

A case of Human Anisakiasis in Korea

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INTRODUCTION

Human anisakiasis is a disease caused by the third stage larvae of the genera, *Anisakis* and *Terranova*. And it is characterized by the invasion of the larval worm in the gastrointestinal wall, causing various degree of inflammatory change. This entity of disease had not been established until 1960, when van Thiel identified the nematode as that belonged to the genus *Anisakis* (van Thiel, 1962). Thereafter, attention have been paid to this disease all over the world particularly those countries like the Netherlands and Japan (Asami et al., 1965; Yokogawa and Yoshimura, 1965, 1967) because it is closely related with the habit of raw eating of marine fishes.

In Korea, where the raw consumption of marine fish is also popular, the occurrence of human anisakiasis has long been speculated by many parasitologists. However, only one case of aberrant parasitism in pharynx has been recorded (Kim et al., 1971), and no additional case was reported since then as far as the literature is concerned.

In this communication, the authors describe

a case of human anisakiasis involving the ileum in a young Korean male who was incidentally found to have the lesion during an exploratory laparotomy.

CASE PRESENTATION AND WORM DESCRIPTION

The patient, Lee, B.C., 23-year old male merchant, living in Seoul, was admitted to the Emergency Room of the Seoul Red Cross Hospital on November 1, 1979, because of the acute abdomen. He had one year history of peptic ulcer pain. Shadow of free gas was revealed under the diaphragm on simple abdominal x-ray on admission. The patient was diagnosed as hollow viscus perforation and underwent an emergency laparotomy. Opening the peritoneal cavity, there found some food material around the gastric pylorus, and 2mm perforation in the duodenum. Ulcer crater was palpated around the perforation. The perforation was closed with omentopexy.

During the close observation of the entire visceral organs in peritoneal cavity, a mass on the ileum was recognized at 10cm proximal from the ileocaecal valve. It was located at the mesenteric side, and its size was about 1cm in diameter. It was elevated about 0.5~0.7cm from the serosal surface. The surface of the mass was grayish yellow, and rough reminiscent

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of strawberry. The mass was excised and submitted to the Pathology Department.

Pathological examination(S79-1505):

Grossly the submitted specimen consisted of an irregular fragment of bowel wall. The serosa was edematous and thickened with somewhat nodular appearance. The mucosa could not be easily identified in the specimen although definite muscularis externa was recognized. The entire piece measured less than 1.5cm in maximum dimension.

Microscopically, the lesion consisted basically of inflammatory change extending transmurally from submucosa to serosa. This inflammatory change, however, showed patchy distribution in the muscularis layer. The serosa and subserosa were most heavily involved. The lesion was characterized by necrotizing granulomatous inflammation around central cavitations and surrounding inflammatory cellular infiltration. The central cavitations appeared to be abscess cavities as the space was partly filled with necrotic debris and pus cells(Fig. 1). However, the margin was irregular and the inner wall consisted of palisading histiocytes and some numbers of multinucleated giant cells together with fibrinous exudation (Fig. 2). A heavy infiltration of mature eosinophils was another prominent feature of this lesion. They were particularly numerous just outside the necrotizing granuloma, and in the subserosa, they were rather diffusely permeated along with lymphoplasmacytic elements (Fig. 3).

In one section, there was worm structure in the necrotic centre that was described above (Fig. 1). Pools of necrotic debris and inflammatory cells were seen around the cross sectioned worm which was found in the subserosa. Charcot-Leyden crystals were seen in and around the degenerated cuticular structure (Fig. 4). They were variously sized and shaped and were

faintly eosinophilic. And here some leukocytes were seen inside the worm. No ova were found in and around the worm. For the identification, it was referred to the Department of Parasitology.

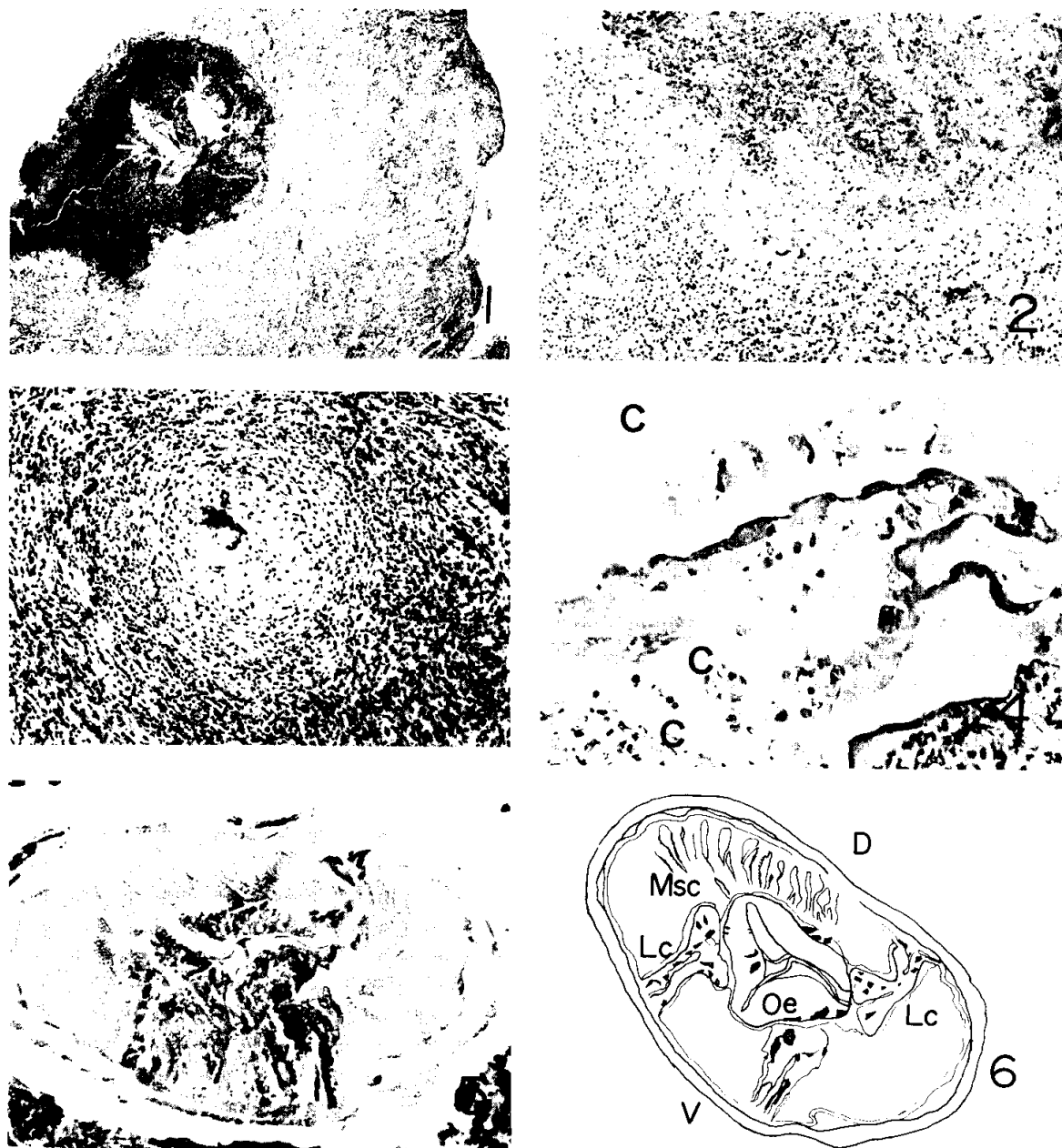
Worm description:

At the centre of the abscess, there was just one section of nematode(Fig. 1 & 5). The worm was sectioned rather obliquely and its size was $208 \times 118 \mu\text{m}$. The tissue of the worm was moderately degenerated. However, the basic structure was still maintained.

The outermost layer of the worm section was cuticle of $6 \sim 7 \mu\text{m}$ in thickness. There were neither ornamental structure nor appendage on the cuticle. Beneath it, there was a layer of hypodermis ($1 \sim 2 \mu\text{m}$ in thickness), frequently detached from cuticular layer. Dorsal and ventral chords were hardly identifiable. There were two lateral chords protruding deeply to the pseudo-coelomic cavity, which were characteristically Y-shaped. In the lateral chords, there scattered chromatin-like dots, and a slit-like space.

At the centre of the worm, there situated a large oesophagus($73 \times 49 \mu\text{m}$ in diameter), which occupied $37 \sim 40\%$ of whole worm dimension. The lumen was lined by triradiated cuticle which made us to confirm it as the oesophagus. By its morphology, dorsal and ventrolateral portion of the worm could be identified as in Fig. 6. The cell distribution pattern of the oesophageal wall were only appeared as scattered pycnotic material.

Though mostly degenerated, the worm musculature were relatively well preserved in dorsal part of the section as ghost structure. In the ventral part, the muscle was markedly degenerated and coagulated. They were tall and narrow, and numerous in a quarter of the section though the exact number was uncountable. Thus this nematode was ccelomyarian, polymyarian



Figs. 1-6. The lesion and anisakid worm section from the present case.

1. A portion of the ileum wall, harbouring a large focus of necrosis in the left upper corner. In the centre of abscess cavity are two portions of the worm. The vertical arrow indicates cross-sectioned nematode and horizontal arrow indicates cuticle, both of which are shown in Figs. 4 & 5. H&E×40.

2. The margin of the necrotizing granuloma showing epithelioid cell proliferation as well as some multinucleated giant cell reaction. The upper portion of this picture is toward the centre of the necrotic foci that are scattered. H&E×100.

3. Focal granuloma with multinucleated giant cell in the centre, and surrounded by diffuse infiltration of eosinophils and lymphoplasmacytic cells. H&E×100.

4. A fragment of degenerated cuticle seen in Fig. 1. Charcot-Leyden crystals(C) are seen in and near the structure. H&E×360.

5. Cross-sectioned anisakid worm, H&E×360.

6. Diagram of the Fig. 5. D: dorsal part, V: ventral part, Lc: Lateral chord, Oe: Oesophagus, Msc: Muscle cells.

nematode. Diffuse, relatively granular, eosinophilic material occupied the narrow space between the muscle and oesophagus. At the centre of ventral part, there was markedly degenerated, hyperchromatic region, which was suggestive of renette cell.

Beside the worm section, there appeared a fragment of cuticle (Fig. 1 & 4), which was degenerated and infiltrated with leukocytes.

Based on the size of the sectioned worm, thickness of cuticle, morphology of lateral chords and muscle cells, this worm section was identified as that of anteriormost oesophageal part of a anisakid larva.

Past history of the case:

The enquiry of the patient's past history was done after the operation. The patient had been a voracious eater and liked everything in raw, such as snakes, oyster, marine fishes and other sea-food.

He made an excursion with his friends on early October, 1979, to a coastal village (Banwol) in Kyunggi Do, and ate two dishes of raw marine fish with wine. The patient recalled that the dishes were mixed with fresh muscle of *Anago anago*, *Raja kenojei* etc. Repeated enquiry failed to remind him any episode of gastrointestinal trouble after the trip, up to the time of operation, other than usual ulcer pain.

DISCUSSION

The main problem in diagnosing anisakiasis in the present case was the apparent limitation of available specimen. We had only one histological section of the worm embedded in the inflamed intestinal tissue. This was the only sectioned worm in it among many serial sections done on the same block. This curtailed our attempt to prove renette cell in the worm. We had to rely on morphometry, internal structure

of the worm and host reaction. The remainder the worm, we believe, was probably lost during surgical excision or maybe during trimming.

We knew that this section represented an oesophageal part of a nematode, and this worm was invading muscle layer of human ileum causing inflammation extending from mucosa to serosa. Having these two obvious facts in mind, the nematodes which possibly cause such lesion at such habitat were listed as follows: Anisakid larva, *Enterobius vermicularis*, immature *Ascaris*, second stage larva of *Toxocara*, hookworm, trichostrongyle, filaroid and spiruroids.

Among these we excluded the possibilities of *Enterobius* and *Toxocara* because our section was lacking of lateral alae. We could further exclude hookworms and trichostrongyles because the present section was polymyarian rather than meromyarian nematode. The possibility of filaroids especially of *Dirofilaria*, and of incidental penetration of immature *Ascaris* could be neglected as well, because of lateral chord morphology. Among the spiruroids, *Gnathostoma* and *Physaloptera* were considered. However, *Gnathostoma* should possess cuticular spines at such high section. *Physaloptera* differed from ours in the size of section and shape of lateral chords (Chitwood and Lichtenfels, 1972). Another unknown spiruroid nematode described by Otsuru et al. (1974) differed in morphology of lateral chords from ours.

After excluding all these possibilities, we came to the conclusion that we were dealing with anisakiasis. The reasons for this could be summarized as follows: Morphologically, all of the findings seen in the section were compatible with those described in *Anisakis* (Oshima, 1972) although we reserved the generic diagnosis; secondly, the history of the patient, consumption of marine fish about one month before the operation, is strongly suggestive for anisakiasis; thirdly, the degree of degeneration seen in the

worm goes along one month history(Oshima, 1972).

The peculiarity of the present case was the apparent lack of directly referable symptom of anisakiasis during the course of infection. Since the lesion was a quite localized in a small portion of ileum at the time of discovery, it could have been silent or easily masked by the mild ulcer symptoms. This suggests that *Anisakis* infection has wider range of clinical manifestation than previously considered. Though hitherto known anisakiasis were mostly manifested by violent acute abdomen(Yokogawa and Yoshimura, 1967; Smith and Wooten, 1978), Ishigura(1969) reported that 5 out of 77 anisakiasis cases were found incidentally during surgery of other causes. Discussing serodiagnostic trials on anisakiasis, Suzuki et al. (1970) discriminated two clinical forms of anisakiasis; fulminant and mild forms; the latter being usually primary infection and failed to manifest the clinical symptom. In this respect, the present case who was found fortuitously during the laparotomy must be the primary infection case. If he did not receive the surgery, the lesion might most probably be unnoticed, and no further trouble by this lesion would be expected.

Another point that we would like to make is on the probable prevalence of anisakiasis in Korea. As stated above, there is only one recorded case of anisakiasis (Kim et al., 1971) in this country. Considering epidemiological and parasitological facts of anisakiasis, it is expected that we have far more clinical cases in Korea. We understand that it is common practice among Japanese physicians, particularly in endemic areas, to do endoscopic examination for the detection of anisakiasis in cases of "acute abdomen". It is the authors' opinion that many infected cases probably went by unnoticed in our country. Among the reports on inflammatory fibroid polyps (Kim et al., 1973; Koh and Kim,

1976) and eosinophilic gastroenteritis (Jung et al., 1974; Chang et al., 1978), at least two cases (Jung et al., 1974) should be the candidates of the intestinal anisakiasis as far as clinical course and some histological features are concerned. However, in all of them, they could not find out any worm in their lesions though serial sections were not performed.

The favorite eating habit of raw marine fish together with reports of high prevalence of anisakid larvae in fishes caught along the coast of the Korean peninsula(Chun et al., 1966) and frequent reports of eosinophilic gastroenteritis suggest that more attention should be paid for the detection of the anisakiasis in Korea.

SUMMARY

A case of human anisakiasis involving terminal ileum was reported. The lesion was incidentally found in a 23-year old Korean male during an operation for duodenal ulcer perforation. As far as one could detect no symptoms directly referable to the lesion were present.

Histopathologically, the lesion was characterized by transmural necrotizing and granulomatous inflammation with central cavitations. One section disclosed a sectioned anisakid larval worm, which was measured $208 \times 118 \mu\text{m}$ in diameters and was characterized by thick cuticle, thin hypodermis with Y-shaped lateral chords, large oesophagus, and polmyarian, coelomyarian musculature.

The present case is the second human anisakiasis reported in Korea.

==국문초록==

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23세의 서울 거주 상인 남자가 소화성 궤양 천공으로 인하여 1979년 11월 서울 적십자병원에서 응급 개복술을 받던 중 회장 말단부에서 우연히 작은 육아종성 병변이 있음이 발견되었다.

이 병변은 주로 장점막하층에 부종성 염증을 일으키면서 장벽에까지 이르는 전 장벽에 한국성(限局性) 염증을 일으키고 있었는데 병변 증상의 괴사성 육아병변에서 선충류의 절단면이 발견되었다.

이 절단면은 직경 208×118μm, 각피 두께 6~7μm, Y-자 모양의 측선 및 체강형, 다근육형 근층을 특징으로 하는 식도부 절단면이었고 그 옆에는 변성된 각피가 발견되었다.

이러한 병리학적 및 기생충학적 소견을 기초로 이 병변은 아니사키스(고래회충)과(科)에 속하는 유충에 의한 병변으로 진단하였다.

환자는 약 1개월전 바닷생선을 날로 먹은 일이 있었으나 특징적인 아니사키스증의 복통은 나타나지 않았었다. 이 증례는 우리나라에서 문헌상 두번째로 기록되는 아니사키스증이며 총체로 인해 흔히 유발되는 심한 증상이 나타나지 않은 초감염형 아니사키스증임을 특징으로 한다.

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