

**BURULI DISEASE IN A DISTRICT OF UGANDA**

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Chronic necrotising skin ulcers caused by *Mycobacterium ulcerans* are a common and serious disorder in certain parts of the tropics. Usually known as Buruli disease this infection was first described in Australia and subsequently reported in Uganda, the Congo, West Africa, Malaya, Mexico and New Guinea. The method of transmission of the disease remains unknown. Following its original description in Uganda (Clancey, Dodge, Lunn and Oduori, 1961) it was found to occur in several parts of the country, all of which were close to the River Nile (Lunn, Connor, Wilks, Barnley, Kamunvi, Clancey, Bee, 1965; Connor and Lunn, 1966). At this time the restriction of the disease to sparsely populated areas, and the usual occurrence of only one patient in a family, gave rise to the suggestion that the disease was not transmitted from one person to another. Evidence from the subsequent intensive study of the disease at Kinyara refugee camp strongly supported this suggestion (Uganda Buruli Group, 1970).

Complete knowledge of the non-human source of *Myc. ulcerans* must wait upon its successful isolation from the environment. Meanwhile epidemiology offers an alternative method by which the source might be identified. This paper describes one of a series of epidemiological surveys which are being carried out in Uganda in an attempt to infer the distribution of the organism in nature by study of the distribution of the lesions in human populations.

## Methods

Busoga District lies on the East side of the Victoria Nile during its course from Lake Victoria in the South to Lake Kyoga in the North (Fig. 1).

The total population at the 1969 census was approximately 900,000 and the southern part of the district is more densely populated than the north. Patients with chronic skin ulcers can receive surgical treatment at either of the two government hospitals in the district, or at the mission hospital (Fig. 1). The records in

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these three hospitals were used to ascertain the name, age, sex, address and clinical details of all patients treated for Buruli disease up to the end of 1970.

Hospital records were found for 88 patients with Buruli disease, but six records were incomplete. Of the remaining 82 patients 59 were treated at the mission hospital and 23 at the main government hospital. All the mission hospital cases were treated by one doctor, except during her leave periods, and all the government hospital cases were under the care of one consultant surgeon. For this reason it has been possible to make use of hospital records in this study. The first cases of the disease were recorded in 1965 at the mission hospital and in 1967 at the government hospital. In 17 of the mission hospital cases and all of the government hospital cases the diagnosis was confirmed by biopsy.

Out of the 82 patients with complete records a sample of 27 was selected for home interview. One was untraceable and of the others in only one case was the address given in the hospital records different from the address at which the patient stayed during the presumed incubation period of the disease, that is from two to five months before the first appearance of the lesion (Uganda Buruli Group, 1970). A further 10 patients were in hospital at the time of the survey, during 1970, and their addresses were verified there.

Each of the 15 dispensaries in Busoga (Fig. 1) was visited after the local people had been informed that a doctor was coming to treat skin ulcers of all kinds. One hundred and ninety-seven patients with skin ulcers, mainly tropical ulcers, were treated but only two cases of Buruli disease not already known from hospital records were found. The leprosy assistants were informed about the disease and asked to notify suspicious lesions among patients attending the leprosy posts in the district. One new case was found in this way. There was therefore no evidence that there were large numbers of unrecognised cases in the district who were not being referred to hospital. No records of Buruli cases from Busoga were found in the hospitals in adjacent districts.

TABLE I. DISTRIBUTION OF CASES BY AGE, SEX, AND SITE OF LESION ON BODY.

Site of lesion	Below 5		5 - 9		10 - 14		15 and over		All ages	
	M	F	M	F	M	F	M	F	M	F
Arms	2 (50)	5 (50)	3 (23)	5 (36)	2 (25)	3 (60)	3 (33)	7 (37)	10 (29)	20 (42)
Body and head	0 (-)	4 (40)	3 (23)	3 (21)	0 (-)	0 (-)	2 (22)	3 (16)	5 (15)	10 (21)
Legs	2 (50)	1 (10)	7 (54)	6 (43)	6 (75)	2 (40)	4 (44)	9 (47)	19 (56)	18 (38)
All sites	4	10	13	14	8	5	9	19	34	48

The figures in parentheses are the percentages of lesions at a given site out of all lesions within each age and sex group.

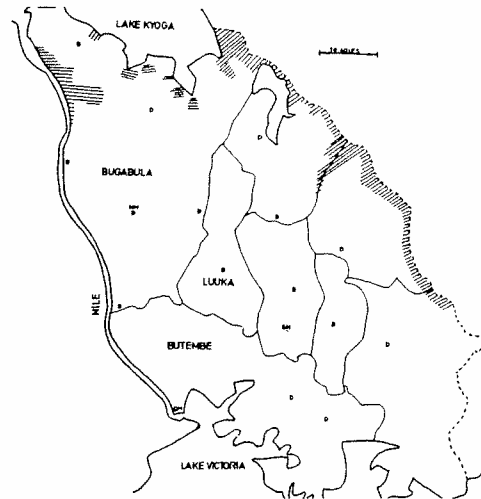


Figure 1.

Busoga District

MH=Mission Hospital.  
GH=Government Hospital.  
D =Dispensary.

### Results

#### Age, sex and site of lesion on the body

Table 1 shows the age and sex distribution of the patients and the site of the lesions on the body. The disease occurred in all age groups but was most common among children, 66 per cent of the patients being less than 15 years old. There were more females affected (48 cases) than males (34 cases) with more than twice as many females as males in the 15 and over age group. Eighty-two per cent of the lesions were on the limbs; but whereas in males there was a preponderance of leg lesions, with 19 leg lesions compared to 10 arm lesions, in females there were 18 leg lesions and 20 arm lesions.

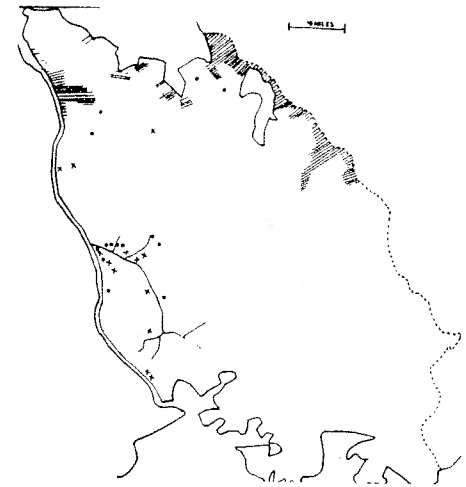


Figure 2.

Distribution of patients with Buruli disease treated during 1965 - 68.  
0 cases in 1965 - 66.  
x cases in 1967 - 68.

#### Geographical distribution

The distributions of the homes of patients treated in the years 1965 - 68, and 1969 - 70, are shown in Figures 2 and 3 respectively. Buruli disease is confined to three of the eight counties into which Busoga is divided. With the exception of two patients living near to Lake Kyoga the disease does not occur at more than 20 miles distance from the River Nile. Most cases come from the north, where the land is flat, swampy, and sparsely populated rather than the south where the land is hilly and more densely populated. Seventy-two out of the 82 cases occurred in Bugabula County (Figure 1), making a total incidence of 34/100,000 population in the county, whereas in Butembe County there were six cases, total

DDY.

all ages

F  
20  
(42)  
10  
(21)  
18  
(38)  
48

each age

TABLE II. AGE OF PATIENTS ACCORDING TO THE DISTANCE OF THEIR HOMES FROM THE RIVER NILE.

Distance from Nile	Below 5	5 - 9	10 - 14	15 and over	All ages
Less than 5 miles	3 (8)	12 (32)	5 (14)	17 (46)	37
More than 5 miles	11 (24)	15 (33)	8 (18)	11 (24)	45

The figures in parentheses are the percentages in each age group out of all patients living at that distance from the Nile.

incidence 4/100,000 population, and in Luuka County four cases, total incidence 5/100,000 population. The 23 cases treated at the government hospital were not confined to those patients living nearest to the hospital, but were drawn from the entire area. Seventeen of the 26 patients interviewed at home had developed the disease within the previous year. They were interrogated in detail about their activities during the five month period before the disease was first noticed. Only one of them had been to the Nile during this period. Therefore although the distribution of the disease is related to the Nile its occurrence is not dependent on the patient visiting the river. This finding is supported by the analysis shown in Table II, where patients are divided into two groups according to whether their homes are more or less than five miles from the Nile. Among the group living more than five miles away there is not the preponderance of older, and therefore more mobile, people one might expect if it was necessary for these people to have travelled to the Nile before contracting the disease.

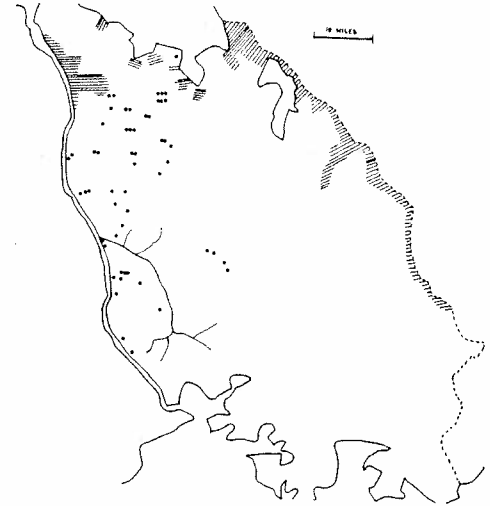


Figure 3. Distribution of patients with Buruli disease treated during 1969-70.

*Annual variation in distribution*

In 1965-66 14 cases of Buruli disease were admitted to hospital (Fig. 2). They all lived within a few miles of the Nile, Lake Kyoga or the River Kiko, the only large river entering the Nile in Busoga. The distribution of the 13 cases treated in 1967-68 was very similar (Fig. 2). But in 1969-70 the number of cases increased sharply to 55, and many of these cases came from areas in North Busoga which are up to 20 miles from the Nile, Kiko or Lake Kyoga.

**Discussion**

The distribution of age, sex and site of lesion among Buruli patients in Busoga is in accord with that observed at the Kinyara refugee camp in Bunyoro District (Uganda Buruli

Group, 1970). Buruli disease is more frequent among adult females than adult males; and the majority of lesions occur on the limbs, with a preponderance of leg lesions in males, and equal number of leg and arm lesions in females. The incidence of the disease in Busoga is low; and the annual incidence of 0.02 per cent for Bugabula County in 1970 may be compared with that of 1.6 per cent for Ibutje sub-county, Lango (Revill, 1970) and 3.2 per cent among Tsetse Control workers in North-east Bunyoro (Barker 1971).

The early reports of Buruli disease in Uganda all came from areas within a few miles of the River Nile (Connor and Lunn, 1966). The centre of Kinyara refugee camp was only two miles from the Nile. Among refugees who developed the disease some said that they had never been to the river; but one



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could not be sure about the truth of this information since it was difficult to communicate with the Ruandans, whose language is foreign to Uganda, and all the refugees lived within easy walking distance of the river. But in Busoga patients lived up to 20 miles from the river; among those subject to detailed interviews at home only one said he had been to the river; and there is no difference between the age distribution of cases living less than five miles or more than five miles from the river. These findings give strong support to the conclusion that the environmental source of *Myc. ulcerans* is not confined to the river's edge.

The observations on Buruli disease in Busoga have revealed three hitherto unrecorded features of the epidemiology of the disease in Uganda.

First, the disease was not recorded in the district before 1965, except for two isolated cases who were treated in a hospital outside the district but whose recorded addresses were in Busoga, close to the Nile (Clancey, Dodge, Lunn and Oduori, 1961). There is no evidence to suggest that this sudden appearance of the disease was due either to changes in the staffing or diagnostic practice in the Busoga hospitals, or to a new willingness of the people to accept modern rather than traditional medicine.

Secondly, all the early cases lived within a few miles of the River Nile, or its main tributary the Kiko, or Lake Kyoga. It seems unlikely that this can be accounted for in terms of the hospitals being more accessible to people in these areas, or the people being more willing for other reasons to seek hospital treatment. Some of the cases treated in 1965-68 came from remote and sparsely populated places in the north, whereas the places in which the disease appeared for the first time in 1969-70 have easy access to the mission hospital.

Thirdly, there was a marked increase in the number of patients in the district during 1969-70, associated with an apparent extension of the disease area away from the rivers and lake. The number of recorded cases rose from 13 in 1967-68 to 55 in 1969-70. This increase cannot be attributed to a change in the diagnostic or record keeping procedures in the hospitals, and it seems likely that it was due to an increase in the incidence of the disease in the population. In Lango District, Revill (1970) observed a similar recent

increase in the number of patients with Buruli disease seeking medical attention. He concluded that this reflected a real increase in the incidence of the disease; but it is not yet known whether this increase was due to an extension of the area in which the disease occurred, or an increase in the number of cases occurring in the same disease area. In Busoga the findings suggest that the incidence has remained constant in the areas where the disease first appeared, but there has been a recent extension of the disease area into the swampy land in the north.

In an attempt to explain these findings one can speculate that the appearance of Buruli disease in Busoga in 1965, in areas related to the Nile, Kiko and Lake Kyoga was related to the unprecedented flooding of the rivers and lakes of Uganda in 1962-64. At that time there were heavy rains, and the water level on the Busoga shore of Lake Victoria, for example, rose from 10-11 metres, which had been the constant levels since 1899, to 12-13 metres; and these elevated levels have persisted until the present time. In low-lying areas such as North Busoga there was flooding of the land with consequent marked ecological changes, of which one obvious manifestation was the sharp increase in the number of tsetse flies (Wooff, 1970). We have interviewed groups of local people in 21 villages in Busoga in order to find out what changes the floods caused. Many seasonal swamps became permanent and poorly drained soil became wetter, resulting in an enhancement in soil fertility for the growth of certain crops, and a marked increase in the height and density of grass growth. All except one of the 27 cases of Buruli disease treated in Busoga hospitals during 1965-68 (Fig. 2) lived within easy walking distance of areas affected by the flooding in 1962-64. Despite the high population density near to the Nile in South Busoga only six cases of the disease have been recorded from Butembe County (Fig. 1), which is mainly upland that was unaffected by the floods.

The hypothesis that some change produced by the flooding in 1962-64 led to the appearance of Buruli disease in Busoga can be related to a recent suggestion that *Myc. ulcerans* occurs on certain species of grass growing in poorly drained soil, and that Buruli disease is contracted by contact with grass and subsequent penetration of the organism through small scratches or other traumatic lesions in the skin. (Barker, Clancey, Morrow, and Rao, 1970). There is evidence in support

of this hypothesis. Buruli lesions usually occur on parts of the body which are unclothed and therefore come into contact with vegetation. Among adults the disease is more frequent in the women, who have to spend more time than the men in the bush in order to carry out such activities as collecting firewood and fetching water from swamps, streams or pools. In Ibuje sub-county, Lango District, there is a lower incidence of the disease within trading centres, which are usually clear of grass. Grasses collected from Buruli areas have been shown to be an abundant source of mycobacteria. Most of the mycobacteria so far isolated have not yet been identified, although some of them are obviously dissimilar to *Mycobacterium ulcerans* in their temperature and media requirements for growth in the laboratory.

There is no conclusive evidence to support the hypothesis that Buruli disease is contracted by contact with grasses. However, one may speculate that before the flooding *Mycobacterium ulcerans* was confined to habitats such as grass swamps, where it was closely related to the rivers and lake and relatively inaccessible to man. After the floods had subsided in 1964 it became established on grasses which grew up on the land that had been inundated. It thereby became more accessible to man and, in consequence, cases of Buruli disease began to appear in hospitals in Busoga in 1965. Having become established in areas of poorly drained soil on the rivers' edge and lake shore it subsequently extended its distribution onto grasses growing in similar areas away from the rivers and lake, so that in 1969-70 there was an increase in the endemic area of the disease.

These findings in Busoga may have implications for the other districts of Uganda where Buruli disease occurs, Lango, Bunyoro, Madi and East Mengo. Here the disease also occurs in low-lying areas within 20 miles of the Nile and large numbers of cases have only been reported since the 1962-64 flooding. If the disease has extended its distribution in Busoga one must wonder whether this has not also occurred in the other districts, and whether the disease will not continue to spread in the coming years.

#### Summary

This paper describes the epidemiological findings on 82 patients with Buruli disease, who represent all cases of the disease that have

been treated in the hospitals of Busoga District. The findings suggest that the disease became endemic in the district after the flooding of the rivers and lakes in 1962-64. All the early cases lived near to the Nile, its main tributary, or Lake Kyoga. In 1969-70 there was a marked increase in the number of cases in the district, associated with an apparent extension of the disease area into the swampy land in the north of the district. These findings may have implications for the other districts of Uganda where Buruli disease occurs, and they can be related to the hypothesis that the disease is contracted by contact with grasses growing in poorly drained soil.

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