

Female Worm *Hysterothylacium aduncum* Excreted from Human: A Case Report

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Abstract

A case of anisakidosis caused by a young adult nematode of *Hysterothylacium aduncum* (Rudolphi, 1802) was reported. The patient was a 55-year-old man who had been suffering from diarrhea and abdominal pain for a month. A living worm was found in his feces. The worm was examined morphologically using a phase-contrast microscope, a light microscope against the sections of nematode and a scanning electron microscope. The excreted worm was identified as *H. aduncum*. The morphological findings were compared with those of a female adult of *H. aduncum* (infected Host: *Cleisthenes pinetorum herzensteini*) caught in the North Pacific Ocean off Kushiro, and reported by Moravec and Nagasawa (1985) and Moravec *et al.* (1985). The present case, therefore, represents a rare example of anisakidosis caused by a passage of *H. aduncum* through a human alimentary canal.

Key words: Anisakidae; anisakiosis; *Hysterothylacium aduncum*; adult worm.

Introduction

Anisakidosis, which now includes anisakiosis, pseudoterranovosis and contractaecosis, was first reported in the Netherlands (van Thiel *et al.*, 1960); that case suffered anisakiosis (previously anisakiasis) caused by *Anisakis simplex* (Rudolphi, 1809). In 1967, Rausch *et al.* (1967) reported the first case of pseudoterranovosis due to *Pseudoterranova decipiens* (Krabbe, 1878) larva in the United States. Subsequently, a human case caused by *Contractaecum osculatatum* (Rudolphi, 1802) larva was

reported by Schaum and Müller (1967) in Germany, but in this case, the excretory system of the larva was not morphologically described, so that the identification cannot be accepted as definite. *Contractaecum* sp. was suspected to be the causative worm in two patients in Korea (Im *et al.*, 1989). In Japan, rare cases of anisakiosis caused by *Anisakis physeteris* Baylis, 1932, have been reported by Kagei *et al.* (1978) and Asato *et al.* (1991). During the past 28 years, 1,825 reports dealing with 20,582 cases of anisakiosis have been published in Japan. This report deals with a rare example of parasitism of *H. aduncum* passed through a human alimentary canal. The worm was ultimately excreted in his feces.

Case Report

A 55-year-old Japanese man residing in Sapporo, Japan, visited a hospital complaining of chronic

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abdominal pain and diarrhea. He brought a nematode, 5 cm in length, which had been evacuated with his feces. No objective symptoms were observed. Physical examination revealed no abnormality. Two days after the patient evacuated the nematode, the abdominal pain and diarrhea disappeared.

The patient had eaten a raw seafood dish named "Tara no kobujime" (codfish, *Gadus macrocephalus*, meat pressed between kelp) about a month before the onset of symptoms. He had also eaten "Ruibe" (sliced quick-frozen salmon, *Oncorhynchus nerka*) a week before the onset; both codfish and salmon are known as paratenic hosts of *H. aduncum* (Shiraki, 1974).

Material and Method

A worm excreted from the patient and the worms collected from *Cleisthenes pinetorum herzensteini*, caught in the Pacific Ocean off Kushiro, and from *Oncorhynchus masou*, caught in Tsugaru Straits, Hokkaido, Japan, were investigated.

These worms were fixed with 10% formalin and cleared in lactophenol solution for phase contrast microscopy. For light microscopy against of worm, the worms were fixed with 10% formalin solution, dehydrated with alcohol, and stained with hematoxylin and eosin. For scanning electron microscopy, these worms from the patient and *Cleisthenes pinetorum herzensteini* were re-fixed with 1% glutaraldehyde for 2 hours and post-fixed

with OsO₄ for 2 hours. They were then dehydrated and critically dried. Ion sputter coating was performed, and the coated worms were observed under a scanning electron microscope at 15 kV.

Results

Gross findings

The worm was yellowish-brown and non-transparent, 53.3 mm in length, and 1.29 mm at the greatest width; its cuticle had fine transverse striations; cervical alar, indistinctive; lips, approximately equal in size, 0.278 mm in length, bearing transparent cuticular flanges on lateral margins; flanges without indentation; dorsal lips with two lateral double papillae; each subventral lip with amphid, adjacent mediolateral double papillae and single lateral papillae; dentigerous ridge, absent; interlabia present, 0.102 mm in length; esophagus, narrow and long, 5.29 mm in length; the ventriculus, globular; ventricular appendix, sac-like and approximately as long as the anterior intestinal caecum, 1.88 and 1.91 mm in length, respectively; nerve ring, 1.01 mm from the anterior end; excretory pore 1.15 mm from the anterior end; vulva situated 19.50 mm (37% of body length) from the anterior end; tail, conical, 0.30 mm in length, ending in a small process covered by fine rudimentary spines; and a pair of small lateral papillae present near the posterior end of the tail (Table 1, Fig. 1 and Fig. 2A–C).

Next we described the morphological features

Table 1 Comparison of *Hysterothylacium aduncum* from human (present material) and from pothead flounder (*Cleisthenes pinetorum herzensteini*)

	human (present material)	<i>Cleisthenes pinetorum herzensteini</i> (after Moravec and Nagasawa, 1985)
Length of body (mm)	53.3	60.87–72.90
Width of body (mm)	1.29	0.979–1.455
Length of lips (mm)	0.278	0.258–0.272
Length of interlabia (mm)	0.102	0.095–0.109
Length of esophagus (mm)	5.29	5.28–7.14
Length of ventricular appendix (mm)	1.88	1.292–1.360
Length of intestinal caecum (mm)	1.91	2.120–2.380
Distance of excretory pore (mm)	1.15	1.034–1.251
Distance of nerve ring (mm)	1.01	0.966–1.183
Distance of vulva (mm)	19.50	–
Length of tail (mm)	0.30	0.326–0.369

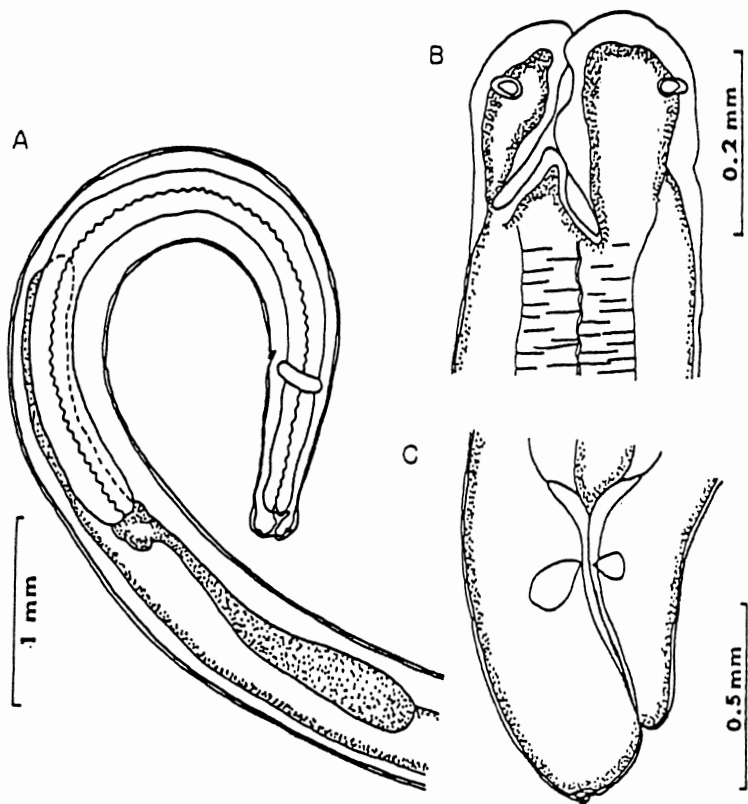


Fig. 1 Female worm of *Hysterothylacium aduncum* (Rudolphi, 1802) excreted from a human.

A, head end; B, anterior extremity; C, tail

of a *H. aduncum* female adult collected from *Oncorhynchus masou* caught in Tsugaru Straits, Hokkaido, Japan. We used the pressed transparent method to observed the worm (Fig. 3A–C).

Anterior extremity of this nematode (Fig. 3A): Three lips and three interlabia well developed. The whole view of the anterior extremity resembled a hare-lip of an infant. The excretory pore was situated anteriorly at the level of the nerve ring.

Midgut cuticle (Fig. 3B): Intestinal caecum and ventricular appendix were seen; ventriculus was a globular form.

Posterior extremity (Fig. 3C): cactus tail, and small process called spines were seen. The tail seemed to be the same as that in the worm from the human case.

Light Microscopic Findings in cross section of nematode

The following characteristics were found: The cuticle was thin and smooth. At the level through the esophago-intestinal junction, there was intestinal caecum together with the ventricular appendix. Lateral cords were observed, and they appeared to be classified as either the so-called 'gourd seed' or 'hillock' type (Fig. 4A, B). The renett cell was a common type of hoe, either half-moon-like or sausage-like; however in our case, the renett cell of the worm was irregular due to postmortal tissue-degenerative destruction. The reversive triangular formic spindle-cell of the muscular layer had abundant chromatin. The arrangement of the columnar epithelial muscle cells was somewhat complicated, and

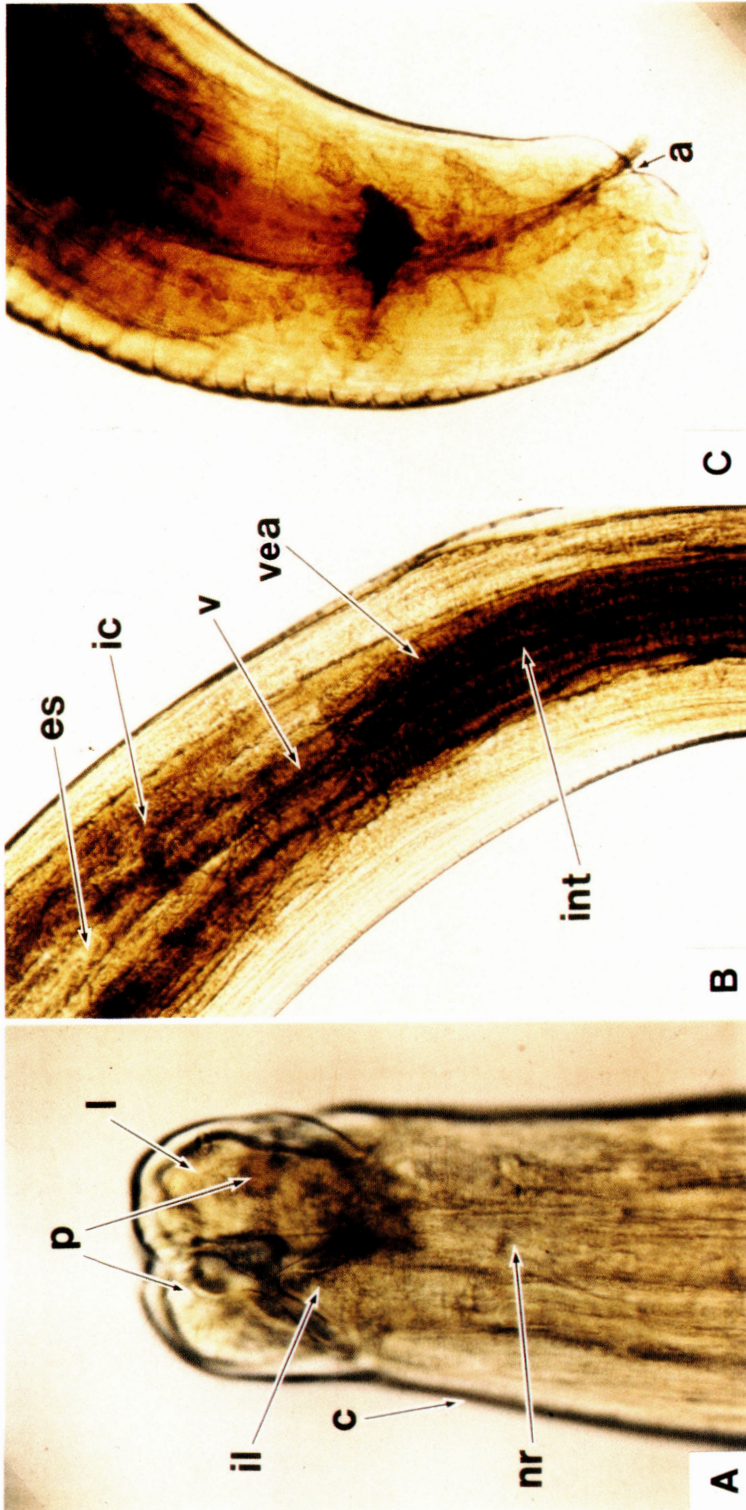


Fig. 2 *Hysterothylacium aduncum* female young adult excreted from a man in Sapporo City, Japan.
 A, Anterior extremity; B, Midgut cuticle; C, Posterior extremity
 p: papilla, l: lip, c: cuticle, il: interlabia, nr: nerve ring, es: esophagus, v: ventriculus, int: intestine, vea: ventricular appendix, ic: intestinal caecum, a: anus

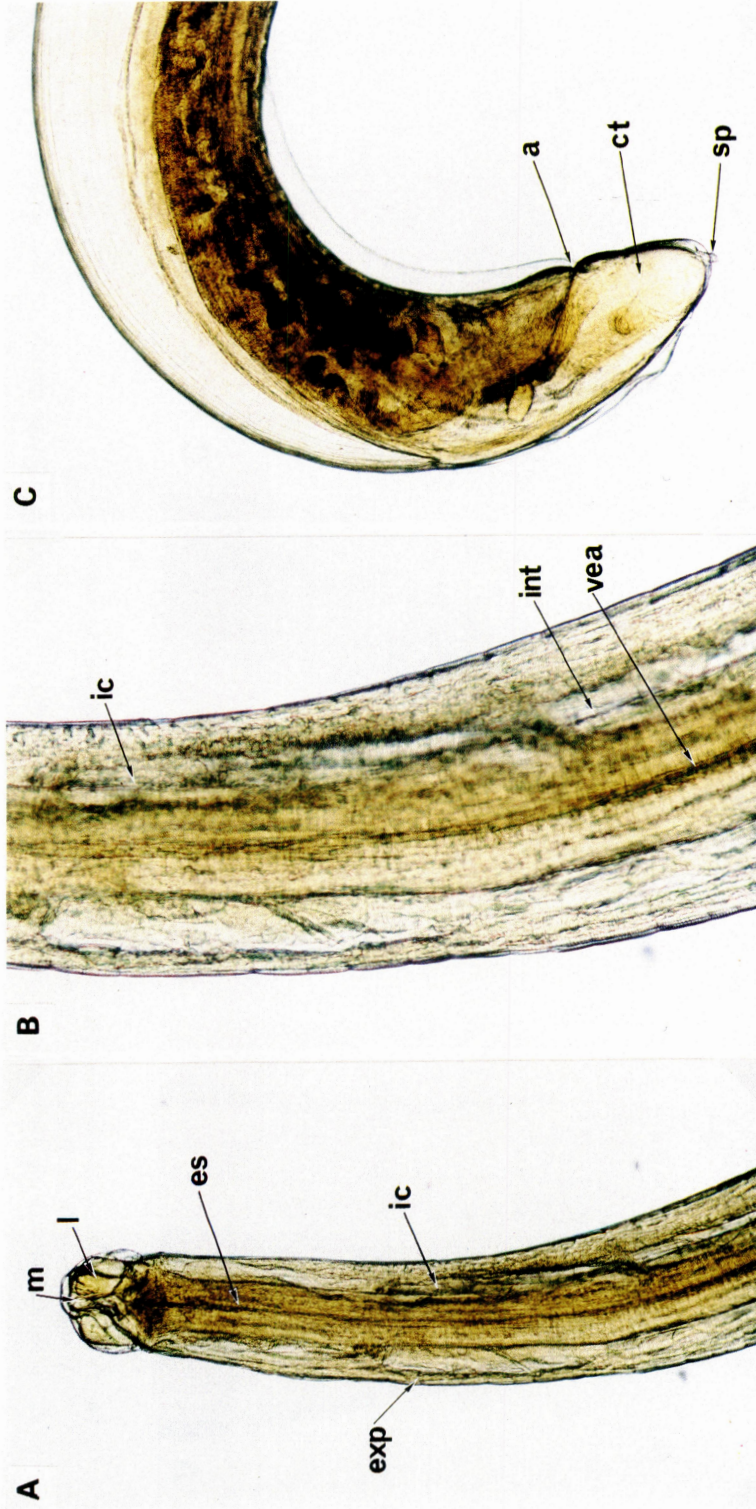


Fig. 3 *Hysterothylacium aduncum* female adult collected from *Oncorhynchus masou* caught in Tsugaru Straits, Hokkaido, Japan.
A, Anterior extremity; B, Region of stomach; C, Posterior extremity
m: mouth, l: lip, es: esophagus, exp: excretory pore, int: intestine, vea: ventricular appendix, a: anus, ct: cactus tail, sp: spine

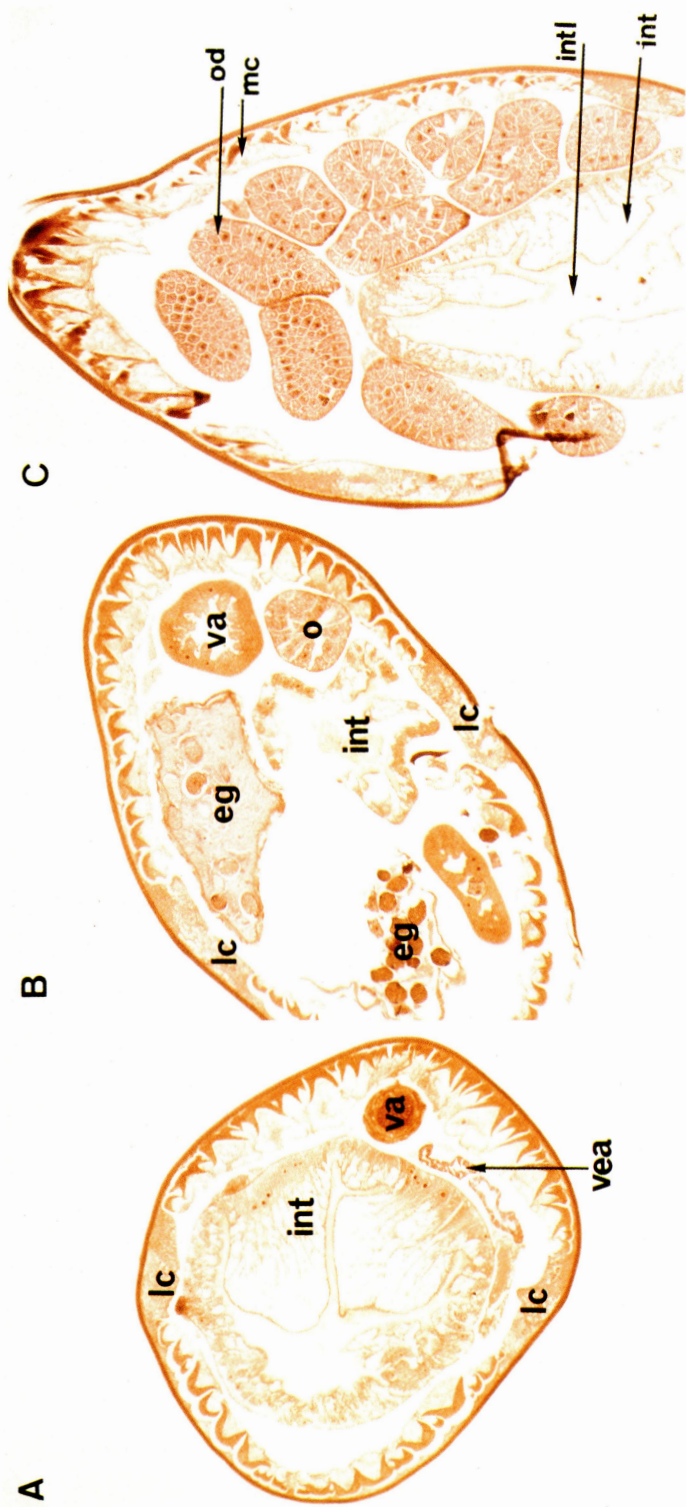


Fig. 4 Histopathological findings of *Hysterothylacium aduncum* female young adult excreted from a man in Sapporo City, Japan.

A, Upper part of intestine; B, Middle part of intestine; C, Lower part of intestine

lc: lateral cord, int: intestine, va: vagina, vea: ventricular appendix, eg: egg, o: ovary, od: oviduct, mc: muscle cell, intl: intestinal lumen



Fig. 5 *Hysterothylacium aduncum* female adult collected from *Cleisthenes pinetorum herzensteini* caught at Kushiro Bay in Hokkaido.
c: cuticle, mc: muscle cell, lc: lateral cord, int: intestine, e: egg, od: oviduct, ov: ovary, ut: uterus

the number of muscle cells was from 10 to 12 per quadrant. The open vulva was situated at the level 1/3 part from the head. The uteri were filled with eggs. The ovaries formed several coils. The eggs were spherical with thin translucent walls. One-, two-, or four-cell stage developing eggs were admixed.

The histological features in the cross section of *H. aduncum* female adult collected from *Cleisthenes pinetorum herzensteini* caught in the Pacific Ocean off Kushiro, Hokkaido, Japan, were investigated as follows: Both Fig. 5A and 5B depict cross sections of the worm at the intestinal level. The lateral cord in Fig. 5A appeared to have a hillock-like form. The number of the muscle cells was about 12 per quadrants. Columnar epithelial muscle cells were V- or cup-form, as in the present case. Intrauterine eggs were immature (Fig. 5B).

Scanning Electron Microscopic Findings

The smooth surface of the excreted worm was divided into three regions, as follows:

1) Anterior end (Fig. 6A): The mouth of *H. aduncum* was surrounded by the three large separated lips, which consisted of a dorsal lip and two subventral lips (approximately equal in size). Dorsal lips had a pair of ear-shaped papillae. At the separate margin of the three large lips, an interlabia which was shorter than the lips was observed. The excretory pore opened at a spot just under the 1/3 of the whole length.

2) Midgut cuticle structure (Fig. 6B): The cuticular surface of this worm had abundant deep ordinary spaces and continuous transverse striations over the whole body surface. Between the two transverse striations, light fine longitudinal ridges were found. At the junctions with the transverse striations the longitudinal ridges were branched, and feather-like structures were elevated above the surface of the body. A vulva without verge opened at the ventral body cuticle, about 1/3 of the body length from the head end.

3) Posterior end (Fig. 6C): The tail was conical, and, at the tail ending, a small process with cactus-like structure was present. Several pairs of phasmids were observed around the end of the tail.

SEM observation of *H. aduncum* female adult collected from *Oncorhynchus masou* caught in Tsugaru Straits, Hokkaido, Japan was shown in

Fig. 7.

Anterior extremity (Fig. 7A): From bird's eye view of the tip of head, we were able to distinguish the three lips that were approximately equid in size, and from the frontal face of interlabia, it looked like a children's hare-like face.

Midgut cuticle (Fig. 7B): Transverse striations were parallel and well-regulated. Longitudinal ridges were fine and both endings resembled feathers and separated five or six parts.

Posterior extremity (Fig. 7C): Tail was cactus-like; at the tip of tail scores of spines were observed.

Discussion

At the beginning of the last century, Rudolphi (1802) distinguished a new nematode *Ascalis adunca* from the other ascarid nematoda. This was the first description of *H. aduncum*. *H. aduncum* is now well-known as a common parasite of marine and fresh water fish throughout most of the world (Moravec *et al.*, 1985; Moravec and Nagasawa, 1985). Fish are considered to serve as the final, intermediate and paratenic hosts of this parasite (Norris and Overstreet, 1976).

This nematode was considered a species of *Contracaecum* (Railliet and Henry, 1912) in the past as it had both a ventricular appendix and intestinal caecum. However, Hartwich (1957) resurrected *Thynnascaris* (Dollfus, 1933), distinguishing it from *Contracaecum* as some of its species lack a ribbon-like widening of the excretory pore near the level of the nerve ring rather than ventral interlabiae, and because it matures in fish rather than in birds or mammals.

Deardorff and Overstreet (1980) reexamined the type-species of *Hysterothylacium* (Ward and Magath, 1917) and revealed that it has the same genetic features as a species of *Thynnascaris*. Therefore, *Hysterothylacium* is theoretically the oldest available name of "Contracaecum-like" species from fish.

In Japan, several investigations on the intermediate and paratenic hosts of anisakid nematodes have been carried out (Yamaguti, 1935, 1941; Koyama *et al.*, 1969; Kikuchi *et al.*, 1970; Koyama *et al.*, 1970; and Shiraki, 1974). In those reports, the larvae of *Hysterothylacium* spp. were described as those of

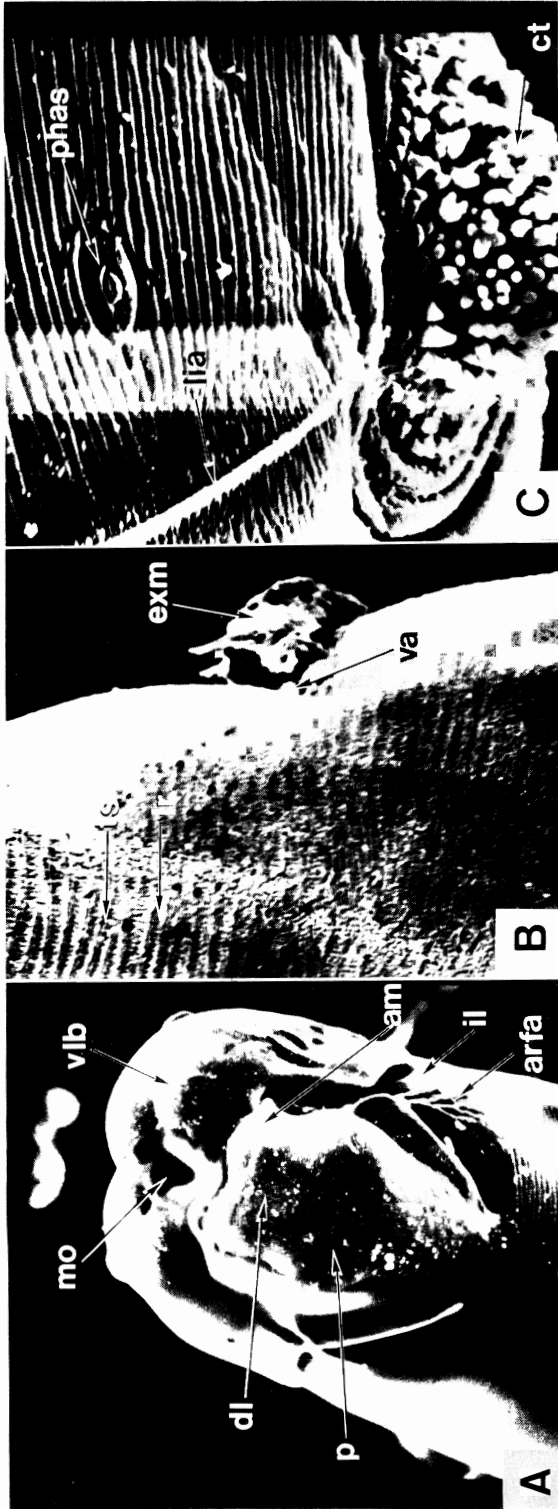


Fig. 6 *Hysterothylacium aduncum* female adult evacuated from a man in Sapporo City, Japan.

A, Anterior extremity; B, Midgut cuticle; C, Posterior extremity

vlb: ventro lateral lip, mo: mouth, am: amphid, dl: dorsal lip, p: papilla, il: interlabia, arfa: arifact, is: transverse striation, lr: longitudinal ridge, va: vagina, exm: excretory mass, phas: phasmid, lia: linia alba, ct: cactus tail

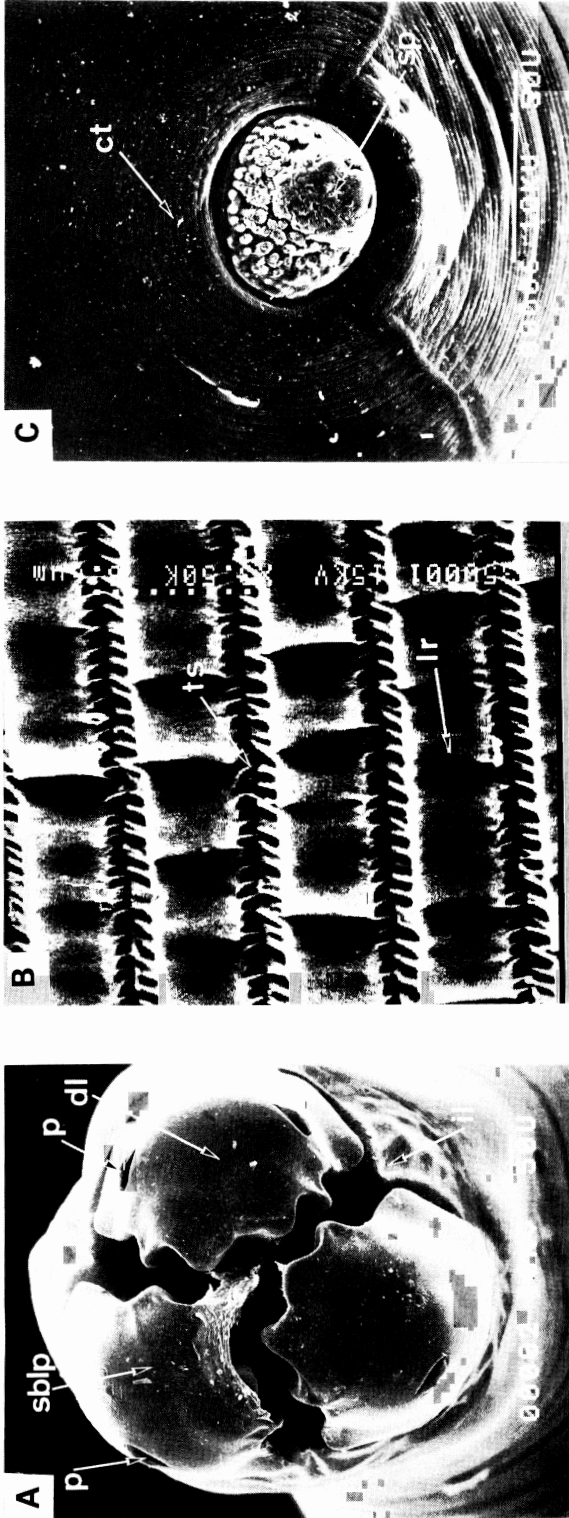


Fig. 7 *Hysterothylacium aduncum* female adult collected from *Oncorhynchus masou* caught in Tsugaru Straits, Hokkaido, Japan.
 A, Anterior extremity; B, Middle part of the body; C, Posterior extremity (in this material, anus and phasmid were not photographing)
 p: papilla, sbp: subventral lip, dl: dorsal lip, il: interlabia, ts: transverse striation, lr: longitudinal ridge, ct: cactus tail, sp: spine

Contraecum spp. and *Thynnascaris* spp.

It is a critical question whether *H. aduncum* is a causative agent of human anisakidosis or not; several experimental studies have taken this up. Jackson (1975) suggested that several kinds of *Hysterothylacium* sp., such as *Hysterothylacium* type MB (Norris and Overstreet, 1976), were infectious to humans. Norris and Overstreet (1976) showed that *Hysterothylacium* type MB could penetrate the gastrointestinal wall of mice, but *Hysterothylacium* type MA not. Vermell *et al.* (1975) were unsuccessful in experimentally infecting *Thynnascaris aduncum* in rabbits. In spite of this, Petter (1969) and Overstreet and Meyer (1981) still hypothesized that *H. (Thynnascaris) aduncum* from *Sardina pilchardus* was one of the causative worm for human anisakidosis.

In this study, we compared the worm expelled from a man with *H. aduncum* caught off the coast of Kushiro, and in Tsugaru Straits, Hokkaido, Japan, and also with a *H. aduncum* female adult described by Moravec and Nagasawa (1985). Our observation clearly demonstrated that the present nematode corresponded to the description of a *H. aduncum* young adult (female) provided by Moravec and Nagasawa (1985), and our comparative study using the naturally infected worm in *Cleisthenes pinetorum herzensteini* or *Oncorhynchus masou*.

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