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D. L. KITARA, P. O. BWANGAMOI and E. I. ODONGO-AGINYA

SUMMARY

Buruli ulcers (BU) is a disease caused by infection with *Mycobacterium ulcerans*. It is one of the most neglected but treatable tropical diseases. Cases of Buruli ulcers are extremely rare in Gulu District. It is because of this reason that we report a case of a 25 year old male patient who presented with Buruli ulcers on the right thigh.

INTRODUCTION

Buruli ulcers (BU), is a disease caused by infection with *Mycobacterium ulcerans* (1). It is one of the most neglected but treatable tropical diseases (1,2). The causative organism is from the family of bacteria which causes tuberculosis and leprosy but Buruli ulcer has received least attention than these other two diseases (1,2). Infection leads to extensive destruction of skin and soft tissues with the formation of large ulcers usually on the legs or arms. Patients who are not treated early often suffer long-term functional disability such as restriction of joint movement as well as the obvious cosmetic problems (1-5). Early diagnosis and treatment are vital in preventing such disabilities (6-9). Buruli ulcer has been reported in over 30 countries mainly in the tropical and sub-tropical climates but it may also occur in some countries where it has not yet been recognised. Limited knowledge of the disease, its focal distribution and its occurrence mainly amongst poor rural communities contribute to low reporting of cases (9, 10). In 1897, Sir Albert Cook, a British physician working at the Mengo Hospital in Kampala, Uganda, described skin ulcers that were consistent with Buruli ulcer (BU)(1).

In 1948, Professor Peter MacCallum and his colleagues in Australia provided a detailed description of a similar disease among six patients from the Bairnsdale area near Melbourne (9,10). They were the first scientists to isolate the causative organism, *Mycobacterium ulcerans* (9). In Southern Australia, the disease is still referred to as the Bairnsdale ulcer (9). In the 1960s, many cases occurred in Buruli County (now called Nakasongola District) in Uganda, giving rise to the most widely used name for the disease – Buruli ulcer (8,9). Since 1980, the disease has emerged rapidly in several parts of the world, particularly in West Africa (9). Given the increasing geographical spread, severe consequences and limited knowledge of the disease, there is need for improved surveillance methods and control.

We discuss in this paper the characteristic clinical features, investigations, management and control of Buruli ulcer.

CASE REPORT

A 25 year old male peasant farmer from Koch Goma Sub County in Gulu District presented to Gulu Hospital Casualty unit during the night of February 2006 with a one month's history of an ulcer in the right thigh. He had moved to several health facilities for treatment but without any improvement. He gave a history that the ulcer started with a small itchy nodule at the upper right thigh. Because he was scratching on the nodule, three weeks later it developed into an ulcer. He reported to have been cultivating crops in the nearby the swamps. He also admitted that very often he made sleeping mat from papyrus reeds which he normally collected from the swamps. The proceeds from the sale of the mats was his additional family income. On examinations, he was in good general condition with a large ulcer in the upper right thigh, measuring over 25 centimetres in the longest diameter and was covering the anterior and medial portions. The wound was painless, was not causing him fever, the inguinal lymph nodes were not enlarged. The ulcer had undermining edges and the base was not fixed to the underlying structures and was not tender. The floor of the ulcer was dirty

with some scabs and debris (Figure 1).

Swabs were taken from the floor and edges of the ulcer. One was used for microscopy. Gram stain was performed for general bacterial infections while the Zhiel Neelsen (ZN) was performed specifically for the Acid Alcohol Fast (AAFB) mycobacterium. The other swabs were used for culture both for general bacteria on Blood agar and MacConkey agar while the third swab was inoculated on Lowenstein Jensen medium for mycobacteria. Both Gram and ZN stains revealed no mycobacterium. However the culture results revealed the presence of mycobacterium ulcerans. Similarly, biopsy of the ulcer edges was done and the histology result was comparable with the culture results.

Treatment: The patient was started on rifampicin and streptomycin for eight weeks and after three weeks of treatment, the ulcer was skin grafted and the rehabilitation process successfully conducted and patient discharged healed physically in ten weeks.

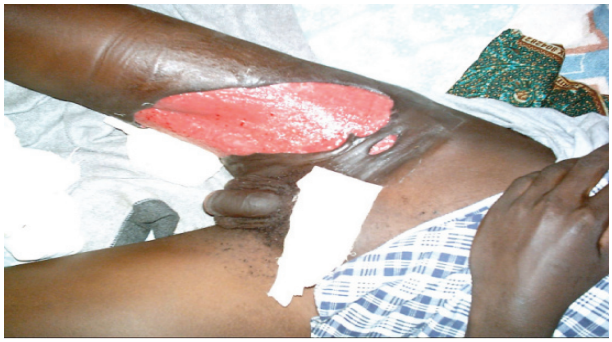
DISCUSSION

The true incidence of Buruli ulcers is not well known and although it was first described in Uganda, in the sixties, it had literally been eradicated out from the country. This case is particularly unique and the fact that it is a rare occurrence in this particular area in Uganda (1-3). *M. ulcerans* is an environmental mycobacterium. Recent information suggests that the organism does not live freely in the environment, as previously thought, but is likely to occupy a specific niche within aquatic environments (for example small aquatic animals, biofilms) from where it is transmitted to humans by an unknown mechanism(9). Although slow growing, *M. ulcerans* can be cultured from human lesions on Lowenstein Jensen medium for mycobacteria, at incubation temperature between 29–33°C. *M. ulcerans* produces a destructive toxin, mycolactone, which causes tissue damage and inhibits the immune response. The toxic effects of mycolactone explain most of the virulence of this organism (9, 10). The exact mode of transmission is still under investigation. Some patients state that lesions develop at the site of antecedent trauma (10). Research in Africa suggests that, some aquatic insects of the order Hemiptera (Naucoridae and Belostomatidae) can harbour *M. ulcerans* in their salivary glands and transmit the disease to experimental animals (9,10). More recent data from Australia suggest that salt marsh mosquitoes test positive for *M. ulcerans* DNA, although transmission by this type of mosquito has not been established(9). Further research is in progress to establish the exact role of insects and other factors in the transmission of the disease to humans. Buruli ulcer frequently occurs near water bodies – slow flowing rivers, ponds, swamps and lakes; cases have also occurred following flooding (9,10). Exposure risk factors of economic and social activities that take place near water bodies are the major source of infections. 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. 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. There is also no evidence that the disease can be transmitted from person to person (9, 10). Buruli ulcer has been reported from many countries in tropical and subtropical regions. In Côte d'Ivoire 24,000 cases recorded between 1978–2006 (9). In Benin, 7,000 cases recorded between 1989-2006; in Ghana more than 11,000 recorded since 1993. In Australia, more cases of BU are being reported 25 in 2004, 47 in 2005 and 72 in 2006. Most of the recent cases have come from the State of Victoria and the town of Point Lonsdale. Increasing number are being reported from West Africa and Uganda (9). Some patients have been reported from China, but the extent of the disease is not known (9,10). Recent reports suggest, for the first time, that Brazil may be endemic in the areas bordering French Guyana (10). These numbers may only be an indication of the presence of the disease but not the magnitude of the problem. For these and other reasons, it is difficult to establish the exact number of people affected by the disease and the size and location of all endemic areas (10). BU often starts as a painless, mobile swelling in the skin called a nodule. The disease can present as a large area of induration or a diffuse swelling in the legs and arms (9,10). Strains of *M. ulcerans* isolated from the different clinical forms of the disease in a particular geographical region appear identical, suggesting that host factors may play an important role in determining the different clinical presentations. 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. However, without treatment, massive ulcers result, with the classical, undermined edges. Sometimes bone is affected causing gross deformities. When lesions heal, scarring may cause restricted movement of limbs and other permanent disabilities in about a quarter of patients (10).

In Uganda, socio-cultural beliefs and practices strongly influence the health-seeking behaviours of people affected by BU. The first recourse is often traditional treatment. In addition to the high cost of surgical treatment, fear of surgery and concerns about the resulting scars and possible amputations may also prevail (1-5). Disfiguration stigma is a problem that also prevents people from seeking

early treatment. The long hospital stay, huge losses in productivity for adult patients which affects children educational opportunities. Bacille Calmette–Guérin (BCG) vaccination appears to offer some short-term protection from the disease although the protection is limited. Improved BCG-based vaccines, rational attenuation of a live *M. ulcerans* isolate or sub-unit vaccines, based on surface proteins or the toxin itself, are potential avenues for vaccine research. A safe and effective vaccine that can be targeted to newly emerging endemic areas may be the most effective way to combat BU in the long term (6-8).

Figure 1



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