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Sequelae of *Mycobacterium ulcerans* infections (Buruli ulcer)

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Abstract Buruli ulcer, a type of mycobacterial infection, has recently emerged as an increasingly common and major cause of skin ulceration and associated morbidity, especially in the West African sub-region. In 1993, there was an outbreak of the disease in the Amansie West District of Ghana. From 1994 to 1998, 360 ulcers (already healed or active disease) were seen. Some of them were managed surgically at the Komfo Anokye Teaching Hospital (KATH), Kumasi and at two district hospitals all in the Ashanti Region. Many of the victims lost limbs (amputated), sight, breast, genitalia or they developed contractures of the toes, ankle joints, knee joints, wrists, fingers, elbows, etc. with subsequent lymphedema. Surgical excision of the ulcers with skin grafting resulted in wound closure. Secondary problems such as contractures were corrected by standard reconstructive methods.

Key words *Mycobacterium ulcerans* infection · Buruli ulcer · Sequela · Scar contractures

Introduction

Buruli ulcer or Bairnsdale ulcer is a chronic necrotizing cutaneous and subcutaneous ulcer caused by *Mycobacterium ulcerans* [2,3,4]. This microorganism belongs to the group that causes tuberculosis and leprosy. The name Bairnsdale ulcer originates from the district of Bairnsdale, Australia, where the disease was first observed in 1948 [3,4]. The disease is now popularly known as Bur-

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Fig. 1 Extensive ulceration (due to the Buruli ulcer) involving the chest and abdominal walls, the pubic region, the penis and the scrotum. Post excision and ready for grafting

Fig. 2 Post split-thickness skin grafting with a good result. This girl's left breast was totally destroyed by the *Mycobacterium ulcerans* infection. She will need a breast reconstruction

Fig. 3 Contracture release with extended physiotherapy gave acceptable function of the elbow. Note the persistent edema

Fig. 4 Contractures of the elbow and wrist with gross necrosis of the fingers. Apart from minor debridement, this child had no further operation as his parents refused to give their permission

Fig. 5 Above wrist amputation due to *Mycobacterium ulcerans* infection

uli ulcer, after the Buruli county in Uganda where it was frequently observed in 1953 [3]. Buruli ulcer is found mainly in tropical and subtropical regions, especially in swampy areas where the water is slow-flowing [5,6]. The disease often starts as a painless nodule or papule in the skin which, if not excised, leads to massive skin ulceration and fatty necrosis followed by debilitating sequelae such as contracture, amputation of extremities, and the loss of such organs as the nose, the breast, the genitalia and the eye.

Patients and methods

From 1994 to 1998, 360 Buruli ulcer cases were managed at the Komfo Anokye Teaching Hospital (KATH) in Kumasi and two nearby district hospitals, all in the Ashanti Region of Ghana. Some of these cases were diagnosed clinically (Fig. 1): ulceration with undermined edges, chronicity, failure to respond to traditional wound management – daily eusol dressings and oral antibiotic therapy. Some of them were confirmed by a positive Ziehl-Neelson test for Acid-Fast-Bacilli and/or by histopathology. Sixty-four percent were in males, 36% in females. The majority, 238, were in children up to 14 years of age, with 122 in adults. About 60% of the patients were of school age, with 33% aged 10–15 years (120) and 26% aged 5–10 years (96).

Table 1 Sequelae of Buruli ulcers

Sequela	Number	%
Lower extremity contracture (toes, ankle joint, knee of various degrees)	64	41.3
Upper extremity contracture (fingers, wrist joint, elbow of various degrees)	41	26.5
Amputations within the lower extremities (foot, toes, below/above knee)	18	11.6
Amputations within the upper extremities (fingers, wrist joint, upper arm)	16	10.3
Loss of eye (s)	5	3.2
Loss of eyelids (upper and/or lower, partly or fully)	6	3.9
Loss of nose (or part if it)	3	1.9
Loss of genitalia (or part ifthem)	2	1.3
Total	155	100

Results

Of the 360 patients seen and/or treated, 205 were treated by surgical excision followed by split-thickness skin grafting and healed uneventfully. The remaining 155 had the various sequelae outlined in Table 1.

Many of the patients who came into the hospital with long-standing ulcers required treatment of the sequelae. The elbow, wrist, and knee contractures were released and the defects covered appropriately, usually with thick/medium split-thickness skin coverage or a radial forearm (Chinese) flap in the case of elbow and wrist joint defects. The eyelid defects required sophisticated reconstructive procedures (Figs. 2–5).

Discussion

The severity of sequelae following *Mycobacterium ulcerans* infection is related to multiple factors. A major cause is the late presentation of patients to health facilities for treatment. In a survey conducted in one of the endemic areas, it was found that 60% of the victims attributed their delay in seeking medical treatment to poverty [1]. This contradicts the previous suggestion that the late reporting was mainly due to witchcraft and/or other local beliefs. Indeed, the latter reasons were given by only 6.5% of the patients [1].

Most of the sequelae could have been prevented by early management. For example, early physiotherapy and splinting in the case of the extremities can prevent contractures and limit the surgery necessary to treat these deformities.

Many patients suffered from malnutrition and severe anemia, causing delay in their surgery. During this time, the patient needed conservative treatment: physiotherapy, splinting, and local wound care.

Prevention of large ulcers could have been achieved if the Buruli ulcer nodules or early ulcers were adequately excised and grafted initially. Facial lesions, if excised and grafted early, can prevent spread to the eyes or eyelids.

Conclusion

The sequelae of *Mycobacterium ulcerans* infections cause significant deformities. Those patients who receive corrective surgery need some form of rehabilitation. Early diagnosis and subsequent surgical treatment coupled with physiotherapy and splinting of affected limbs will prevent these deformities.

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