

Case report

A case report of small bowel ileus possibly caused by *Gnathostoma doloresi*

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Accepted 25, September 2007

Abstract: Small bowel ileus due to the parasitic infection caused by omophagia of freshwater fish is relatively rare. We present a case of small bowel ileus possibly caused by inflammatory change associated with *Gnathostoma doloresi* infection. A 62-year-old man underwent partial resection of the small bowel under a diagnosis of ileus due to complete obstruction of the small bowel. He had eaten a few slices of raw freshwater fish four weeks before abdominal pain appeared, and he contracted creeping disease with several welts on the abdominal wall. Chronic inflammatory change suggestive of parasite infection was observed in the resected specimen. An immunoserodiagnostic study using microenzyme-linked immunosorbent assay led to a diagnosis of *Gnathostoma doloresi* infection. The postoperative course was favorable, and the patient was discharged 12 days after surgery. Only two cases of ileus due to *Gnathostoma doloresi* infection have ever been reported.

Key words: ileus, *Gnathostoma doloresi*, freshwater fish, gnathostomiasis

INTRODUCTION

Small bowel mechanical ileus caused by volvulus or a tumor inside and outside of the bowel is not rare. Most cases of mechanical ileus require surgery when there is no improvement by conservative treatment including nil per os, nasogastric suction and parenteral feeds. Sometimes inflammatory changes due to parasite infection such as anisakiasis caused bowel obstruction. *Gnathostoma*, which was initially reported by Tamura in 1921 [1], migrates into a wide range of tissues and organs causing larva migrans, although the typical clinical feature of gnathostomiasis is characterized as a creeping eruption and/or mobile erythematous eruption of the skin. *Gnathostoma* can migrate throughout the body including the small bowel, but an inflammatory change leading to bowel obstruction is relatively rare. We present a patient with small bowel mechanical ileus possibly caused by the inflammatory change of *Gnathostoma doloresi* infection.

CASE REPORT

A 62-year-old man was admitted to our hospital complaining of abdominal pain. Four weeks prior to the onset of abdominal pain, he had eaten a few slices of the raw

freshwater fish, *Oncorhynchus masou*, which is a kind of brook trout, but he had never eaten snakeheads or loaches. Three days later, he became aware of an erythema about 3 cm in diameter with a slight itch near the umbilicus. On physical examination several welts, elevated linear erythemas, were noted on the right upper quadrant of the abdominal wall. Abdominal X-ray photography demonstrated multiple gas-fluid levels with distended small bowel. The patient underwent nil per os and parenteral feeds. Eosinophilia was not detected in the peripheral blood. Biopsy of one welt on the abdominal wall under local anesthesia revealed inflammatory change with massive infiltration of eosinophil, but we were unable to identify any worms in the skin specimen. An immunoserodiagnostic study by microenzyme-linked immunosorbent assay using monoclonal antibodies specific for *Gnathostoma doloresi* antigens led to a diagnosis of *Gnathostoma doloresi* infection (Table 1). The patient was discharged after the symptoms disappeared.

Four days later, he was re-admitted because of abdominal pain and vomiting. He underwent conservative treatment again on the basis of a diagnosis of ileus, but small bowel series through an ileus tube demonstrated complete obstruction of the small bowel (Fig.1). Partial resection of the small bowel was performed. The site of obstruction was

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Table 1: Multiple-dot enzyme-linked immunosorbent assay showing positive binding of the serum to only *Gnathostoma doloresi* antigen and weak binding to other parasites antigen which cause creeping disease.

| | |
|---------------------------------------|-----|
| <i>Human Serum (positive control)</i> | + + |
| <i>Dirofilaria immitis</i> | ± |
| <i>Toxocara canis</i> | - |
| <i>Ascaris suum</i> | ± |
| <i>Anisakis simplex</i> | ± |
| <i>Gnathostoma doloresi</i> | + |
| <i>Strongyloides ratti</i> | ± |
| <i>Paragonimus westermani</i> | ± |
| <i>Paragonimus miyazakii</i> | - |
| <i>Fasciola hepatica</i> | ± |
| <i>Clonorchis sinensis</i> | ± |
| <i>Spirometra erinacei</i> | ± |
| <i>Cysticercus cellulosae</i> | ± |

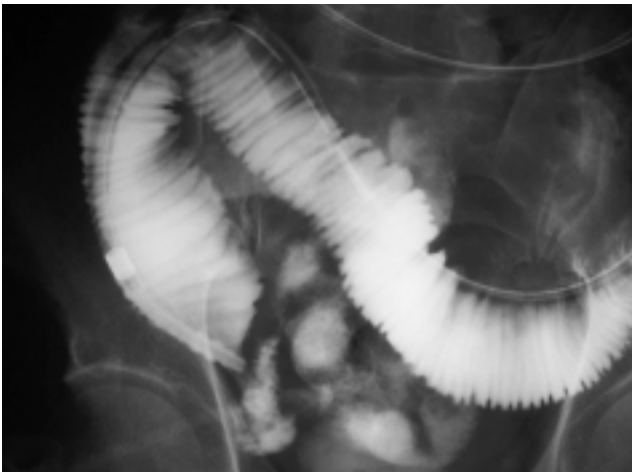


Fig. 1: Small bowel series through an ileus tube showing complete obstruction of the small bowel.

on the oral side 60cm from the end of the ileum (Fig.2). Microscopic examination of the resected small bowel showed a chronic ulcer consisting of a fibrotic granuloma with lymphocyte infiltration. This suggested chronic inflammation, but we were unable to identify any worms in the small bowel specimen (Fig.3).

The postoperative course was favorable. We administered albendazole 600mg/day for one week after the patient restarted oral intake, because the effectiveness of albendazole for gnathostomiasis was proved by Kraivichian et al [2]. The patient was discharged 12 days after surgery.

DISCUSSION

The genus *Gnathostoma* includes twelve species [3].



Fig. 2: Macroscopic appearance of resected specimen of small bowel showing ulcer formation in the center of the obstructive site.

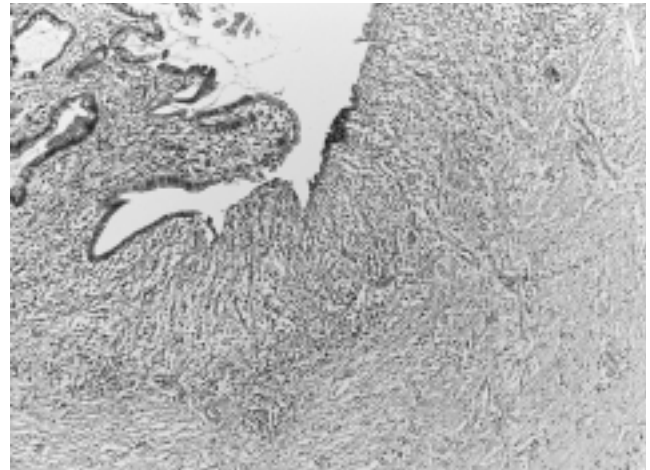


Fig. 3: Microscopic appearance of the lesion showing a chronic ulcer consisting of a fibrotic granuloma with lymphocyte infiltration.

Gnathostoma doloresi was reported in 1925. For this worm, the pig and wild boar of Southeast Asia and Japan, particularly the Kyushu District, are last hosts. Gnathostomiasis is often contracted by eating the raw flesh of a kind of brook trout living in freshwater, *Oncorhynchus masou*, which is an intermediate host. The first confirmed case of human gnathostomiasis by *Gnathostoma doloresi* was reported in 1989 by Nawa *et al* [4] in Miyazaki Prefecture. This case was caused by omophagia of *Oncorhynchus masou*. Our patient lived near Miyazaki Prefecture and ate raw *Oncorhynchus masou* four weeks before the onset of abdominal pain.

The life cycle of *Gnathostoma* is essentially identical within the genus, with only slight variations in the secondary and paratenic host species and also in the sites of infection in definitive host animals. After being hatched from eggs in freshwater, the first-stage larvae are ingested by Cy-

clops in which they molt twice to become the early third-stage larvae in the second intermediate host (fishes and amphibians). The distribution is further disseminated into paratenic hosts, such as large carnivorous fishes and reptiles along the food chain. Infection in humans occurs when these second intermediate or paratenic hosts are ingested [5].

A recent survey revealed that most wild boars in Miyazaki Prefecture were infected with *Gnathostoma doloresi*, which parasitizes the stomach wall of domestic pigs and wild boars causing ulcerative and/or granulomatous lesions [6]. In our case, microscopic examination of the resected small bowel showed a chronic ulcer consisting of a fibrotic granuloma with lymphocyte infiltration.

The ileus due to the infection of *Gnathostoma doloresi* is relatively rare. Only two cases have ever been reported [5,7]. In both cases, ingestion of the flesh of a raw snake (*Agkistrodon halys*) caught in a local mountainous area was the cause of colonic ileus. In one case [5], colonic ileus due to malignant tumor was strongly suspected, and a simple colonic resection was performed. The final diagnosis in this case was made by pathologic examination of the resected specimen, which included the entire body of the worm.

Detection of the larvae in the specimen biopsied from the lesion is ideal, but this is frequently unsuccessful. In most cases, therefore the diagnosis is made on the basis of the combination of clinical manifestations, history of ingesting intermediate and/or paratenic hosts, and results of immunoserological examinations [8]. Similarly, we reached a final diagnosis on the basis of the combination of physical examination, history of omophagia of freshwater fish and immunoserodiagnostic study.

ACKNOWLEDGEMENT

The authors thank Ms. F. Nakamura (Uchiyama) of the Department of Parasitology, Faculty of Medicine University of Miyazaki, for her technical assistance in the serodiagnostic study using microenzyme-linked immunosorbent assay.

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