manuscript. JNC and KNU are guarantors of the paper.

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