

## A Clinical Case of Cutaneous Gnathostomiasis having Creeping Eruption

Ryo HATSUSHIKA, Shigeru MIMURA\* and Kaoru MIYOSHI\*

Department of Parasitology, Kawasaki Medical School,  
Kurashiki 701-01, Japan

\*Department of Dermatology, Kawasaki Hospital,  
Kawasaki Medical School, Okayama 700, Japan

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**ABSTRACT.** A clinical case of cutaneous gnathostomiasis having creeping eruption on the right buttock skin region of the infected patient is reported. The patient, a 58-year-old male living in Okayama Prefecture had eaten raw catfish, *Parasilurus asotus* prior to the occurrence of definite symptoms. Intestinal planes of nematode larvae were recognized on the upper dermis. The results of the morphological examination of the sectioned larva revealed that the arrangement and shape of epidermal muscle cells, lateral cords, intestinal wall as well as cuticular spines were similar to the characteristics of those of larval *Gnathostoma*.

**Key words :** Cutaneous gnathostomiasis — Creeping eruption —  
Larva migrans — *Gnathostoma* — Nematoda

Human gnathostomiasis has frequently been occurring in Asia, especially, in Far East, Middle East, China and Japan, and this disease is well known as the names of cutaneous or visceral larva migrans. Three species of genus *Gnathostoma* are distributed in Japan, i. e., *G. spinigerum* Owen, 1836, *G. doloresi* Tubangui, 1925 and *G. nipponicum* Yamaguti, 1941 in which only one of them *G. spinigerum* is known to be an infectious agent for human gnathostomiasis.

Adults of *G. spinigerum* almost completely develop in the gastric wall of several carnivorous animals, mainly domestic and wild felines, dogs and foxes. Two intermediate hosts, cyclops and freshwater fishes or amphibious animals are essential for the development of *G. spinigerum*, and the hatched larvae develop to the 2nd-stage larva in the former host and the 3rd-stage larva in the latter host. The human gnathostomiasis is to be induced by consuming raw meat of infected animals. The 3rd-stage infective larva entered orally to human body will never be reached its full adult development because it is not the definitive host for the parasite, but occasion may arise when some larvae invade into subcutaneous tissue region of human skin or into lung, liver or other tissues of human body and cause creeping eruption or Quinke-type edema. Many cases of human gnathostomiasis occur by consuming raw meat of snake-headed fish, mainly *Ophicephalus argus* harbouring 3rd-stage larva of *G. spinigerum* within their muscles.

For last 5 years however, human gnathostomiasis resulted by eating raw loaches *Misgurnus anguillicaudata* imported food from Formosa, China and Korea has been increasing considerably, and phenomenon has been focused as a problem from medical and public health points of view.

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## CASE NOTES

The patient (C. O.) was a 58-year-old male residing in Sanyo-Cho, Akaiwa County, Okayama Prefecture, Japan. On February 1st, 1983, the patient first noticed the presence of linear erythema progressive of about 10 cm long on the skin surface of his right buttock region with mild itchiness. After a week he visited our hospital because of the erythema gradually spread over his buttock. The erythema was found moving zigzag direction a few centimeter per day.



Fig. 1. Clinical picture of the skin lesion of creeping eruption (arrows) caused by larval *Gnathostoma* infection, right buttock region.

The creeping eruption appeared on his buttock skin region with several edematous erythema and spread along a line (Fig. 1). The lesions are brownish in color and slightly elevated from skin surface. On palpation of the cutaneous lesion, induration and erythema were noted. The cutaneous impression seemed to be creeping eruption induced by animal parasites. On the same day the patient was admitted to our hospital for further examination.

Blood tests disclosed  $523 \times 10^4$  red blood cells and 7,800 white blood cells, with a differential count of 69% segmented neutrophils, 22% lymphocytes, 1% monocytes, 8% eosinophils and  $15.7 \times 10^4$  thrombocytes. Serum proteins were 5.4 g/dl, of which 3.2 g and 2.2 g were albumin and globulin, respectively. By serum immunoglobulin assay IgG, IgA, IgM and  $C_3$  were found all normal with an exception of IgE of 3,536 IU/ml, a definite increase.

Two days after his admission to the hospital the patient was submitted to resection of the cutaneous lesion. The apical point of linear erythema of the right buttock region about  $6.0 \times 4.0$  cm was removed from the slightly deeper

skin surface. The removed tissue was divided into several blocks, and were fixed with formalin, embedded in paraffin, sectioned and stained with hematoxylin and eosin.

Histological study revealed that focal edematous or inflammatory cells with multiple eosinophils are infiltrated in dermis of subcutaneous area. The transverse sections of nematode larva were found in the upper dermis. The larva was about 0.14 mm in diameter (Fig. 2). Although the larva found in the dermis was cut close to intestinal region, the morphological appearances of the transverse sections were similar to those of larval stage of the genus *Gnathostoma*; lateral cords showed a Y-shape structure, the epidermal muscle cells arranged in single row and showing the polymyarian type of nematode, 3 to 5 granules exist in each epithelial cell of the intestinal wall, and the cuticular spines are found on the body surface (Fig. 3).

The patient stated on his eating habits that he had been eating raw meats

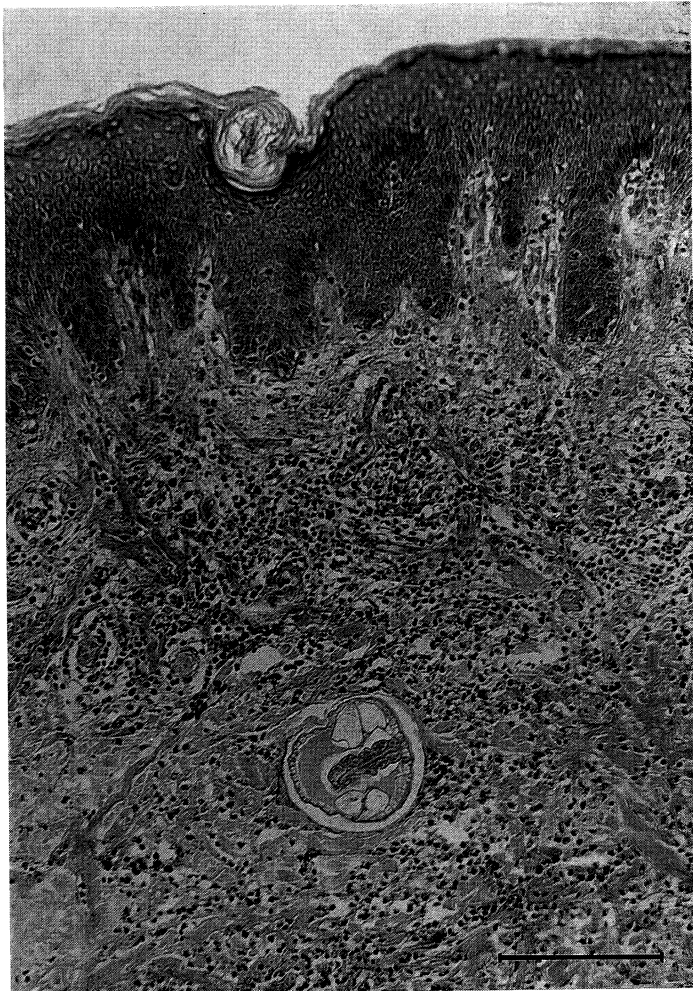


Fig. 2. Section of embedded skin lesion, a larva of *Gnathostoma* in the upper dermis (Scale=0.2 mm, H-E).

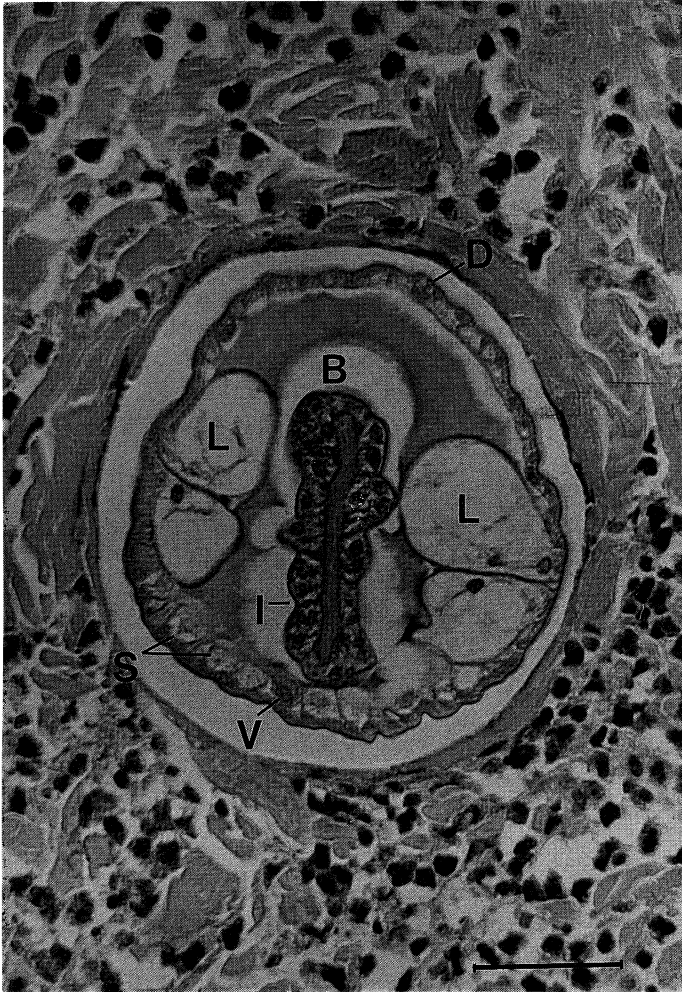


Fig. 3. High power view of the embedded worm, same as Fig. 2; morphological features of epidermal cells and intestinal wall showed structural characteristics (Scale=0.05 mm, H-E). B : body cavity, D : dorsal cord, I : intestine, L : lateral cord, S : somatic muscle layer, V : ventral cord.

of various kinds of water-living animals such as carp, *Carassius* fish, ayu, snake-headed fish, bullfrog, tadpole and also the liver of snakes for nearly 30 years, but had not tried loaches uncooked. Recently, the patient had a chance to eat raw catfish, *Parasilurus asotus* which was caught in a stream near his residence. The creeping eruption appeared on his right buttock skin about 20 days later. The results of immunological serum tests of the patient by immunodiffusion and immunoelectrophoresis performed before and after the operations of skin lesion showed all negative to antigen of adult *Gnathostoma*. Thus species of an infected worm is still unknown.

## DISCUSSION

Human gnathostomiasis has often been found among residents living in Asia from the earliest period of time, and the classical names of the disease in China were 'Quincke's edema', 'Shangchiang edema', or 'Shanghai rheumatism', and those names may be derived from the larva migrans of causative parasites.

Although about 19 species of the genus *Gnathostoma* hitherto has been reported in the world,<sup>1)</sup> of which 3 species are distributed in Japan and human gnathostomiasis is related only to *G. spinigerum*. The first case of human gnathostomiasis in Japan was reported by Kinoshita (1924),<sup>2)</sup> as with an immature worm from a palm. Since gnathostomiasis has been distributed in all areas of Japan, west of Kanto district during the 1950th. At that time, snake-headed fish, *Ophicephalus argus* and *O. tedianus* seemed to play the most important role as an infective agent. But the patients have been then gradually decreased in number. At the present time however, the snake-headed fish is not recognized as a vector of human gnathostomiasis because the *Gnathostoma* infection tended to increase in recent years, and in most cases onset appeared to be more intimately related to imported loaches, which are briskly imported into Japan from outside countries.

The diagnosis of the patient infected with *Gnathostoma* is extremely difficult since the larval worms orally entered to human body keep migrate everywhere all over the body. Many larval worms in human body migrate in the subcutaneous tissues of deep surface of the host skin, and the causative worms can not hardly be detected in the latter case. The numerous cases of human gnathostomiasis are diagnosed by an immunological test, because it has been very hard to find causative worms in the skin lesions and removed tissues. Therefore, the present finding of the causative worm is rather new.

Thus the immunological test is applied usually for diagnosis of larva migrans but not all of the cases show positive results like this case. In the present patient the reason why the immunological test showed negative result is rather unclear, but conceivably immunological reaction may not be detectable because of the test was performed so soon after infection.

It is thought that the population of gastronomers with unusual habits seem to grow among Japanese in recent years, thus many infections have been increasingly reported. Judging from the present knowledge, it should be further noted that the ingestion of raw meats of the animals containing infective agents is absolutely dangerous.

The data concerning human gnathostomiasis with relation to the discovery of causative worms are shown in Table 1. In Table 1, 22 cases from the literatures since 1980 are listed,<sup>2-21)</sup> of which 7 cases<sup>14-16, 18-21)</sup> are definitely related to ingestion of raw meat of imported loaches (*Misgurnus* fish). The present patient had not eaten raw meat of loaches, but he preferred to eat various kinds of vertebrate animals such as fishes, amphibians, reptiles and some wild mammals, whatever, he may be one of typical unusual gastronomers. Among the people consumed raw catfish, the patient was only a person who claimed the definite onset of the disease.

Morita (1955)<sup>22)</sup> reported the detailed innerstructure of the 3rd stage larva of *G. spinigerum* in rats in order to elucidate developmental mechanism of human gnathostomiasis. He stated that the epidermis was bulged out into the

TABLE 1. Cases of human gnathostomiasis with creeping eruption reported in Japan

Case No.	Patients		Locality (Prefecture)	Lesion sites	Sources of infection	Authors (year)
	age	sex				
1.	41	F	Gifu	chest*		Tamura (1919)
2.	43	M		palm		Kinoshita (1924)
3.	65	F	Hyogo	forearm*	<i>Carassius</i> fish	Miyazaki <i>et al.</i> (1951)
4.	30	"	Fukuoka	abdomen*	<i>Ophicephalus</i> fish	" (1954)
5.	55	M	Kumamoto	shoulder	"	Nagao (1955)
6.	57	"	Aich	forearm*	"	Morishita (1957)
7.	34	F		chest		Nozaki <i>et al.</i> (1960)
8.	62	M		"	"	Yoshida <i>et al.</i> (1961)
9.	17	F		mamma		Izawa <i>et al.</i> (1962)
10.	53	"	Aich	ear lobe*	<i>Carassius</i> fish	Tanaka (1967)
11.	47	M		neck*		" (1967)
12.	54	F		abdomen*		" (1967)
13.	46	"		loin	"	" (1968)
14.	62	"		shoulder		Murakami <i>et al.</i> (1971)
15.	27	M		abdomen*	<i>Misgurnus</i> fish	Endo <i>et al.</i> (1980)
16.	26	"	Tokyo	chest	"	Kitajima <i>et al.</i> (1981)
17.	19	F	Tottori	shoulder	"	Nakayama <i>et al.</i> (1981)
18.	27	"	Kanagawa	abdomen*	<i>Agkistrodon</i> snake	Matsuoka <i>et al.</i> (1981)
19.	49	"		buttock	<i>Misgurnus</i> fish	Hosoi <i>et al.</i> (1981)
20.	33	"		chest	"	Kobayashi <i>et al.</i> (1982)
21.	35	M		"	"	Ota <i>et al.</i> (1982)
22.	35	"	Osaka	"	"	Takada <i>et al.</i> (1982)

\* = Extirpation case of causative worms.

pseudocoel at 4 places to from 4 longitudinal cords, the muscle layer consisting of single-layered fusiform muscle cells showed the polymyarian type in arrangement and shape, and the intestine was one-layered columnar cellular epithelium packed with a large number of granules in the cytoplasm. The morphological structure of the sectioned larva found in our study is very similar to that of larval *G. spinigerum* previously described by Morita (1955).<sup>22)</sup>

Sagara (1953),<sup>23)</sup> on the other hand, reported migration routes of larval *G. spinigerum* in rats and found that histological changes followed along the route. According to his detailed experiments, the larvae which given orally to rats began to show the routes beneath the skin about 20 days or more after feeding. In the present study the patient had been consuming raw meats of various water-living animals, but considering from our examination, it would be safe to say that cutaneous gnathostomiasis must be caused by the consumed raw meat specifically from catfish, *Parasilurus asotus*.

The remained catfish meat was checked for presence of the causative worms, but no worm was found.

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