INTRODUCTION

Buruli ulcer is a devastating infectious disease caused by Mycobacterium ulcerans. It is named after a region called Buruli, near the Nile River in Uganda, where in 1961 the first large number of cases was reported. This disease has dramatically emerged in several west African countries, such as Ghana, Côte d’Ivoire, Benin, and Togo during the last decade. The first stage of Buruli ulcer starts as a firm, non-tender nodule. Other possible pre-ulcerative lesions in the first stage are plaques or edema. In the second stage, ulceration of the skin with an undermined edge can be seen. Occasionally, osteomyelitis complicates the course of illness. In stage three, a granulomatous healing response occurs. In stage four, fibrosis, scarring, calcification, and contractures with permanent disabilities may result (Fig. 1). Surgery is the present standard treatment. The average hospital admission time for Buruli ulcer in Ghana is approximately three months. Patients often come to the hospital in an advanced stage of the disease, leading to more extensive disfiguring and costly treatment.

The mode of transmission of this disease is unclear. Mycobacterium ulcerans may be acquired from the natural environment.1 A skin prick during farming or an insect bite could transmit the microorganism into the subcutaneous fat.2,3 Person-to-person transmission has rarely been reported.4 A study by Asiedu and Etuaful that examined the socioeconomic implications of Buruli ulcer in Ghana showed severe disabilities at high treatment costs.5 Costs could be reduced by treatment in an earlier stage of the disease. They hypothesized that factors including geographic access, lack of funds, superstitious beliefs about the illness, and stigma were important determinants for health-seeking behavior, and suggested possible consequences of the disease such as social isolation and problems in family life.

The severity of Buruli ulcer may be underestimated if only mortality and morbidity of skin lesions are measured. The use of medical anthropologic data may be a good way to assess the impact that Buruli ulcer has on the lives of affected people. These data may also be a necessary tool in developing disease control programs appropriate for the community and compatible with traditional perceptions of etiology, prevention, and control.

The purpose of this study was to explore the beliefs and attitudes of people in Ghana towards Buruli ulcer. The study focuses on three aspects: the ideas on the cause of the disease, help-seeking behavior and views on treatment, and stigma. Stigma can be divided into enacted stigma and felt stigma. Enacted stigma refers to episodes of discrimination against people on the grounds of social unacceptability or inferiority; felt stigma is based on a deep sense of inferiority and refers to an oppressive fear of enacted stigma.6 Stigma determines to a great extent the individual suffering caused by the disease and influences the view on the disease of people, both as individuals and community members.

METHODS

Data were collected from three different sites in Ghana: Agogo Presbyterian Hospital at Agogo in the Ashanti Akim North district, the Governmental Hospital in Dunkwa, Upper Denkyira District, and St. Martin’s Hospital at Agroyesum in the Amanse West district. These areas in the forest zone of Ghana are highly endemic for Buruli ulcer.1

Verbal informed consent was obtained from all adult participants and from the parents of minors. The study protocol was reviewed by the Investigations Review Board of the Groningen University Hospital (The Netherlands), and was approved by the local hospitals and district health authorities in Ghana.

One native, local language-speaking interpreter conducted the interviews at all sites. Before the interviews started, the reasons for and goals of the questions were discussed with interviewer and one of the authors (YS). The first series of interviews were conducted by interpreters and this author and discussed afterwards. Both patients with Buruli ulcer and age-and sex-matched controls were interviewed. Controls were unaffected by Buruli ulcer. Conceivably, unaffected people may yield more relevant information about stigmatizing attitudes7 than affected people. In Agogo, 16 patients admitted with Buruli ulcer and 16 age- and sex-matched patients admitted for other diseases on a different ward were interviewed.

Twelve cases and 12 controls were interviewed in Agroyesum, and five cases and five controls were interviewed in Dunkwa. In the case of children, questions were asked to the child, as well as well as to the attendant who was invariably one of the parents. Interviews started with open questions, followed by more specific, semi-structured questions. We
chose to interview only admitted patients. Our perception was that they would give more reliable answers, since they are not under time pressure.

The ideas on the cause of the disease were assessed by open interviews combined with semi-structured questions about the influence of 10 items on Buruli ulcer. These items were work/school, hygiene, environment, family/friends, behavior, mood, religion, curse, witchcraft, and ancestors. We categorized the answers in relation to biomedical explanations and magico-religious influences.

Ideas about treatment were assessed by questions on help-seeking behavior of patients with Buruli ulcer, the reasons and expectations of this help seeking, and experiences with help-seeking behavior of friends/family. Photographs of three different stages of Buruli ulcer (a nodule, an early ulcer, and a late ulcer) from the pamphlet Recognizing Buruli Ulcer by the World Health Organization were shown to unaffected subjects. Questions were asked about their help-seeking behavior for a skin lesion in three stages of disease. Answers were categorized by reasons for patients’ delay, patients’ experiences with helping-seeking behavior of friends and family, help-seeking behavior of the patients, and patients’ expectations of treatment in the hospital.

The stigma of Buruli ulcer was studied by open questions and quantitatively by a stigma query adapted from Vlassoff and others, who studied stigma of onchocercal skin disease in Africa, including Ghana. Questions were asked to the affected and unaffected subjects (Table 1) and responses to

![Figure 1. Young patients with Buruli ulcer in highly endemic area (near the Offin River in Ghana).](image)

| Table 1 | Distribution of respondents by disease status, sex, age, and education |
|---------|-------------------------|------------------|-----------------|-----------------|-----------------|-----------------|-----------------|
|         | Number of respondents   | Mean ± SD age of respondents (years) | Level of education |
|         | M           | F           | M           | F           | M ± F         | None | Primary school | Middle school | Junior secondary school | Post secondary schoo/college |
| Affected (n = 33) | 20 | 13 | 20.1 ± 12.3 | 22.2 ± 9.9 | 20.9 ± 11.3 | 3 (9%) | 14 (42%) | 8 (24%) | 7 (21%) | 1 (3%) |
| Unaffected (n = 33) | 20 | 13 | 20.4 ± 14.3 | 23.2 ± 10.8 | 21.5 ± 12.9 | 6 (18%) | 14 (42%) | 6 (18%) | 5 (15%) | 2 (6%) |
| Total (n = 66) | 40 | 26 | 20.3 ± 13.2 | 22.7 ± 10.1 | 21.2 ± 12.0 | 9 (14%) | 28 (42%) | 14 (21%) | 12 (18%) | 3 (5%) |
each question were scored as 3 for yes, 2 for possibly, 1 for uncertain, and 0 for no. A stigma score per individual was computed by adding the responses to each question. Each question contributed equally.

Items were evaluated for internal consistency with Cronbach's alpha. Comparison of stigmatizing scores of affected and unaffected subjects was done by responses to questions asked to both affected and unaffected respondents, and considering internal consistency. Stigma scores were analyzed for the influence of gender, education, and the difference in scores between affected and unaffected subjects. The influence of perceived cause on stigma and help-seeking behavior was studied.

The scores of the respondents in Agogo were compared with the scores of respondents in Agroyesum and Dunkwa. This distinction was made because hospital records indicate that Buruli ulcer is less endemic in the Agogo district. The open questions complimented the quantitative part of the study, providing explanations for stigmatizing aspects. The combination of quantitative and qualitative methods enables looking for a consistent pattern among socially desired answers.

RESULTS

Respondents' characteristics. There were no differences in age distribution among the different groups (Table 1). The unaffected and affected respondents were equally divided among the educational levels. The reasons for admission of the 33 respondents not affected by Buruli ulcer were malaria (n = 9), trauma (n = 3), tuberculosis (n = 3), heart disease (n = 2), and others with cellulitis, enteric fever, abscess, nephrotic syndrome, peptic ulcer, abortion, childbirth, kidney failure, urticaria, typhoid perforation, anal fissure, goiter, pyelonephritis, osteomyelitis, pneumonia, and snakebite.

Perceived causes of the disease. Causes of the disease as mentioned by the respondents can be divided into both biomedical and magico-religious thinking. An answer in the biomedical domain does not exclude an answer in the magico-religious domain.

Thirty-nine respondents (59% of the total number of respondents) mentioned personal hygiene, but some patients denied this possibility. “Hygiene cannot be of influence because I have good hygiene and I have the disease and other people without good hygiene don’t have it.”

The environment was mentioned as a risk factor by 32 respondents (48% of the total number of respondents). These thoughts are illustrated by the following statements. “When walking in swampy areas and if wearing sandals, insects or worms can bite you and cause the disease.” “Some of the rivers will have swampy areas, walking or bathing here could cause the disease.” “When walking with open toe shoes in swampy areas, you’ll scratch yourself and an organism or insect can get in and it can develop in a big ulcer.”

Four persons mentioned drinking water and three-mentioned food as the bacteria-containing source of the disease. Four respondents referred to an insect bite or snakebite. One person thought Buruli ulcer is a relapse of the disease known as a tropical ulcer.

During the interview, 15 respondents (23% of the total number of respondents) mentioned the fear of becoming infected by close contact with patients with Buruli ulcer. “If a friend is infected and you continue to play with him you might be infected also. Wearing dressing is not protecting, if a fly comes from this wound and then to you, you can get it." “The disease can be transferred via breathing.” “Family and friends can spread this disease through clothing, housing and feeding.”

Forty respondents were asked if they thought the disease was sexually transmitted. Five (13%) said it can be sexually transmitted, 14 (35%) did not know, and 21 (53%) said it is not. “More women have the disease because they usually engage in prostitution.”

Different magico-religious factors were given, of which witchcraft was most common. This was mentioned by 39 respondents (59% of the total number of respondents). Witchcraft sometimes seems to be mentioned as the only explanation as long as no other explanation can be found for the disease. “Witches are known mainly for spreading mysterious diseases in Ghanaian societies, like tuberculosis, Buruli and leprosy.” “In my case, I think it’s caused by witchcraft because I have good hygiene.” “I hope it is only a witch who can bring such a strange sickness or disease to someone because God the almighty loved us and will not bring us such a sickness.”

Other reasons to be bewitched are conflicts in the family, and the witch is usually someone from your own family. “When I got the disease, my sister asked me to give her 5,000 cedis (75 cents, US) and a cock and then it would heal. She’s a witch. We went to my sister to tell her she gave me the disease and she told me I’ll be dead before I’ll be free of Buruli.” “I had only a small injury before the Buruli ulcer developed. Usually such a small wound does not become this big, so it must be witchcraft that caused this severe disease.

I think it’s because I had a fight with my mother’s sister’s daughter, she might have given the disease to me. I’m afraid people in the village think I’m bewitched. People in my village will think witchcraft is probably involved if I’m admitted for a longer period because witches usually cause diseases that are difficult to cure. I’m also afraid the witchcraft will cause my leg to be amputated.”

In addition to a long admission as proof of witchcraft, the clinical presentation and lack of effective drug treatment was another reason to think of witchcraft, as illustrated by the following quote. “If I see the patient, I think it’s a disease, but if a patient says she’s surprised by its size and if medical treatment fails, I think it’s caused by witchcraft.”

A witch can also develop the disease. “Witches can develop the disease because they eat women’s flesh and if that flesh has Buruli, she can get it.”

Thirty-one respondents mentioned a curse as a cause of the disease (47%, 16 were patients with Buruli ulcer), six respondents said they did not know whether a curse has an influence. Patients with Buruli ulcer who said a curse could cause the disease denied that a curse was the cause of their own disease. “It is possible that a curse causes this disease, but not in my case, I did not do anything wrong to get a curse.” “If you continue doing something that someone else does not want, you can get cursed. A person can do that by performing rituals, like talking to an egg and breaking it. I’ve never been cursed. I would have known if I were cursed, outside I would have heard and otherwise the person who cursed you would have told you.”

The scores of respondents in Agogo were compared with the scores of respondents in Agroyesum and Dunkwa. This distinction was made because hospital records indicate that Buruli ulcer is less endemic in the Agogo district. The open questions complimented the quantitative part of the study, providing explanations for stigmatizing aspects. The combination of quantitative and qualitative methods enables looking for a consistent pattern among socially desired answers.
Five patients regarded the disease as God’s will. “God protects us all. Someone who gets the disease might have done something wrong and therefore not get protected by God. I do have Buruli ulcer myself, but I don’t know why God did not protect me.”

The evil eye was mentioned by two respondents as an influence factor. “If a lot of people see my wound, it might not heal. If all people start to talk about it, stare at it and are surprised by it, it would not heal either.” “I always want the door closed during changing of dressings, to protect it from evil eyes.”

Even though ancestors play an important role in the daily life of Ghanaians, no one attributed the disease to problems in the relationship with ancestors.

Witchcraft and cursing were equally often mentioned in the different districts (by chi-square test).

**Help-seeking behavior and views on treatment.** Ten (30%) of 33 patients mentioned financial difficulties as a reason not to come to the hospital earlier. Patients have to gather money first, which sometimes means they have to wait for the harvest season.

Another reason was fear of treatment in the hospital, and especially of amputation: “My mother told me not to go to the hospital. They were afraid my leg would be amputated, because of the size they thought the disease had gone to the bone.” “My sister’s boy’s son had Buruli, his leg’s amputated. He now walks with crutches. My parents don’t know it’s not compulsory to have an amputation for Buruli ulcer.” One respondent mentioned a fear of therapy with injections in his village.

Eleven patients said they expected the skin defect to heal by itself, indicating that they occasionally neglected the skin defect. A mother of a 30-year-old patient, who had Buruli ulcer on his right leg, with the entire limb being involved, said “Young men don’t want to go to the hospital with something small.” Others showed a trust in herbal treatment and were of the opinion that Buruli ulcer is ‘not a hospital disease,’ but something to be managed within the community.

Of 66 respondents, 14 (10 with Buruli ulcer) said that they have at least one family member or friend with Buruli ulcer. There was no significant difference in the percentage of people having a family member or friend with Buruli ulcer among the different locations. These 14 respondents indicated that they had a total of 16 family members and friends with Buruli ulcer, of which 11 had gone to the hospital without any use of herbal treatment, two had gone to the hospital only after the failure of herbal treatment, and three had been treated with herbal medicine. Only one respondent said his friend’s treatment with herbs was not effective; all other respondents regarded the treatment of their friends/family as effective.

Anticipated help seeking was tested by showing unaffected respondents a Buruli ulcer nodule in the pamphlet. Eighteen (55%) of the 33 respondents said they would use an herbal medicine if they had a boil like this, and the remaining 45% said they would go to a hospital. “I would use the herbal medicine that’s commonly used for a boil. If there was no improvement after herbal medicine, I would go to the hospital.” “If you treat a boil with herbs, it will burst and leave only a small scar, if you go to the hospital, you’ll have surgery and the wound will be very big.” In the treatment of the early ulcer shown in the pamphlet, two respondents (6%) said they would use hot water dressings and antibiotics, five persons (15%) said they would use herbs, and the others (79%) said they would go to the hospital. Only one person said she would treat the late ulcer itself with hot water and penicillin; all others said they would go to the hospital.

Patients with Buruli ulcer were asked about their help seeking behavior. After noticing their skin defects for the first time, four patients (12%) did nothing, four respondents (12%) applied hot water and antibiotics, five patients (15%) went to the hospital, three patients (9%) to the village health worker, and 17 patients (52%) applied herbs. Patients decided to come to the hospital when the wound had become of an unexpected size and after failure of herbal treatment. The perceived cause of the disease did not have an influence on the help-seeking behavior (by chi-square test).

Seventeen patients (52%) expected to be treated by surgery, six patients (18%) did not know how they would be treated, and 10 patients (30%), nine patients were from Agroyesum) expected to be treated with medication. Five patients mentioned they did not expect to be admitted for such a long period and three patients were surprised by the extent of the surgery. “I expected a small surgery, removing the boil, not a big surgery. The first time I saw the wound, I became afraid and realized I would be here for much longer than a week. A nurse on the ward explained me afterwards that the tissue around it was also affected.”

**Stigma of Buruli ulcer.** Patients said they were hindered by their disease in functioning as a leader. Controls confirmed this. Patients may feel ashamed or embarrassed because of Buruli ulcer, and think less of themselves. Frequently, patients said they were avoided by others. Approximately half of the controls avoided patients with this disease. Patients sometimes tried to hide their disease from others, although in some cases it was difficult to hide. Others preferred that their community members know about their disease. Patients in Agogo perceived more marital problems than the patients in Agroyesum and Dunkwa. These marital problems included getting married and in continuation of marriage and sexual functioning. Controls expected patients with Buruli ulcer to have even more marital problems; this expectation was more common in Agroyesum than in Agroyesum and Dunkwa. These stigmatizing aspects are confirmed by both quantitative (Tables 2 and 3) and qualitative data.

**Factors influencing stigma scores.** Of the 11 questions that were given to both affected and unaffected participants (marked with an asterisk in Table 2), eight questions were used to compare stigmatizing scores between affected and unaffected respondents. This selection was based on the internal consistency. The alpha value of the 11 items was 0.65. By eliminating the items “think less of yourself,” “others pity you,” and “others think less of family,” the internal consistency could be increased to $\alpha = 0.77$.

The median score of affected respondents was 6.0, which is significantly lower than the median score of 10.0 of unaffected respondents ($P = 0.035$; by Mann-Whitney U test). Location influenced stigma scores for patients, as shown in Tables 2 and 3. Educational background did not have any effect on the stigma scores. Stigmatizing scores for Buruli ulcer were higher for men than for women, although this difference was not significant (median scores: affected men = 7.0, affected women = 5.0). The median scores were 11.5 for unaffected men and 10.0 for unaffected women. Having a friend or a
family member with Buruli ulcer (as 16 participants did) also had no influence.

When witchcraft or cursing was given as a perceived cause, stigmatizing scores were higher ($P = 0.008$ and $P = 0.017$, respectively; by Mann-Whitney U test). Stigmatizing scores did not have an influence on the help-seeking behavior of both patients with Buruli ulcer and control patients.

**Reasons for stigmatizing aspects of Buruli ulcer reported by patients.** Fear of acquiring the disease is the basis of many of the stigmatizing aspects. It is the main reason for avoiding contact with other patients with Buruli ulcer, especially for non-relatives. A 17-year-old woman with Buruli ulcer on her foot (a minor deforming aspect) said: “If I go to a place where there are many people, they’ll all go away, because they think I can transmit the disease, by witchcraft, if they come close.” The same fear can make people think less of the patient. For some respondents, fear of transmission is a reason for people to show their disease instead of hiding it from others. Fear of transmission is a reason for people to show their disease instead of hiding it from others. Another reason for people to show their disease instead of hiding it from others is help-seeking: “I prefer people not to see it, but if you show it to others, they might be able to bring you to the hospital.”

The difficulties in functioning as a leader are diverse. A leader should display an undamaged and ideal performance and therefore cannot function as a leader. An 18-year-old woman said she would not have an influence on the help-seeking behavior of both patients with Buruli ulcer and control patients.

<table>
<thead>
<tr>
<th>Reason for Stigmatization</th>
<th>Agogo (n = 16)</th>
<th>Agroyesum and Dunkwa (n = 17)</th>
<th>Total (n = 33)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Keep others from knowing if possible*</td>
<td>5 (31) 0 (0) 0 (0) 11 (69)</td>
<td>3 (18) 0 (0) 0 (0) 14 (82)</td>
<td>8 (24) 0 (0) 0 (0) 25 (76)</td>
</tr>
<tr>
<td>Would like someone to know*</td>
<td>10 (63) 1 (6) 0 (0) 5 (31)</td>
<td>10 (60) 1 (6) 2 (12) 4 (24)</td>
<td>20 (61) 1 (3) 3 (9) 9 (27)</td>
</tr>
<tr>
<td>Think less of yourself†</td>
<td>5 (31) 0 (0) 2 (13) 9 (56)</td>
<td>7 (41) 8 (47) 0 (0) 2 (12)</td>
<td>12 (36) 8 (24) 2 (6) 11 (31)</td>
</tr>
<tr>
<td>Difficult to function as a leader‡</td>
<td>13 (81) 0 (0) 2 (13) 1 (6)</td>
<td>4 (24) 2 (12) 2 (12) 9 (53)</td>
<td>17 (52) 2 (6) 4 (12) 10 (30)</td>
</tr>
<tr>
<td>Opposite sex would think less of you</td>
<td>2 (13) 0 (0) 6 (38) 8 (50)</td>
<td>2 (12) 1 (6) 10 (59) 4 (24)</td>
<td>4 (12) 1 (3) 16 (49) 12 (36)</td>
</tr>
<tr>
<td>Others would think less of you*</td>
<td>1 (6) 0 (0) 4 (25) 11 (69)</td>
<td>2 (12) 4 (24) 5 (29) 6 (35)</td>
<td>3 (9) 4 (12) 9 (27) 17 (52)</td>
</tr>
<tr>
<td>Others pity you*</td>
<td>10 (63) 1 (6) 3 (19) 2 (13)</td>
<td>12 (71) 4 (24) 0 (0) 1 (6)</td>
<td>22 (67) 5 (15) 3 (9) 3 (9)</td>
</tr>
<tr>
<td>Others have avoided you*</td>
<td>4 (25) 0 (0) 0 (0) 12 (75)</td>
<td>2 (12) 0 (0) 3 (18) 12 (71)</td>
<td>6 (18) 0 (0) 3 (9) 24 (73)</td>
</tr>
<tr>
<td>Felt shamed or embarrassed</td>
<td>8 (50) 1 (6) 0 (0) 7 (44)</td>
<td>8 (47) 4 (24) 0 (0) 5 (29)</td>
<td>16 (49) 5 (15) 0 (0) 12 (36)</td>
</tr>
<tr>
<td>Others think less of family*</td>
<td>1 (6) 1 (6) 4 (25) 10 (63)</td>
<td>2 (12) 4 (24) 4 (24) 7 (41)</td>
<td>3 (9) 5 (15) 8 (24) 17 (52)</td>
</tr>
<tr>
<td>Problem in getting married*</td>
<td>6 (38) 1 (6) 4 (25) 5 (31)</td>
<td>3 (18) 1 (6) 4 (24) 9 (53)</td>
<td>9 (27) 2 (6) 8 (24) 14 (42)</td>
</tr>
<tr>
<td>Problem in ongoing marriage‡</td>
<td>7 (44) 2 (13) 4 (25) 3 (19)</td>
<td>0 (0) 3 (18) 3 (18) 11 (65)</td>
<td>7 (21) 5 (15) 7 (21) 14 (42)</td>
</tr>
<tr>
<td>Problem with sexual functioning†</td>
<td>5 (31) 0 (0) 7 (44) 4 (26)</td>
<td>0 (0) 0 (0) 3 (18) 14 (82)</td>
<td>5 (15) 0 (0) 10 (30) 18 (55)</td>
</tr>
<tr>
<td>Problem with fertility‡</td>
<td>3 (19) 0 (0) 10 (63) 3 (19)</td>
<td>0 (0) 0 (0) 3 (18) 14 (83)</td>
<td>3 (9) 0 (0) 13 (39) 17 (52)</td>
</tr>
<tr>
<td>Problem in relation to marriage‡</td>
<td>4 (25) 1 (6) 7 (44) 4 (25)</td>
<td>0 (0) 0 (0) 4 (24) 13 (77)</td>
<td>4 (12) 1 (3) 11 (33) 17 (52)</td>
</tr>
</tbody>
</table>

**Median score of patients**

| No. (%)| 16.5 | 11.0 | 12.0 |

* Questions asked to both affected and unaffected respondents of which eight were selected for comparison.
† Significant difference between Agogo compared with Agroyesum and Dunkwa; $P = 0.008$, by Mann-Whitney-U test.
‡ Significant difference between Agogo compared with Agroyesum and Dunkwa; $P = 0.045$, by Mann-Whitney-U test.
§ Significant difference between Agogo compared with Agroyesum and Dunkwa; $P = 0.001$, by Mann-Whitney-U test.
¶ Significant difference between Agogo compared with Agroyesum and Dunkwa; $P = 0.031$, by Mann-Whitney-U test.

Discussion

This is the first study to examine the impact that Buruli ulcer has on people, apart from its physical and socioeconomic aspects. We analyzed the beliefs and attitudes of patients with this disease and patients admitted for other reasons.

The cause of the disease is a curse or witchcraft. If there's another cause, I would not think less of the family.

In communities where Buruli ulcer is not endemic, a patient can be accused of transferring the disease to their community. “If someone in our community would get this disease, people might say: That's the family who always brings diseases to our community. Because I think we don't have this disease in our community.”

Getting married is difficult when one has Buruli ulcer, especially when the disease shows long-term consequences and deforming aspects. An 18-year-old woman said she would only find a husband when her wound was completely healed.

Another young woman blamed Buruli ulcer for her still being single. Three patients (one man and two women) who participated in this study said that they were divorced because of the disease. Their partners left them because of the financial burden of the disease. According to other affected and unaffected respondents, fear of getting infected would be a reason for getting a divorce.

Sexual performance can be limited by Buruli ulcer. A 24-year-old patient said he would not have sexual intercourse with a woman because he tries to hide himself and his disease.

Five (7.6%) of 66 respondents indicated that patients with Buruli ulcer cannot be fertile because the disease can make their blood dirty.

**DISCUSSION**

This is the first study to examine the impact that Buruli ulcer has on people, apart from its physical and socioeconomic aspects. We analyzed the beliefs and attitudes of patients with this disease and patients admitted for other reasons.
Many respondents indicated factors in the environment as causes of this disease. The environmental factors mentioned are consistent with environmental factors reported in the current scientific literature. However, one can question the validity of these responses since although they may either be the patient’s actual observations, they may also be influenced by health education programs.

Magico-religious explanations were very common. These were more probable if the disease took a long time to heal, resulted in a long stay in the hospital, and were difficult to cure. A study on illness management in northern Ghana pointed out that perceptions on etiology of the disease change if the situation worsens. The respondents in our locations appeared to think in a similar way about Buruli ulcer, with witchcraft and curses being more probable explanations if the course and outcome of the disease worsens.

One factor in a delay in treatment is financial difficulties. This is not surprising, since Buruli ulcer results in very high indirect costs for patients and their families. Another reason the patients mentioned is fear of treatment. This fear may be underestimated in our study since the respondents were admitted to a hospital and do not represent the fear of patients who were not admitted. Ambivalence and a fear of biomedicine was previously reported in Ghana. Neglect of a disease is another reason for patients’ delay in seeking treatment. Patients did not mention stigma of the disease as a reason for neglecting it, and quantitative data showed no influence of stigma on help-seeking behavior. Other investigators have found that stigma significantly influenced the neglecting of a disease (tuberculosis) by patients.

Buruli ulcer is not considered a “hospital disease” in its early stage. It has been shown that different stages of a disease are associated with different help-seeking behavior. Visiting a hospital is an option if the disease gets out of control. In our study, 28 (85%) of 33 patients first tried to solve their health problem within their community by self-treatment with herbs, or by visiting local healers or health facilities. The expectations of treatment by the patients often did not correspond with reality. Lack of information on treatment not only frightens the patient himself, but might also have a detrimental effect on help-seeking behavior of others with Buruli ulcer.

The mystery of Buruli ulcer, i.e., not knowing the mode of transmission and not having proper treatment, is a major factor in determining the stigma of this disease. Both qualitative and quantitative data show a considerable impact of stigma on the daily life of the patient; therefore, we conclude that social stigma for patients with Buruli ulcer are huge. The stigma in the communities is probably even worse than that observed in this study, since socially desirable answers might have been given by the respondents, and people not admitted to a hospital might reveal a more stigmatizing behavior than that of the admitted respondents.

A study of patients with leprosy showed that this disease has a more detrimental social impact in women than in men. Conversely, a study on onchocercal skin disease by the Pan-African Study Group found a higher stigma score for men. In another study on gender and stigma of onchocercal skin disease, no quantitative difference between men and women was found, but stigma did affect men and women in different ways.

<table>
<thead>
<tr>
<th>Table 3</th>
<th>Responses to stigmatizing items of controls</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Agogo (n = 16)</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
</tr>
<tr>
<td>Keep others from knowing if possible</td>
<td>6 (38)</td>
</tr>
<tr>
<td>Would like someone to know</td>
<td>6 (56)</td>
</tr>
<tr>
<td>Think less of himself/herself*</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Difficult to function as a leader*</td>
<td>16 (100)</td>
</tr>
<tr>
<td>Others would think less of him/her</td>
<td>8 (50)</td>
</tr>
<tr>
<td>Others pity him/her</td>
<td>8 (50)</td>
</tr>
<tr>
<td>Others have avoided him/her*</td>
<td>12 (75)</td>
</tr>
<tr>
<td>You would avoid him/her†</td>
<td>10 (63)</td>
</tr>
<tr>
<td>Others might shame or embarrass him/her</td>
<td>8 (50)</td>
</tr>
<tr>
<td>You might make fun of him/her</td>
<td>2 (13)</td>
</tr>
<tr>
<td>Others think less of family</td>
<td>3 (19)</td>
</tr>
<tr>
<td>You would think less of family</td>
<td>4 (25)</td>
</tr>
<tr>
<td>Problem in getting marriage†</td>
<td>12 (75)</td>
</tr>
<tr>
<td>Problem with sexual functioning‡</td>
<td>8 (50)</td>
</tr>
<tr>
<td>Problem with fertility‡</td>
<td>1 (6)</td>
</tr>
</tbody>
</table>
| Median score of controls‡‡ | 24 | 13 | 18

* Significant difference between Agogo compared with Agroyeum and Dunkwa; P = 0.001, by Mann-Whitney-U test.
† Significant difference between Agogo compared with Agroyeum and Dunkwa; P < 0.05, by Mann-Whitney-U test.
‡ Analysis of internal consistency with Cronbach’s alpha indicated an α coefficient of 0.76. When the items ‘think less of yourself’ is α coefficient increases to 0.79. Median scores are calculated on the remaining 13 items with an α coefficient of 0.79.
† Significance difference between Agogo compared with Agroyeum and Dunkwa; P = 0.014, by Mann-Whitney-U test.
ways. In our study, there was no difference between stigma scores of men and women. The qualitative data also did not suggest distinctions between gender in stigma. The Pan-African Study Group also found a higher stigma for patients with more education. In a study on gender and tuberculosis control, the investigators described the level of education as resulting in different attitudes on the disease. A low level of education makes people think tuberculosis is dangerous and incurable, whereas people with more education regard it as a curable disease. The cause of Buruli ulcer and the best treatment are still unclear. This might explain why educational background did not have an influence in our quantitative and qualitative data analysis.

The difference in stigma scores in Dunkwa and Agroyesum, when compared with those in Agogo, was significant. This might be explained by the fact that people in highly endemic areas such as Dunkwa and Agroyesum have an increased chance in becoming acquainted with the disease and therefore adjust their attitude about it. However, our study found no relationship between having friends or family members with the disease and the level of stigma. Another explanation could be the increased health education efforts in the more endemic areas, e.g., information on videos. In addition, both quantitative and qualitative data show that involvement of witchcraft or curses leads to higher stigmatizing scores.

Since this study involved patients admitted to hospitals in Ghana, its findings cannot be extrapolated to patients in communities, which would require specifically targeted research. Whether the results of this study can be applied to other countries remains to be determined. However, this study has demonstrated other aspects of the ordeal of Buruli ulcer, apart from physical effects, and should stimulate more research on this disease.

Additional educational programs for Buruli ulcer should be developed, not only because they could help in the detection of cases at an earlier stage of the disease, but because they might also lower stigma. These programs should be used in areas with low endemicity, where the disease tends to have higher stigma, and in areas with high endemicity, where the overall impact of stigma could be greater in these areas with more patients. Educational programs should not frighten communities with images of amputated limbs, but should emphasize treatment at early stages of the disease. In addition, the possible causes of the disease should be explained. Witchcraft and curses cause higher stigma, and participation in disease control is more difficult if communities have a magical feeling about the disease, which puts responsibility for transmission outside human control. Based on the information obtained from the patients in this study, improvements are needed in the dissemination of information on the need for surgery and the duration of hospital admission.

Based on our stigma results, another recommendation to limit stigma and fear in patients would be that active case surveillance in the field should be conducted in the presence of as few people as possible. This would lessen fear and superstition among the community population. Treatment and prevention might be more effective at the community level, since our data show that people try to solve their health problems more frequently at this level. According to Bowman and others, surgery performed in villages resulted in better acceptance, lower costs, more time saved, and less fear of this procedure.

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Authors’ addresses: Ymkje Stienstra, Department of Internal Medicine/ICB, Groningen University Hospital; PO Box 30.001, 9700 RB Groningen, The Netherlands, Fax: 31-50-361-3216, E-mail: y.stienstra@int.azg.nl. Winette T. A. van der Graaf and Tjip S. van der Werf, Department of Internal Medicine, Groningen University Hospital, PO Box 30.001, 9700 RB Groningen, The Netherlands, E-mail: t.s.van.der.werf@int.azg.nl. Kwame Asamoah, Ministry of Health, National TB and Buruli Ulcer Control Program, Korle Bu Accra, Ghana.

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