

Rare Occurrence: Buruli Ulcers in Gulu, Northern Uganda. A Case report.**D.L.Kitara¹, P.B. Bwangamoi²**¹Senior Lecturer, Department of Surgery, Faculty of Medicine²Professor of Histopathology, Director of the institute of Research, Graduate studies & Staff Development, Gulu University,**Correspondence to:** David L Kitara, Email: klagoro@yahoo.co.uk

A Buruli ulcer (BU) is a disease caused by infection with *Mycobacterium ulcerans*. It is one of the most neglected but treatable tropical diseases. The causative organism is from the family of mycobacteriaceae which causes tuberculosis and leprosy. Buruli ulcer has received the least attention than these other two diseases. Its infection leads to extensive destruction of skin and soft tissue and formation of large ulcers usually on the legs or arms. Early diagnosis and treatment are vital in preventing such disabilities. We describe a case report of Buruli ulcer diagnosed using culture and histology and successfully managed in Gulu Regional Hospital using medical and surgical methods.

Introduction

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. Buruli ulcer is caused by infection with *Mycobacterium ulcerans*¹. It is one of the most neglected but treatable tropical diseases^{1,2}. Infection leads to extensive destruction of skin and soft tissues with the formation of large ulcers usually on the limbs. Patients who are not treated early often suffer long-term functional disability such as restriction of joint movement as well as cosmetic problems^{1,2,3,4,5}. Early diagnosis and treatment are vital in preventing such disabilities^{6,7,8,9}. Buruli ulcer has been reported in over 30 countries mainly in the tropical and subtropical climates but it may also occur in some countries where it has not yet been recognized. 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 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 ulcer⁹. 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 ulcers^{8,9}. Since the 60s, the disease has apparently disappeared from Uganda with the exception of few cases seen in the West Nile region. But since 1980, the disease has emerged rapidly in several parts of the world, particularly in West Africa⁹. We discuss in this paper, a case report of BU which was diagnosed and managed in Gulu Regional Hospital and make some suggestions on control strategy for the disease.

Case Report

DK a 25 year old male peasant farmer from Koch Goma Sub County in former 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 right upper thigh. Because of scratching the nodule, it developed into an ulcer three weeks later. He reported to have been cultivating crops in the nearby swamps in his area. He also reported that very often he made sleeping mats from papyrus reeds which he normally collected from the swamps. The proceeds from the sale of the mats was his additional family income. He was in good general condition, afebrile and with a large elliptical ulcer measuring over 25cm in the longest diameter and was covering the anterior and medial portions of the thigh, non-tender, with undermining edges, the base was not fixed to the underlying structures and the floor of the ulcer was clean with just some few scabs and debris and the inguinal lymph nodes were not enlarged and non tender.

Table 1. Characteristic Features of Buruli Ulcers in DK

Characteristics	Present
Painless ulcer	+
Undermining edges	+
Lower limb (site)	+
No palpable inguinal nodes in the area	+
No generalized symptoms	+

+ = is present in the case under review

Investigations

Swabs were taken from the floor and edges of the ulcer. One was used for general microscopy. Gram stain was performed for general bacterial infections while the Zhiel Neelsen (ZN) was performed specifically for the acid alcohol fast bacilli (AAFB). 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 and incubated at 30°C. Both Gram and ZN stains revealed no mycobacterium. However the culture 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 treated with Rifampicin (450mg) and Streptomycin (15mg/kg) daily for eight weeks. When six weeks of treatment had elapsed, the ulcer was skin grafted and the rehabilitation process successfully conducted and patient discharged healed physically in 12 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 this country. This case is particularly interest to us because BU is a rare occurrence in this particular area of Uganda^{1,2,3}. Mycobacterium Ulcerans is an environmental mycobacterium. Recent information suggests that the organism does not live freely in the environment, as was previously thought, but is likely to occupy a specific niche within aquatic environments (e.g. small aquatic animals, biofilms) from where it is transmitted to humans by an unknown mechanism⁹.

Buruli ulcer often starts as a painless, mobile nodular swelling in the skin or may present as a large area of induration or diffuse swelling in the legs and arms^{9,10}. 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 patients¹⁰. 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}. This typically explains the experience DK had throughout the course of the disease up to the diagnosis.

The exact mode of transmission of BU is not known. Some patients state that lesions develop at the site of antecedent trauma¹⁰. Buruli ulcer frequently occurs near water bodies – slow flowing rivers, ponds, swamps, lakes and 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 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}. In Uganda, sociocultural 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 prevail^{1,2,3,4,5}. 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 also affects children educational opportunities.

Recommended control strategies

Early clinical detection of cases at the community level, and community participation, Laboratory confirmation of cases, training of health workers and village health workers, case management (a combination of antibiotics, surgery and prevention of disability /rehabilitation), strengthening of health facilities, and monitoring and evaluation of control activities.



Figure 1. The case we reported for DK, 25 year old (Picture above reproduced with patient's permission)

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