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Studies on Intestinal Trematodes in Korea I. A Human Case of *Echinostoma cinetorchis* Infection with an epidemiological Investigation

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INTRODUCTION

The species *Echinostoma cinetorchis* was firstly described by Ando et Ozaki (1923) from the rats, *Rattus norvegicus* and several human infections have been reported mainly in Japan (Takahashi et al., 1930 a & b; Kawahara et Yamamoto, 1933 and Moriyama, 1952). On the *E. cinetorchis* in Korea, the natural infection of a wild rat of Chungpyong area, Kyunggi Do was firstly described by Seo et al. (1964). However, no other reports including human case have been made until present.

On the other species of the genus *Echinostoma*, a few reports have been made in Korea. Isshiki (1934) reported the infection of a wild duck of Pusan with *E. revolutum*, Park (1938) and Seo et al. (1964) recorded *E. hortense* from rats of Seoul and other areas, and Chu et al. (1973) described *E. revolutum*, *E. gotoi* and *E. miyagawai* from wild birds from various parts of Korea.

The recovery of large, immature, operculated trematode eggs from human stool, evidently different from those of *Fasciola* sp. or *Paragonimus* sp. was sometimes at the disposal of some workers in Korea. However, such cases were never confirmed as authentic infection.

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In the present paper, the authors proved an authentic human infection with *E. cinetorchis* by obtaining adult worms, from the area of Hwasung Gun, Kyunggi Do, Korea. An epidemiological investigation was made in the area to find out the source of infection.

HUMAN CASE AND WORM DESCRIPTION

The patient, 60-year old house-wife, was residing in a rural area of Korea since birth; Hwariheun-l-Ri, Hyangnam Myon, Hwasung Gun, Kyunggi Do. Stool examination on January 1977 revealed positive for large, immature, straw-colored trematode eggs together with eggs of *Ascaris lumbricoides* and *Trichuris trichiura*. Spurious infection by *Fasciola* sp. etc. was denied by consistent appearance of the eggs in stool on November 1977 and February 1978. *Ascaris* infection was treated with pyrantel pamoate at the dose of 10mg/kg after the first examination.

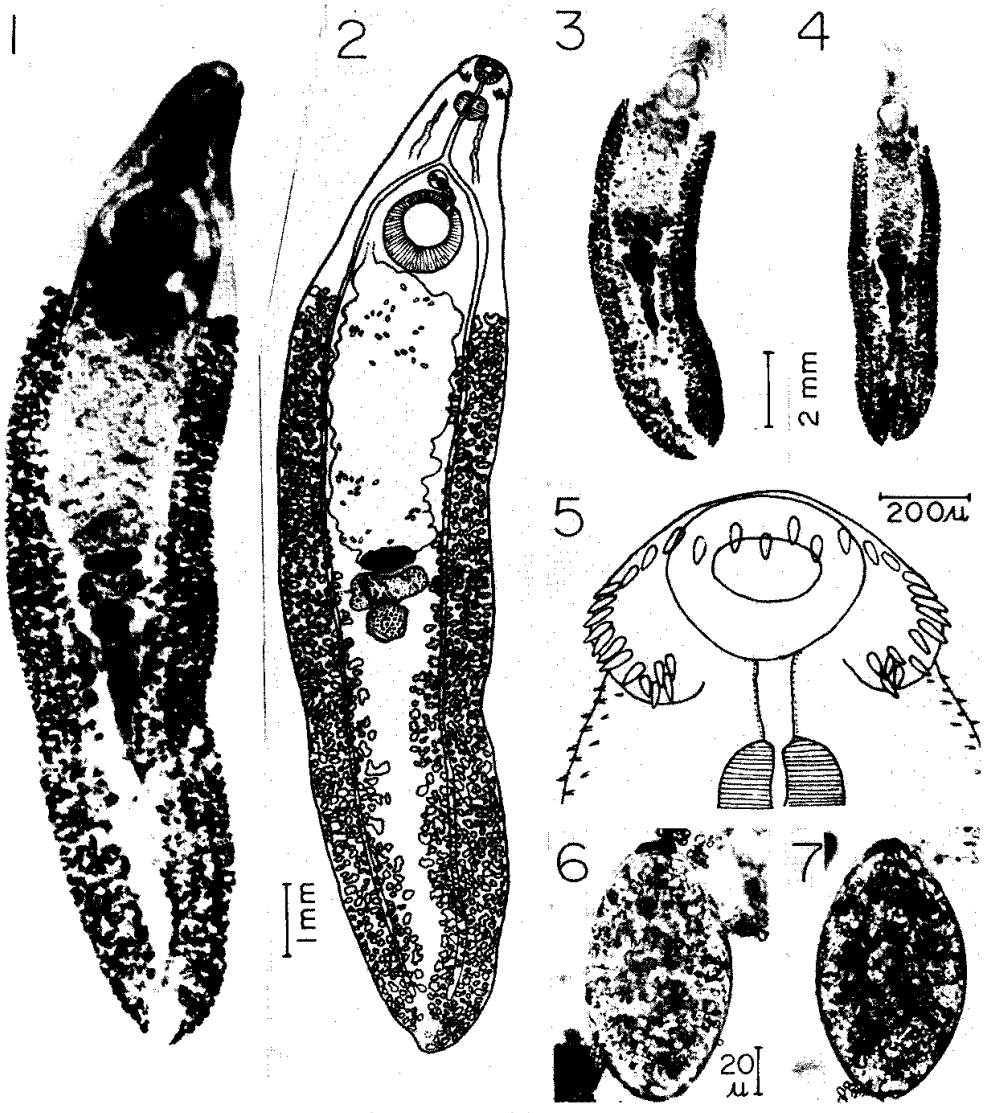
On February 1978, the patient was treated with bithionol at the dose of 35mg/kg after premedication with cathartics. One and half hour after the treatment, eight worms were collected from the watery stool of the patient. The worms were about 1 cm in length, reddish at the marginal part of the body and C-shaped in coiled state, leaf-like on pressed state. They were distinct with head crown around oral sucker and

had protruded acetabulum. Collected worms were fixed in Alcohol-Formalin-Acetic acid fixative and stained with Semichon's acetocarmine.

The patient had experienced intermittent abdominal pain since one year before the treatment and the episode of pain appeared nearly every month with the duration of 2 or 3 days. Other gastrointestinal symptoms such as nausea, vomiting, indigestion, diarrhea or constipation were not noted consistently. Laboratory tests were not performed.

Follow-up stool examination was done on March 1978 again and complete cure was assured by the disappearance of the trematode eggs. Abdominal pain also disappeared up to one year follow-up period.

Description of the worms: *Echinostoma cinetorchis* Ando et Ozaki, 1923(Figs. 1, 2, 3 and 4)



Figs. 1–7. *Echinostoma cinetorchis* from presented human case.
1. Adult worm with single testis. 2. *Ibid*, drawing of Fig. 1. Main structures are described. 3. Another adult worm with smaller size of testis than Fig. 1. 4. *Ibid* without testes. 5. Head crown of adult worm in Fig. 1 showing total 36 collar spines and 6 end group spines. 6&7. Eggs from human stool.

Eggs: Oval in shape (Figs. 6 and 7) and 99—116 ×65—76 μm in size (Table 1). Operculum small and most distal in position measured 16~21 μm in width and 2~4 μm in height. Egg shell thin and refractile measuring about 8 μm.

Adults: Measured data of the worms are presented in Table 2 and compared with other workers' results.

Body elongated, dorsoventrally flattened. Head crown distinct, bearing 36 collar spines not interrupted dorsally in one best specimen (Fig. 5) and more than 30 in other worms. Number of end group spines counted 6 on each side. Tegumentary spines distinct and extended from collar to posterior one-fifth level of the body.

Oral sucker subterminal and pre-pharynx present. Pharynx well developed, muscular and longer than width. Ventral sucker very large and well developed. Esophagus slender and intestine bifurcating at anterior end portion of cirrus sac. Ceca terminating near the posterior end. Excretory bladder Y-shaped and extended from the level of testis to excretory pore. Excretory pore small and subterminal.

Table 1. Comparison of the size of the eggs of *E. cinetorchis*

Author	Size of eggs(μm)	Source of eggs	Host
Present authors(1980)	99~116×65~76	Stool	Human
Kawahara et Yamamoto(1933)	105×68	Stool	Human
Sugimoto(1933)	96~99×47~53	*Intrauterine	Dog
Hirazawa(1926)	97~100×69~82	**Intrauterine	Dog, Rat
Ando et Ozaki(1923)	96~100×61~70	**Intrauterine	Rat
Takahashi(1927)	105×65	Stool	Rat
Seo et al. (1964)	89~96×53~59	*Intrauterine	Rat

* Measured at stained specimens
** Measured at fixed specimens

Table 2. Comparison of the measurements of *E. cinetorchis* adults by authors(mm)

	Present authors (1980)	Takahashi et al. (1930 a&b)	Sugimoto (1933)	Ando et Ozaki (1923)	Seo et al. (1964)
Host	Human	Human	Dog	Rat	Rat
Body length	10.8~12.6	12.0~14.0	9.5~10.4	9.5~14.6	12.2
Body width	2.6~3.2	about 2.0	2.0~2.5	1.7~2.2	2.8
Oral sucker	0.39~0.43 ×0.43~0.61	—	0.32	0.26~0.30	0.27×0.28
Ventral sucker	1.07~1.21 ×0.84~1.15	—	0.92	0.72~0.73	0.85×0.76
Diameter of head collar	0.75~0.83	—	0.74	0.44~0.53	0.50
No. of collar spines	36	37	35	37	37
Pharynx	0.37~0.43 ×0.31~0.37	—	0.31	0.19~0.25 ×0.20~0.21	0.21
Ovary	0.51~0.80 ×0.20~0.37	—	0.56~0.60 ×0.35~0.45	0.41~0.62 ×0.22~0.30	0.52×0.42
Cirrus sac	0.26~0.43 ×0.17~0.40	—	0.56	0.47~0.54 ×0.26~0.30	0.29×0.25
Testes anterior	0.35~0.68 ×0.20~0.66	—	0.42×0.19	0.44~0.61 ×0.44~0.61	0.34×0.25
posterior	none	—	0.38×0.42	0.58~0.91 ×0.38~0.62	0.36×0.27

Cirrus sac anterior to ventral sucker, nearly median, globular in shape and not so elongated. Seminal vesicle elongated with some sacculations. Ejaculatory duct short and slender. Testis single in 6 worms and none in other 2 worms. Remaining one testis median, not lobulated, post-equatorial in position and variable in size (Figs. 1 and 3).

Ovary elongated horizontally and on median line of the body. Mehlis' gland large and immediately behind ovary. Vitelline glands well developed, follicular and laterally extending from the level of acetabulum to posterior end of the body. Uterus long and wound, locating between acetabulum and ovary.

EPIDEMIOLOGICAL INVESTIGATION

To investigate the source of infection in the area, the possible intermediate and final hosts study was done. As shown in Table 3, the snails, 191 *Lymnaea* sp. and 26 *Viviparus* sp. and the fishes, 12 *Acheilognathus* sp. and 84 *Misgurnus anguillicaudatus* were collected from the ponds adjacent to rice fields. Collected snails were examined by crushing method and the fishes by muscle compression or flesh grinding method. Grinding method was used in loaches, *M. anguillicaudatus*. Ground flesh was dipped in saline solution, filtered repeatedly through coarse gauze, then examined under dissecting microscope.

As in Table 3, all of the snails examined were negative for echinostomatid cercariae and metacercariae. The fishes, *Acheilognathus* sp. were also negative for metacercariae. However, a total of 5 echinostomatid metacercariae were collected from the 5 loaches out of 84 examined.

Table 3. Results of intermediate hosts study in the patient's village

Intermediate host	No. exam.	No. positive (%) for echinostomatid	
		Cercariae	Metacercariae
Lymnaea sp.	191	0(0.0)	0(0.0)
Viviparus sp.	26	0(0.0)	0(0.0)
Acheilognathus sp.	12	—(—)	0(0.0)
Misgurnus anguillicaudatus	84	—(—)	*5(6.0)

* All of the infected loaches had one metacercaria each.

Each metacercaria was separately fed to 5 albino rats to identify the species, but only one adult worm was recovered from the ileum of a rat on 38th day after infection. The worm was fixed in Alcohol-Formalin-Acetic acid solution followed by acetocarmine staining. The morphology of the adult worm and the size of the eggs from the rat stool were, however, not agreeable to *E. cinetorchis* but identified as *E. hortense*.

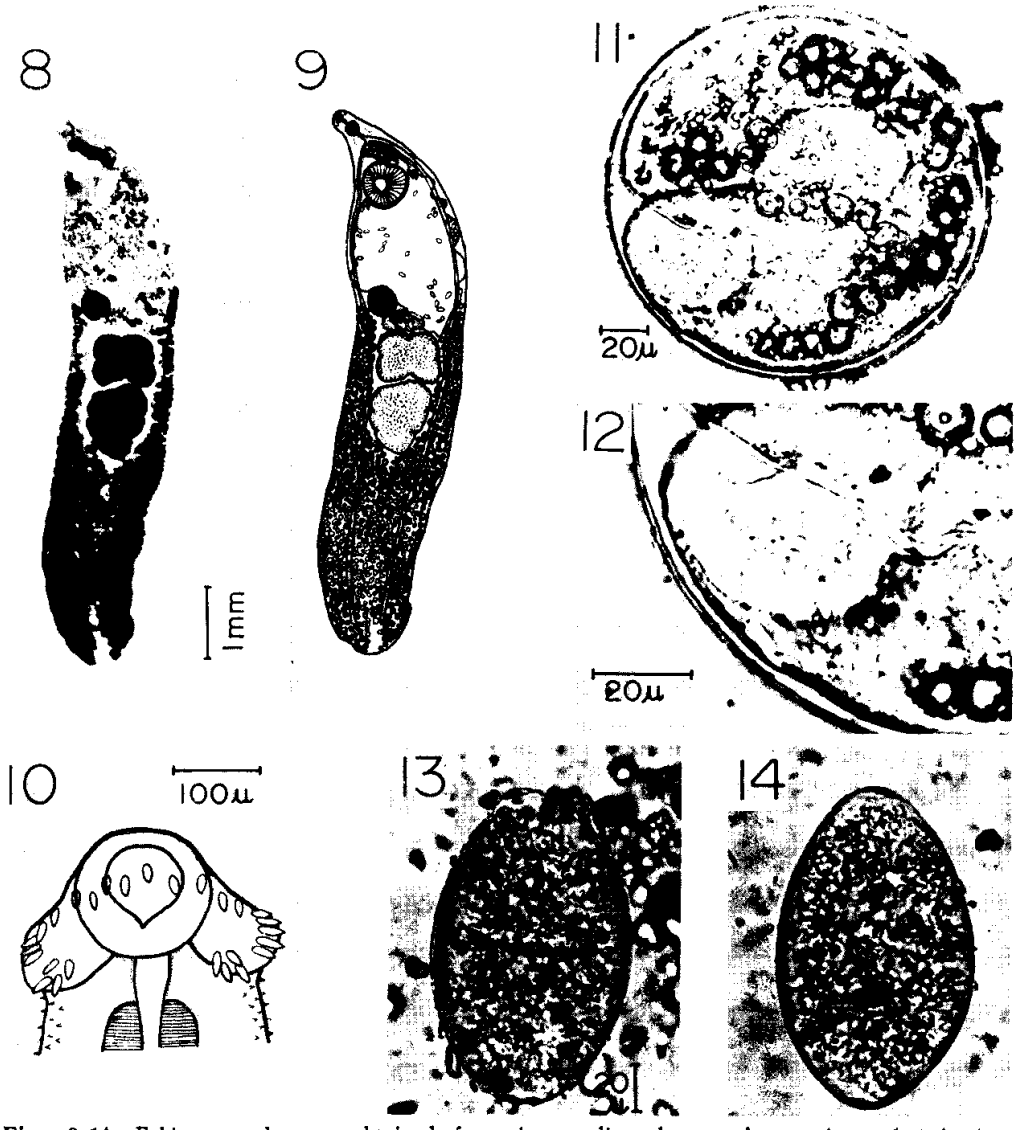
For reservoir host study, stool specimens of 10 cats and 6 dogs bred in the area were examined for trematode eggs, but none of them revealed echinostomatid eggs. Fifty people in that area including the patient's family were subjected to stool examination to detect any other infection case, however, the result was also negative in all cases.

Description of the worm: *Echinostoma hortense* Asada, 1926 (Figs. 8 and 9)

Metacercaria: Cyst nearly round (Fig. 11), 160~165×140~163 μm in size. Collar spines distinct around oral sucker (Fig. 12) but the number hardly countable in encysted state. Excretory bladder Y-form, extending longer than posterior half of the body and filled with many excretory corpuscles. Ventral sucker located largely at middle portion of the body.

Eggs: Morphology very similar to those of *E. cinetorchis*, although larger in size(Figs. 13 and 14), 126~140×73~86 μm (132×79 in average). Height of the operculum ranged from 3 to 7 μm and the width from 16 to 35 μm.

Adult: Body elongated, dorsoventrally flattened, 7.6mm in length and 1.3mm in greatest width, and attenuated at anterior end (Figs. 8 and 9). Head crown distinct, 0.30mm across, bearing 27 collar spines not interrupted dorsally. Dorsal spines arranged



Figs. 8–14. ***Echinostoma hortense*** obtained from intermediate host and experimental infection. 8. 28-day adult worm from experimental rat **9.** *Ibid*, drawing of Fig. 8. **10.** Head crown of adult worm in Fig. 8 showing total 27 collar spines with 4 end group spines. **11.** Metacercaria from the loach, ***Misgurnus anguillicaudatus.*** **12.** Magnification of Fig. 11 showing 4 end group spines. **13.** & **14.** Eggs from experimental rat.

in two rows and the end group spines 4 in number on each side (Fig. 10). Tegumentary spines covered from the anterior end to the level of posterior testis.

Oral sucker subterminal, 0.17 by 0.15mm and prepharynx present. Pharynx well developed and nearly round in shape, 0.17 by 0.18mm in size. Ventral sucker well developed and 0.45 by 0.50mm in size. Esophagus slender and muscular, and intestine bifurcating midway between two suckers. Ceca terminating near the posterior end of the body. Excretory bladder extended from the posterior testis to excretory pore. Excretory pore small and subterminal.

Cirrus sac closely behind intestinal bifurcation, submedian and elongated, and 0.41mm in length and 0.13mm in width. Seminal vesicle prominent in cirrus sac. Testes tandem in position, large and lobulated, measuring 0.83 by 0.70mm in anterior one and 0.80 by 0.96mm in posterior one. The position of the testes to the body nearly equatorial.

Ovary spheroidal, 0.37 by 0.40mm in size and deviated to right side of the body. Mehlis' gland large, submedian and behind ovary. Vitelline follicles well developed, laterally extending from the level of ovary to posterior end. Uterus somewhat wound and located between acetabulum and ovary. Metraterm connected to female genital opening and parallel with cirrus sac.

DISCUSSION

According to Ando et Ozaki (1923), the specific characteristic of *E. cinetorchis* was the abnormal location or moving tendency of one testis from normal position. They also recorded some worms with single testis or without testes among *E. cinetorchis* they collected. Seo et al. (1964) recorded one *E. cinetorchis* worm both testes of which migrated to bilateral sides to ovary. However, as shown in Table 4, reduction in number of testes seems another characteristic of this species. Takahashi(1927) and Takahashi et al. (1930 a & b) mentioned that the majority of *E. cinetorchis* they collected from human cases and from experimental rats had single testis and sometimes no testes. Hirazawa(1926), Sugimoto (1933) and Kawahara et Yamamoto (1933) also recorded worms with single testis. Our specimens did not show any abnormal location of testis but revealed reduction in number and variation in size of testes.

Major morphological characteristics of *E. cinetorchis* in our specimens are as follows:

1. variation or reduction in number and size of testes
2. total 36 collar spines and 6 end group spines
3. location of ovary on median line of the body
4. anteriormost level of the distribution of vitelline follicles
5. maximum width of the worms near the middle portion of the body
6. the size of the eggs in stool, about 100~115×65~75 μm

E. hortense is different from *E. cinetorchis* in the items 1, 2, 3, 4 and 6. *E. revolutum* is the most similar species to *E. cinetorchis*, although differs in the items 1, 2 and 5, i.e., no variation in testes, end group spines 5 in number and maximum width of the body at the acetabular level. Beaver (1937) regarded *E. cinetorchis* as

Table 4. Comparison of number and location of testes in *E. cinetorchis* worms according to authors

Author	No. obtained worms	Host	No. worms with						
			No testis	Single testis		Two testes			Uncertain
				*A	*B	*C	*D	*E	
Present authors (1980)	8	Human	2	6	0	0	0	0	0
Kawahara et Yamamoto(1933)	3	Human	0	1	0	0	1	0	1
Takahashi et al. (1930a)	17	Human	1	6	1	1	1	0	7
Takahashi et al. (1930b)	24	Human	4	12	3	0	0	0	5
Ando et Ozaki (1923)	38	Rat	4	3	0	15	16	0	0
Takahashi (1927)	40	Rat**	7	26	0	1	6	0	0
Seo et al. (1964)	1	Rat	0	0	0	0	0	1	0
Sugimoto (1933)	3	Dog	0	2	0	1	0	0	0

* A: behind Mehlis' gland, B: other location, C: two testes behind Mehlis' gland in tandem position, D: anterior one behind Mehlis' gland and another one migrated, E: both testes migrated

** Experimental infection with metacercariae.

a synonym of *E. revolutum*, but Yamaguti(1958) did not agree to the synonymy. Anyhow, present authors regarded our specimens as *E. cinetorchis*, considering the testes variation as the most characteristic feature of this species.

The second intermediate hosts of *E. cinetorchis* are known to be the large snails, the tadpoles of the frogs, the larva of the salamander and the loaches (Komiya, 1965), which are largely overlapped with those of *E. hortense* or other species of *Echinostoma*. Although not all of the above hosts were collected and examined, the life cycle study in this paper resulted in negative for *E. cinetorchis* in snails and fishes, so the infection source is not clear at present.

Interestingly, a part of life cycle of *E. hortense* was incidentally found during the intermediate hosts study in that area. Obtained adult worm was identified as *E. hortense* because of total 27 collar spines with 4 end group spines on each side, laterally deviated ovary, distribution of vitelline follicles from the level of ovary to the posterior end and the size of the eggs about 130 by 80 μm The measured data of the worm were clearly agreed to other records (Park, 1938 and Seo et al., 1964).

The fact that the loaches, *M. anguillicaudatus* serve as the second intermediate host of *E. hortense* was confirmed again in this study following the reports of Okamoto (1953) and others. Tani(1976) included Japanese bitterlings, *Acheilognathus morikae* as a new second intermediate host. Large snails are not included until present (Komiya, 1965).

In the treatment of human infection with various species of *Echinostoma* and *Echinochasmus*, Japanese workers have used thymol, carbon tetrachloride, tetrachlorethylene and kamala (Tani, 1976). In this study bithionol was firstly tried and single dose of bithionol was also effective and safe for the satisfactory removal of *E. cinetorchis* worms.

According to Yamashita (1964), human infection with echinostomes had been recorded by 13 species of the family Echinostomatidae including 5 species of *Echinostoma*. Recently human infection with *E. hortense* was found in Japan(Tani et al., 1974; Arizono et al., 1976), hence 14 kinds of echinostomes are recorded from human. Considering the echinostomatid eggs were found sporadically from inhabitants of Korea, more human cases of echinostomiasis would be detected if attention is paid to these kinds of flukes in Korea.

SUMMARY

A case of human infection with *Echinostoma cinetorchis* was found firstly in Korea on February 1978. The patient was a 60-year old house-wife residing in Hwasung Gun, Kyunggi Do and suffered from intermittent abdominal pain for about one year. Stool examination revealed positive for echinostomatid eggs three times consistently during about 1 year. Eight worms were collected after bithionol treatment with cathartics. Collected worms had distinct head collar with 36 spines, and revealed marked variation in number and size of testes. So they were identified as *E. cinetorchis*.

Although an intermediate host study was made to investigate the source of infection of *E. cinetorchis*, not a relevant finding to the human case was obtained. Some of the loaches, *Misgurnus anguillicaudatus* had echinostomatid metacercariae but one adult worm, obtained successfully from animal experiment, was identified as *Echinostoma hortense*.

The echinostomatid eggs have been sometimes found from Koreans, so close observation and tracing of the positive cases would be helpful to understand further details of human echinostomiasis in Korea.

=國文抄錄=

韓國의 腸吸蟲類에 關한 硏究
I. 國內最初의 移轉睾丸棘口吸蟲(*Echinostoma cinetorchis*)의 人體感染 1例 및 疫學調査

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우리나라에서는 처음으로 移轉睾丸棘口吸蟲의 人體感染 1例를 1978年 2月에 成蟲을 얻어 確認하였다. 환자는 京畿道 華城郡에 거주하는 60세 主婦로 1年前부터 때때로 腹痛을 경험하였으며 1977년 1월의 대변검사에서 크기 99~116×65~76μm인 吸蟲類의 大形蟲卵이 나타났었고 그후 10개월 및 13개월째에도 계속 蟲卵을 排出하고 있었다. 治療를 위해 비치오놀(bithionol) 35 mg/kg을 투여하고 下劑로 蟲體排出을 시도한 결과 泄瀉便으로부터 8마리의 蟲體를 얻을 수 있었다. 蟲體는 頭冠이 뚜렷하였고, 모두 36개의 頭棘을 가지고 있었으며, 睾丸의 數와 크기에 變異를 나타내고 있어서 이전고환극구흡충으로 同定하였다.

이지역에서 환자의 感染源을 찾기 위하여 중간숙주 및 종숙주에 대한 疫學調査를 실시하였으나 本 蟲種과 부합되는 결과를 얻을 수 없었다. 中間宿主로 알려져 있는 미꾸라지중 5마리(6.0%)에서 棘口吸蟲類의 피낭유충을 발견하였으나 動物實驗에서 成功的으로 얻은 1마리의 성충은 호르텐스棘口吸蟲(*Echinostoma hortense*)으로 판정되었다.

棘口吸蟲類의 蟲卵은 우리나라의 집단대변검사에서 때때로 발견되어 왔으므로 蟲卵의 상세한 관찰 및 연속 陽性여부등을 확인하고 적당한 치료를 행한다면 성충을 기초로 한 人體感染例는 더 찾아낼 수 있을 것으로 생각하였다.

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