

## Treatment of elephantiasis in a community with timorian filariasis

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### Abstract

To elucidate the issue of lymphoedema and elephantiasis in our previous reports, all data on the two clinical conditions have been reviewed and analysed. The various characteristics of the swelling are described and factors that may influence the outcome of treatment with diethylcarbamazine (DEC) analysed. The results showed that all cases with lymphoedema could be treated with DEC, irrespective of the size of the swelling, and that in most cases the swelling disappeared within one year. On the other hand, it required at least two to four years for most swelling to disappear in people with elephantiasis. Elephantiasis of the arms was easier to treat than of the legs. Bilateral elephantiasis of the legs were more difficult to treat than unilateral elephantiasis. Elephantiasis of less than three to five years' duration were easier to treat than that of longer duration. Individuals with a higher grade of elephantiasis were more difficult to treat than those with a lower grade of elephantiasis. The age and sex of patients did not influence the outcome of treatment.

### Introduction

Long term evaluation of filariasis control using diethylcarbamazine (DEC) was initiated in 1977 in several small villages in West Flores, Indonesia, endemic for *Brugia timori*. Initial results have been published earlier (PARTONO *et al.*, 1981, 1984) with the major emphasis on control strategies. The basic concept of treating individuals each time they had an attack of adenolymphangitis during the control programmes was to prevent future development of chronic lesions and microfilaraemia. The issue of treating those with elephantiasis was originally not considered. It happened that most people with these lesions experienced episodic attacks of adenolymphangitis and had therefore been repeatedly treated with DEC for the last four to five years. This paper reports the results of treatment of these individuals with emphasis on describing the various characteristics of the swelling and analysing the various factors that may influence the outcome of treatment with DEC.

### Materials and Methods

#### Study population

The study population consisted of 74 persons with persistent lymphoedema or elephantiasis, 36 from Karakuak, six from Sengari, 16 from Wangkung and 16 from Waemanis, on the island of Flores in the lesser Sundas, Indonesia. Individuals with intermittent lymphoedema were not included in this evaluation. During the study period, five persons left the area or died; of the remaining, 36 were males and 33 were females ranging in age from 6 to 80 years.

#### Location and duration of swelling

43 persons had swelling of one leg, 20 had swelling of two legs, two swelling of one leg and one arm, two swelling of two legs and one arm, one swelling of two legs and two arms and one swelling of one arm. The total number of affected extremities were 98. At the beginning of the study, the swelling had been established on the affected extremities for a period of three months to 22 years. Six extremities had been swollen for less than one year, 47 between one to two years, 16 between three and four years, 15 between five and nine years, 13 between 10 and 19 years and one for more than 20 years.

#### Lymphoedema and elephantiasis

A distinction was made between lymphoedema and

elephantiasis. The clinical diagnosis of lymphoedema was based on the following criteria.

The skin on the affected extremity was thin, shiny and of normal texture. There was obvious pitting oedema and the swelling had a smooth contour.

The criteria for elephantiasis were thickening of the skin, swelling with slight or no pitting oedema, or local bulging of the extremities. Local bulging was thought to be an indication of subcutaneous fibrosis, which locally bound the skin to the underlying tissues. Elephantiasis was further categorized into three subgroups; grade I with slight swelling, grade II with obvious swelling and grade III with obvious swelling and local bulging of the affected extremity. A person who had lymphoedema in one extremity and elephantiasis in another was considered as having elephantiasis. There were 20 persons with lymphoedema and 49 with elephantiasis. 25 extremities had lymphoedema, and 36 had grade I, 21 grade II and 16 grade III elephantiasis.

#### Association with adenolymphangitis and microfilaraemia

63 of the 69 persons with lymphoedema or elephantiasis had episodic attacks of adenolymphangitis. The attacks ranged from one to 12 per year during the study period. There were nine persons with microfilaraemia.

#### Treatment

In Karakuak most people had received two courses of DEC (5 mg/kg/day for 10 days) during two rounds of mass treatment. In the other villages DEC was distributed weekly in low dosage on a mass scale by the villagers for a period of 18 months (PARTONO *et al.*, 1984). In addition, motivated persons in the communities were trained to treat all cases with acute adenolymphangitis each time they had an attack. The dosage was  $3 \times 100$  mg DEC per day for 10 consecutive days for each person 10 years old and above. Children younger than 10 years received half of the above dosage. Within a period of four to five years, the number of DEC courses a person received varied from one to 21. 34 persons received one to four courses, 24 between five and nine, six between 10 and 14, four between 15 and 19, and one over 20 courses of DEC.

### Results

All individuals with lymphoedema had complete regression of swelling within a period of three years, irrespective of the size of the swelling (Table I). In most cases the swelling disappeared within one year. However, the swelling of persons with elephantiasis

Table I—Number of persons whose swelling disappeared after repeated treatments with DEC

Swelling	Disappeared within					Did not disappear
	1 year	2 years	3 years	4 years	5 years	
Lymphoedema	16	1	3	—	—	—
Elephantiasis	7	13	8	6	1	14

Table II—Effects on lesions in the 14 persons with elephantiasis who remained affected after repeated treatments with DEC

Case No.	Elephantiasis	No. of DEC courses	Results
K2	RL-6*-III**	8	unchanged
K63	RL-3-III, LL-6-III, RA-1-II	11	RL-II, LL-III
K66	RL-10-II, LL-10-III	4	unchanged
K92	RL-4-III, LL-1-III	18	RL-II, LL-III
K111	RL-7-III, LL-13-II, RA-7-I	12	RL-III, LL-I
K136	RL-5-II, LL-7-III	9	unchanged
K173	RL-2-II, LL-12-III, RA-1-III LA-1-III	21	RL-II, LL-III
Y12	RL-1-I, LL-1-I	6	RL-I
Y43	RL-2-1, LL-2-II	9	RL-I, LL-I
S150	RL-13-I, LL-13-II	13	unchanged
W37	RL-3-III, LL-1-I	19	unchanged
W63	LL-4-III	14	unchanged
W100	RL-10-III, LL-9-II	9	unchanged
W160	RL-3-II	14	unchanged

RL = right leg; LL = left leg; RA = right arm; LA = left arm

\*Duration of swelling in years

\*\*Degree of elephantiasis

Table III—Factors that may influence the outcome of treatment with DEC among individuals with elephantiasis

Factors		$\chi^2$	p
<i>No. of affected extremities</i>			
extremity v.	more than 1		
24*/27	11/22	8.98	<0.01
<i>Duration of swelling</i>			
less than 3 years v.	3 or more		
25**/31	24/42	4.46	<0.05
less than 5 years v.	5 or more		
36/47	13/26	5.37	<0.05
less than 10 years v.	10 or more		
42/59	7/14	2.30	>0.10
<i>Grade of elephantiasis</i>			
grade I v.	grade II		
32**/36	13/21	5.81	<0.02
grade II v.	grade III		
13/21	4/16	4.97	<0.05
grade I + II v.	grade III		
45/57	4/16	16.48	<0.001
grade I v.	grade II + III		
32/36	17/37	15.25	<0.001
<i>Sex of patients</i>			
males v.	females		
20*/28	15/21	0.00	>0.99
<i>Age of patients</i>			
less than 20 years v.	20 or more		
11*/16	24/33	0.08	>0.70
less than 30 years v.	30 or more		
22/31	13/18	0.01	>0.90

\*No. of persons cured/No. of persons affected

\*\*No. of extremities which regressed to normal/No. of extremities affected

disappeared more slowly. It required two to four years for most swellings to regress completely.

There were 14 persons with elephantiasis who remained affected after treatment with DEC; 13 of this group continued to suffer from episodic adenolymphangitis. The characteristics of the swelling, the number of treatment courses and the results of treatment are shown in Table II. Swelling of the arms disappeared completely in all affected cases. In some cases with bilateral involvement, one leg returned to normal appearance or became smaller while the other remained unchanged.

Table III shows the various factors that may influence the outcome of treatment of people with elephantiasis. Bilateral involvement of the legs were more difficult to treat than unilateral elephantiasis. Elephantiasis with a duration of less than three to five years were easier to treat than that of three to five years duration or more. The higher the grade of the elephantiasis, the more difficult it was to treat. Grade III elephantiasis was the most difficult to treat. The age and sex of the treated individuals did not influence the results of treatment.

During mass treatment with DEC, four persons developed lymphoedema one year after treatment. However, in all cases the swelling disappeared on subsequent follow-up evaluations.

#### Discussion

In previous reports, which mainly emphasized control strategies of filariasis, we had classified lymphoedema and elephantiasis into one group, although on the data recording sheet a distinction was made between these clinical conditions. There were several considerations for this decision. Both conditions were caused by the same pathological process and there were no distinct boundaries between them. In addition, villagers who worked knee-deep in the mud were prone to constant mechanical trauma resulting in thickening of the skin on the extremities. Consequently, the differentiation of lymphoedema and elephantiasis among this group was more difficult. However, the main reason for collectively grouping the two clinical conditions was a retrospective decision. Following treatment with DEC, several individuals who were recorded as having elephantiasis previously, had no swelling of the extremities on subsequent clinical evaluation. The number of persons belonging to this group continued to increase on each successive follow up study.

Subsequent to our published reports and discussion, we were asked to elucidate the issue of

lymphoedema and elephantiasis since each clinical condition denoted a different prognostic connotation. An outstanding issue is whether elephantiasis with fibrosis can be treated with DEC. The answer to this question is affirmative based on the following results:

- (i) The response to treatment of those with lymphoedema was different from those with elephantiasis. Most lymphoedematous swelling disappeared within one year, irrespective of the size of the swelling. However, it required two to four years for the swelling to disappear in cases with elephantiasis, and the degree of swelling was an important factor in determining the outcome of treatment.
- (ii) Four persons with grade III elephantiasis became normal after treatment.
- (iii) Swelling which lasted for 10 years or more disappeared in 7 of 14 extremities.

It was not clear why DEC was so much more effective in treating cases of elephantiasis than found in most previous trials. It could be that *B. timori* is more susceptible to treatment. Repeated treatment with DEC given immediately during each attack of adenolymphangitis over a long period of time might also be responsible for the effective results.

#### Acknowledgements

This study was supported by the Filariasis component of the UN Development Programme/World Bank/WHO Special Programme for Research and Training in Tropical Diseases. The author wishes to thank Professor Bintari Rukmono, Chairperson, Mr. Purnomo and Professor Sri Oemijati of the Department of Parasitology, University of Indonesia, Dr. H. Fernandez, Chief, Nusa Tenggara Timur Provincial Health Office, and Dr. A. Soewarta for supporting this study, and Dr. John H. Cross, Scientific Director, U. S. Naval Medical Research Unit No. 2 for reviewing this manuscript.

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Accepted for publication 20th February, 1984.