CASE REPORT

BURULI ULCER: THE FIRST REPORTED CASE FROM ETHIOPIA

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ABSTRACT

We report a 22 years old female patient from Southern Ethiopia with ulcerative form of Buruli ulcer on the left leg. The case suffered for more than 10 years without proper diagnosis and treatment until she was last diagnosed and treated in Jimma University Specialized Hospital in 2009. She received anti-tuberculosis drugs for six months and showed marked improvement. This case demonstrated the diagnostic and management difficulties of BU in a country where no previous case was reported.

INTRODUCTION

Buruli ulcer was first described by a British physician working in Uganda. It has got the name Buruli ulcer since many cases occurred in the 1960s in Buruli County (now called Nakasongola District) in Uganda (1).

Buruli ulcer is the third most common mycobacterial disease worldwide. It has been reported in over 30 countries mainly with tropical and subtropical climates. These numbers may only be an indication of the presence of the disease but do not reveal the magnitude of the problem. The reasons for the growing spread of BU remain unclear. All ages and sexes are affected, but most patients are among children under 15 years. In general, there is no difference in the infection rate among males and females (2,3). There is little seasonal variation in the incidence of the disease.

It is caused by infection with *Mycobacterium ulcerans* which is likely to occupy a specific niche within aquatic environments from where it is transmitted to humans by an unknown mechanism. Activities that take place near water bodies, such as farming are risk factors, and wearing protective clothing appears to reduce the risk of the disease (8,9).

Buruli ulcer is often diagnosed and treated based mainly on clinical findings. Laboratory diagnoses are infrequently used to make decisions about treatment because of logistic and operational difficulties. However, direct smear examination, culture, polymerase chain reaction (PCR) and histopathology are recommended investigation means (4).

To the best knowledge of the authors, there is no of case Buruli ulcer reported from Ethiopia. We present the history, physical, laboratory, radiologic and pathologic findings are presented below and discussed.

CASE REPORT

SK, a 22 years old female patient from Arbaminch Zuria district of Gamogofa Zone presented with swelling and wound on left leg of 10 years duration. She came from one of the Chamo lake plane villages where year round irrigation based farming taken place (fig 1). SK had small prick by wood which latter swollen and ulcerated with gradual increment in size. She had no fever, pain or puss discharge from the wound site but had swelling in the left groin. Had no change in appetite weight and any cough or contact with chronically coughing person. Soon she visited Arba-Minch Hospital where she received amoxicillin and seven unknown injections to no avail. Four years latter she was referred to Black Lion Hospital where she was investigated. At the same time she also visited St. Paul Hospital where biopsy from the wound and skin was taken. In both hospitals, she was not told the diagnosis or given specific treatment but was scheduled for admission. Because of the long queue for bed, she went back home. There after, since she lost hope on modern medicine, she visited several traditional healers and holly waters. She reported as she usually applies tetracycline powder on the wound. As she is blood relative to one of the authors, she was brought to Jimma University Specialized Hospital for better investigation and management in 2009.

On examination her blood pressure was 118/78 mmHg, pulse rate 76 beats per minute, temperature 36.7°C, weight 61 kilograms and height 172 centimeters. Had pink conjunctivae, resonant and clear chest and no palpable abdominal organ but has left inguinal and femoral non-tender enlarged lymph nodes measuring 3 by 4 centimeters.

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The main finding was on the left leg which was grossly swollen with indolent, undermined ulcer on the lateral aspect and raised edges. The ulcer measured 8 by 12 centimeters. The skin on the non-ulcerated part appeared dark (fig 2). No sinus discharge but yellowish thin coating was visible on the surface.

Hematologic examination: White blood cell count was 10,000/mm^3 (neutrophil 49.8%, lymphocyte 35.6%, monocyte 14.6%), hemoglobin 14.8gm/dl, platelet 480,000/mm^3 and erythrocyte sedimentation rate 17 millimeter in the first hour.

Roentgenogram: X-ray of left leg showed cortical thickening with some lytic lesions on fibula (fig 3).

Biopsy: showed chronic inflammatory change.

With the diagnosis of Buruli ulcer, she was given anti-tuberculosis drugs. She took five drugs as it was difficult to initiate with Streptomycin and Rifampicin since the four anti tuberculosis drugs were combined. No therapeutic surgery was done for her.

At two months follow-up, the swelling and the ulcer decreased significantly. Unfortunately, no picture was taken during this visit.

DISCUSSION

The presented case was 12 years old by the time she acquired the disease which is in line with the fact that Buruli ulcer affects primarily children less than 15 years of age (2,3, 5-7). The presented case came from year round wet area goes with the fact that *Mycobacterium Ulcerans* is likely to occupy a specific place within aquatic environments from where it is transmitted to humans (8,9).

Figure 1. Lake Chamo and the surrounding, 2009

Though the exact mode of transmission is unsettled, some patients state that lesions develop at the site of antecedent trauma (10); like the present case where she reported history of trauma to her left leg before the disease. Study also indicated that some aquatic insects could transmit (10). There is no evidence that the disease can transmit from person to person. The incubation period of Buruli ulcer varies from approximately 2 weeks to 3 years, with an average of 2 to 3 months (11). Buruli ulcer starts as a painless swelling in the skin. In early or pre-ulcerative lesions, *M. ulcerans* produces a lipid toxin, mycolactone, which is responsible for necrosis of the dermis, panniculus, and fascia, culminating in extensive ulcers (12). Infection leads to extensive destruction of skin and soft tissue with formation of large ulcers usually on the legs or arms (13). Reports showed that there are different forms of Buruli ulcer. Though the ulcerative type is the commonest form; nodular, edematous,

contagious disseminated and metastatic forms/ stages are also well described (11,14). The disease can affect any part of the body, but in about 90% of cases the lesions are on the limbs, with nearly 60% of all lesions on the lower limbs which is true in the present case too (15,16). Because of the local immunosuppressive properties of mycolactone or perhaps as a result of other unknown mechanisms, the disease progresses with no pain and fever, which may partly explain why those affected often, do not seek prompt treatment (12). Without treatment, it results in massive ulcers like the presented case, with the classical, undermined borders. Sometimes, bone is affected causing gross deformities. When lesions heal, scarring may cause contracture of limbs and other permanent disabilities.

As there were no cases of Buruli ulcer reported from Ethiopia before, the diagnosis was delayed even though SK sought treatment early. As a result she stayed long with the disease which caused disfiguring of her leg and damaging the bones mainly the fibula. Though WHO recommendation is to administer streptomycin and rifampicin to treat Buruli Ulcer, we gave her five drug regimen as we couldn't get rifampicin alone. On the other hand, we continued the treatment for 6 months, the shortest duration of tuberculosis treatment as per the national guideline otherwise to be labeled as defaulter. She responded to the chemotherapy satisfactorily. The case of SK could be the tip of the iceberg where other cases either remained at home or not properly managed because of misdiagnoses.

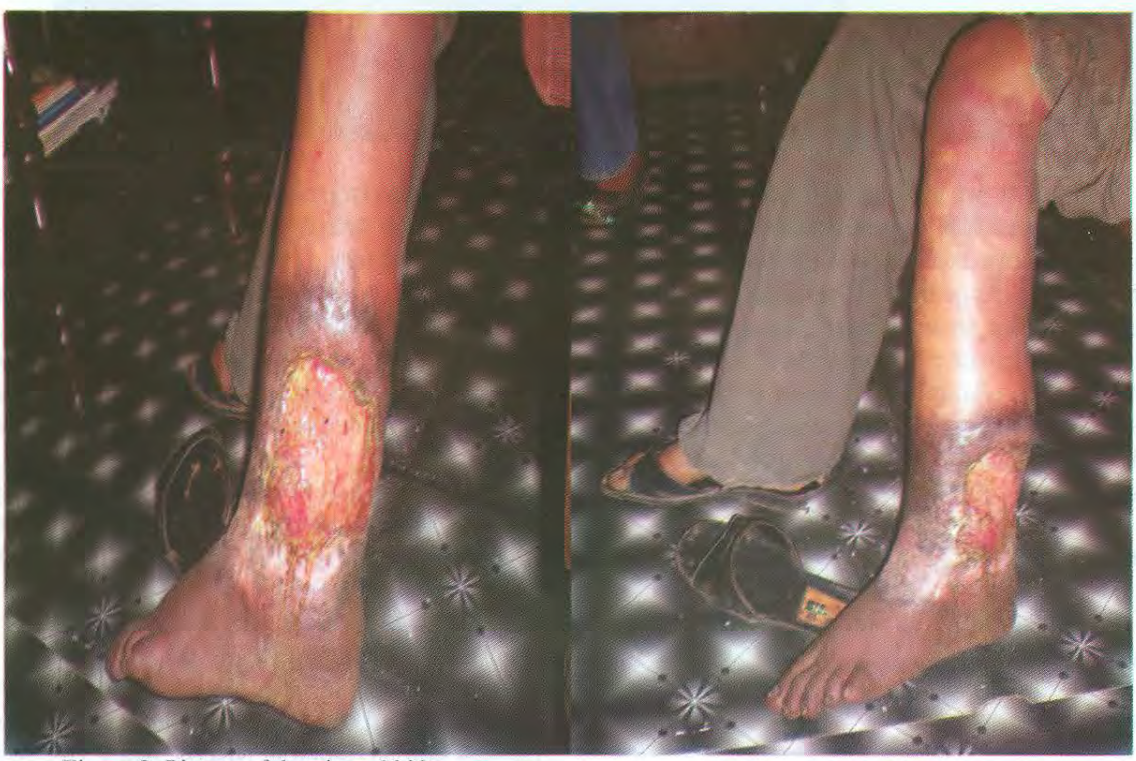

Figure 2. Picture of the ulcer, 2009.

Figure 3. Left leg X-ray showing soft tissue swelling, cortical thickening and lytic lesions of the fibula, 2009.

In conclusion, this case demonstrated the diagnostic and management difficulties of Buruli Ulcer in an area where there was no previous reported cases. Our observations highlighted the importance of the history and clinical presentation in considering Buruli Ulcer. Finally, all clinicians treating Buruli ulcer patients should stay well informed of developments in the antimicrobial treatment of the disease.

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