

Case Report

**A Case of Hepatic Anisakiasis with a Literal Survey for  
Extra-gastrointestinal Anisakiasis**

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**Abstract**

In the course of a histopathological examination of a liver abscess extirpated for gastric cancer, a cross section of nematode larva was detected in an eosinophilic granuloma. The section revealed the following distinctive morphological characteristics of larval *Anisakis*: a polymyarian-type muscle layer, a renette cell, and Y-shaped lateral cord. Although many human cases with several types of extra-gastrointestinal anisakiasis in Japan have been reported including a hepatic anisakiasis diagnosed by immunodiagnosis, this is the first case of *Anisakis* larva found in the liver.

**Key words:** extra-gastrointestinal anisakiasis; liver; Japan.

**Introduction**

Since larval *Anisakis* infections in human were first reported in Holland (van Thiel *et al.*, 1960) and Japan (Asami *et al.*, 1965), the disease has come to be regarded as a public health problem also in Japan, and more than 2,000 cases of gastrointestinal anisakiasis are reported by Japanese physicians every year.

With regard to the extra-gastrointestinal involvement due to *Anisakis* infection, van Thiel and van Houten (1967) were the first to find a living larva in the abdominal cavity of a patient undergoing a surgical operation to treat an acute abdomen. Since then, such cases involving extra-gastrointestinal tissues have been reported in sev-

eral Japanese papers.

The present report dealt with special reference to the first case of extra-gastrointestinal anisakiasis in which a larval *Anisakis* nematode was found in human liver.

**Case Report**

The male patient was a company employee, aged 51 years old, and living in Saitama Prefecture. He had never been abroad. Complaining heart burn, he was referred to the Kan-etsu Hospital on June 1994 for a detailed endoscopic examination, which resulted in the diagnosis of gastric cancer. During his admission, an adenocarcinoma was found in the mucous membrane collected from his stomach by gastrofiberscope. Subtotal gastrectomy and partial hepatectomy were performed on September 25, 1994. A postoperative pathological examination revealed gastric cancer (M, I1c, stage I, T<sub>1</sub>NoPoHoMo). An examination of the liver abscess by partial hepatectomy showed no tumor

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cells. However, a cystic mass filled with necrotized materials surrounded by thick fibrous connective tissue and an unidentified nematode was found in

the center of this abscess.

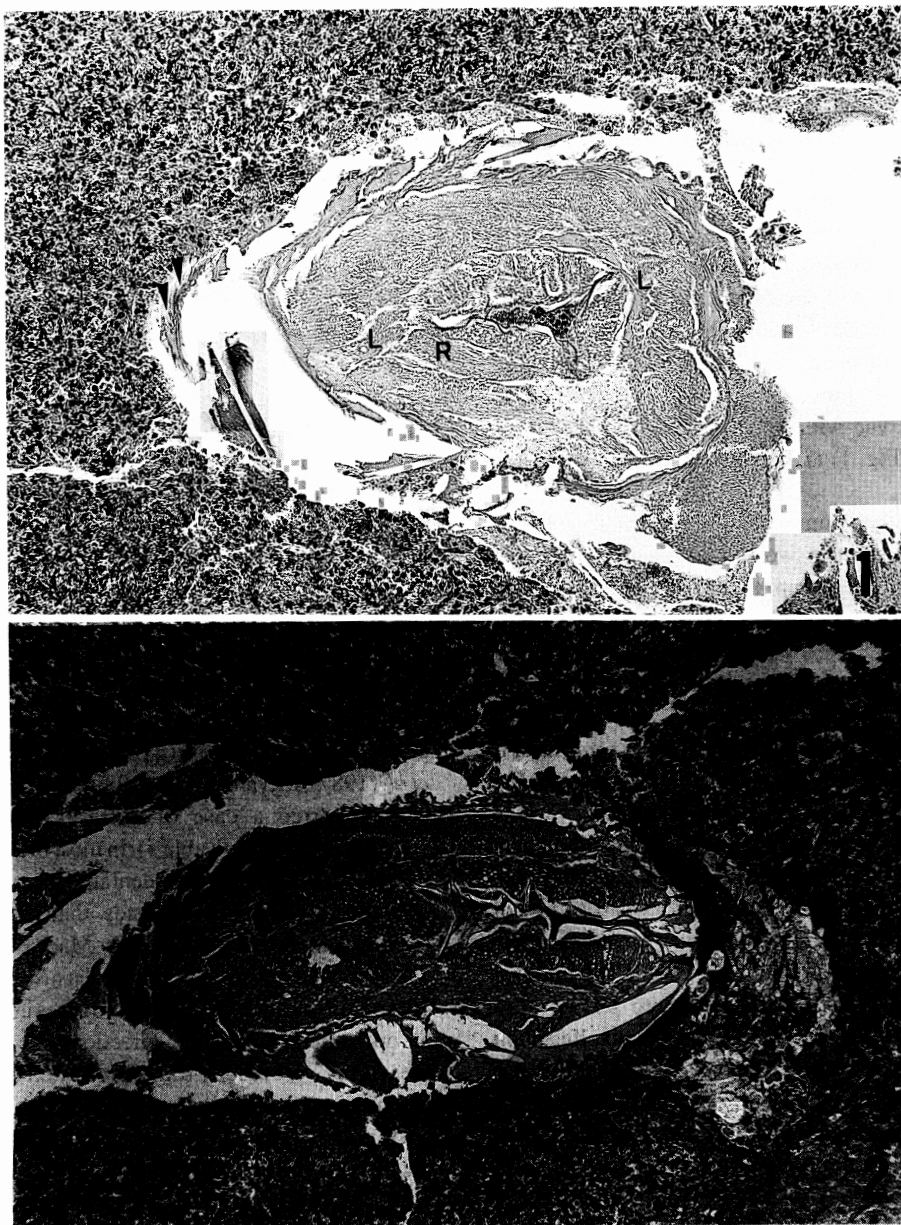


Fig. 1 Cross stump of *Anisakis* larva found in a section of liver abscess (van Gieson's stain. R, renette cell; arrow head, cuticular striation; L, lateral cord).

Fig. 2 Longitudinal section of the anterior part of the same larva (Haematoxylin and eosin stain. Arrow head, lip).

### Histopathological Findings

The sections of excised liver (3.5×2 cm) were stained with hematoxylin and eosin, and van Gieson's stain. A histopathological examination revealed part of an unidentified nematode in a thin section of the liver abscess (1.5 cm in diameter). The stump of nematode was ellipsoidal in shape, measuring 0.36–0.48 mm by 0.26–0.45 mm in width (Fig. 1). The inner structure of the abscess had disintegrated into eosinophilic materials, and the inner structure of the worm was a little degenerated. However, it appeared to be a filarial worm of zoonotic origin because of its polymyarian-type muscle cells and cuticle-like structure consisting of several layers, with wider, more distinct cuticular striations (the arrow in Fig. 1) on the surface of some areas. The inner structure of the body cavity showed the distinctive features of *Anisakis* larvae, with a muscular esophagus, a renette cell (R in Fig. 1) and Y-shaped lateral cords (L in Fig. 1).

In the other stump, we observed that the lips of the anterior part of worm had dentigerous ridges with an attachment cap (Bier and Raybourne, 1988) covering on the lips (the arrow in Fig. 2).

### Discussion

Anisakiasis, a kind of visceral larva migrans, is caused by *Anisakis* larvae. This type of larvae are harbored in various marine fish and squid, of which mackerel, horse mackerel, cod, sardine, and squid are epidemiologically important in Japan. When these parasitic hosts are digested as part of raw or undercooked fish, the freed larvae penetrate or invade the human gastrointestinal wall. The immune response to repeated infection with larvae, alloprotein, produces an acute or chronic manifestation of anisakiasis. Gastric anisakiasis can be readily diagnosed by detecting the worm in a roentgenographic, ultrasonographic or gastrofiberscopic examination. In contrast to this, clinical diagnosis of intestinal anisakiasis is relatively difficult to detect, although the worm is occasionally found by chance during a histopathological examination of the intestinal submucosa resected for ileus. Larvae that have penetrated the gastrointestinal wall migrate into various organs in the abdominal cavity or into

the abdominal and thoracic organs. Most larvae are found dead in eosinophilic abscesses or granulomas during histopathological examinations for different diseases. Rarely the migrating worms have been found alive in the abdominal cavity in the course of emergent laparotomy for acute abdomen.

According to the recent statistical survey of anisakiasis by Ishikura *et al.* (1990), a total of 14,162 cases were reported up until June in 1990 in Japan, of which 13,474 (95.1%) were gastric (including 357 cases of pseudoterranoviasis), 617 (4.4%) were intestinal, and only 52 (0.4%) were extra-gastrointestinal.

Table 1 is a list of extra-gastrointestinal anisakiasis reported by many Japanese physicians in Japan. However, it is preferable for at least some of these cases to be classified as gastrointestinal anisakiasis in a wide sense, because they contain circumstantial evidences for the migration/penetration of *Anisakis* larva through gastrointestinal wall. Hatsushika *et al.* (1990) reported a case, in which the worm produced a granuloma in the cyst of femoral hernia, and listed 15 cases of extra-gastrointestinal anisakiasis reported in Japan. Another two cases from their list, in which a worm was found alive in the abdominal cavity after penetrating the intestinal wall, producing enteritis, should also be preferably included in the group of gastrointestinal anisakiasis. Furthermore, Watanabe *et al.* (1987), Miyahira *et al.* (1989), and some others each reported a case of extra-gastrointestinal anisakiasis in Japan (Table 1). In most of these cases the worms were found accidentally during surgical operations for other diseases than anisakiasis (Kagei and Sakaguchi, 1977; Maesawa *et al.*, 1991; Watanabe *et al.*, 1987; Hatsushika *et al.*, 1990; Kajiya *et al.*, 1992). In those cases undergoing an operation shortly after infection with *Anisakis*, the worms were readily found grossly on the greater omentum or mesentery, as seen in the cases of Nishimura *et al.* (1974), Yoshimura *et al.* (1979b), and Miyahira *et al.* (1989).

In the present case, the worm was detected on the occasion of a histopathological examination of the liver extirpated during radical surgery for gastric cancer. Ueki *et al.* (1989) reported on a human hepatic anisakiasis where several white spots were found on the liver by laparoscopy, though they

Table 1 Extra-Gastrointestinal Anisakiasis in Japan

Location of the larva recovered	References [age, sex (M: male; F: female) and residence of patients]
pulmonary	Kobayashi <i>et al.</i> , 1985 (37, M, San Francisco)*; Matsuoka <i>et al.</i> , 1994 (2, M, Miyazaki)*
lung	Kagei, 1993 (80, F, Nagasaki)
abdominal cavity	Furukawa, 1974 (28, M, Tokyo); Sasaki <i>et al.</i> , 1978 (32, F, Wakayama); Yoshimura <i>et al.</i> , 1979b (36, M, Kanazawa); Kawano <i>et al.</i> , 1990 (58, F, Okayama)
gastrocolic omentum	Yamaguchi <i>et al.</i> , 1967 (-, -, Aomori); Yoshimura <i>et al.</i> , 1979a (22, M, Kanazawa); Yoshioka <i>et al.</i> , 1981 (30, M, Fukuoka); Maesawa <i>et al.</i> , 1991 (11, M, Iwate); Takeuchi, 1993 (37, F, Tottori)
gastrohepatic omentum	Nishikawa <i>et al.</i> , 1992 (39, -, Hokkaido)
mesenterium	Yamaguchi <i>et al.</i> , 1967 (-, -, Aomori); Nishimura <i>et al.</i> , 1974 (43, F, Oosaka); Yoshimura <i>et al.</i> , 1979b (36, M, Kanazawa); Konishi <i>et al.</i> , 1979 (38, M, Ishikawa); Sasaki <i>et al.</i> , 1984 (60, M, Yamaguchi); Miyahira <i>et al.</i> , 1989 (26, M, Tokyo)
peritoneum	Watanabe, 1987 (36, F, Iwate); Maesawa <i>et al.</i> , 1991 (23, F, Iwate)
abdominal wall	Kagei & Sakaguchi, 1977 (30, M, Kanazawa); Matsuki <i>et al.</i> , 1979 (30, M, Ishikawa); Yamamoto <i>et al.</i> , 1990 (39, M, Chiba)
serosa	Chiba <i>et al.</i> , 1971 (22, F, Aomori); Maruyama <i>et al.</i> , 1981 (73, M, Aomori); Furukawa, 1985 (36, M, Chiba)
liver	Ueki <i>et al.</i> , 1989 (62, M, Shimane) <sup>†</sup> ; Kagei <i>et al.</i> , 1995 (51, M, Saitama)
pancreas	Otsuru <i>et al.</i> , 1965 (51, F, Niigata)
ovary	Mori <i>et al.</i> , 1982 (23, F, Nagasaki)
utero-cervix	Takao <i>et al.</i> , 1993 (47, F, Saga)
oviduct	Shiozaki <i>et al.</i> , 1993 (47, F, Tokyo)
hernia	Hatsushika <i>et al.</i> , 1990 (74, F, Okayama); Kajiyama <i>et al.</i> , 1992 (1, M, Kagoshima)
lymphonodulus	Otsuru <i>et al.</i> , 1965 (13, F, Niigata); Yoshimura <i>et al.</i> , 1979b (78, F, Ishikawa); Maesawa <i>et al.</i> , 1991 (54, M, Iwate); Yazaki <i>et al.</i> , 1991 (65, M, Tottori); Hashiguchi <i>et al.</i> , 1992 (46, M, Nagasaki)
subcutaneous tissue	Yoshimura <i>et al.</i> , 1980 (42, F, Ishikawa; 3, M, Ishikawa)

\*diagnosed by immunodiagnosis; <sup>†</sup>diagnosed by laparoscopy.

could not found a worm. The present report is the first human case in which an *Anisakis* larva was found inside of a human liver. At first, identification of the worm was difficult because of its oblique cut as well as its slightly degenerated inner structure. Thus, we have made the right transverse sections which revealed the distinctive characteristics of *Anisakis* larva for the diagnosis of this case. This suggests that more cases of extra-gastrointestinal anisakiasis will be found by careful attention and examinations by pathologists and others concerned.

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