Factors Associated to Diagnosis Delay of Patients with Buruli Ulcer in Akonolinga District Hospital-Cameroon

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ABSTRACT

Introduction: Buruli Ulcer (BU) is the most common human mycobacterial disease worldwide after tuberculosis and leprosy. The early detection and immediate recourse to the treatment centre are decisive for the progression of the disease. Despite international and national efforts for early management, there are still BU patients with disabling lesions. It is within this context that our study aims to describe the factors associated with the diagnosis delay of BU patients in Akonolinga District Hospital (ADH).

Materials and methods: The cross-sectional, retrospective, mixed descriptive study was carried out from 2015-2019 among 291 BU cases and 6 informants admitted to ADH. Quantitative data from a survey sheet were analyzed using CSPRO 7.1, SPSS 21, SPAD, and STATA 13 software while content analysis of qualitative data collected using an interview guide was investigated manually.

Results: 63.6% of patients arrived late at the centre for diagnosis and the average age was 21 years. Associated factors were female sex (68.4%), the first resort to traditional practitioners (88.6%), ulcerated forms (80.8%), and disability (91.7%). Female patients were 3.64 (95% CI. 1.15-11.53; P-value = 0.028) times more likely to have a delayed diagnosis compared to their male counterparts. Patients that resort to traditional practitioners usually experience psychological problems as they suffer from a negative self-image and self-conscience of themselves when faced with social rejection, stigma, and a downfall period from a healthy status to a BU status.

Conclusion: Emphasis should be placed on raising awareness among household heads as well as strengthening collaboration with traditional practitioners to further reduce the stigma around the disease and delays in diagnosis.

Keywords: Akonolinga District Hospital, Associated Factors, Cameroon, Diagnostic Delay, Buruli Ulcer, Patients.

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Introduction

Buruli Ulcer (BU) is a skin Neglected Tropical Disease (NTD) caused by Mycobacterium ulcerans.[1] It is the third most frequent mycobacterial infection after tuberculosis and leprosy.[2] This disease has been reported in 33 countries, predominantly tropical and subtropical regions, but is mostly prevalent in sub-Saharan countries.[3] In 2016, almost 80% of all new BU cases were reported in West Africa, mainly Ivory Coast, Ghana, Benin, and Nigeria.[3] The epidemiologic pattern is defined by the presence of confined foci where BU is endemic.[4] The preventive and therapeutic means for reducing the impact of this disease are still very limited.[5] Recently published data suggest that antibiotic therapy with Rifampin and Streptomycin is effective in reducing and even eliminating BU lesions when initiated during the early phases of the disease, decreasing the extent of surgical intervention and drastically reducing recurrence rates.[6] If left untreated, the infection can then lead to more or less extensive ulcerations. These can cause a limitation of function through severe physical deformities, especially if the infection has spread to the bone. Early diagnosis and treatment are therefore crucial to minimise the adverse consequences of this disease.[7]

The skin infection it develops is a mysterious disease in the communities where it occurs, resulting in significant morbidity in children due to physical incapacity and disfigurement.[8] Children under 15 years of age are predominantly affected by the disease resulting to an emerging public health threat in many humid tropical rural areas. Foci of this mycobacterium are geographically circumscribed, almost always around an aquatic ecosystem, along low-flow rivers (ponds, swamps, artificial or natural lakes, swampy areas, irrigation systems, rice cultivation, etc.). Despite extensive studies to determine the mode of transmission, it is still not clear how humans contract the disease from the environment. Determining the mode of transmission remains a key research priority in order to design an effective public health intervention.

Faced with this pathology, in 1998, WHO set up the Global Initiative against BU at the Yamoussoukro Conference, in order to coordinate, control and research activities specific to this pathology. With the support of international and national control institutions, they recommend to the populations of endemic zones early detection and immediate recourse to treatment centres as soon as the first signs appear. However, some patients continue to attend treatment centres late due to stigmatization in their daily environment, leading to the use of various therapeutic routes. Consequently, their recovery requires long-term hospitalisation which requires medical and sometimes surgical treatment. According to the WHO[9], about 90% of patients in Africa present too late, with extensive lesions that cause severe disabilities. The late referral of patients to care centres and the predominance of African medicine in their therapeutic itinerary has been addressed by several authors in the case of observations related to several pathologies. Thus, traditional healers occupy a very important position in the community.

According to Kibadi[10], consultations at the care centre sometimes occur more than 6 months after the first symptoms appear. The main reasons cited are economic variables and accessibility to care as well as representations of the disease. Moreover, these detours from African medicine are causing complications and long-term hospitalization for populations of rural areas that are often deprived.

In Cameroon, BU occupied close to 6000 cases with only 30 % that benefit from medical treatment with antibiotics while the others remain in isolated regions and resort to traditional healing.[11] According to the WHO, service delivery is the primary function of any health system and entails the provision of "effective, safe, good quality care to those that need it with minimal waste"[12], and to address health care needs through promotion, prevention, treatment, and rehabilitation.

Despite the cumulative efforts that have led to better care for victims of BU in Cameroon and although it is regressing, it is noted that new endemic communities are constantly increasing year after year causing a handicap to development and an emerging threat to the population at the socio-demographic, socio-economic and psychosocial level in view of the consequences that the disease generates, thus constituting a thorny public health problem. For instance, the social discrimination, stigma, and rejection that affected patients experience demonstrate the difficulties and issues that they are confronted with in order to be given adequate care and help.

Having considered this, it appears that observations made in the current Cameroonian context link the disease to spirituality issues, superstition, and witchcraft in our environment by others. It is within this perspective that we conducted a study with the aim to describe the factors associated with diagnosis delay of patients with BU in the Akonolinga District Hospital.

Materials and Methods

We conducted a cross-sectional, retrospective, mixed descriptive study at the Akonolinga District Hospital. Briefly, it was carried out in two phases: A quantitative and a qualitative phase. The retrospective, quantitative descriptive study was conducted on 291 BU cases admitted to Akonolinga District Hospital from 2015-2019. The sampling technique was essentially based on an exhaustive non-probabilistic sampling method by convenience while the technique used to collect patient information was a documentary review of medical records with the aid of a survey sheet. The survey sheet was made up of the WHO BU clinical and treatment form of the new case which allowed to have a global view of the epidemiological profile of this case. Some minor modifications were made to this sheet so that it best matches our context and our objectives.

The cross-sectional, qualitative descriptive study was performed onsix hospitalized interviewers who agreed to participate. Data collected through a semi-structured interview guide was designed essentially for the socio-cultural factors and verbatim transcription was carried out word to word on Microsoft Word before the content analysis was performed manually.

Measures

Diagnosis delay

It is the time interval between the onset of symptoms and confirmation of the illness. This includes patient delay and health system delay. Diagnostic delay was categorized by considering an acceptable patient delay and health system delay of 4 weeks. This was done by considering the observations made by Capela *et al.*[13] where measures of median time delay related to clinical forms revealed that for non-ulcerated forms (nodule, edema, and plaque) the median time-delay was 32.5 days (IQR 30.0–67.5), while for ulcerated forms it was 60 days (IQR 20.0–120.0) (P-value =0.009). Furthermore, results from the study of Coutts *et al.*[14] outlined an overall median presentation delay of 30 days (IQR 14–60 days). Subsequently, for data analyses purposes, we recoded into two (02) modalities our levels of time delay by BU patients: Early delay (≤ 30 days) and Late delay (> 30 days).

Independent variable

The independent variables considered in this study have allowed to study factors associated with diagnosis delay. Socio-demographic and clinical variables were presented quantitatively while the socio-cultural variable was described qualitatively. Concerning socio-demographic variables, we have included age, gender, marital status, place of residence, distance from health areas, and profession. Age was spread into three categories (≤ 15; 16-30; >30 years old). Gender was measured as a dichotomous variable (male/female). Marital status was given as single, free union, married, divorced, and widowed. The place of residence was either a rural or urban area. Distance from health areas was subdivided into three distances according to the Akonolinga sanitary card (Short distance ≤ 7 Km; Medium distance 8-59 Km; Long distance ≥ 60 Km). Profession included (unemployment;….). With regards to clinical variables, patient consultation delay was considered dichotomous (Early consultation ≤ 20 days; Late consultation > 20 days), clinical forms were subdivided into two (Non-ulcerated forms (Papule; Nodule; Plaque; Oedema) and Ulcerated forms (Ulcer; Osteomyelitis)); Existence of disability (yes/no); length of hospitalization (short < 3months; medium 3 – 6 months; long > 6 months) and recourse to surgery (yes/no). Socio-cultural variables were regrouped into four main components namely Perception, understanding, and interpretation; Personal and social representation; Patient experience before and after diagnosis and Adaptation strategies. Perception, understanding, and interpretation took into account (the definition of BU, the cultural name given to the disease, and its significance). The personal and social representation described how the patient represented the disease and the attitudes developed by the surrounding and society towards them. The patient experience before and after diagnosis related to how the disease started, what the patient did, the different routes taken, and the feelings perceived after diagnosis. The adaptation strategies describe how the patients and their counterparts overcome the disease after experiencing negative life events.

Data Analysis

Quantitative Analysis

Binary logistic regression models were used to determine factors that were associated with diagnosis delay of BU patients. In this model, diagnosis delay was given by the dichotomous nature of the dependent variable, early or late diagnosis. In univariate analyses, percentages determining the sample's distribution for each variable were presented. With regards to bivariate analyses, probabilities associated with chi-square test results were used to indicate the statistical significance of the relationship between diagnosis delay and each independent variable. The clinical variables predict more their influence on diagnosis delay of BU patients, in contrast to the socio-demographic factors that had only age and profession as associated variables. We considered the threshold probability for a statistically significant association at 5%. Thus all independent variables with a P-value < 0.05 were considered to have a statistically significant association with diagnosis delay. To measure the strength of the association between each independent variable that was significantly associated to diagnosis delay, we conducted Cramer's v statistics (Cramer's v value ranging from 0 to 1; $v < 0.10$ = null intensity, $0.10 \leq v \leq 0.20$ = weak intensity; $0.20 \leq v \leq 0.30$ = average intensity; $v \geq 0.30$ = high intensity). We equally tested for the presence of interdependence relationships among all independent variables, using the factorial analysis of multiple correspondences to highlight individuals with a lower or greater susceptibility to experience an early or late diagnosis delay. To examine the association between diagnosis delay and each category of factors (socio-demographic and clinical), we conducted a series of binary logistic regression analyses.

Diagnosis delay was entered as the dependent variable while each component of the socio-demographic and clinical factors was entered as the independent variable. Early diagnosis was considered as the reference modality for the dependent variable (diagnosis delay) as respondents with the late diagnosis were the most represented.

We excluded from the multivariate analysis, the type of lesion and surgical intervention since they had an under-represented modality. All analyses were conducted using the statistical software CSPRO 7.1, SPSS 21, SPAD, and STATA 13.

Qualitative Analysis

The qualitative analysis is oriented around socio-cultural factors. Each of the six (6) interviews was unique as each of the participants agreed to participate. The analysis process started from the first interview and continued after each new data collection. The analysis was based on the transcribed verbatim and on the audio recording of the interviews. A thematic analysis was carried out from this database. Each interview was first analyzed individually. Meaningful words and expressions were identified and grouped into themes. The interviews were then analyzed a second time with this list of themes. Which made it possible to flesh it out. The common and most relevant themes were developed in the results section. The analysis also made it possible to draw up the profile of the actors of the community in relation to the subject matter.

Ethical Considerations

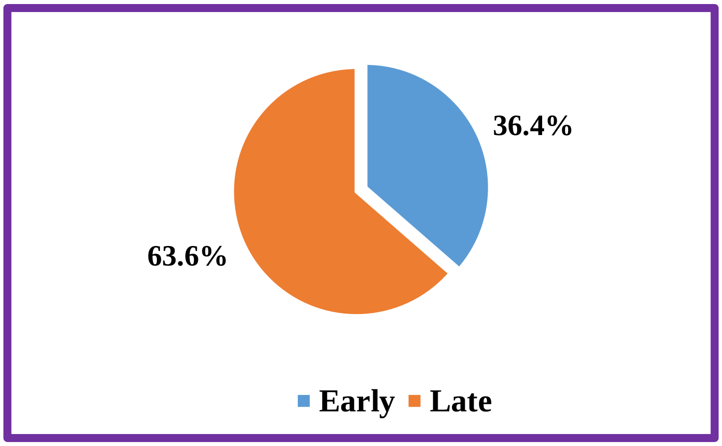
This study has received ethical approval (N°2020/020179/CEIRSH/ESS/MSP) from the Institutional Ethics Committee of Research for Human Health. It was approved institutionally by the Director of the Akonolinga Health District (N° 3350/AP/MINSANTE/SG/DRSPC/CRERSH).

Patients' agreement was obtained from those who signed an informed consent form before starting each interview and only the investigator had access to their data, in order to respect patients' confidentiality. Patients under the age of 21 have the parental consent form and the minor's assent form.

Results

Diagnosis delay

Globally, Graph 1 shows that the majority of patients with BU had a late diagnosis (63.6%, n=185). Only 36.4% had an early diagnosis delay.



Graph 1: Diagnosis delay of patients with Buruli Ulcer.

Socio-demographic characteristics of patients

Of the 291 patients included in our sample, 53.3% were males and 46.7% were females. The study sample was dominated by patients aged at most 15 years old (59.5%). About 38.5% were students while 25.4% reported being farmers/traders. 67.4% lived in rural areas, mainly within 7 km (36.8%) of the Akonolinga District Hospital.

Socio-demographic Characteristics	Frequencies (N=291)	Percentage (p= 100%)
Gender		
Male	155	53.3
Female	136	46.7
Age (years)		
≤15	173	59.5
16 – 30	76	26.1
>30	42	14.4
Marital status		
Single	199	68.4
Other status*	83	28.5
Place of residence		
Urban	95	32.6
Rural	196	67.4
Distance to HDA**		
Less than 7 km	107	36.8
8-59 km	96	33.0
60 km and more	88	30.2
Profession		
No profession	24	8.2
Farmers/traders	74	25.4
Housewife	37	12.7
Civil servants	15	5.2
Student	112	38.5
Other professions***	29	10.0

*__Other status:__ Married, free union, widowed, divorced, **__Distance of health areas form HDA (Akonolinga District Hospital):__ Less than 7 km (Akonolinga urbain), 8-59 km(Abem, Djoudjoua, Ebolakounou, Endom and Zalom) and 60 km and more (Akak, Edjom, Ekoudou, Emvasse-sso, Mengang and Mengueme-SSI) and ***Other professions**: Driver, Hairdresser, Shoemaker, Seamstress, Electrician, Bricklayer, Carpenter, Fisherman, Welder and Technician.

Table 1: Distribution of patients according to socio-demographic characteristics

Measure of diagnosis delay of BU according to the patients' socio-demographic characteristics

Table 2 below shows that only age (P-value = 0.020) and occupation (P-value = 0.000) were significantly associated with the delay in diagnosis of BU patients at 5%. In other words, young people aged at most 15 years (57.2%) were less likely to have a late diagnosis of BU, in contrast to their counterparts aged more than 30 years (69.0%) and especially 16 to 30 years (75.0%). The same is true for patients with a household occupation (21.6%), in contrast to farmers/traders (71.6%) and especially students (81.3%), who have a high susceptibility to late diagnosis.

Socio-demographic characteristics	Diagnosis delay of BU			P-value
	Early N (%)	Late N (%)	Total N (%)	
Gender				
Male	63 (40.6)	92 (59.4)	155 (100.0)	0.110
Female	43 (31.6)	93 (68.4)	136 (100.0)	
Age				
≤15	74 (42.8)	99 (57.2)	173 (100.0)	**0.020****
16 – 30	19 (25.0)	57 (75.0)	76 (100.0)	
>30	13 (31.0)	29 (69.0)	42 (100.0)	
Marital status				
Single	79 (39.7)	120 (60.3)	199 (100.0)	**0.088***
Other status*	27 (29.3)	65 (70.7)	92 (100.0)	
Place de résidence				
Urban	19 (20.9)	72 (79.1)	91 (100.0)	**0.055***
Rural	12 (26.1)	34 (73.9)	46 (100.0)	
Distance to the Health District				
Short	38 (35.5)	69 (64.5)	107 (100.0)	0.237
Medium	30 (31.3)	66 (68.8)	96 (100.0)	
Long	38 (43.2)	50 (56.8)	88 (100.0)	
Profession				
No profession	14 (58.3)	10 (41.7)	24 (100.0)	**0.000*****
Farmers/traders	21 (28.4)	53 (71.6)	74 (100.0)	
Housewife	29 (78.4)	8 (21.6)	37 (100.0)	
Civil servant	8 (53.3)	7 (46.7)	15 (100.0)	
Student	21 (18.8)	91 (81.3)	112 (100.0)	
Other professions	13 (44.8)	16 (55.2)	29 (100.0)	

***Other statuses:** Married/free union/widowed/divorced

***significant at 1%;** significant at 5 %;* significant at 10%.

Table 2: Association between diagnosis delay of BU and socio-demographic characteristics of patients.

Measure of diagnosis delay of BU according to the patients' clinical characteristics

According to table 3 below, late consultation delays are observed in 73.9% of patients, yet most of them present a severe state of health (55.3%), with lesions of the unifocal type (96.9%) and ulcerated forms (73.0%). The latter are at risk of prolonged hospitalization (32.6%) due to disability, of which they are victims (16.5%).

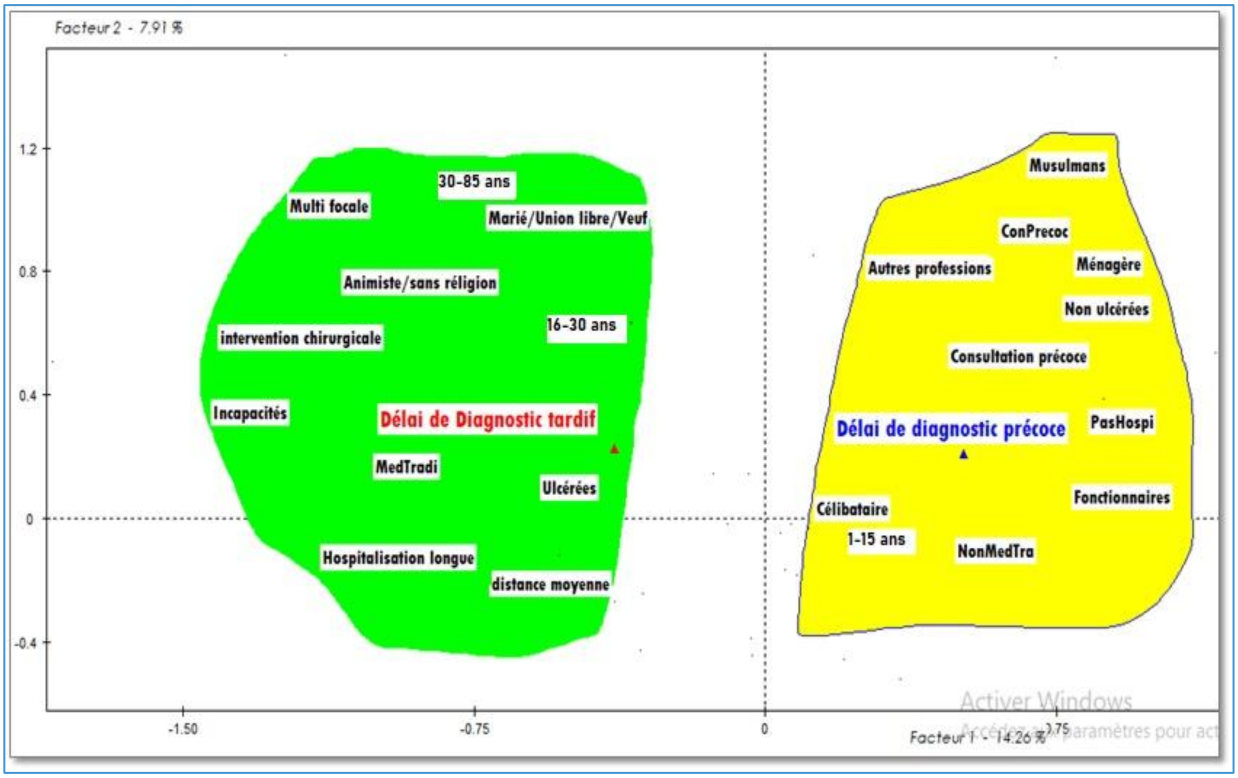
Clinical characteristics	Diagnosis delay of BU			P-value
	Early N (%)	Late N (%)	Total N (%)	
Consultation delay				
Early	72 (94.7)	4 (5.3)	76 (100.0)	**0.000*** **
Late	34 (15.8)	181 (84.2)	215 (100.0)	
Type of lesion				
Unifocal	104 (36.9)	178 (63.1)	282 (100.0)	0.368
Multifocal	2 (22.2)	7 (77.8)	9 (100.0)	
Clinical forms				
Non-ulcerated	65 (84.4)	12 (15.6)	77 (100.0)	**0.000*** **
Ulcerated	41 (19.2)	173 (80.8)	214 (100.0)	
Having a disability				
Yes	4 (8.3)	44 (91.7)	48 (100.0)	**0.000*** **
No	102 (42.0)	141 (58.0)	243 (100.0)	
Length of hospitalization				
None	95 (72.5)	69 (64.5)	107 (100.0)	**0.000*** **
Short/Medium	10 (15.4)	66 (68.8)	96 (100.0)	
Long	1 (1,1)	94 (98.9)	95 (100.0)	
Surgical intervention				
Yes	0 (0.0)	10 (100.0)	10 (100.0)	**0.015** **
No	106 (37.7)	175 (62.3)	281 (100.0)	

***significant at 1%; ** significant at 5 %; * significant at 10%.

Table 3: Association between diagnosis delay of BU and clinical characteristics of patients

Factorial analysis of patients with early or late diagnosis of BU

The analysis of the figure below highlights two major groups: those with a late diagnosis delay (**group I**), compared their counterparts with an early diagnosis delay (**group II**). We can clearly see that patients using traditional medicine usually experience a long diagnosis delay (**group I**) and are generally faced with complications such as ulcerated lesions and incapacities that usually lead to surgical intervention and long-term hospitalization. On the contrary, those that do not use traditional medicine tend to have an early diagnosis delay (**group II**) with non-ulcerated lesions and are generally not hospitalized.



Graph 2: Profil of patients with early or late diagnosis of BU in Akonolinga Health District.

Factors associated to diagnosis delay among BU patients

Results from the binomial logistic regression (**Table 4**) depict that among all studied variables, four were associated with diagnosis delay. These were gender, distance to the health district, use of traditional medicine, and consultation delay. We note that female patients were 3.64 (95% CI. 1.15-11.53; P-value = 0.028) times more likely to have a delayed diagnosis compared to their male counterparts. The same was true for those whose distance from their place of residence to the Akonolinga District Hospital was remote (RR=10.22; 95% CI. 1.47-20.12; P-value = 0.019) at the 5% threshold compared to those living nearby. Patients using traditional medicine were 4.43 times (RR=4.43; 95% CI. 0.94-10.77; P-value = 0.049) more likely to have a late diagnosis delay compared to their counterparts that did not use it at all. Finally, those with an early consultation delay for BU had a 95% (95% CI. 0.01-0.45; P-value = 0.000) lower risk of a late diagnosis compared to their counterparts with a late consultation.

Characteristics (variables)	Relative Risk	C.I (95%)	P - value
Gender			
Female	3.64	1.15 – 11.53	**0.028****
Male	(Réf.)		
Distance to the Health District			
Medium	4.67	0.85-25.8	0.077
Long	10.22	1.47-20.12	**0.019****

Short	(Réf.)		
Use of traditional medicine			
Yes	4.43	0.94 -10.77	**0.049****
No	(Réf.)		
Consultation delay			
Early	0.05	0.01 – 0.45	**0.000*****
Late	(Réf.)		

***significant at 1%; ** significant at 5 %; * significant at 10%.

Table 4: Binary logistic regression.

Socio-cultural characteristics of patients

Perception, understanding and interpretation

The interviews carried out revealed that the disease was considered in the community mystical and difficult to explain. Some define it as a superstitious curse while others described flies around the river as being the principal cause. "It's a mystical plague, or it's also invoked during the day or it's the fly or something that happened to you like that" (Informant 4). "Buruli Ulcer is caused by a fly and especially in swampy areas by people who live near swamps" (Informant 3). Furthermore, the cultural name given to the disease by all the participants was "Atom" but the interpretation differed. Some did not know what it meant while others described it as a negative signification caused by a troublesome person, by water, by the river, or by flies. "It was explained to us that you get it in two ways that are in the water when you cross it and mystically when you cross it. Locally, it means something has been thrown at you" (Informant 3). We notice here that the cultural name was given to the disease still relates it to a curse that is either transferred to someone or provoked by an individual.

Personal and social representation

Most patients were of the opinion that BU has a mystical and spiritual representation: "Here in the village, everyone knows it is something thrown at someone. It means that he has touched something that did not belong to him or he was mystically trapped without him knowing it by stepping on something on the ground" (Informant 6). Socially, all participants shared a positive interaction and support with regard to their family members: "My family has been very supportive...my family is always by my side" (Informant 3). While they expressed different opinions like stigma, odor, witchcraft, school and work rejection as negative attitudes from the society: "Despite the thoughts of others, it is an incurable wound, it is a way of doing witchcraft since the "Atom" smells a lot, it has an embarrassing smell. In a way that most people think you have been attacked or that you yourself know what is happening to you" (Informant 4). Globally, we can see here that the way people represent the disease is diverse leading to different interpretations that make patients adopt frustrated behaviors that highly affect their therapeutic route.

Patient experience before and after diagnosis

Most of the participants took different therapeutic routes as the first intention such as traditional healers, auto medication, and pastors, before reaching the hospital at a late stage: "When it started, I was doing the little dressings, I was putting Betadine... I went to the old mothers who knew the treatment... we had prohibitions on food intake... I left their treatment there...the disease was sucking my blood and I was losing weight...I had to come here" Informant 4). Others rather went to the hospital first before going to traditional healers before coming back after being discouraged: "I went to the hospital... I have prescribed the remedies, I drank... but, much later, it started, it was a little swollen and it hurt too much...sleeping there was no way... then I was told that there is someone who treats it... I went there and spent three months there....he told me that he does not treat this, he thought it was "biyè"...my elder sister took me to Nkongsamba to see a guy, who could not clearly find a solution, so we went to the hospital'' (Informant 2). After diagnosis, most of the patients felt sad, most had family charges, and others had financial difficulties due to unemployment and hospital stay: ''I was touched because given the state of the family when you notice that you are in such a poor family at first and that you were the one who was everything for your family, even for the children, I start to think a lot'' (Informant 3).

Adaptation strategies

Patients face different challenges and strategies to overcome the disease and accept it. These include strong personal mentality, children, other patient experiences, family and religious support: ''In the hospital, it was my daughter who got me through it...I was always crying, she always said we will always be next to you and we always like to see you like this...then she takes her phone, puts on the music and I see how she starts to dance with her brothers. When I see her dancing like that, I also try to forget'' (Informant 3).

For most of the participants, former patients they met at their arrival were of the opinion that psychological assistance, acceptation, counselling, and prayers helped them to overcome their situation: ''He tells you how the treatment can go, because there were steps he explained that first of all, it smells a lot, he advises you that the disease doesn't end as you think, you just have to endure, you also follow the treatment they tell you and you have to pay for all the medicines they ask you to pay for'' (Informant 4).

Discussion

Our study's main objective was to study the factors that are associated with the diagnosis delay of patients with BU in Akonolinga District Hospital. With regards to this, our results have shown that the delay in diagnosis of patients with BU was late (63.6%, n=185) than early (36.4%, n=106). Results from Agbenorku et al.[15] also corroborated with ours, as they reported that the majority of patients 76.7% presented a late diagnosis delay. Consequently, the longer a BU patient stays at home, the more difficult it is to treat him/her and the longer the duration when admitted, leading to an increase in the impoverishment of affected persons and their households. This clearly highlights the fact that a long time delay to diagnosis indicates the need for interventions that could help reduce it and alleviate the menace of BU sequelae and multiple surgical interventions in BU endemic areas.

Socio-demographic Factors

With regards to age, young people aged 15 years and below (57.2%) are less likely to have a late diagnosis of BU, in contrast to their counterparts aged more than 30 years (69.0%) and especially 16 to 30 years (75.0%). However, the age group below 15 is still seen to be more vulnerable to experiencing BU than the others as they constitute 59.45% of the total sample. These results are in line with those from Agbenorku et al.[16] in Ghana who found out that more than fifty percent of patients (56.90%) were children below 15 years. Quentin et al.[17] in Cameroon also highlighted the fact that the incidence rate was twice as high for children less than 15 as for adults. This can be explained by the fact that several factors such as different exposures of children along swampy playing grounds, behaviours, immunity, and treatment-seeking attitudes are likely to be expressed more in the younger age group when compared to the other age groups. Another reason could be the fact that children have a late delay because their parents take them late to the treatment centre since their health and wellbeing solely depend on the responsibility of their parents. In contrast, our study's findings do not corroborate with those from Georgia et al.[18] in Australia who showed that children ≤15 years of age were the least represented with 9.2%, followed by adults aged 16–64 years with 51.2% and lastly those aged ≥65 years with 39.6% of cases. Among the children, half were female and the median age was 8.0 years (interquartile range 4.8–12.3

years). This decline in cases among the young age group might have been mostly driven by improving living conditions, prophylactic recommendations, and access to health care. Another difference observed here with our findings could be the fact that we did a retrospective study compared to theirs that was prospective.

Globally, male patients were the most represented in our study with a proportion of 53.3% and the mean age was 21±21.11 years old. Results from our study showed that the female gender 93 (68.4%) were more likely to experience late diagnosis delay as compared to their male counterparts 92 (59.4%). All things being equal, female patients are 3.64 (95% CI. 1.15-11.53; P-value = 0.028) times more likely to have a delayed diagnosis compared to their male counterparts. The significance of these results demonstrates that the differences observed between both genders are due to behaviors and requirements that are specific to men on the one hand and women on the other hand. In fact, the available literature provides consistent evidence that women tend to be more affected by BU than their male counterparts with a majority of 66.7 % of cases in the cross-sectional study conducted by Ukwaja et al.[19] in Nigeria. Such findings could be attributed to the fact that women living within this cultural environment are still dependent and the management of their illness for most of them depends on their spouse's authorization. Most studies outlined the fact that risk categorisation related to gender is relevant for epidemiological studies and public health programmes such as prevention or case-detection campaigns.

Clinical Factors

Our results suggest that clinical forms are significantly associated with diagnosis delay (P-value = 0.000). Specifically, patients who had ulcerated forms of BU (80.8%) tend to have a longer delay in diagnosis than their opposite counterparts with non-ulcerated forms (15.6%). Similar associations have also been reported by Dégboé et al.[20] in Benin, Yeboah et al.[21] in Ghana, Agbenorku et al.[15] in Ghana, Meka et al.[22] in Nigeria, and Mondjo[23] in Gabon who found higher percentages of ulcerated forms that were respectively 69%, 69.2%, 76.7%, 82.1%, and 90%. These findings are far above the measure of early detection recommended by WHO that outlines that the proportion of ulcerative lesions at diagnosis reported from any district or country should be 60% or lower.[24] From this recommendation, there is evidence that a high percentage of ulcerative lesions plays a role in the occurrence of long-term hospitalization, hereby implying medical and surgical treatment. In effect, the more advanced and destructive ulcerated forms and osteomyelitis are associated with longer delay-periods, while non-ulcerated forms are more common in patients with recent infection, justifying the importance of early diagnosis and treatment for the disease.

According to our results, 91.7% of patients with a disability had the tendency of presenting a late diagnosis delay when compared to their opposite counterparts with no disability (58%). Our results corroborate those from Dégboé et al.[20] in Benin and Ukwaja et al.[19] in Nigeria who also registered higher cases of movement limitation at diagnosis greater than 50% that were respectively 61% and 82.9 %. These results were in contrast with those of Agbenorku et al.[16] who found that 33.6% of cases presented disabilities. Although this last result is far lower than ours, it still remains far above the WHO recommendation concerning the proportion of patients presenting with limitation movement at diagnosis from any district or country that should be below 15%.[24]

If applied in this context, the theory of social representation allows understanding of how a person may fall into a state of distress and confusion as a result of being chronically ill. As a result of their disabling condition, they will consider themselves responsible for the illness they suffer from (negative internal attributions) and will tend to stay isolated from the community they feel deprived of any special attention. They equally perceive having a serious illness as an event that will transpose onto other things they do (global causal attributions) and that the situation they find themselves in (being ill) is permanent. The combination of all these pessimistic thoughts will therefore result in hopelessness, as they would think that the situation is out of their control.

These high rate results show that BU is endemic but its severity is underestimated at least in the study setting and a need to identify and map BU endemic regions is necessary. Ahorlu et al.[25] on his part mentioned that with a little more investment in early case detection, diagnosis, and treatment, coupled with free transportation and breakfast for patients, most cases could be treated effectively with the available antibiotics to avoid disability and complications from the disease. For Simon et al.[26], the large and deep wounds caused by BU are often a major source of disability, and thus rehabilitation is an important component of care. To optimise results, rehabilitation should begin early and patients should be taught basic exercises appropriate to their condition. Research on how to improve the access and effectiveness of rehabilitation programmes could contribute to reduced disability.

Socio-cultural factors

With regards to the socio-cultural factors that could have a health impact on the way that BU patients perceive, understand, and interpret their disease, the different social representation and therapeutic itinerary they experience as well as how they overcome their situation with adaptation strategies will be discussed here convoking the theory of self-esteem.

⇒ Perception, understanding and interpretation

Overall the perception, understanding and interpretation of the disease are still expressed differently in the community. Most expressed it as being a superstitious treat while others defined it as being caused by flies around the river. As such, the cultural name given to the disease "Atom" was mostly interpreted as a superstitious treat that could be transferred from one person to another mystically as a result of a bad habit or troublesome people.

Linking such fact with the theory of self-esteem by Smith-Lovin's[27], it clearly appears that the individual's perception and interpretation of a situation is a reflexive emotion that has developed over time in social processes of the invention, that individuals learn to experience and to talk about, that arises in predictable social circumstances, and that is subject to social control. As such, it clearly appears that people's perception of the disease is a major hindrance to the use of modern therapies and in addition, the individual's consciousness of the pathology may be consistently influenced by collective representations.

⇒ Personal and social representation

The attitude of respondents with respect to their personal representation of the disease on one hand and that represented by the society on the other hand, seems to make them adopt a negative opinion of themselves. Concerning their personal opinion, they express more of a spiritual disease that can be transferred mystically, some have a psychological representation of the disease that weakens the moral and makes them doubt while others have a physical representation of it as they see it as a wound that does not heal for a long time after a motorbike accident.

From the social point of view, the attitudes are expressed differently. The family and relatives show a lot of sympathy, assistance, and support towards their affected ones while the society expresses negative opinions such as stigma and rejection as they interpret their situation as being a manner of practicing witchcraft while others feel embarrassed and prefer to run away from them due to the characteristic bad odor that is being expelled by the wound. The explanation behind our findings may lie with the theory of self-esteem that considers the individual's perception or subjective appraisal of one's own self-worth, one's feelings of self-respect and self-confidence, and the extent its positive or negative views about self is related to personal beliefs about skills, abilities and social relationships. As such, the individual self-esteem rises when he or she experiences another's love, making self-esteem dependent on not only one's perceptions of himself but also other's perceptions of him that appear to be important for self-regulation and quality of life, and for good personal and social adjustment. The presence of a parent or relative may act as a reassuring factor for their mental effect, preventing them from having pessimistic cognitions.

⇒ Patient experience before and after diagnosis

Most participants declared to relay first on different therapeutic routes such as traditional healers, auto medication, and pastors, before reaching the hospital at a late stage. This is probably due to the mystical nature around the uncured wound that leads them to several trials. Although it was also observed that some patients went to the hospital as a primary resort, they further consulted traditional healers due to the fact that they did not have a certain follow-up by some medical practitioners that rather interpreted the state of their disease as harmless at that period of time. At this stage, the patient left on his own, feeling the gravity of the disease as time passes begins to be in a state of confusion, distress, and uncertainty and will try to find out his own answers that could match his or her situation by relying on other therapeutic routes which significantly affect healing according to Nzilanye et al.[28] in Nigeria. For the author, late reporting, use of herbs, and access to health care further impeded wound healing, thus the necessity for the provision of accessible health care and education are warranted to improve early reporting.

In fact, the current African context highlights that in the event of illness, the choice of therapeutic routes is generally oriented by the individual's social status. Thus, the upper and middle-class individuals resort primarily to modern medicine while the lower class rather refers to alternative medicine. In this perspective, the usage of traditional medicine still finds its place. In fact, Yap Boum[29] and Ngo Nsoga et al.[30] outlined that collaboration between communities as well as between traditional and biomedical carers remains essential to improve BU management and reduce stigma. Furthermore, results from their findings demonstrate that keeping such collaboration would strongly influence therapeutic routes chosen by the community and therefore could contribute to reducing consultation time in a specialized treatment centre.

After diagnosis, most of the patients felt sad, most thought of their family charges, and others had financial difficulties due to unemployment and hospital stay. If transposed to the present study, the theory of self-esteem provides an explanation to the observation that the individual is faced with a negative self-image and self-conscience of himself with regard to the disease as he or she experiences a downfall period from a healthy status to a BU status, from one social or professional status to another and the change in tasks and responsibilities.

Consequently, they tend to have a self-protective approach to life, a shortage of positive resources in the self, and a chronic internal conflict of who they are, thus inducing psychological problems such as depression, social anxiety, loneliness, and alienation since they have less competency to overcome difficulties. Such a cognitive style explains why such people may reach a hopeless state, yet feeling their life was falling apart, and finally, fall into a depressive state. The patients in this study, especially those hospitalized, experienced more grief, and sadness in connection with the loss of an ideal life as they tend to express a series of transition stages such as negation (shock and denial of reality); anger at the situation; projection of the future and risk measures; reflection and self-reflection, as well as the acceptance expressed by the affective elements.

⇒ Adaptation strategies

The choice of adaptation strategies differs according to the patients interviewed. The relational interaction of patients with their peers such as family and loved ones, as well as other patients have been identified as highly beneficial as most patients used social support in order to learn, accept and overcome their hard times. Regarding interactions with family or loved ones, the elements of comfort and resilience strategies offered by the family or loved ones especially words of motivation and encouragement helped them gain sympathy, help from others, and listening and made them more open to information. Some optimistic patients in interviews tend to use strategies such as religious practices by carrying out through prayers. For them, it provided faith, comfort, and hope during this difficult time. While others appeared to be more passive in nature as they used their personal mental support to control their mind with positive emotions.

Concerning the experience of other patients, most were of the opinion that psychological assistance, acceptation, counselling, and prayers helped other patients to overcome their situations. They expressed the fact that over time and with the help of their family and the situation of other patients they met, they avoided thinking about their situation and changed their outlook on this issue. Thus, most ended up using more problem-focused strategies which also corresponds to the acceptance phase. Each individual goes through certain intermediate stages before reaching acceptance of the new situation in the sense of Kubler - Ross, and Kessler[31] in their theory of mourning. This aspect may help understand why in our context, the positive effect of self-esteem may express more resilience to the vicissitudes of life as individuals will have the ability to accept happy moments, handle unpleasant situations, cope effectively with challenges, engage in close relationships, and to improve their strengths. Thus, considered at the acceptance stage to positively moderate the expression of dysfunctional schemata and depressive symptoms at the experience of negative life events.

Conclusion

Buruli Ulcer remains a chronic debilitating infection that affects the skin and bones and can lead to permanent deformities and disabilities in Cameroon, Africa, and other countries worldwide. Our study has revealed that gender, age, the use of traditional medicine, the presence of ulcerated forms as well disability represent factors that are associated with the diagnosis delay of patients with Buruli Ulcer. Some of the main issues noted from our findings are negative self-image and self-conscience of patients as they face social rejection and downfall period from a healthy status to a BU status, psychological problems that induce less competency to overcome difficulties, and low self-esteem in terms of confusion or uncertainty in self-knowledge of the disease.

Consequently, such an observation calls for an immediate response implicating key actors at the political, family, social, academic, clinical, psychological, traditional as well as the economic level to ensure early case detection and a better quality of life through the expansion of specialized treatment centers in affected health districts. In conclusion, emphasis should be placed on raising awareness among household heads as well as strengthening collaboration with traditional practitioners to further reduce the stigma around the disease and delays in diagnosis.

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Contribution of authors

Edouardo S. Nana, Louise J. L. Ngo, and Yvonne O. M. Bassong, have designed the study. Edouardo S.

Nana took part in data collection and wrote the manuscript. Louise J. L. Ngo and Yvonne O. M. Bassong, supervised the work. Cedric F. Tchinda has made the critical reading of the manuscript. All authors have given their approval for publication.

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