

The 12th Human Case of *Clinostomum* sp. Infection in Japan

AKIO ISOBE¹⁾, SHIN KINOSHITA²⁾, NOBUMASA HOJO¹⁾, TETSUHITO FUKUSHIMA¹⁾,
KUNINORI SHIWAKU¹⁾ AND YOSUKE YAMANE¹⁾

(Accepted for publication; June 3, 1994)

Abstract

The genus *Clinostomum* is one of the causes of parasitic laryngo-pharyngitis, "Halzoun". We reported the 12th case of *Clinostomum* sp. infection and reviewed all cases in Japan. The patient is a 68-year-old male fish-wholesaler who eats raw fishes every day. He complained of an discomfort sensation in the pharynx and a worm was removed, which was identified as *Clinostomum* sp.

Key words: *Clinostomum* sp., parasitic laryngo-pharyngitis, halzoun

Introduction

In Japan, eleven cases of parasitic laryngo-pharyngitis, "Halzoun", caused by the genus *Clinostomum* (Trematoda; Clinostomatidae) were reported in the past (Yamashita, 1938; Hori *et al.*, 1942; Kamo *et al.*, 1962; Sakaguchi *et al.*, 1966; Sano *et al.*, 1980; Kumada *et al.*, 1983; Hirai *et al.*, 1987; Furukawa and Miyazato, 1987; Yamane *et al.*, 1989; Umezaki *et al.*, 1989; Yoshimura *et al.*, 1991). We report here the 12th case and reviewed all the 12 cases in Japan.

Case Report

A 68-year-old man was admitted to the Oto-Rhino-Laryngology Unit, the Hirata Municipal Hospital on March 14, 1992, because of an unusual sensation in the pharynx, which was like a sticking fish bone and sometimes it gave a pain.

The patient was a fish-wholesaler, living in Hikawa-cho, Shimane Prefecture, Japan. People in Hikawa-cho have traditionally ingested raw fishes from the Lake Shinji, for example, silver carp, *Carrasius carrasius*, carp, *Cyprinus carpio*, common goby, *Acanthogobius flavimanus* and Japa-

nese sea perch, *Lateolabrax japonicus*.

The patient had taken raw silver carp every day before the onset of unusual sensation in the pharynx. A foreign body was observed attached to the dorsal wall of the left posterior palatum and it was removed with a pharyngoscope. This foreign body was fixed in 10% formol saline and sent to the Department of Environmental Medicine of Shimane Medical University for identification. After its removal, the discomfort in the pharynx disappeared. The patient had no history of headache, dyspnea and fever.

There were no special findings in the personal and the family history of the patient. The hematologic and blood chemical values are presented in Table 1. The serum IgE value is slightly elevated to 548.1 IU/ml. White cell count is 6,100 per mm³ without eosinophilia. No eggs are found by the fecal examination.

Morphological findings of *Clinostomum* sp.

The worm body, fixed in 10% formol saline, is white, flat and leaf-like, being 4.0 mm long and 1.65 mm wide (Figures 1 and 2). There are no spines on the body surface. The oral sucker, located on the frontal end of the body, has a collar-like form, being 0.34 mm long and 0.19 mm wide. The oral sucker is surrounded by a collar-like fold of the body wall. Then it was fixed in 70% alcohol again, and stained with aceticalum carmine. The ventral sucker (acetabulum) is located in the one fifth anterior part of the body, looking like a saucer of 0.40 mm long and 0.44 mm wide. The esophagus continues to the both intestinal canals. The intestinal caecum is ter-

¹⁾Department of Environmental Medicine, Shimane Medical University, 89-1 Enya-cho, Izumo, Shimane, 693, Japan.

²⁾Oto-Rhino-Laryngology Unit, Hirata Municipal Hospital, 613 Nadabun-cho, Hirata, Shimane 691, Japan.

磯邊 顯生 北條 宣政 福島 哲仁 塩飽 邦憲 山根 洋右 (島根医科大学環境保健医学教室)
木下 心 (平田市立病院耳鼻咽喉科)

Table 1 Laboratory data of the present case

Variable	Value
Red cell count (per mm ³)	544×10 ⁴
Hematocrit (%)	49.2
Hemoglobin (g/dl)	16.7
White cell count (per mm ³)	6,100
Differential count (%)	
Neutrophils Band	1
Neutrophils Segmented	49
Eosinophils	3
Lymphocytes	37
Monocytes	10
Platelet count (per mm ³)	188×10 ³
Glutamic oxaloacetic transaminase (IU/l)	17
Glutamic pyruvic transaminase (IU/l)	23
Total bilirubin (mg/dl)	1.58
Direct bilirubin (mg/dl)	0.26
Amylase (IU/l)	236
Urea nitrogen (mg/dl)	11.3
Creatinine (mg/dl)	1.0
Alkaline phosphatase (IU/liter)	145
Lactate dehydrogenase (IU/liter)	277
IgE (IU/ml)	548.1
C-reactive protein (mg/dl)	0.3

minated at the posterior extremity, and does not have long lateral branches. There are uterus, anterior testis and posterior testis beneath the ventral sucker. Testes are tandem. Genital pore is anterior to the testis, with vitellaria confined to the hind body. The size of eggs in the uterus is 0.100–0.115×0.054–0.065 mm (Fig. 3).

Discussion

This specimen appeared to be *C. complanatum* (Rudolphi, 1819) or *C. marginatum* (Rudolphi, 1819). However, the morphological relationship between caeca and excretory vesicle and also between ovary and testes were obscure in this specimen, we identified it as *Clinostomum* sp.

The symptom caused by *Clinostomum* sp. infection is known as "Halzoun", one of the parasitic laryngo-pharyngitis. "Halzoun" is also caused by the leech, *Limnatis nilotica*, *Fasciola hepatica* and *Clinostomum complanatum* (Witenberg, 1944).

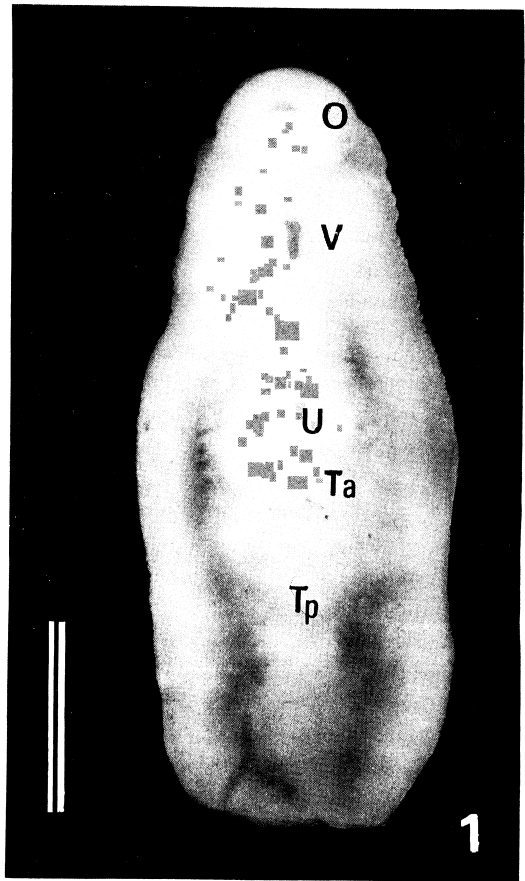


Fig. 1 Ventral view of *Clinostomum* sp. obtained from the present case. O: oral sucker, Ta: anterior testis, Tp: posterior testis, U: uterus, V: ventral sucker. Bar = 1 mm.

In Japan, 12 cases of "Halzoun" caused by *Clinostomum* sp. including the present case, have been reported (Tables 2 and 3). The causes of five cases of them were identified as *C. complanatum*. The age of the patients were 30, 50 and the rest over 50. Eight females were included, occupying 67% of all the patients. Chief complaints were mainly pain or abnormal sensation in the pharynx. In the 6th case (Kumada *et al.*, 1983), the patient had cough, blood sputum, hoarseness and fever in addition to pain in the pharynx. The symptom of the present case is applicable to "Halzoun".

The infectious source of *Clinostomum* sp. is commonly fresh-water fishes, which are the second



Fig. 2 Ventral view of *Clinostomum* sp. stained with acetic almaline. O: oral sucker, Ta: anterior testis, Tp: posterior testis, U: uterus, V: ventral sucker. Bar = 1 mm.

intermediate hosts of *Clinostomum* sp. Clinostomiasis occurs by eating these fishes with metacercariae of the parasite. The intermediate hosts of *Clinostomum* sp. reported in Japan are *Carassius carassius*, *Pseudogobio esocinus*, *Rhodeus (Aceilognathus) lanceolatus* (Yamaguti, 1933), *Misgurnus anguillicaudatus* (Kagei *et al.*, 1984), *Carassius gibelio langsdorfi*, *Carassius cuvieri*, *Cyprinus carpio*, *Pseudorasbora parva* and *Rhodeus ocellatus* (Aohagi *et al.*, 1992a). The infectious sources of the 12 cases in Japan are *Carassius carassius* 7, *Cyprinus carpio* 4, *Acanthogobius*

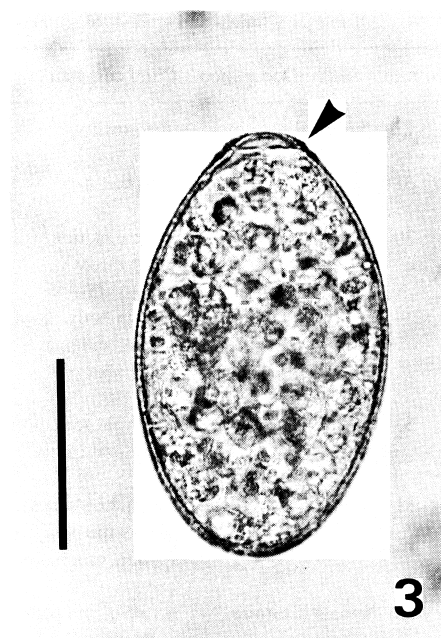


Fig. 3 Egg in the uterus showing an operculum (arrow). Bar = 50 μ m.

flavimanus 2, *Hypomesus japonicus* 1, *Lateolabrax japonicus* 1. The patient of Case 2 ate *Carassius carassius* captured in the Lagoon Junicho (Hori, 1942). The patient of Case 4 ate fishes captured at a pond nearby his house (Sakaguchi *et al.*, 1966). In Case 11, the patient ate raw fishes, especially roach (*Carassius carassius*) captured in the Lake Hachirogata and also at ponds nearby her house (Yoshimura *et al.*, 1991). It is more important for the infection of this parasite to eat natural fishes than raised fishes. Among the Japanese cases, Case 3, Case 9 and the present case occurred in the eastern part of Shimane prefecture. There are two brackish water lakes, Lake Shinji and Nakaumi, in this district. The fishes in these lakes include the second intermediate hosts of *Clinostomum* sp. Aohagi *et al.* (1993a) found metacercariae of *C. complanatum* from *C. cuvieri*, which were caught from three lake including the Lake Shinji. As the present patient ate raw fishes every day, particularly *Carassius carassius*, we suspect *Carassius carassius* is the cause of infection. The infectious sources of Case 3 and Case 9 are exactly unknown.

Table 2 Clinical and epidemiological data of human cases of *Clinostomum* sp. infection in Japan

Case	Age	Sex	Occupation	Chief complaints	Infectious source	Locality	Author	Year
1	22	female	unknown	unknown	unknown	Osaka	Yamashita	1938
2	38	female	unknown	dysphagia	<i>Carassius carassius</i>	Toyama	Hori <i>et al.</i>	1942
3	30	male	office worker	acute irritation in the throat	<i>Carassius carassius</i> <i>Acanthogobius flavimanus</i>	Shimane	Kamo <i>et al.</i>	1962
4	34	male	farmer	foreign body feeling and pain in the pharynx	<i>Carassius carassius</i>	Nagasaki	Sakaguchi <i>et al.</i>	1966
5	53	male	farmer	abnormal sensation in the oral cavity	unknown	Gifu	Sano <i>et al.</i>	1980
6	31	female	unknown	cough, hoarseness, pain in the pharynx, sputum, cruentum	<i>Cyprinus carpio</i>	Aichi	Kumada <i>et al.</i>	1983
7	35	female	farmer	irritation and pain of the pharynx	<i>Carassius carassius</i>	Kumamoto	Hirai <i>et al.</i>	1987
8	57	female	housewife	abnormal sensation in the pharynx, dysphagia	unknown	Shiga	Furukawa <i>et al.</i>	1987
9	15	female	student	abnormal sensation in the pharynx	<i>Carassius carassius</i> <i>Cyprinus carpio</i>	Shimane	Yamane <i>et al.</i>	1990
10	54	female	unknown	abnormal sensation in the pharynx	<i>Cyprinus carpio</i>	Saga	Umezaki <i>et al.</i>	1990
11	70	female	farmer	irritation and pain of the pharynx	<i>Hypomesus japonicus</i> <i>Carassius carassius</i>	Akita	Yoshimura <i>et al.</i>	1991
12	68	male	fish-wholesaler	abnormal sensation in the pharynx	<i>Carassius carassius</i> <i>Cyprinus carpio</i> <i>Acanthogobius flavimanus</i> <i>Lateolabrax japonicus</i>	Shimane	Present Authors	1994

The final hosts of *Clinostomum* sp. are frogs, cats and waterfowls such as herons and egrets. The final hosts of *Clinostomum* sp. reported in Japan are *Nycticorax nycticorax* (Yamaguti, 1933; Kagei *et al.*, 1988), *Ardea cinerea*, *Egretta garzetta*, *Egretta intermedia* (Aohagi *et al.*, 1992b) and *Egretta alba* (Aohagi *et al.*, 1993b). The Lake Shinji is a brackish water lake and a treasury of wild birds. This seems to be the reason why the life cycle of *Clinostomum* sp. is maintained. It is important to pay attention to

these "Halzoun" for physicians in the district.

Acknowledgments

The authors wish to thank Mrs. T. Yoneyama and Mr. M. Ohshita, for their technical supports.

Table 3 Morphological data of *Clinostomum* sp. obtained from human cases in Japan

Case	1	2	3	4	5	6	7	8	9	10	11	12
Body length (mm)	3.7×1.3	ND	5.2×2.2	3.03×1.25	4.7×2.0	2.1×1.2	3.02×1.44	8.45×2.95	6.2×1.8	4.9×1.9	7.00×2.43	4.00×1.65
					immature							
Body surface	unarmed	ND	unarmed	armed	ND	ND	ND	unarmed	ND	ND	unarmed	unarmed
Collor	+	ND	+	ND	ND	+	+	-	+	+	ND	+
Oral sucker (mm)	0.44	ND	ND	0.15×0.13	0.38×0.33	0.23	0.21×0.27	0.50×0.38	0.30×0.35	+	0.273×0.444	0.34×0.19
Ventral sucker (mm)	0.95	ND	0.86×0.88	0.63×0.36	0.76×0.67	0.570	0.50×0.69	0.93	0.75×0.75	+	0.848×0.859	0.40×0.44
Testes (mm)	tandem	ND	tandem	tandem	tandem	ND	degenerated	tandem	tandem	tandem	tandem	tandem
anterior	ND	ND	0.500×0.750	0.350×0.225	0.2×0.5			0.80×0.60	0.80×0.33	+	0.450×0.727	0.28×0.41
posterior	ND	ND	0.325×0.750	0.385×0.290	0.3×0.4			0.93×0.45	0.63×0.50	+	0.394×0.990	0.27×0.54
Ovary (mm)	0.27	ND	0.200×0.275	0.125×0.081	uncertain	ND	degenerated	0.43×0.35	0.30×0.30	+	0.200×0.430	uncertain
Union of intestine & bladder	ND	ND	+	+	+	ND	ND	+	ND	ND	ND	uncertain
Egg (µm)	108.0–127.0	ND	120.0–129.0	106.0–119.0	absent	120.0–130.0	115.0–123.0	130.0×80.0	ND	+	118.0–125.0	100.0–115.0
	×		×	×		×	×				×	×
	70.0–80.0		67.0–75.0	63.0–70.0		80.0–85.0	59.0–69.0				75.0–80.0	54.0–65.0
Species	<i>C. complanatum</i>	<i>C. complanatum</i>	<i>C. sp.</i>	<i>C. sp.</i>	<i>C. sp.</i>	<i>C. sp.</i>	<i>C. complanatum</i>	<i>C. sp.</i>	<i>C. sp.</i>	<i>C. complanatum</i>	<i>C. complanatum</i>	<i>C. sp.</i>

ND; no description

References

- 1) Aohagi, Y., Shibahara, T., Machida, N., Yamaga, Y. and Kagota, K. (1992a): *Clinostomum complanatum* (Trematoda: Clinostomatidae) in five new fish hosts in Japan. *J. Wild. Dis.*, 28(3), 467–469.
- 2) Aohagi, Y., Shibahara, T., Machida, N., Yamaga, Y., Kagota, K. and Hayashi, T. (1992b): Natural infections of *Clinostomum complanatum* (Trematoda: Clinostomatidae) in wild herons and egrets, Tottori Prefecture, Japan. *J. Wild. Dis.*, 28(3), 470–471.
- 3) Aohagi, Y., Shibahara, T. and Kagota, K. (1993a): *Clinostomum complanatum* (Trematoda) infection in freshwater fish from fish dealers in Tottori, Japan. *J. Vet. Med. Sci.*, 55(1), 153–154.
- 4) Aohagi, Y., Shibahara, T. and Kagota, K. (1993b): A newly recognized natural definitive host of *Clinostomum complanatum* (Rudolphi, 1814) in Japan. *Jpn. J. Parasitol.*, 42, 44–46.
- 5) Furukawa, T. and Miyazato, T. (1987): A case of *Clinostomum* sp. infection in human pharynx. *Med. J. Kinki Univ.*, 12, 665–669 (in Japanese).
- 6) Hirai, H., Oiso, H., Kifune, T., Kiyota, T. and Sakaguchi, Y. (1987): *Clinostomum complanatum* infection in posterior wall of the pharynx of a human. *Jpn. J. Parasitol.*, 36, 142–144.
- 7) Hori, K. (1942): *Clinostomum complanatum* (Rud. 1819) an der Rachenschleimhaut des Menschen shmarotzend. *Jibi Inkoka* (Otolaryngology), 15, 249–252 (in Japanese).
- 8) Kagei, N., Yanohara, Y., Uchikawa, R. and Sato, A. (1984): On the yellow grubs, metacercariae of *Clinostomum complanatum* (Rudolphi, 1819), found in the cultured loach. *Jpn. J. Parasitol.*, 33, 59–62.
- 9) Kagei, N., Yanohara, Y., Uchikawa, R. and Sato, A. (1988): Natural infection with *Clinostomum complanatum* (Rud., 1819) in the birds of Southern Japan. *Jpn. J. Parasitol.*, 37, 254–257.
- 10) Kamo, H., Ogino, K. and Hatsushika, R. (1962): A unique infection of man with *Clinostomum* sp., a small trematode causing acute laryngitis. *Yonago Acta Medica*, 6, 37–40.
- 11) Kumada, N., Mizuno, S., Kawamoto, F., Fujioka, H. and Nakanishi, K. (1983): A case of *Clinostomum* sp. infection in Aichi Pref., Japan. *Jpn. J. Parasitol.*, 32, Supple, 17 (in Japanese).
- 12) Sakaguchi, Y., Yamamoto, T. and Yamada, N. (1966): *Clinostomum* sp., a trematoda parasite, from larynx of a man. *End. Dis. Bull. Nagasaki Univ.*, 8, 40–44 (in Japanese).
- 13) Sano, M., Mogi, K. and Kamegai, S. (1980): A case of *Clinostomum* sp. throat infection. *Jibi Inkoka* (Otolaryngology), 52, 1037–1039 (in Japanese).
- 14) Umezaki, T., Shin, T., Oda, M., Kifune, T. and Mogi, M. (1989): A case of acute laryngitis caused by *Clinostomum complanatum* with a complaint of throat irritation. *Jibi To Rinsho*, 36, 665–668 (in Japanese).
- 15) Witenberg, D. G. (1944): What is the cause of the parasitic laryngo-pharyngitis in the Near East ("Halzoun")? *Acta Med. Orient. (Jerusalem)*, 3, 191–192.
- 16) Yamaguti, S. (1933): Studies on the helminth fauna of Japan, Part 1, trematodes of bird, reptiles and mammals. *Jap. J. Zool.*, 5, 66–72.
- 17) Yamane, Y., Osaki, Y., Fujino, N. and Nagasako, R. (1989): The 8th human case of *Clinostomum* sp. infection in Japan. *Jpn. J. Parasitol.*, 39, 117 (in Japanese).
- 18) Yamashita, J. (1938): *Clinostomum complanatum*, a trematode parasite new to man. *Annot. Zool. Jap.*, 17, 563–566.
- 19) Yoshimura, K., Ishigooka, S., Satoh, I. and Kamegai, S. (1991): *Clinostomum complanatum* from the pharynx of a woman in Akita, Japan. A case report. *Jpn. J. Parasitol.*, 40, 99–101.