PACIFIC JOURNAL OF MEDICAL SCIENCES
{Formerly: Medical Sciences Bulletin}
ISSN: 2072 – 1625

Pac. J. Med. Sci. (PJMS)
www.pacjmedsci.com. Email: pacjmedsci@gmail.com.

BURULI ULCER IN THE PROXIMAL RIGHT THIGH AND GROIN: A CASE REPORT

***David Lagoro Kitara and Paul Okot Bwangamoi**

Gulu University, Faculty of Medicine, Department of Surgery, Gulu, Uganda

***Correspondence author: klagoro@yahoo.co.uk**

BURULI ULCER IN THE PROXIMAL RIGHT THIGH AND GROIN: A CASE REPORT

***David Lagoro Kitara and Paul Okot Bwangamoi**

Gulu University, Faculty of Medicine, Department of Surgery, Gulu, Uganda

***Correspondence author: klagoro@yahoo.co.uk**

ABSTRACT:

We report a case of histopathologically proven Buruli ulcer (BU) in a 25-year-old man which was found at the proximal right thigh and groin – an unusual site of occurrence. Laboratory results including Gram and ZN stains were negative while a culture on Lowenstein Jensen media at 33^{oC} from the tissues produced a positive growth of Mycobacterium ulcerans. Histology of the edges of the ulcer showed a granulomatous lesion consistent with BU. This highlights the differentiation of Buruli ulcer from tropical ulcer and, to a lesser extent other forms of skin malignancies and benign skin lesions. The ulcer presented by the young man was Buruli ulcer.

Key words: Buruli Ulcer, Gulu Hospital, Diagnosis, tropical ulcer

Received: January 2012; Accepted: March 2012

INTRODUCTION

Buruli ulcer (BU) is caused by Mycobacterium ulcerans [1]. It is one of the most neglected but treatable tropical diseases [1,2]. The causative organism is from the family of Mycobacteriaecae which causes tuberculosis and leprosy but Buruli ulcer has received the least attention compared to 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 [2]. 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 [1,2,3,4,5]. Early diagnosis and treatment are vital in preventing such disabilities [6,7,8,9]. 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 Mengo Hospital in Kampala, Uganda described skin ulcers that were consistent with Buruli ulcer (BU) [1]. 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 1980, the disease has emerged rapidly in several parts of the world, particularly in West Africa [9]. We discussed in this paper the characteristic clinical features, investigations and compared Buruli ulcer with tropical ulcer.

CASE REPORT

A 25 year old male peasant farmer from Koch Goma Sub County in Nwoya district presented to Gulu Hospital in 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 nodule at the upper right thigh which he kept on scratching because it was itchy and three weeks later it developed into an ulcer. He also reported to have been cultivating crops in the nearby swamps for many months and that very often he made sleeping mats from papyrus reeds which he normally collected from the swamps for which proceeds from their sale were his additional family income. On examination, he was in good general condition with a large ulcer in the proximal right thigh, measuring over 25cm in the longest diameter and was covering the anterior and medial portions of the thigh. The wound had an undermining edge, non tender, no palpable inguinal lymph nodes and the base was not fixed to the underlying structures, the patient was afebrile and the floor of the ulcer was clean. Laboratory examination was conducted on swabs which 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 Acid Alcohol Fast (AAFB) Bacilli. 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 mycobacterium. Both Gram and ZN stains revealed no mycobacterium. However the culture result revealed the presence of mycobacterium ulcerans. Similarly, biopsy of the ulcer edges was done and the histology result was comparable with the culture results. The patient was treated with Rifampicin (10mg/kg once a day) and Streptomycin (15mg/kg once a day) because previous studies showed that the 2 drugs were bactericidal to tubercle bacilli [9, 21]. This treatment was for eight weeks and thereafter, the ulcer was skin grafted and the rehabilitation process completed.

Table 1: Clinical characteristic comparison between Buruli and Tropical ulcers [10,13,16,17]

Characteristics	Buruli ulcer	Tropical ulcer
Pain	–	+
Undermining edges	+	–
Lower limb (site)	+	+
Inguinal nodes	–	+
Generalized symptoms e.g. Fever	–	+

+ present; – absent

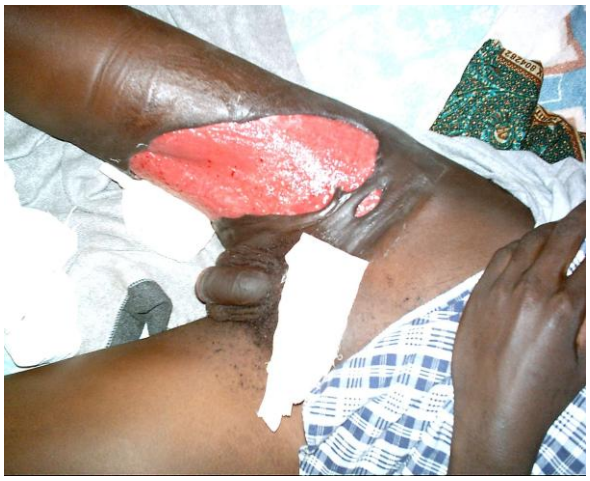

Fig. 1: Buruli ulcers in Gulu Regional Referral Hospital (Uganda)
This patient was a 25 year old boy who presented with Buruli ulcer to Gulu Regional Referral hospital and was managed in surgical department. He was treated with anti TB drugs (Rifampicin (10mg/kg once a day and Streptomycin 15mg/kg once a day) and wound dressing until when it was clean enough for skin grafting. Skin grafting was undertaken and patient's wound healed completely. The patient's condition could not be diagnosed by most health workers he visited before coming to Gulu Hospital. Secondly the ulcer occurred at an unusually uncommon site in the body of this patient (Normally, buruli ulcer occurs at exposed parts of the limbs.

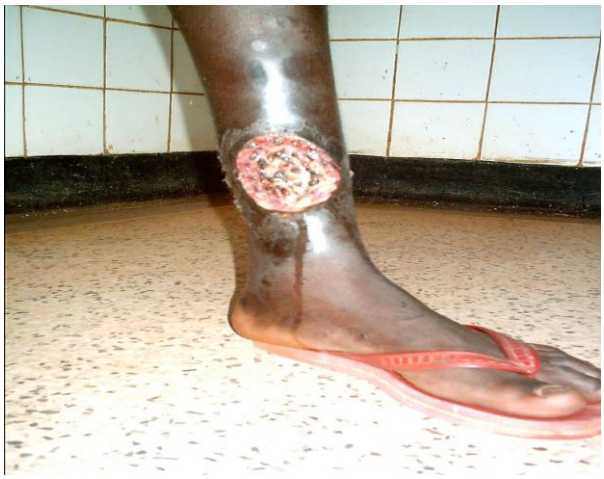

Fig 2: Tropical Ulcers in a 17 year old in Gulu Regional Referral Hospital
This patient was a 17year old boy who presented with tropical ulcer to Gulu Regional Referral hospital and was being managed in the outpatient surgical department. He was treated with broad spectrum antibiotics and daily dressing of the wound until when it was clean enough for skin grafting. His wound was skin grafted and it healed completely.

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 from the country [1]. This case report presents a particularly unique fact that it is a rare site (groin) of occurrence of the disease in this particular area of Uganda [1, 2, 3]. M ulcerans produces a destructive toxin, mycolactone, which causes tissue damage and inhibits the immune response [2,9,10]. The toxic effects of the toxin explain most of the virulence of this organism [9, 10]. 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 [9, 10]. 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 [9]. In Uganda, socio-cultural beliefs and practices strongly influence the health-seeking behaviours of people affected by BU [8, 9]. 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, 9, 10]. Disfiguration stigma is a problem that also prevents people from seeking early treatment and the long hospital stay, huge losses in productivity for adult patients which affects children's educational opportunities [1, 9, 10].

The possible differentials in this case report could be tropical ulcer and other rare skin lesions. Tropical ulcer though has distinct clinical features, such as presentation with painful ulcers below the knee, usually the ankle [11, 12, 13]. They are often initiated by minor trauma, and subjects with poor nutrition and poor hygiene are at higher risks [12, 14, 16, 17]. Once developed, the ulcer may become chronic and stable, but also it can run a destructive course with deep tissue invasion and osteitis [12,13,16]. Unlike Buruli ulcer in acute stage, tropical ulcers are very painful and aggravated by standing up [14,16,17]. Walking often causes venous congestion, leading to bleeding which can be quite severe. Pain and bleeding are both relieved by firm bandaging [16,17]. The pain and rapid rate of spread are characteristic of tropical ulcer; the thick rolled edge and the continuation into a chronic condition complete the clinical picture [15,16,17]. Lesions begin with inflammatory papules that progress into vesicles and rupture with the formation of an ulcer [16,17]. One of the major problems with tropical ulcers is the spread of infection leading to generalized septicemia or infection with tetanus and gas gangrene which are further complications [16,17]. The majority of tropical ulcers develop in the lower leg, overlying the tibia or the fibula and in the first 5 days, until a pustule discharges, there are usually no significant

radiological findings [16,18]. When the ulcer has developed there is usually marked soft tissue thickening, visible in profile radiography or with ultrasonography [15,16,17]. As the ulcer spreads, the earliest radiological finding in bone is a periosteal reaction which is localized immediately beneath the ulcer [16,17,18]. In most cases this begins as a minimal layered, fusiform periosteal reaction, but occasionally "sunray" spicules develop [16,17,18].

The periosteal reaction then blends with the original cortex to produce a thickened sclerotic layer which may be as much as 2.5 cm in thickness [18].

This ivory periosteal reaction can eventually involve the whole circumference of the bone and extend above and below the site of the ulcer and as it does so; it often becomes wavy and irregular, varying in width and in outline [18]. In some patients the next development is "ulceration" or, more strictly, sequestration of a localized superficial area of the periosteal new bone [18,19]. The sequestrum is sharply defined and may be separated from the underlying cortex and may eventually be discharged, resulting in a clear-cut, saucer-shaped defect [18,19]. The sharpness of this deficit is of considerable importance, because it is the main clue from which the radiologist can try to differentiate an infective process from malignant degeneration: when there is malignancy, the edge of the bone defect will be less regular and less well defined [18,19]. The same bone or adjacent bones may be affected some distance from the ulcer and may show periosteal reaction, with irregular widening and bony spicules quite remote from an ulcer in the anterior soft tissues and this presumably results from the soft tissue reaction in the leg [19,20]. A characteristic finding in some cases is calcification of the interosseous membrane between the tibia and the fibula [19]. Septic arthritis may occur depending on the locality of the ulcer [18,19,20]. When healing occurs there is considerable residual deformity, not only due to contraction of the skin and underlying soft tissues, but also because there may have been tendon and joint damage [13,14,19,20].

A flexion deformity of the knee or “talipes equinovarus” can develop and if scarring continues, the tibia and fibula may be drawn together by contraction and the bones may become thin and atrophied [19,20]. When ulceration is extremely chronic, deformity of the lower leg occurs and the bones may elongate and bow anteriorly or laterally [20]. The greatest convexity lies beneath the ulcer, the site of the osteoma and lymphoedema may result from the scarring [19,20].

Radiologically, the differential diagnosis of tropical ulcer in its early stages will include almost any cause of a localized periosteal response, but tropical ulcers may be distinguished because of the local soft tissue ulceration overlying them [18,19,20].

ACKNOWLEDGEMENT

We wish to acknowledge the support from Gulu Regional Referral Hospital and appreciate the willingness of the 2 patients to participate in this study at their successful management in Gulu Hospital.

Competing Interest: None

REFERENCES

1. Lunn HF, Connor DH, Wilks NE, et al. Buruli (mycobacterial) ulceration in Uganda. (A new focus of Buruli ulcer in Madi district, Uganda): Report of a field study. East Afr Med J.1965; 42:275.
2. Clancey JK, Dodge OG, Lunn HF, Oduori ML. Mycobacterial skin ulcers in Uganda. Lancet.1961; 2:951.
3. Clancey JK. Mycobacterial skin ulcers in Uganda: description of a new mycobacterium (Mycobacterium buruli). J Pathol Bacteriol.1964; 88:175.
4. Barker DJ. Buruli disease in a district of Uganda. J Trop Med Hyg. 1971; 74:260.
5. Barker DJ. The distribution of Buruli disease in Uganda. Trans R Soc Trop Med Hyg.1972;66:867.
6. Smith PG, Revill WD, Lukwago E, Rykushin YP. The protective effect of BCG against Mycobacterium ulcerans disease: a controlled trial in an endemic area of Uganda. Trans R Soc Trop Med Hyg. 1977; 70:449.
7. Portaels F, Aguiar J, Debacker M, et al. Mycobacterium bovis BCG vaccination as prophylaxis against Mycobacterium ulcerans osteomyelitis in Buruli ulcer disease. Infect Immun. 2004; 72:62.
8. BCG working Group. BCG vaccination against mycobacterium ulcerans infection (Buruli ulcer). First results of a trial in Uganda. Lancet 1969; 1:111.
9. Resolution WHA57.1 Surveillance and control of Mycobacterium ulcerans disease (Buruli ulcer). In: Fifty-seventh World Health Assembly, Geneva, 17–22 May 2004. Resolutions and decisions. Geneva, World Health Organization, 2004 (WHA57/2004/REC/1):1–2
10. Johnson PD, Hayman JA, Quek TY, et al. Consensus recommendations for the diagnosis, treatment and control of Mycobacterium ulcerans infection (Bairnsdale or Buruli ulcer) in Victoria, Australia. Med J Aust. 2007; 186:64.
11. Adriaans B, Hay R, Drasar B, Robinson D. The infectious aetiology of tropical ulcer--a study of the role of anaerobic bacteria. Br. J. Dermatol. 1987; 116(1):31–7.
12. Aribi M, Poirriez J, Breuillard F. Guess what! Tropical phagedenic ulcer. Eur J Dermatol.1999; 9(4):321–2.

13. MacDonald P. Tropical ulcers: a condition still hidden from the western world. J Wound Care. 2003; 12(3):85–90.
14. Gill, Geoffrey V, Geoff G, Beeching N. Lecture notes on tropical medicine. Oxford: Blackwell Science. 2004. ISBN 0-632-06496-X.
15. Odom RB, Davidsohn I, James WD, Henry JB, Berger TG. Clinical diagnosis by laboratory methods; Dirk M. Elston. Andrews' diseases of the skin: clinical dermatology. Saunders Elsevier.2006; 276–267. ISBN 0-7216-2921-0.
16. Adamson PB. Tropical ulcer in British Somaliland. J Trop Med Hyg. 1949; 52:68-75.
17. Blank H: Tropical phagedenic ulcer. Am J Trop Med Hyg. 1947; 27:383-396.
18. Brown JS, Middlemiss JH: Bone changes in tropical ulcer. Br J Radiol. 1956; 29:213-217.
19. Ennis JT, Gueri MC, Serjeany GR: Radiological changes associated with leg ulcers in the tropics. Br J Radiol.1972; 45:8-14.
20. Middlemias H: Tropical ulcer. In: Cockshott P, Middlemias H: Clinical Radiology in the Tropics. Churchill Livingstone, Edinburgh, 1979.
21. Tjip. Antimicrobial treatment for buruli ulcer is effective in early limited disease. Lancet. 2010. www.thelancet.com.