

A REVIEW OF SCHISTOSOMIASIS IN INDONESIA WITH REFERENCE TO *SCHISTOSOMA JAPONICUM*

Lim Boo Liat¹ and M. Sudomo²

ABSTRAK

Tinjauan tentang *Schistosoma* di Indonesia ini mencakup *Schistosoma japonicum*, *S. incognitum*, *S. spindale* dan *Trichobilharzia brevis*. Tinjauan dibuat atas dasar laporan-laporan penelitian yang telah diterbitkan. Di dalamnya dapat dijumpai uraian singkat tentang *S. spinale* dan *T. brevis*. Dari banyak publikasi tentang *S. japonicum* dan *S. incoganitum* dapat disajikan uraian tentang peranan kedua parasit tersebut sebagai penyebab penyakit baik manusia maupun hewan.

INTRODUCTION

Schistosomiasis, sometimes called bilharziasis, is one of the most widespread of all human parasitic diseases. It ranks second in terms of social economic and public health importance in tropical and sub-tropical areas, immediately behind malaria. In terms of prevalence, it takes first place among the waters borne diseases, representing one of the major health risks in rural areas of developing countries.

Schistosimiasis is a three-factor complex, involving and agent, an intermediate molluscan host and a mammalian definitive host (Fig. 1).

It is caused by certain digenetic trematodes which choose as their habitat the venous system of man. There are

two clinical forms of the disease, the vesical and the intestinal. In the oriental the disease, which is an intestinal forms, is known as oriental schistosomiasis, Katayama fever, bilharziasis japonica and Yangtze Valley fever. The disease is accompanied by toxic and dysenteric symptoms, as well as loss of appetite and weight, emaciation and retarded growth of young patients. The most characteristic of the disease, are enlarged liver and spleen, and often ascites usually more pronounced in the oriental form than in intestinal schistosomiasis of Africa, Central and South America.

Four species of *Schistosoma* have been reported from this archipelago. They are the classical *Schistosoma japonicum*, *S. incognitum*, *S. Schistosoma japonicum*, *S. Spindale* and *Trichobilharzia brevis*.

1. C/O Institute for Medical Research, Kuala Lumpur, Malaysia.

2. Health Ecology Research Centre, National Institute of Health Research and Development, Jakarta, Indonesia.

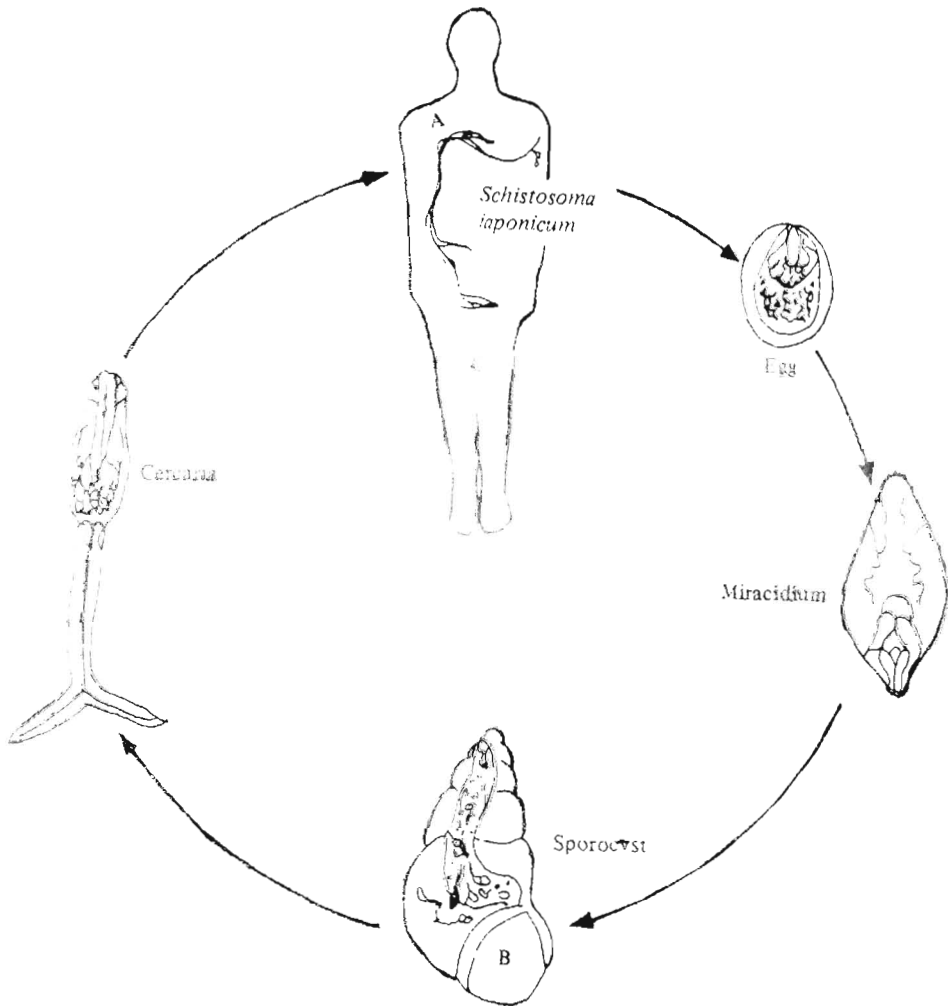


Fig. 1. Life cycle *Schistosoma japonicum*.

A. Copulating pair of *S. japonicum* inhabiting the upper mesenteric veins of definite host.
 B. Intermediate molluscan host, *Oncomelania hupensis lindoensis*. C. Definitive host represented by man, although any mammal that enters infested waters will provably serve as a definite host. Eggs of *S. japonicum*, after being passed from female worm into small veins, make their way into bowel of host, pass out with faeces, and, if deposited in aquatic habitat, hatch, and release a motile miracidium. If miracidium comes in contact with an oncomelaniid snail, it will penetrate and go through a series of asexual multiplications resulting in the production of larval worms, known as cercariae. These escape into an aquatic environment, and penetrate the skin of mammals frequenting that habitat. Within the definitive mammalian host the cercaria metamorphoses into an adult schistosome, mates and, thus, completed the life cycle.

Of these, *S. japonicum* is the most important in terms of human health and its biology is best understood. In this review, brief mention is made of what is known about *S. Spindale* and *T. brevis* and then the discussion concentrates on *S. japonicum* and *S. incognitum*. The classical form of *S. japonicum* from *S. japonicum* like schistosomes of Indonesia and their respective roles in the etiology of human and animal disease are also discussed.

CLASSICAL SCHISTOSOMA JAPONICUM

Preliminary studies of schistosomiasis in Indonesia were made in the late 1930's and the early 1940's. The first human case of *S. japonicum* was discovered by Muller and Tesch¹ from Lindu valley of Central Sulawesi (Celebes). Early epidemiological studies prior to World War II demonstrated that, in addition to man, wild deer and domestic dogs served as reservoir hosts, and subsequent microscopic examination of adult worms from these mammals confirmed them to be *S. japonicum*^{2,3} Although extensive snail surveys were conducted at that time, the molluscan host was not found. The schistosomiasis problem in Lindu Valley virtually remained dormant until 1956. Epidemiological studies were made, which confirmed the findings of previous investigators⁴, and since then lake Lindu Valley has been known as the only area of endemic *S. japonicum* in Indonesia⁵. *S. japonicum* in Indonesia⁵

In the 1970's there was a resurgence of interest in the epidemiology of schistosomiasis in Indonesia. A new schistoso-

miasis area in the Napu valley was discovered⁶. During these periods, the intermediate host, *Oncomelania hupensis* was found in the Lake Lindu valley. Thus confirmed that the disease situation in Indonesia was, in fact, a form of classical oriental schistosomiasis similar in its biology and transmission to that found in the Philippines, Japan and China. The molluscan host of *S. japonicum* in the Lake Lindu valley was subsequently described as a new species, *O.h. linduensis* and is most similar to *O.h. quadrasi*, the vector host in the Philippines⁸. The disease occurs now only in two very isolated areas, the Lake Lindu valley and Napu valley in Central Sulawesi.

DESCRIPTION OF THE LINDU AND NAPU VALLEYS

Detailed descriptions of both these valleys were given by Clarke et. al.⁹ and Carney et. al.⁶ However, it will be useful to summarize some of the salient features of the areas.

The Lindu valley is located in the highlands of Central Sulawesi (Fig. 2). The valley has an area of approximately 100 km² in the Takolekadju mountain range. In the northwestern portion of the valley lies Lake Lindu, which is 10 km in length and average 5 – 6 km in width. Numerous streams originating from the surrounding mountains empty into the lake. At the northwest corner, the Gumbasa river, which is the only outlet, flows into the adjoining Palu valley. There are flat stretches of low-lying marshy areas on the eastern and

northern sides as well as at the northwest corner of the lake. These open swampy areas are utilized by the village population for cultivating rice and grazing cattle. Climatologically the Lindu valley is situated in a tropical rain forest characterized by high temperatures and humidity.

Human population numbering about 1,500 – 2,000 are spread among the villages there, most of which lie on the western shore of the lake.

Napu valley is also located in the highlands of Central Sulawesi, about 50 km southeast of Lake Lindu (Fig. 2).

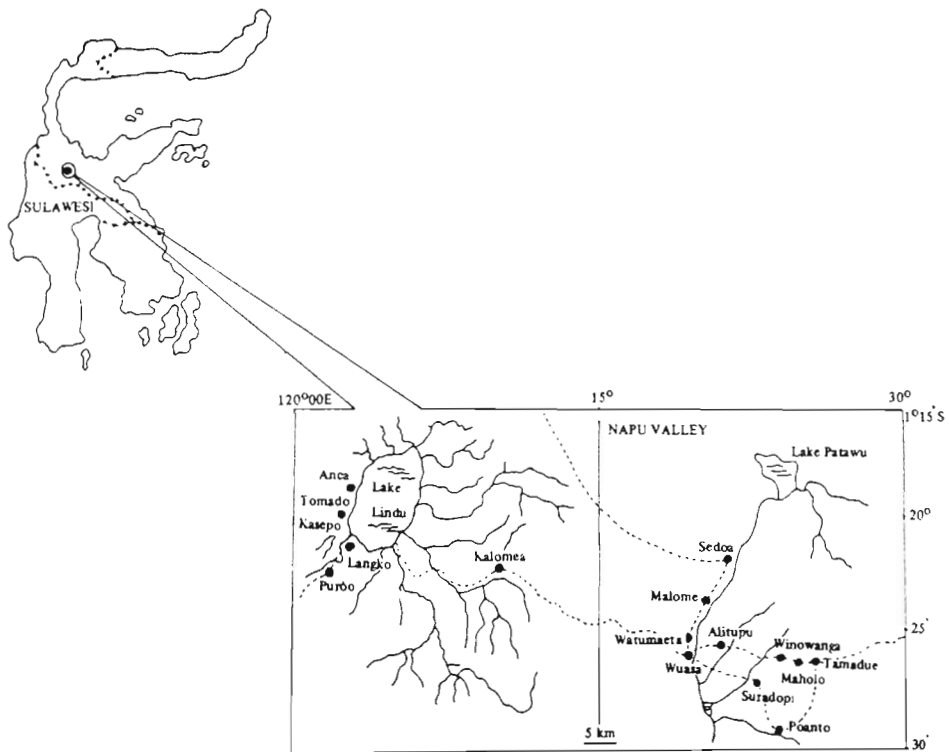


Fig. 2. Map showing the two known endemic areas for *Schistosoma japonicum* in Central Sulawesi, Indonesia : Palu Valley and Napu Valley.

The valley lies between the Takolekedju mountains on the west and the Tineba mountains on the east, and is now a dried-out lake bed of more than 1,000 km². It is surrounded by tropical rain forest characterized by high temperatures and humidity. The valley floor is a well-drained grassy plain. A small body of water, Lake Wanga, is situated 5 km south of Wuasa, the district capital. The Napu valley is drained by the Lariang which river empties into the strait of Makassar.

Presently more than 4,000 people inhabit the Napu valley in villages located along the Lariang river and its tributaries.

ECOLOGY OF *ONCOMELANIA* SNAILS AND INFECTIVITY

Following the discovery of a single specimen of *Oncomelania* snail in 1971 by Carney et. al.⁷, further intensive surveys revealed over 70 foci of this snail were found on the lowlands surrounding the lake Lindu area¹⁰ and 15 foci in Napu valley about 50 km southeast of Lindu^{11, 12}. The ecology of this snail is well documented by these investigators, and later also by Pinardi Hadidjaja and Sudomo¹³. The subspecific status of the *Oncomelania* in the Napu valley has not been determined as yet, and is presently referred to at the Napu geographic strain of *O. hupensis*.

In the Lake Lindu valley, most of the *Oncomelania* colonies isolated have been found on the western and northern side of the lake. Colonies of this snail were found near cultivated fields, abandoned farming areas and in the uncultivated virgin forests surrounding the lake.

In the Napu valley, the habitats are mainly swampy fields with dense, short or tall marsh grasses, usually adjacent to actively worked paddy fields. Forested areas or along the edge of the forest have not yet been searched for the snails.

Potential oncomelamid habitat in Napu valley is far greater than that available in Lindu valley. In Lindu valley less than 50 km² are lowlands or moors needed for *O. hupensis* habitat, whereas in Napu valley there are 7,500 km² of such terrain⁶.

Infection rates of *S. japonicum* in *O.h. lindoensis* varied considerably between foci and between sampling periods. The overall average infection rate in the Lindu valley was 2.4 % in large disturbed area foci, and about 7.0 % in small natural foci bordering the lake shore or in the virgin lowland forests. Infection rates were higher in females (2.70 %) than in male snails (1.95 %). By age (length) snail infection rates were quite consistent in snails two months of age or older¹⁴.

EXPERIMENTAL INFECTION STUDIES

Studies on experimental animals, using mice, Wistar rats, Long Evans rats, Mongolian gerbils, wild rats (*Rattus norvegicus*), hamsters and guinea pigs showed that mice were found to be the most susceptible experimental host for the Indonesian strains of *S. japonicum*¹⁵.

Experimental studies on subspecies of *O. hupensis* exposed to infection with zoophilic and anthrophilic strains of *S. japonicum* to determine the ability

of geographic strains of the parasite were carried out by Cross et. al.¹⁶ The snails, *O.h. quadrasi* from Philippines, *O.h. formosa* and *O.h. chiui* from Taiwan, *O.h. nosophora* from Japan and *O.h. lindoensis* from Indonesia were used for these experiments. Anthropophilic strains of *S. japonicum* from Philippines, China, Japan and Indonesia, and zoophilic strains from Taiwan were used to infect these different geographic strains of snails. The results show the diversity specificity of *O. hupensis*

subspecies to infection with geographic zoophilic and anthropophilic strains of *S. japonicum*. Most species of snails can be infected with geographic strains of the parasite from other areas, but usually in low numbers. Others such as *O.h. quadrasi* from Philippines and *O.h. formosa* from Taiwan are essentially resistant, but *O.h. chiui* from Taiwan seems to be a universal host easily becoming infected with all strains of *S. japonicum*. (Table 1).

Table 1. Development of geographic strains of *Schistosoma japonicum* in various subspecies and strains of *Oncomelania hupensis*

<i>Oncomelania hupensis</i> subspecies	Geographic strain of <i>Schistosoma japonicum</i>						
	Philippines		Taiwan		China	Japan	Indonesia
	Bohol	Leyte	Changhua	Ilan			
<i>O.h. quadrasi</i> (Bohol)	4,028	554	558	100	447	396	419
No. exposed	2,148	345	430	65	253	290	354
No. survived	634	100	35	0	6	5	43
No. positive (%)*	(30)	(29)	(8)		(2)	(2)	(12)
<i>O.h. quadrasi</i> (Leyte)	717	2,715	631	90	368	386	469
No. exposed	576	2,271	532	77	230	321	394
No. survived	2	483	6	6	0	0	1
No. positive (5)	(0,4)	(21)	(1)	(8)			(0.3)
<i>O.h. quadrasi</i> (Mindanao)	767	496	470	—	392	250	526
No. exposed	606	320	329	—	395	230	378
No. survived	1	0	1	—	0	0	0
No. positive (%)	(0.2)		(0.3)	—			
<i>O.h. formosana</i> (changhua)							
No. exposed	639	621	3,255	490	677	470	594
No. survived	543	502	2,451	53	465	395	417
No. positive (%)	0	7 (1)	490 (19)	0	0	0	(0.2)
<i>O.h. formosana</i> (Ilan)							
No. exposed	400	396	420	1,440	486	90	290
No. survived	272	356	362	894	305	89	259
No. positive (%)	9 (3)	4 (1)	12 (3)	46 (5)	3 (0.9)	0	4 (2)

<i>Oncomelania hupensis</i> subspecies	Geographic strain of <i>Schistosoma japonicum</i>						
	Philippines		Taiwan		China	Japan	Indonesia
	Bahol	Leyte	Changhua	Illan			
<i>O. h. chiui</i> (Alilo)							
No. exposed	939	818	245	—	669	835	504
No. survived	814	634	187	—	532	792	418
No. positive (%)	405 (50)	327 (52)	66 (35)	—	279 (52)	355 (45)	175 (42)
<i>O. h. quadrasi</i> (China)							
No. exposed	531	460	251	—	3,322	—	448
No. survived	411	414	251	—	2,445	—	412
No. positive (%)	55 (13)	48 (12)	2 (0.4)	—	240 (9)	—	11 (3)
<i>O. h. nosophora</i> (Japan)							
No. exposed	—	—	—	—	—	1,872	—
No. survived	—	—	—	—	—	1,056	—
No. positive (%)	—	—	—	—	—	326 (31)	—
<i>O. h. lindoensis</i> (Indonesia)							
No. exposed	—	—	90	—	—	—	717
No. survived	—	—	22	—	—	—	417
No. positive (%)	—	—	2 (9)	—	—	—	180 (43)

* Numbers in parentheses are percentage positive for cercariae to nearest whole number (Cross et. al., 1984).

These experimental studies clearly show that the anthropophilic strain of *S. japonicum* from Indonesia can infect a wide diversity of geographic strains of *Oncomelania* snails.

Subsequent experimental studies on four subspecies of *O. hupensis*, namely, *O. h. lindoensis*, *O. h. hupensis*, *O. h. nosophora* and *O. h. quadrasi* exposed to miracidia of four geographic strains of *S. japonicum*, i.e. The Chinese, Japanese, Philippines and Indonesia were carried out by Sudomo¹⁷. He found that *O. h. lindoensis* from Indonesia and *O. h. hupensis* from China to be the most susceptible among all the four geogra-

phic strains of *S. japonicum* while *O. h. quadrasi* from the Philippines were susceptible to the Philippines strain of *S. japonicum*, and refractory to the other three strains of *S. japonicum*.

The results of this study differs from Cross et. al.¹⁶ in that the *S. japonicum* strain of Indonesia was shown to be not a good experimental host for *O. h. hupensis* from China, while the reverse was found in Sudomo experimentati-
on.¹⁷

The survival infectivity of the Indonesian strains of *S. japonicum* at different temperatures to its natural host, the *O. h. lindoensis* was carried

out by Sudomo¹⁸. He showed the miracidia were active and infective at three different temperatures at 18°C, 23°C and 28°C from 0 – 8 hours. At 18°C the miracidia were still active at the age of 24 hours, although the infection rate was comparatively low. At 23°C the miracidia were still infective from 0 – 12 hours whereas at 28°C miracidia were not able to withstand the temperature longer than 8 hours.

MAMMALS RESERVOIR HOST

Man, of course, is one of the predominant mammals in the Lake Lindu valley serving as a reservoir host. Surveys on wild mammals revealed *S. japonicum* naturally infected 13 of the 23 mammal species examined. They are the field and forest rats: *R. exulans*, *R. hoffmanni*, *R. chrysocomus rallus*, *R. marmosurus*, *R. celebensis*, wild deer (*Cervus timorensis*), wild pigs (*Sus scrofa*), wild civet (*Viverra zibetha*), forest shrew (*Crocidura nigripes*), domestic cow, dog, horses, and water buffalo^{19, 10}. Mammal surveys have been geared to determine the distribution and infection rates in rodents, principally *R. exulans*, which inhabit the cultivated lowlands of the disease foci in the Lindu area. Routine fecal examinations of wild and domestic mammals were shown to be significant

in the transmission of this disease, because humans, domestic and wild animals all frequent areas, which harbour the molluscan host and leave their excreta in the amphibious or aquatic habitats.

DISEASE PREVALENCE IN HUMANS

The distribution of schistosomiasis throughout the Indonesian archipelago was studied extensively in the 1970's, especially on the island of Sulawesi. Although more than 50,000 stools specimens were examined and extensive surveys conducted *Schistosoma japonicum* and *Oncomelania hupensis* appear limited in their respective distribution to two contiguous drainage systems of Central Sulawesi, i.e. the Lindu valley, 1,000 meters in elevation at the headwaters of the Gumbasa river drainage system and the Napu valley, more than 1,000 meters in elevation at the headwaters of the Lariang river drainage system. Carney²⁰ speculated that at least 7,000 individuals are continuously exposed to schistosomiasis in confirmed endemic areas.

Since 1971, extensive and intensive surveys were narrowed down to the two foci in the Lindu and Napu valleys of Central Sulawesi, and a summary report is presented in Table 2. In Lindu valley

Table 2. Summary of *Schistosoma japonicum* infection in villages of Lindu and Napu valleys, Central Sulawesi, Indonesia.

Area	Villages surveyed	Total population	Number of stools examined	Percentage positive	References
Lindu valley, Central Sulawesi	Tomado, Langko and Anca	1,458	126	53	Pinardi Hadidjaja et al., 1972.
Lindu valley, Central Sulawesi	Anca, Tomado, Langko and Puroo	1,500	1,417	37.9	Clarke et. al., 1974
Lindu valley, Central Sulawesi	Anca, Tomado, Langko, Puroo and Paku	1,515	1,423	37.5	
Napu valley	Watumeta, Wuasa, Alitupu, Winowonga and Maholo	1,843	583	43	Carney et.al., 1974
Napu valley	Kaduwa, Tamadue, Sedoa and Watutau	1,214	1,003	31	Carney et.al., 1977
Napu valley	Maholo, Winowanga and Tamadue	708	241	65.9	Pinardi Hadidjaja et.al., 1985.
Palu valley and Lindu valley, Central Sulawesi	Ruwali and Pakuli	7,000	2,433	0.8	Cross et.al, 1975
Rampi and Seko valley, South Sulawesi	Singkalong, Eno and Dodolo	640	640	0.9	Carney et.al., 1977

93.9 % of 1,515 inhabitant among the villages surveyed in 1971, 1972, 1974, the infection rates were 53 %, 37.9 % and 37.5 % respectively^{21, 9, 22}. In the Napu valley, a coverage of 50.4 % of 1,843 inhabitants among the villages surveyed in 1972, 1974, 1980, the infection rates were 43 %, 31 % and 65.9 %^{6, 23, 24}. The results showed that the prevalence rates vary at different periods of surveys, and they are relatively high for each of the period examined. It is important to emphasize that these numbers and percentages refer to persons passing schistosome eggs in their stools, a much smaller number and percentage of persons are really

clinically compromised by this infection.

In Palu valley which is nearby the lake Lindu where waters from lake Lindu enter the Gumbasa river which joins the Palu river flowing through the lower Palu valley, 2,433 stools from an estimated population of 18,700 in the seven villages along the Palu drainage system were examined in 1971. Less than 1.0 % of these stools were positive with *S. japonicum* eggs and these were from two of the seven villages²⁵. In 1973, the Rampi and Seko valleys of South Sulawesi with similar mountain valleys and geological history to that of the Lindu and Napu valleys; human parasites were surveyed in seven

villages (3 in Seko village and 4 in Rampi village). Only six persons of 640 inhabitants in the 3 villages at Seko valley were found with *S. japonicum* eggs in their stools²⁶. A follow-up investigation of these positive cases in the Seko valley by these investigators, revealed that all had recently migrated from the Napu valley, a confirmed schistosomiasis area.

Study on the clinico-pathologic of infection showed that the positivity of liver biopsy for eggs from 52 *S. japonicum* patients in the Lindu valley foci was 96 %²⁷. This finding indicates that although diagnosis of *S. japonicum* is generally made by stool examination and rectal biopsy, live biopsy also proved to be a valuable method of diagnosis as well.

Since 1971, all these studies have conclusively demonstrated that the disease and its molluscan host are well established in the undisturbed lowland forest. The presence of the disease and large population of the snail in disturbed and cultivated areas appear to be secondary adaptations.

CONTROL PROGRAMME

Since 1975, there have been a number of developments that have changed the epidemiological picture—some for the better and some, regrettably, for the worse.

A pilot control project was initiated by the National Institute of Health Research and Development (NIHRD) in conjunction with the World Health Organization (WHO). This pilot project which focussed on the Paku-Anca area involved selective mass treatment, agro-

engineering, mollusciciding, improved sanitation and health education. The results were good as the prevalence of schistosomiasis was reduced from 70 % to 25 % in the intervention area over a two year period^{22, 28, 29}. Regrettably, however, the control effort has not been maintained and one indicator of increased transmission, the prevalence of infection in rodents, suggests that the disease will return to its previous level of endemicity in a short period.

Other schistosomiasis control projects were conducted throughout the Lindu valley during the same time frame and subsequent to the NIHRD — WHO pilot control projects. The non-intervention area of one study was accidentally "controlled" by another project obviously compromising the results of both studies. In addition 4 — 5 villages in the Lindu valley have been treated by a number of agencies on more than one occasion using more than one anti-schistosomal compound making it difficult to determine who has been treated, with what, by whom or when.

DRUG TRIALS

Mass and selective treatments with different antischistosomal compound, such as Niridazole, Stibophen and Praziquantel were carried by various investigators at different time period in the focus area. The results show that 11 patients treated with Stibophen did not pass ova in their stools 2 and 6 months after treatment, and 11 months post treatment follow-up, one out of 10 patients passed ova. In the case of Niridazole treatment to 31 patients, 4 passed ova after 2 to 6 months post treat-

Table 3. Reservoir and intermediate host of *Schistosoma* species in Indonesia.

Schistosoma species	Reservoir hosts (mammals & bird)	Intermediate hosts (mollusks)	Locations	Reference
<i>Schistosoma spindale</i>	Domestic mammal Water buffalo	? Planorbid snails	North Sumatra	Burggraaf, 1935
<i>Trichobilharzia brevis</i>	? ducks	<i>Radix (Lymnaea) javanica</i>	Jakarta, West Java	Margono, S.S. 1968
<i>Schistosoma incognitum</i>	Field & forest Rodents <i>Rattus argentiventer</i>	<i>Radix (Lymnaea) auricularia rubiginosa</i>	Cikurai, West Java	Carney et.al., 1977
<i>Schistosoma incognitum</i>	<i>Rattus exulans</i> <i>R. hoffmani</i> <i>R. nitidus</i> Wild deer <i>Cervus timorensis</i>		Lake Lindu and Napu valleys, Central Sulawesi	Carney et.al., 1977
<i>Schistosoma japonicum</i>		Lymnaeid snails	Paso valley Central Sulawesi	Bonne et.al., 1942. Laust & Bonne 1948.
<i>Schistosoma japonicum</i>	Primate Man		Lindu valley, Central Sulawesi	Muller & Tesch, 1937 Bonne et.al., 1942 Brug & Tesch, 1937 Bruck & Urman, 1956.
<i>Schistosoma incognitum</i>	Primate Man Rodents <i>Rattus exulans</i> <i>R. hoffmani</i> <i>R. chrysocomus</i> <i>R. marmosurus</i> <i>R. clebensis</i> Insectivore <i>Crosidura nigripes</i> Wild deer <i>Cervus timorensis</i> Wild pig <i>Sus scrofa</i> Wild vicet <i>Viverra tangalunga</i> Domestic mammals Water buffalo cow, dog, Horses	<i>Oncomelania nupensis lindonensis</i>	Lindu & Napu valleys, Central Sulawesi.	Carney et.al., 1973c Sudomo & Carney, 1974

ment, and 11 months follow-up treatment 5 were still observed to pass ova³⁰ Mass treatment of 708 people in three villages of Napu valley with praziquantel showed significant reduction of prevalence rate from an average of 15 % pretreatment to 2 % six months post treatment²⁴. The side effects of the 3 antischistosomal drugs to treated patients, showed praziquantel has the least side effects as compared to the other two drugs. It was apparent from these studies, praziquantel is the drugs of choice for the treatment of *S. japonicum* in the endemic area.

POTENTIAL SPREAD OF SCHISTOSOMIASIS BY ENVIRONMENTAL CHANGES

A logging road has been constructed from the Palolo valley close to Bamba which is confirmed schistosomiasis transmission area on the northern shore of lake Lindu. The only reason schistosomiasis is not a major health problem in Indonesia today is because of its very focal and limited distribution. It has the potential to be a much more serious health problem. Increased contact with the outside world – as by a commercial road to the valley – would be the most sure way of spreading schistosomiasis to other areas of Central Sulawesi. The present isolation of the Lindu and Napu valleys is the single most important location for its limited distribution and its relative low importance as a national health problem.

In the absent of a more effective method in the control of the transmission cycle between the molluscan intermediate and wild mammal reservoir

hosts, caution should be taken not to introduce any new elements, particularly translocation schemes of people within the local province and country transmigration programs in both the Lindu and Napu valleys endemic schistosomiasis areas.

Schistosoma japonicum like trematodes

There have been several records of *Schistosoma japonicum* like infection reported in Java outside the lake Lindu and Napu endemic foci. Cases of *S. japonicum* infection from Indonesia usually area considered non autochthonous as almost all have occurred in residents of Chinese heritage who have previously migrated from or made trips to mainland China^{31,32, 33, 34, 35, 36} One case, reportedly made numerous trips to Central Kalimantan (Borneo), staying there for 3-4 month periods. However, this individual, also of Chinese heritage, denied every having been in endemic areas of Sulawesi or having travelled outside of Indonesia³⁷. Another case that has recently been reported involved a man of Chinese heritage who denied ever leaving the island of Java³⁸. All these cases occurred in middle or older age male between 34 and 68 years of age and each case was diagnosed following histological examination of tissue samples which were recovered from surgical reasons or at autopsy. Essentially then, these occult cases were due to *S. japonicum* or *S. japonicum* like trematodes. The possible explanations for the etiology of these cases are:

1. They may have come in contact with a classical strain of *S. japonicum* in a transmission area of Indonesia that

has yet to be identified.

2. They may have visited on endemic area of classical *S. japonicum*, such as China, but choose not to admit the fact, and.

3. They may have been exposed to an unknown *S. japonicum* like trematode that is endemic in Java and/or Kalimantan.

In the cases involving individuals who had migrated from China or who had visited that country, the most probable explanation is that these individuals were exposed to a classical strain of *S. japonicum* in China. However, the two cases who never visited known endemic areas either in or out of Indonesia, are more difficult to explain. All three of the above options must be considered.

In light of recent studies in Malaysia where *Schistosoma japonicum*-like eggs have also been reported from the liver of 10 aborigines³⁹; where adult worms indistinguishable from *S. japonicum* were reported in monkeys (*Macaca fascicularis*) in Ranau area of Sabah⁴⁰ and three imported cases of a similar etiology for cases in Indonesia, particularly Java and Kalimantan, takes on a new credibility.

Schistosoma incognitum.

Schistosoma incognitum was described from human fecal specimens collected in India⁴¹. Since then, Indian parasitologists have found it naturally infected dogs and pigs, as well as from a wide range of experimental hosts including pigs, sheep, goats, cattle, rabbits, guinea pigs, rats, mice, dogs and cats⁴². Most recently, Lee and Wykoff^{4,5} found *S. incognitum* to be enzootic in field

and seaport rats (*Bandicota indica* and *Rattus norvegicus*) from Thailand, and Harinasuta and Kruatrachue⁴⁴ added forest rats (*Rattus berdmorei* and *Rattus r. thai*) to the mammalian host list in Thailand.

In Indonesia, a mammalian schistosome, appropriately called "The Lake Poso Blood Fluke" was isolated from lymnaeid mollusks in the Poso valley of central Sulawesi, but its specific status was not determined^{3,45}. During 1973/1974, *S. incognitum* was reported in ricefield rats (*Rattus argentiventer*) from Cikurai, West Java, and in Rawasari in Jakarta^{46,47}. Subsequently, the parasite was again found naturally infected in three species of field and forest rats (*R. exulans*, *R. hoffmanni* and *R. nitidus*) and in a wild deer (*Cervus timorensis*) from the schistosomiasis endemic area of lake Lindu and Napu valley, Central Sulawesi⁴⁸. Although a wide variety of rodents and a wild deer have been found naturally infected with the parasite in Indonesia, human infections have not been diagnosed by stool examinations in area where it is enzootic.

Lymnaeid mollusks are the only confirmed molluscan hosts of *S. incognitum*. *Lymnaea luteola* (*Radix luteola*) was incriminated as an intermediate host of *S. incognitum* in India⁴⁹ and this subsequently was confirmed⁴². In Thailand, *Lymnaea auricularia rubiginosa* (= *Radix a. rubiginosa*) was considered as an intermediate host of *S. incognitum*⁴⁴. In Indonesia, natural infections with *S. incognitum* larval stages were observed only in *R.a. rubiginosa*. This

was further confirmed by laboratory experiments in which miracidia of *S. incognitum* were exposed to laboratory-bred *R.a. rubiginosa*, and shed cercariae 41–48 days post exposure⁴⁸.

This further confirmed that the intermediate host of *S. incognitum* in Indonesia is the same molluscan host species (*R.a. rubiginosa*) as that established in Thailand.

In Indonesia, *S. japonicum* and *S. incognitum* have been shown to share the same geographic area and hosts in the Lindu and Napu valleys schistosomiasis endemic areas of Central Sulawesi. In two cases both schistosomes species were concurrently found in the same rodent host (*R. exulans*) and in one of these cases a heterologies pair—a *S. incognitum* male and a *S. japonicum* female were found in copula⁴⁶. Subsequently, this same phenomenon was observed in experimental animals. Laboratory mice were exposed to 50 *S. incognitum* cercariae from West Java and challenged with 50 *S. japonicum* cercariae from Central Sulawesi 40 days later. When perfused 48 days after challenge female *S. incognitum* were found in the gynecophoral canal of *S. japonicum* males and vice versa. Heterologously paired females of both species contained eggs which were of maternal origin⁵⁰. The viability of these eggs, unfortunately was not determined. Thus, even though these two mammalian schistosomes have been isolated through a high level of intermediate host specificity, their sympatric distribution, both geographically and in regards at least some definitive hosts, presents a natural opportunity for recombination of genes that may favour-

ably affect the hybrid's ability to infect a wide range of intermediate or definite hosts. If these sympatric schistosomes which share the same geographic region in Indonesia hybridize successfully, the results could be offspring capable of infecting humans yet also capable of utilizing mollusks, such as lymnaeids. In that even human schistosomiasis could become a problem throughout all of Asia.

Schistosoma spindale

Schistosoma spindale was the first reported schistosome from Indonesia. In 1935, this blood fluke was recovered from water buffalos in Northeast Sumatra⁵¹ and to our knowledge this is the only written documentation of this blood fluke in Indonesia. It is most likely that the distribution of this parasite is much wider and probably includes Sumatera, Java and Kalimantan. Besides Sumatera, *S. spindale* is known to occur in India, Ceylon and Thailand. The parasite is responsible for cercarial dermatitis "swimmers itch" in any area of Indonesia where it is endemic and it is also elsewhere in Asia. It is a cause of bovine nasal granuloma in areas of Asia where it occurs. Larval stages of *S. spindale* develop in planorbid mollusks elsewhere in Asia, and it is most likely that planorbids are responsible for the development of the larval stages of *S. spindale* in Indonesia.

Trichobilharzia brevis

Trichobilharzia brevis is an avian blood fluke. In Indonesia the parasite was reported only once in lymnaeid mollusks from Jakarta area of West Java⁵². The definite hosts area usually

ducks. The larval stages develop in lymnaeid mollusks which are common throughout the Indonesian archipelago, especially in rice growing areas. It causes cercarial dermatitis in areas where it is endemic. Its distribution is most likely widespread throughout the western half of Indonesia as it was originally described from ducks and lymnaeid mollusks of western Malaysia^{5,3}.

CONCLUSION

The animal reservoir and molluscan intermediate hosts of the four schistosomes in Indonesia is presented in Table 1. Among these four *Schistosoma* species, the classical *Schistosoma japonicum* is the only one which affect man in addition to infecting a vast variety of domestic and wild mammalian hosts. The distribution of classical *S. japonicum* is limited to the distribution of *O. hupensis* and this species is very fastidious in its ecological requirements. *S. incognitum*, on the other hand, although it has not been found in man, but like *S. japonicum*, it lacks definitive host specificity as it can develop in at least six orders of mammals in Asia, and in Indonesia it has been reported in four species of rodents and a species of wild deer. It is adapted to ubiquitous lymnaeid snails potentially has a much wider distribution in Asia and elsewhere than classical *S. japonicum* because of the latter snails dependence on *O. hupensis* for transmission. In Indonesia, this schistosome share the same definitive hosts in the same geographic region and heterologous mating of both *S. japonicum* and *S. incognitum* was found in naturally infected rodent as

well as in experimentally infected rodents. Human are constantly exposed to the cercarial stage (the infective stage) of *S. incognitum* in rice fields of Asia where lymnaeid snails and commensal rodents maintain this sylvatic cycle of this blood fluke. Evolutionary pressure is always present and with any favorable changes in its gene pools, *S. incognitum* may be able to exploit the second most common mammals in Asia—man. *Schistosoma spindale* and *T. brevis* are very host specific. The former species appears to be restricted to domestic mammal, and the latter is an avian blood fluke.

It is most likely that there are still unidentified schistosomes in addition to the four known schistosomes in the Indonesian archipelago. In regards to human health the primary concern is the classical oriental schistosomiasis which presently is limited to two remote mountain valleys of Central Sulawesi. It is of utmost importance that a rational control or eradication program be developed before economic exploitation of Sulawesi spreads this disease to other areas. The potential second public health concern involves schistosome of mammals other than man which may adapt themselves such that they exploit or utilize the human population in areas where they are now enzootic for their own survival. The schistosome species which is of particular importance is *Schistosoma incognitum*. If this ever occurs, schistosomiasis in Indonesia could become a major public health problem.

Classical oriental schistosomiasis in Indonesia is controllable or managable and possibly eradicable disease because of its current limited and isolated

distribution in an underpopulated island of the Indonesian archipelago. Before any other attempts are made to control, manage or eradicate this disease in Indonesia, it is imperative to re-examine and re-evaluate past experiences of control programmes already carried out in regards to the feasibility and probability of success of those programmes.

a. In 1974, pilot control multi-disciplinary approach in Anca and Langko foci of Lindu valley which consist of: (1) reduction of one source of infection by treating positive human cases; (2) reduction of the intermediate molluscan host population by spraying foci of transmission with a molluscicide and by modification of snails habitat by agro-engineering technology; (3) reduction of human contact with infectious cercariae in infested waters by promoting environmental sanitation and health sanitation was carried out jointly by the National Institute of Health Research and Development, Ministry of Health, Republic of Indonesia and the World Health Organization.

This pilot control project was conducted over a two years period and the results were considered a success as the prevalence in the human population that was theated decreased from 71% to 26% and the incidence of new cases was approximately one-third that of the non-interventioned control area. A more sensitive indicator of transmission changes, e.g. infection rates in commensal rodents, also decreased during this period by 50%. However, as this control project was only a pilot project intervention methods were relaxed following the trial, and schistosomiasis transmission, as expected, returned to the pre-intervention levels²².

It was better unfortunate that no follow-up of intervention methods being planned after having found that the multidisciplinary approach pilot control project was promising in the control of schistosomiasis in the endemic areas. Based on the experiences on the pros and cons of the successful pilot control project, a better intervention control method could be made to further reduced the transmission levels, and be incooperated into the general vector-control programme by the government.

b. In 1978, two rounds of molluscicide trials, using Bayluscide was carried out in each of the Anca and Paku areas of Lindu valley against moluscan host, *O.h. lindoensis*. The trials were conducted in sequence. In the first round of spraying, the density of snails was suppressed by 30.7% (fom 89.9 to 59.2 snails per m²), and in the second round it was further suppressed by 67.2% (from 59.2 to 19.4 snails per m²). In the Paku area, the density of snails was reduced by 16.1% in the first round of spraying (from 90.3 to 74.2 snails per m²), and substantially reduced by 94.3% in the second round (from 74.2 to 4.2 snails per mm²). The overall suppression of snail density in the combined two rounds of spraying in the Anca area was 78.4% (from 89.9 to 19.4 to 19.4 snails per m²) and that of the Paku area was 95.3% (from 90.9 to 4.2 snails per m²). The infectivity of the snails in the Anca area was reduced by 78.8% (from pre-spray of 3.4% to 0.7% post-spray), while that in the Paku, it was reduced by 32.3% (from pre-spray of 1.30% to 0.88% pos spray). In the control villages of both these areas, the density of snails and the infectivity rates remained consistent

during pre and post spraying round²⁹

The results of these trials provided us with a target molluscicide compound which is effective in the control of the intermediate host of schistosomiasis in the endemic areas. Mollusciciding would probably be very dramatic in the suppressing of the transmission cycle if the snails habitat is highly restricted in relatively small isolated area. In the Lindu and Napu valleys, the habitats of the snail cover a vast range of habitats, ranging from disturbed to undisturbed forested areas, thus it is not practicable to use molluscicide as the only method of control taking into consideration on the frequency of spraying needed and the cost effectiveness.

c. Drug trials were carried out in 1974 using niridazole and stibophen on 42 cases with *Schistosoma japonicum*³⁰ and in 1983 mass treatment with praziquantel on 241 infected individuals²⁴ in the Lindu and Napu valleys. Among these drugs used, praziquantel was found to be the most favoured and drug of choice for treating *S. japonicum* infection in Central Sulawesi.

Schistosoma japonicum lack devinitive host specificity, and develops in a wide range of mammalian species from insectivores to primates. In Indonesia, besides man as one of the important reservoir hosts, at least 5 species of rodents, a deer including a few genera of domestic mammals also act as reservoir hosts of this schistosome in the endemic areas. Mass chemotherapy treatment with praziquantel to human cases, though it is very tempting and encouraging, and if carried out will obviously have dramatics effect on the human population, but it will not cut the

transmission cycle completely as long as there are other wild and domestic mammalian reservoir hosts, particularly the rodents. However, intermittent use of chemotherapy to masstreat the exposed human and domestic animal population combined with intensive rodent control will drastically raduce the transmission. If this type of control effect is maintained the threshold of schistosomiasis transmission might be reached and the disease eradicated from the human and animal populations in inhabited areas. But, if the disease is left unchecked schistosomiasis would return with a vengeance.

Discussion on the pros and cons of the various control, measure attempts undertaken as above mentioned, it appears that the multidisciplinary approach which include agro-engineering, chemotherapy, mollusciciding, health education and improved sanitation for the control of oriental Schistosomiasis is more adequate than the other methods attempted. Unfortunately this type of control is long term and expensive in terms of manpower and public funds.

In comparison to other health problems in Indonesia, schistosomiasis is relatively unimportant as it involves less than 10,000 of Indonesia's 167 million residents, and most of those infected are not seriously compromised by these blood fluke, but it will remain a health problem in confirmed endemic areas. In addition, as the economy of Sulawesi grows, means of communication between the remote areas where it is endemic will improve, and the spreading of this disease to other areas of Sulawesi where the habitat and mammalian fauna are suitable is foreseeable. Thus, it is important alternative methods

of control which are more economical should be explored, and suggestion of a few alternatives are as follows:

1. Declare the endemic areas as a National Park. This has been suggested by the Indonesian Government as the "Lore Kalamanta National Park" because of the rich faunal and floral reserves in this remote areas. If these endemic areas are closed to human settlements, schistosomiasis will remain as its sylvatic cycle is well established in at least 13 mammalian species and the molluscan host is well established throughout undisturbed, virgin forests as well as disturbed and cultivated areas of the valleys. Human cases of Oriental schistosomiasis would then be limited to isolated cases in visitors whose exposure would be minimal if adequate preventive measures were utilized.

This alternative would necessitate moving (transmigrating) the human population of both the Lindu and Napu valleys to other fertile areas of Central Sulawesi. The Lindu valley residents could be moved to the Poso valley which is free from the disease. As Lindu valley residents have farmed the valley for less than 100 years they most likely would willingly move in a well organised transmigration project. However, as residents of the nearby Napu valley, where schistosomiasis is also endemic, have been settled in that area for many hundred of years, they might resist any attempt to transmigrate them to another area.

2. Drastic alteration of environment in endemic areas such as a permanent

or temporary change in the water levels.

The Lindu and Napu valleys are both geologically situated in partially dried out lake beds. It appears feasible that proper water management might radically reduce habitats suitable for maintenance of oncomelaniids. The Lindu valley appears most suitable for this type of intervention because of the close proximity of the surrounding mountains. A dam which raised the water level in the Lindu valley only 5 – 10 meters would inundate more than 99 % of the habitat now utilized by oncomelaniids. The new shores of lake Lindu would wash against the relatively steep mountains surrounding the valley. Potential habitat remaining would be a small portion of what is available today and could be managed by conventional methods.

The advantages of a permanent raising of the lake's level would be (1) regulation of water for irrigation downstream; (2) hydroelectric power production; (3) expanding fishing industry and most importantly; (4) reduction of schistosomiasis transmission by destroying suitable oncomelaniid habitats resulting in a drastic reduction in these snails and commensal rodents which are considered the principal mammalian reservoirs of schistosomiasis in the Lindu valley.

The disadvantages of such a drastic measure would be (1) the necessity to transmigrate approximately 2,500 persons to another area of Sulawesi; (2) the flooding of many hectares of fertile farm land; (3) the one time, yet initially high, expense

of a hydroelectric dam for power production and water management.

A related measure that should also be considered would be the intermittent changing of the water level of the lake. The principal advantages of this drastic alteration would be (1) inhabitants would only have to move to higher areas of the valley; (2) intermittent flooding would allow for intermittent farming of the valley floor and reduction the population of commensal rodents. This alternative would initially be expensive farming would only be intermittent.

The outcome of either of these drastic measures would probably be very good, if feasible from the engineering standpoint, for the ecological requirements of oncomelanids are very specific at different stages. Even temporary yet drastic changes in the environment – such as flooding – could reduce the oncomelanid populations and the populations of the most important mammalian reservoir of schistosomiasis, rodents, enough to permanently interrupt transmission.

3. Treat only individuals with serious clinical signs and symptoms of schistosomiasis.

Even though schistosomiasis is considered a serious health problem in areas where it is endemic, not everyone is afflicted with disease just because he or she harbours *Schistosoma japonicum*. This alternative involves limited health resources be utilized to treat those who are compromised by the disease and

that schistosomes and the human population of endemic areas be allowed to maintain a balance of compatibility. This is definitely a very economically advantages alternative as it concentrates limited health reserves for individuals most in need. The disease will be managed but the problem will remain and if it is not monitored periodically, the possibility of spread to other areas of Sulawesi is foreseeable as the economy develops.

Among these three alternatives, alternatives 1 and 2 would be most ideal, but in both cases it involves initially very high capital expenditures. In view of the current economic slow – down of the country, alternative 3 would probably be the most suitable temporary. In the long run the best alternative would be to eradicate this snail-borne disease through drastic alteration of its very specific ecological requirements before it has a chance to spread to other areas of the island.

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