

## The rate of cerebral involvement in paragonimiasis an epidemiological study

SHIN JOONG OH

*Institute of the Endemic Diseases, Seoul National  
University, Seoul, Korea*

*Department of Neurology, National Medical Center,  
Seoul, Korea\**

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While pulmonary paragonimiasis is a benign parasitic disease, cerebral paragonimiasis is no longer considered benign, since it produces devastating neurological sequelae and results in an occasional fatal outcome.

Based mostly upon hospital statistics, the rate of cerebral affection among patients with paragonimiasis has been reported to vary from 1.7 (Iwasaki, 1962) to 26.6% (Grauman, 1957). It is apparent that these rates would not represent the true rate for the following reasons:

1. Pulmonary paragonimiasis is so benign that medical help is not sought by most of the infected people. Those patients who seek for medical aid and who are most often hospitalized, are those who have cerebral involvement.

2. Patients with cerebral paragonimiasis are referred to the special centers where neurological or neurosurgical facilities are available while the majority of patients with pulmonary paragonimiasis are treated by the general practitioners.

All these factors tend to increase the rate of cerebral involvement and explain partially the wide range among the reported rates (Table 1).

The present author has attempted to minimize these biases by epidemiological survey and the results are reported in this paper.

### Material and Method

Selected for this survey was an area in Pa Joo, 30 miles north of Seoul, Korea, which was known for its high endemicity of paragonimiasis. As of October 1, 1964, there were 6,738 inhabitants.

This study was carried out by a team consisting of two physicians, two nurses, and five medical students, and conducted by weekly visits to this area in September, 1964.

To determine the rate of *Paragonimus* infection in the survey area the intradermal *Paragonimus westermani* (P.w.) test was given to 379 individuals under four different groups as noted in Table 2. The rate of positive reactors among these 379 is considered to be representative of infection in this population. In an attempt to minimize the bias which would occur from non-randomization procedure these four groups were selected for this study. The value of the intradermal P.w. test was well established as a screening procedure of paragonimiasis (Sadun *et al.*, 1959; Yokogawa *et al.*, 1955; Chung *et al.*, 1955).

To detect the rate of active paragonimiasis, the positive reactors were urged to submit a single morning sputum specimen for P.w. egg

\* Present address: Division of Neurology, Meharry Medical College, Nashville, Tennessee, U.S.A. 37208

Table 1 The rates of cerebral involvement in patients with paragonimiasis

Reporters	Rates	%	Criteria for diagnosis of cerebral involvement	Sources of material
Grauman*	83/311	26.6	Not specified	German Red Cross Hospital, Pusan Korea (and field survey?)
Chang <i>et al.</i>	48/200	24.0	Clinically well proven cases	Chekiang Provincial Hospital, China
Sadun* <i>et al.</i>	15/ 63	23.8	Headache and other neurological symptoms	German Red Cross Hospital, Pusan, Korea
Chyu	7/ 65	10.8	Not specified	Field survey, Ko-Heung, Korea
Sadun* <i>et al.</i>	12/143	8.4	Not specified	German Red Cross Hospital, Pusan, Korea
	19/270	7.0	Headache and other neurological symptoms	Field survey, Kyung Nam and Che Ju, Korea
Park	16/311	5.1	Clininically well proven cases	Pediatrics, Dong San Hospital, Korea
Iwasaki	12/690	1.7	Clinically well proven cases	Ko Chi National Hospital, Japan
Oh	4/500	0.8	Given in the text	Field survey, Pa Joo, Korea

\* These studies were done at the same hospital. Therefore, there might be overlapping of cases.

Table 2 Rate of positive intradermal test in groups studied

Groups	Number of individual examined	Number of positive reactors	Percentage of positive reactors
6th grade primary school children	87	16	18.2
A village	70	22	31.4
B village	113	36	31.8
Clinic visitors	109	38	34.8
Total	379	112	29.6

examination. Only 48 positive reactors responded. The rate of positive sputum in this group is considered to be the typical rate of active paragonimiasis among the positive reactors in this population. When an individual was known to have an active paragonimiasis, the full course of bithionol treatment was given.

To estimate the number of cases of cerebral paragonimiasis, our study team opened a free diagnostic clinic. With the help of local physicians, local public health service and local village chiefs, all suspected cases were located

and examined. Eighteen patients with seizures, 4 with hemiplegia, 2 with sequelae of poliomyelitis, and a few with other neurological problems were examined. Special history taking was also made among the positive reactors to detect the cases with cerebral paragonimiasis.

The criteria for diagnosis of cerebral paragonimiasis were set arbitrarily according to our clinical experiences and are as follows (Oh, 1969):

1. Positive reaction to the intradermal P.w. skin test.
2. History of "rusty sputum" or hemoptysis.
3. Two out of five neurological symptoms: Headache, visual impairment, seizures, weakness of extremities and sensory disturbances.
4. Two of five neurological signs: mental deterioration, hemiplegia, unilateral hypesthesia, homonymous hemianopsia, and optic atrophy.
5. Detection of P.w. eggs in the sputum or positive serum complement fixation test.

These five criteria had to be met by each case diagnosed as cerebral paragonimiasis.

## Results

Among 379 individuals tested, 112 or 29.6% showed a positive reaction to the intradermal test. The ratio between male and female of

Table 3 Distribution of positive reactors according to sex

Sex	Total number examined	Number of positive reactors	Percentage
Male	226	91	40.2
Female	153	21	13.7
Total	379	112	29.6

Table 4 Distribution of positive reactors according to age

Age	Total number examined	Number of positive reactors	Percentage Per-
0-9	12	0	0
10-19	153	26	17.0
20-29	33	11	33.3
30-39	69	19	27.4
40-49	40	21	52.5
50-59	47	25	52.9
Over 60	25	10	40.0
Total	379	112	29.6

these positive reactors was 3 : 1 (Table 3). The rate of positive reactors in different age groups is shown in Table 4 with a higher rate among older people. Of 48 positive reactors who submitted a single sputum samples, 12 or 25.0% had eggs in the sputum.

If these rates were applied to the community as a whole, the number of patients infected with paragonimiasis would be 1,994 and the number of patients with active paragonimiasis would be at least 498.

Four cases of cerebral paragonimiasis were found in this community. This constitutes 0.2% (4/1,994) of positive reactors and 0.8% (4/498) of active paragonimiasis cases. Their case histories are as follows:

Case 1: A 17 year old male with a history of rusty sputum for 10 years had left Jacksonian seizures, headache, and impaired vision for 5 years. Positive physical findings were hyperreflexia in the left extremities, Babinski toe sign on the left and left homonymous hemianopsia. The skin test was positive. The examination

of sputum showed P.w. ova.

Case 2: A 13 year old female had a history of rusty sputum, headache, right Jacksonian seizure for 5 years and repeated episodes of meningitis. Abnormal physical findings were hyperreflexia of the right extremities with slight weakness, right hypesthesia, and Babinski toe sign on the right. Skin test and sputum for P.w. ova were positive.

Case 3: A 22 year old female complained of right hemiplegia, blurring of vision, and headache for 3 months. In the past, she had rusty sputum. Examination revealed dull mentation, right homonymous hemianopsia, impaired visual acuity, Foster Kennedy syndrome on funduscopic examination, right hyperreflexic hemiplegia and right hypesthesia. Skin test was positive but a single sputum test was negative for P.w. ova. Serum complement fixation test for P.w. was positive.

Case 4: A 30 year old male had a history of rusty sputum for 24 years, left Jacksonian seizures and blurring of vision for 4 years. Abnormal signs consisted of left spastic hemiplegia with hyperreflexia, Babinski toe sign on left, left hypesthesia, left homonymous hemianopsia, and left optic atrophy. Skin test was positive. P.w. ova were absent in a single sputum test but he was told previously, after sputum examination at the local hospital, that he had pulmonary paragonimiasis.

### Discussion

Our study had shown that 0.2% of positive reactors and 0.8% of active paragonimiasis cases have cerebral involvement. Since our material was not randomly selected and our estimation was based on the findings obtained from the four selected groups, our results may not be interpreted as a true rate in this community. It can be safely said, however, that our rate of cerebral involvement among active paragonimiasis cases (0.8%) is probably higher than the true rate, since repeated sputum examination would have uncovered more of active cases.

Our figures are remarkably low compared to the rates reported in the literature as shown

in Table 1. We feel that 0.8% is much more realistic. Other factor which influences the higher rate is the criteria for diagnosis. In the literature, only three reports dealt with the clinically well proven cases with cerebral paragonimiasis (Chang *et al.*, 1958; Park, 1963; Iwasaki, 1962). Rates in these reports were much lower than the others except for Chang's (24%). Chang's study was carried out in a hospital where many patients were referred by different hospitals. He stated in his paper that this explained his higher rate. The validity of Grauman's rate (1957) has been doubted by many. The criteria of diagnosing the cerebral paragonimiasis was not given in his paper. It is more likely that "neurological symptoms" were found in 26.6% of active cases with paragonimiasis. The fact that the different criteria of diagnosis and sources of material change the rate of cerebral complication was well shown by Sadun (1960): the rate varied from 7.0 to 23.8%. This suggests strongly that there have to be strict criteria for diagnosis in this kind of study.

Although the rate of cerebral paragonimiasis among positive reactors is small, there are estimated to be 7,448 cases of cerebral paragonimiasis in Korea as of January 10, 1966, if 13% average positive rate reported by Walton & Chyu (1959) is applied to the nation-wide population census.\* This high prevalence emphasizes the importance of cerebral paragonimiasis as public problem in Korea since this disease is treatable and cerebral affection is preventable.

### Summary

To determine the rate of cerebral involvement among active paragonimiasis patients a field survey was conducted in an endemic area, Pa Joo, Korea. The intradermal P.w. test was used as the screening test and the examination of a single sputum for P.w. ova was a mean of detecting active cases.

Our result showed 0.2% of positive reactors

\* Total census, January 10, 1966: 28,647,176

Source: Research & Statistics Bureau, Economic Planning Board, the Republic of Korea.

to skin test and 0.8% of the active paragonimiasis cases had cerebral involvement.

Previous literature was reviewed and compared with our findings. Probable reasons for our lower rates are discussed.

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## 肺吸虫症における脳症状の発生率, その疫学的研究

Shin Joong OH

*(Division of Neurology, Meharry Medical College,  
Nashville, Tennessee, U. S. A.)*

韓国の肺吸虫症流行地の1つである Pa Joo 地方において肺吸虫症として診断された患者の脳肺吸虫症発生率について疫学的な調査をした。

患者のスクリーニングは皮内反応及び喀痰内肺吸虫卵の1回検査法によつた。その結果肺吸虫皮内反応の陽性

者の0.2% (4名/1,994名)に、活動性肺吸虫症患者の0.8% (4名/498名)に肺吸虫の脳内寄生を認めた。

本篇においてはこれらの仔細を報告し、かつ先人の韓国における脳肺吸虫症発生の疫学的調査成績と比較して考察をなした。