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DEVELOPMENT OF A QUESTIONNAIRE ASSESSING BURULI ULCER–INDUCED FUNCTIONAL LIMITATION

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Abstract. Buruli ulcer, a disease with long-term consequences, is emerging in west Africa. Thus, a functional limitation scoring system is needed to assess its nature and severity. A list of daily activities was developed for this disease. Following treatment of Buruli ulcer, persons in Benin (n = 47) and Ghana (n = 41) were investigated. Nineteen items were identified with good internal consistency. Participants (median age = 14 years) could not perform 23% of their daily activities. Twenty-nine participants did not have any functional limitation. The average limitation score was 31% in Benin and 15% in Ghana ($P = 0.006$). The mean limitation score in participants without visible contractures (n = 65) was 13%, whereas patients with visible contractures (n = 20) or an amputation (n = 3) had a score of more than 50%. Validity and reliability should be further analyzed to optimize the scale for use in individual evaluation, as an end point in intervention trials, and in planning of resources needed for the care of patients with functional limitations.

INTRODUCTION

Buruli ulcer is caused by *Mycobacterium ulcerans* and is the third most common mycobacterial disease of humans in the world.[1] This disease has emerged dramatically in west Africa.[2] Prevalence rates in endemic districts in Ghana are reported to be up to 150 per 100,000 persons.[3] The mode of transmission of the disease is unclear, but aquatic insects, notably, *Naucorida* spp., may serve as a vector of *M. ulcerans*.[4] According to the clinical case definition of the World Health Organization (WHO), the pre-ulcerative stage includes nodules, plaques, or edema. In the ulcerative stage, skin ulcers with typically undermined edges can be clinically discriminated from other skin disorders. Later, a granulomatous healing response occurs, and fibrosis, scarring, calcification, and contractures with permanent disabilities may result.[5]

Clearly, Buruli ulcer is a devastating disease. The nodular stage of disease is treated by excision, which can be performed relatively easily. However, most patients do not come to the hospital until the ulcerative stage of disease. Admission of patients in Ghana and Benin takes an average of three months. Treatment is difficult and involves extensive surgery, which is the current standard treatment, but it may result in scarring and subsequent physical limitations due to scar retraction. These physical limitations may result in psychosocial and economic problems.[6–8]

We measured range of motion of joints of treated Buruli ulcer patients in Ghana and found that 58% of the patients had a reduction in the range of motion of one or more joints. We proposed that a simple and functional scoring system should be developed to assess nature and severity of the impairment in carrying out daily activities.[8] In this study, we describe the first steps in the development of such a scoring system.

METHODS

Study population. In 2002, patients treated for Buruli ulcer in Benin and Ghana were contacted based on records kept by hospital administration and community health workers. Data were collected from two different sites: the Center de Santé, Unité de Traitement des Ulcères de Buruli (Couffo region, Lalo, Benin) and the Agogo Presbyterian Hospital (Ashanti-Akim north district, Agogo, Ghana). In Benin, patients who received surgical treatment in the hospital and patients who received traditional treatment in their communities were included. In Ghana, only patients who received surgical treatment in the hospital were included. Individuals could participate in the study if their treatment for Buruli ulcer was completed at least three months earlier. A global functional limitation score was assigned for each subject by the first impression the observer (YS) had before the questionnaire was used. The study protocol was reviewed and approved by the Investigations Review Board of the Groningen University Hospital (Groningen, the Netherlands) and the local hospitals and district health authorities in Ghana and Benin.

Steps in scale development. Standard procedures for scale development were used.[9] First, a list of items was composed of daily activities that might be influenced by Buruli ulcer. The composition was based on a literature review,[8] clinical expertise, and on field observations of the research group. A similar approach is used in measuring functional limitations in the WHO manual on community-based rehabilitation. The list of items was adjusted based on discussions with health workers and interviews with Buruli ulcer patients in Benin. Items that were ambiguous were removed. The attempt to verify answers of the patients with the actual performance of the patients of the same items did not succeed. Performance of the items could not be carried out in the settings in the villages involved. An audience of villagers cannot be avoided, which leads to an uncomfortable situation for the Buruli patient when asked to perform activities.

For calculations of the individual functional limitation score, the number of answers 'with difficulties' and 'not possible at all' were divided by the number of activities applicable for that individual and converted into a percentage. Thus, a higher score indicates more functional limitations. The first study was then performed with 47 treated Buruli ulcer patients. Frequency endorsement was applied to the

results of this study; items answered affirmatively less than 10% or more than 90% of the time were removed from the initially constructed scaling list. Items that were not applicable for more than 10% of the respondents due to age, sex, or cultural beliefs of the activity were also removed. Internal consistency of this list was analyzed. The selected items were then used for a second study with 41 treated Buruli patients in Ghana. Some of the questions omitted from the scale in the first study were included for descriptive purposes only. Frequency endorsement, applicability, and internal consistency were again analyzed. Responses to each item were scored as 1) 'easily, on normal level' if the respondent could perform the activity without difficulties and on a level comparable to other community members of the same sex and age; 2) 'with difficulties' if respondent could perform the activity, but the level of performance was not the same as before Buruli ulcer started, or the level was not comparable to other community members of the same sex and age; and 3) 'not possible at all' if a respondent could not perform this activity (without help of others) because of Buruli ulcer, both if physically impossible and if not possible because respondent was avoiding the activity since he or she was afraid to damage the scar tissue. If the item was not valid for the respondent, e.g., the person was too young or too old to perform the activity, the item was scored as 'not applicable'.

Global impression on the functional limitations of the patients was categorized into no limitation, slightly limited, limited, severely limited, and very severely limited.

Descriptive study. In this part of the study, activities were asked that were not part of the scale due to the criteria of frequency endorsement and applicability, yet are important for daily life. These questions dealt with several aspects of taking care of children. Occupation before and after acquiring Buruli ulcer were also discussed. This part of the study started systematically in Ghana.

Statistical analyses. Internal consistency was analyzed by calculating Cronbach's α. An α value greater than 0.70 was considered sufficient for further use of the test. For statistical analysis, the Mann-Whitney U test and the chi-square test were used as appropriate.

RESULTS

Study subject characteristics. The median age of the participants was 14 years, with an interquartile range of 15 years. There was no difference in age between the participants in Ghana (median age = 13 years) and the participants in Benin (median age = 16 years; $P = 0.165$, by Mann-Whitney U test). Of the participants, 53% were female. Global impression on the functional limitations of the treated patients was the same in Ghana and Benin ($P = 0.150$, by Mann-Whitney U test). The locations of the lesions are shown in Table 1. The group of 88 treated Buruli ulcer patients had 95 Buruli ulcer lesions. Eighty-four lesions were present on the extremities, of which 48 were on the right extremity. This difference did not reach statistical significance ($P = 0.190$, by chi-square test).

Item and scale development. For the first study in Benin, 44 items were selected. After frequency endorsement, 19 activities were maintained in the set. These activities can be grouped into four dimensions: preparation of food/eating (four questions), clothing/personal care taking (three questions), working (five questions), and mobility (seven questions). The 19 items had an internal consistency α value of 0.91. In Ghana, the set of 19 items was administered to 41 treated Buruli ulcer patients. Of the 19 activities, six did not fulfill the frequency endorsement criteria. The set of 19 items had an internal consistency α value of 0.82 in Ghana.

TABLE 1
Locations of lesions in 88 treated Buruli ulcer patients in Ghana and Benin

	No.	Percent
Hand	3	3
Wrist	4	4
Forearm	4	4
Elbow	14	15
Upper arm	5	5
Shoulder	1	1
Entire arm	4	4
Trunk	8	8
Head/neck	3	3
Foot	9	9
Ankle	8	8
Lower leg	8	8
Knee	13	14
Upper leg/thigh	5	5
Entire leg	6	6
Total	95	100

When the results in Ghana and Benin (88 treated Buruli ulcer patients) were combined, the frequency endorsement criteria were met for all 19 items. The results of these items are shown in Table 2. The items had an overall internal consistency α value of 0.89.

Scale characteristics. Global impression of the functional limitations of the patient and their score were strongly correlated ($\rho = 0.837$, $P < 0.001$). On the scale, seven items can be categorized as upper extremity activities, six items as lower extremity activities, and six as both lower and upper extremity activities. Activities correlated more strongly with other activities of the same extremity than with the other activities. Results of the correlation matrix between items are shown in Appendix 1.

Results of the scale. Twenty-three percent of the activities (4.37 of 19 activities) on the scale could not be performed by the 88 treated Buruli ulcer patients (median = 16%). Only 29 (33%) patients did not have any functional limitation. There were no differences in scores between men and women ($P = 0.485$, by Mann-Whitney U test). The average limitation score was 31% in Benin and 15% in Ghana. This difference was statistically significant ($P = 0.006$, by Mann-Whitney U test).

Treated patients without visible contractures (n = 65) had an average limitation score of 13%, patients with visible contracture (n = 20) an average score of 50%, and patients with an amputation (n = 3) a score of 64% (Figure 1). Only one patient with an amputation had a prosthesis.

Descriptive study part. Of the 41 participants in Ghana, eight treated Buruli ulcer patients had children to take care of. Three patients could not provide food for their children by themselves due to their physical impairment caused by Buruli ulcer. Hoeing the field is an activity that was performed by 36 of the 41 treated patients. Fifteen had problems performing or could not perform this activity due to Buruli ulcer.

TABLE 2
Items of scale on functional activities

Type of activity	Activity	Extremity involved	Not applicable	Not at all/with difficulties	Easily, on normal level
Preparation of food/eating	Fetching water from pump	Lower and upper	3 (3%)	30 (34%)	55 (63%)
	Pound fufu (/manioc*)	Lower and upper	8 (9%)	32 (36%)	48 (55%)
	Pouring water from a bottle into a glass	Upper	3 (3%)	17 (19%)	68 (77%)
	Cutting vegetables with a knife	Upper	3 (3%)	8 (9%)	77 (88%)
Clothing/personal care taking	Putting on T-shirt	Upper	0 (0%)	9 (10%)	79 (90%)
	Wash yourself	Upper	1 (1%)	14 (16%)	73 (83%)
	Cleaning yourself after going to the toilet	Upper	2 (2%)	8 (9%)	78 (89%)
Working	Using a cutlass	Lower and upper	6 (7%)	30 (34%)	52 (59%)
	Heavy loads on head	Lower and upper	5 (6%)	32 (36%)	51 (43%)
	Carry harvest home	Lower and upper	5 (6%)	38 (43%)	45 (51%)
	Opening bottle with screw top (/corked bottle*)	Upper	3 (3%)	10 (11%)	75 (85%)
	Tie a knot	Upper	2 (2%)	11 (13%)	75 (85%)
Mobility	Walking level ground	Lower	0 (0%)	18 (20%)	70 (80%)
	Walking uphill	Lower	0 (0%)	25 (28%)	63 (72%)
	Walking downhill	Lower	0 (0%)	21 (24%)	67 (76%)
	Running	Lower	2 (2%)	33 (38%)	53 (60%)
	Squatting	Lower	0 (0%)	14 (16%)	74 (84%)
	Kneeling	Lower	0 (0%)	16 (18%)	72 (82%)
	Standing up from floor	Lower and upper	0 (0%)	9 (10%)	79 (90%)

* As asked in Benin.

Information about the occupation of the patient was acquired from 38 of the 41 treated patients in Ghana. Before Buruli ulcer started, 11 of the 38 participants were farmers. Four farmers had to change their occupation due to physical consequences of Buruli ulcer. They became hairdressers, a trader, or jobless. Other farmers who did not change their occupation did complain of less productivity, e.g., able to take care of only half an acre instead of an acre before Buruli ulcer started. Twenty-four of the 38 participants were going to school before Buruli ulcer started. Five participants stopped going to school due to Buruli ulcer. Embarrassment (n = 2), physical difficulties (n = 1), and financial problems caused by Buruli ulcer (n = 2) were the reasons for stopping. In addition to patients who stopped going to school, the children of Buruli ulcer patients also stopped going to school because the disease led to financial difficulties in paying school fees.

DISCUSSION

To assess the long-term consequences of Buruli ulcer, treated patients were asked about their performance of relevant daily activities. The list of activities was developed and adjusted based on studies in Benin and in Ghana. This resulted in a questionnaire with 19 items with good internal consistency.

FIGURE 1. A woman in Benin with an amputation due to Buruli ulcer using her seat to walk.

Limited number of patients in Ghana and the difference in the study populations between Ghana and Benin may have led to the fact that some items did not meet criteria of frequency endorsement among the 41 treated patients in Ghana. In the overall group, all frequency endorsement criteria were met. We consider the correlation between the global impression and the functional limitations score only as a confirmation of globally assessing the same construct. If the correlation would have been low, the scale should have been reconsidered.

The average functional limitation score was 23%. During the study in Ghana, we did not include patients who had only traditional treatment. Without having consulted a doctor in a hospital or a health center, the diagnosis for these patients cannot be confirmed, especially in the current healed stage of the disease. A selection bias may have occurred since patients tend to go to the hospital in a late stage of the disease.[7,10] The number of limitations due to Buruli ulcer disease may be somewhat less in general than in our study population. Patients recruited from hospital records are those with larger and more extensive ulcerations and are more likely to seek treatment of the disease. In our study population, only two patients had had treatment in the early stage of the disease with a nodule. Patients with less extensive ulcers who did not visit the hospital/health center could not be included in the study because of uncertainty regarding the diagnosis. We believe this estimate of limitations due to Buruli ulcer is more accurate than if persons with non-confirmed Buruli ulcer were included.

Functional limitation scores were higher in Benin than in Ghana. Many factors may have influenced this. However, the current scale can detect differences within patient populations. The patients seemed to be a very homogenous population concerning their daily activities. In addition, all participants lived in the rural areas of their country.

Patients with contractures or amputations had higher limitation scores. New treatment strategies and interventions should be developed to prevent contractures and amputations or to rehabilitate the patients. The disease and its consequence are of long-term importance, resulting in children who stop going to school and farmers with lowered productivity due to Buruli ulcer. Economic studies should be performed to describe the economic affects into detail, especially also on a household level.[11]

Thus, the scale we propose here should be further evaluated with a larger number of patients so that validity and reliability can be analyzed. After this analysis, the scale should be used for individual evaluation, as an end point in intervention trials, and hopefully as a guide in the planning of resources needed for the care of patients with functional limitations. Community-based rehabilitation programs should be developed for Buruli ulcer patients that take into account the stigma of the disease.[12,13]

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APPENDIX 1
Correlation matrix of activities

Area	Activity	Upper extremity						
		Opening bottle with screw top	Cutting vegetables with a knife	Wash yourself	Cleaning yourself after going to the toilet	Putting on T-shirt	Tie a knot	Puring water from a bottle into a glass
Upper extremity	Cutting vegetables with a knife	0.757						
	Wash yourself	0.527	0.400					
	Cleaning yourself after going to the toilet	0.508	0.448	0.618				
	Putting on T-shirt	0.586	0.675	0.570	0.544			
	Tie a knot	0.621	0.716	0.680	0.716	0.893		
	Pouring water from a bottle into a glass	0.456	0.543	0.412	0.342	0.401	0.508	
Lower extremity	Running	–0.056	0.001	0.179	0.163	0.121	0.131	0.102
	Walking level ground	–0.176	–0.161	0.100	0.042	–0.171	–0.103	–0.166
	Walking uphill	–0.222	–0.196	0.010	–0.021	–0.213	–0.161	–0.172
	Walking downhill	–0.196	–0.179	0.058	0.013	–0.189	–0.128	–0.127
	Squatting	–0.148	–0.137	0.167	0.088	–0.147	–0.064	–0.034
	Kneeling	–0.064	–0.044	0.214	0.169	–0.062	0.007	0.016
Both extremities involved	Fetching water from pump	0.342	0.276	0.535	0.352	0.386	0.375	0.185
	Pound fufu	0.474	0.407	0.514	0.408	0.436	0.489	0.434
	Using a cutlass	0.489	0.432	0.530	0.348	0.462	0.518	0.454
	Carry harvest home	0.261	0.280	0.361	0.355	0.380	0.425	0.262
	Heave loads on head	0.401	0.411	0.503	0.412	0.440	0.493	0.470
	Standing up from floor	0.007	0.034	0.294	0.311	0.010	0.117	0.040

		Lower extremity						Both extremities involved				
		Running	Walking level ground	Walking uphill	Walking downhill	Squatting	Kneeling	Fetching water from pump	Pound fufu	Using a cutlass	Carry harvest home	Heave loads on head
Lower extremity	Walking level ground	0.569										
	Walking uphill	0.629	0.743									
	Walking downhill	0.641	0.774	0.889								
	Squatting	0.494	0.627	0.415	0.412							
	Kneeling	0.483	0.637	0.422	0.427	0.762						
Both extremities involved	Fetching water from pump	0.404	0.463	0.437	0.431	0.195	0.269					
	Pound fufu	0.325	0.161	0.093	0.200	0.154	0.157	0.569				
	Using a cutlass	0.297	0.201	0.146	0.183	0.115	0.126	0.518	0.614			
	Carry harvest home	0.580	0.409	0.458	0.420	0.269	0.323	0.611	0.501	0.394		
	Heave loads on head	0.299	0.178	0.118	0.217	0.204	0.143	0.605	0.675	0.583	0.430	
	Standing up from floor	0.277	0.480	0.286	0.251	0.571	0.619	0.184	0.289	0.092	0.273	0.161