Introduction and background

Mycobacterium ulcerans disease or Buruli Ulcer (BU) is a rare chronic bacterial skin disease which presents different forms of lesions[1]. The non-ulcerative form appears as a papule or a firm, subcutaneous nodule which may ulcerate with deep undermined edges[2, 3]. The infection in the ulcerative form can be extensive and may destroy nerves, appendages and blood vessels[1]. An acute oedematous form of the disease may involve a whole limb, or the entire face[4]. BU may cause osteomyelitis when the severe form can lead to amputation[5].Although mortality of BU is low, morbidity and subsequent disability are high. For example all the 27 participants in this study had severe scarring on different parts of their bodies and 15 participants had severe contractures to their arms and legs. Active treatment including wound care, debridement, excision and skin grafting leads to long hospital stays. This study considered these needs within the context of one of WHO's[6] seven recommended research priorities of *cultural and socio-economic research* related to BU. A literature review update conducted since this study (to September 2016) revealed that no other published studies have examined the support needs of people living with BU.

Methods

To understand the psychosocial impact of BU we adopted a qualitative research design using Glaser's[8-9] and Chamaz's[10-11] versions of grounded theory method. The method consist of simultaneous data collection and analysis with each informing the other throughout the research process. It involved the use of in-depth interviewing and observation of patient behaviour, field notes and conversations and discussions with key people in the hospital and the communities.

Access, sampling, interviews and data analysis

Ethical approval was obtained from The Ghana Health Service Ethical Review Committee. Verbal and written consents were obtained from all adult participants and from those with parental responsibilities for minors.

A case based sampling strategy and snowballing technique (a method whereby potential participants are asked to name any one else with the condition who would be prepared to take part in the study) were used to recruit the participants and those providing their healthcare. Thirty five participants were selected initially, however, the actual number of twenty seven (21adults and 6 children) was determined at the point of theoretical saturation (where no new data were derived). Table 1(Appendix 1) depicts the participants' characteristics.

The researcher (AE), stayed in the community for five months employing observation and one-to-one semi-structured interviews which were tape-recorded in the *Twi* language. The recordings were translated into English and transcribed verbatim. An independent person audited the transcripts to ensure accuracy.

Content analysis was employed to examine key themes and then constant comparative method of data analysis was used to examine and reveal the similarities and differences between the data sets.

Development of BU functional limitation score

Disability measurement in BU is largely based on functional disability scores, yet these tend to focus on physical limitations. Therefore we modified Stienstra et al's[12] BU functional limitation scoring system to assess the nature and severity of the impairment on carrying out daily activities as a result of BU. We then included the more profound impact on the participants' *social* and *psychological* well-being. The modified version and the scoring system used are shown at appendix 2.

Findings

The findings here relate to the support needs of the participants. Where appropriate participants' responses are quoted verbatim with brevity to support the finding around the key conceptual themes emerging from the data.

Functional limitations

The study revealed that all of the participants had physical functional limitations. The average physical functional limitation score was 45% [score ranging from no functional limitation (0%) to total limitation (100%)]. However, the average psychological and social functional limitation scores were 52% and 38% respectively. (See appendix 2 for the explanation of these scores). The impact of this is borne out in the following qualitative data.

Fear of recurrence

For many participants their greatest worry was the fear of the ulcer recurring and the need to prevent this. The mean time and the range for the disease to be inactive was 7 and 6-12 months respectively. The mean recurrence rate was also found to be twice per year. The fear of recurrence often led to anxiety and panic which affected the participants' daily routines and social activities. This is encapsulated by a young mother of two,

> *'I spent about four to five hours daily thinking about recurrence and then try to think about how best to minimise any activity that will lead to recurrence'*

Contracture

Contracture leading to disability affected 55% (n=15) of the participants and what they could do. The average physical functional limitation score of those with visible contractures was found to be 52% and it was serious among young mothers and young men who had to walk long distances to their farms for work and for school pupils who struggled to write with the affected hands. A female respondent recounted,

> *'I will always need someone to bath and cream my back because of contracture'*

Lack of sensation and numbness

Apart from the obvious physical losses due to amputation and contracture, 72% (n=19) of the participants described other physical symptoms such as loss of sensation and numbness in diseased areas. Consequently undertaking normal domestic chores such as cooking led to risk of burning and walking to work required protection on the affected area. This 25 years old young man recounts his experience,

> *'I have to be cautious about my surroundings which in itself has been a worry to me'*.

Feeling tired

Participants also reported severe fatigue which they describe as "*feeling constantly tired*" with little or no improvement on resting. This also interfered with their activities of daily living, including leisure, recreation and sexual relationships. A 22 year old man reported that;

> *'Buruli ulcer has restricted my hobbies and what I used to enjoy'*

Lack of leisure and recreational activities resulted in above average social and psychological functional limitation scores of 42% and 57% for all the participants.

Lack of information

Participants were concerned about the lack and reliability of information from the health professionals following their discharge. Some felt that the only information they get about BU was through doubtful sources including the media especially newspapers and the radio which distorted the true nature of their condition. A young lady recounted her experience;

> *'I was given a booklet on Buruli ulcer and that was it. And the information from the radio and newspapers were all disgusting'*.

Poor dietary requirements

Many of the participants confirmed that the hospital staff discussed with them about the importance of balanced diet to help improve ulcer healing but there was no follow up nor professional dietary advice on what precisely to eat to support their immune system and wound healing.

Sexual needs

Many of the participants said that they found it difficult to engage in intimate relationships due to the stigma attached to BU. This was more profound among participants with physical functional limitation scores between 36-45% and psychological and social functional limitation scores of between 39-58% and 34-42% respectively. Others expressed the need to get trusted health professionals to discuss and help them to address their sexual problems, such as feeling tired after sex and genital soreness after masturbating. However, such support was unavailable. A 23 year old man recounted his humiliation when he visited the local clinic with a sore penis after masturbating;

> *'The nurses started giggling and told me it was self-inflicted. I have never had the courage to go there again so I will rely on the dispenser or use indigenous medicine'*.

Insomnia

The thought of ulcer recurrence coupled with intermittent and phantom pains for those who had undergone amputation often led to sleeplessness. The average psychological functional limitation scores for these participants was found to be 47% compared with the 18% of those participants who did not complain of sleeplessness and insomnia. Many also recounted how they have become addicted to *Akpeteshie* (a local Gin) to overcome this, while others took sleeping medication, (*diazepam)* which they purchased on the black market from local dispensers.

Prostheses and aids

In order to feel normal and achieve their potential 51% (n=14) of the participants wanted limb prostheses, from '*artificial legs and arms'* (participants own words), crutches, to protective clothing and a wheelchair; yet these resources were often inaccessible or of inadequate quality and effectiveness.

> '*My crutches were paid for by the Catholic Church, but that was then. I have outgrown them, making life uncomfortable for me'*. (Male, 23 years old).

Effects on education

Many participants recounted the long periods of hospitalisation following surgery, which ranged from 9 months to 2 years. Some younger participants had lost a year or so of schooling and so decided to concentrate on improving their disease recovery instead of school attendance. Others returned from hospital with deformed arms and fingers which affected their ability to write. The average physical functional limitation score for these participants was 36% compared to the 14% of those who were able to go back to school. The average social and psychological functional limitation scores for these participants was found to be 35% and 56% respectively as compared to the 10% and 13% respectively of those who were able to go back to school.

A 15 year old boy recounted his experience;

> *'I spent 19 months in and out of hospital and when I decided to go back to school I realised I was struggling alone. I had to learn how to use my left hand to write for the first time. Then the ulcer would recur and I had to stop, then go back to school when it had healed. There was no end to my struggle so I had to give up.*

All these challenging experiences of living with BU made it difficult for these participants to return to school as they had no support to help them reintegrate into the educational system at the time. The Catholic Church and other charities assisted some to undertake various vocational courses but they often had to drop out due to recurrence of the disease or contracture developing.

Discussion

Two key studies that have investigated the support needs of BU patients[13, 14] focused only on the support needed to remove the barriers to early detection of the ulcer. The study reported here provides unique insights into the support needs of people living with BU following discharge, and how they live and attempt to adapt their lives with disability. Also it highlights that the availability of appropriate interventions and equipment that could help them self-manage their condition and improve access to skin care/dermatological services.

Lack of sensation to the affected limb parts were linked to the fear of the ulcer recurring. The study identified that 28% (n=8) of the respondents had either new wounds next to old sites or old wounds had opened up or had become macerated through work or play with the same symptoms. When this happened it was difficult to ascertain whether the new wound was a relapse or a new infection. There were others whose wounds had become chronic which may need weeks or months of antibiotic therapy and proper dressings to deal with. The findings here support the established belief about recurrent rate in BU during follow-up[15, 16, 17], but in addition they reveal that the repeat recurrences could in fact be new infections or re-infections or indeed both and this new knowledge offers the opportunity to improve BU management.

There are many reasons for the repeat recurrences. The first is that bandages are washed and reused both at the hospitals/clinics and at home for economic reasons. These bandages showed evidence of secondary bacterial contamination and the pathogens found in the bandages have been reported to be comparable to the bacterial profile of wound samples[16]. So lack of proper wound care management after discharge among people living with BU, such as the use of recycled bandages – without adequate washing, is a probable source of wound infection among chronic wounds. Another source could be the indiscriminate use of antibiotic therapy, which can be bought over the counter by people living with BU peri-operatively. There is the tendency for those with BU to develop antibiotic resistance to secondary organisms normally found in their wounds. High levels of *Staphylococcus Aureus* isolate, resistance to *flucloxacillin,* which is the main antibiotic in use in Ghana for treating skin infections, have been found in another study[18]. Similarly the antibiotic- *ciprofloxacin* - was identified to be the antibiotic of choice for our participants and the 'dispensers' who treated them at home. Those living with BU in the study area may have developed antibiotic resistance to *ciprofloxacin.*

The disease specific needs above revealed a lack of suitably organised after care and effective education to enable people living with BU to effectively self-manage wound recurrence. Interviews with the health professionals revealed that the government's restricted budget allocation meant that they could only ensure that people with BU were safely discharged into the community yet without any resource-efficient, follow-up or self-management plan at the time.

The data revealed high level of exhaustion and lethargy. BU is a systemic disease[3, 19] and the large doses of poly-medication during treatment could have adverse effect on the patient's immune system and aggravate their lethargy. The common sleep disruption experienced by the participants is a problem that has been found to have deleterious effects on health [20] and to be a key issue for other dermatological patients[21, 22].

Intimacy and sexuality constitute a major health related quality of life issue among adolescents[23]. The adolescents in this study expressed a lack of self-confidence in approaching the opposite sex and their ‘normal’ peers. The average psychological functional limitation score with these participants was found to be higher than those who did not complain this lack of self-confidence. This could be due to two interrelated issues of the media’s (television and newspapers) portrayal of what constitutes a perfect body and the misconception of BU as a contagious disease. Therefore sexual practice for some participants such as masturbating has physical, psychological and clinical implications as described by the study participants. The lack of professional support for this need is unfortunate as the participants felt neglected and lacked the opportunity to discuss their feelings or their health.

44% (n=12) of study participants had to stop their education due to disease recurrence, re-infection and/or contracture which affected them psychologically. Most expressed the desire to retrain in various vocations or go back to school as a means to improve their chances of finding a job. In a broadly related study those living with acne and psoriasis in resource rich countries were found to be unable to follow their desired career paths due to the stigma attached to their conditions[21].

The stigma of the visible nature of the condition led many participants to avoid certain social situations or functions, recreational pursuits and leisure similar to other dermatological studies that revealed the adverse effects of chronic skin diseases on peoples’ social life[24, 25]. In this study the need to socialise and to enjoy leisure activities to improve quality of life was expressed by all the participants, yet their ability to meet this need was found to be adversely affected due to their disease-related problems such as immobility, social stigma and isolation. This led to average higher social and psychological functional limitation scores.

Satisfactory diet and nutrition are established factors in effective wound healing[26, 27, 28] with fatigue as an early symptom of vitamin C depletion in the body[24]. Such evidence supports what was revealed in the present study as well as a similar Ghanaian study[29] of BU patients whose staple diet was found to be mainly carbohydrate lacking key nutrients vital for the normal wound healing process. The findings indicated that there was also the clinical need for health professionals to provide education about self-care or self-management for the patients and their cares. Participants felt that the paucity of information from health professionals was one of the reasons they felt neglected. They lived in close-knit communities where indigenous knowledge of health needs in the form of medicinal herbs is commonly shared. Traditional healers were found to play important roles in shaping the health belief of the participants and in this cultural context they could not be ignored.

The need for different kinds of prostheses was identified in the study. This ranged from expensive artificial legs/arms and wheel chairs to simple aids, such as crutches and grip aids. From our observations the respondents would have needed a basic level of orthopaedic/physiotherapy assessment prior to discharge to ensure comfortable footwear and suitable splints. This short term measure could then be possibly a followed up to review the patient for an appropriate long term limb prostheses.

The systematically derived qualitative data, combined with a functional limitation score that incorporated psychosocial impacts, helps to advance our understanding of the support needs of those living with BU. However, the functional limitation tool employed requires further validation and development, which the research team intend to undertake and so such scores are a study limitation, but their validity is enhanced by employing the related comparative qualitative data

Conclusion

The key support needs of people living with BU varied from their physical needs such as lack of sensation, feeling tired, the fear of the ulcer recurring and sleeplessness to educational needs such as lack of health information about managing their ulcer and ways of preventing recurrence.. The study findings revealed that the support needs of people living with BU arise from the impact of the disease and inadequacies in the health system as well as a lack of understanding of their needs. Therefore systematic educational intervention is required to respond to the support needs of those living with BU that focuses on improving their ability to self-manage their condition with effective local empowerment within the affected communities.

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