

GNATHOSTOMIASIS : TWO CASES ADMITTED TO NONG KHAI HOSPITAL, THAILAND

Sujet Lertanekawattana¹, Jitra Waikagul², Somsakol Pilomvarakorn¹ and Kamol Saepeung³

¹Department of Medicine, Nong Khai Hospital; ²Department of Helminthology, Faculty of Tropical Medicine, Mahidol University; ³Bung Kan Hospital, Nong Khai Province, Thailand

Abstract. Human gnathostomiasis is a well-known parasitic infection widely-distributed in Thailand, Japan, and Latin America. Since 1985, there have been 65 cases of gnathostomiasis in Nong Khai Hospital. There were two types of clinical manifestations, of which intermittent migratory cutaneous swelling was more common than visceral larval migrans. This is a report of two cases of visceral organ manifestation of human gnathostomiasis admitted to Nong Khai Hospital. The first case presented with hematemesis, endoscopy was done, and fortunately a gnathostome was found. The symptoms disappeared after removal of the parasite. The worm was identified as male *Gnathostoma spinigerum*. The second case presented symptoms of radicular pain on the abdomen and chest, which developed into paraplegia. The parasite was not found in this case but the diagnosis was made by magnetic resonance imaging and immunoblot, which showed a specific band for gnathostomiasis spinigerum. Radicular pain was decreased at the time paraplegia developed and the patient had not recovered, even after a full course of albendazole treatment (800 mg x 21 days).

INTRODUCTION

Human gnathostomiasis has been known for more than one hundred years. The cause of this disease is parasitic infection by a nematode in the genus *Gnathostoma*, where the infective stage (advanced third stage larva) accidentally enters the human body by contaminated food, then migrates to various organs. During the years 1961-1963, an investigation was carried out using report forms sent to 92 provincial and Bangkok hospitals for clinical gnathostomiasis cases. As a result, about 900 cases were reportedly admitted to hospitals, 81% to hospitals in central Thailand, and 10% in the northeast (Daengsvang, 1980; Radomyos and Daengsvang, 1987). At the Hospital for Tropical Diseases in Bangkok, 300-600 gnathostomiasis suspected cases visited the gnathostomiasis clinic during the years 1985-1988, and about 100-400 cases in 1989-2002 (Rojekittikhun *et al*, 2002). During the period 1985-2003, 65 gnathostomiasis cases were admitted for treatment in Nong Khai Hospital, in northeastern Thailand. The common manifestation was cutaneous migratory swelling. Visceral larva migrans also occurred, but in fewer cases. Among the visceral larva migrant cases, two were interesting in terms of location of infection.

Adult *G. spinigerum* have been reported in the stomach of animal definitive hosts, dogs or cats

(Daengsvang, 1980). The specimens obtained from the reported cases comprised advanced third stage larvae and immature adults (Radomyos and Daengsvang, 1987). The organs of involvement in humans reported in the literature include the eyes, lungs, abdomen and brain, but not the stomach. In the first case, we report a worm discovered in the stomach; the adult worm was removed during gastroscopy. This is believed to be the first case of an adult gnathostome found in the stomach of a human. In cerebrospinal gnathostomiasis, it is impossible to get the worm in the living human body at the time symptoms occur. Many reports of this form made diagnosis by clinical profile and exclusion of other parasite involvement. In this second reported case, we have more evidence from MRI findings, and a serologic test confirmed the diagnosis.

CASE REPORTS

Case 1

A 41-years-old Thai female visited the hospital on April 23, 1995. Her presenting symptoms were hematemesis and epigastric pain. She had these symptoms three days before visiting the hospital and without previously taking any specific drug or treatment. She had mild pallor, no sign of liver disease, soft abdomen, and vital signs within normal limits. The laboratory findings: CBC were white blood cell (WBC) 14,200 cells/mm³, hematocrit (Hct) 30%, differential counts were neutrophil 46%, lymphocyte 42%, eosinophil 10%, monocyte 2%, platelet adequate. We performed endoscopy on the 2nd day of admission, and the findings were esophagus clear with no varices, duodenum normal. In stomach, there was no ulcer,

Correspondence: Sujet Lertanekawattana, Department of Medicine, Nong Khai Hospital, Nong Khai 43000, Thailand.

some coffee ground content presented. There was a dirty spot with small oozing of blood around it at the greater curvature and it was rather fixed to the mucosa. This object was removed by the biopsy forcep. This, examined under the microscope found that it was a worm. It was sent to the Department of Helminthology, Faculty Tropical Medicine, Mahidol University, for further identification. The result concluded that this parasite was a male *Gnathostoma spinigerum* (Fig 1). We followed up the patient for a few days after the worm was removed, and the patient fully recovered with no further complication. She had a habit of taking raw food especially freshwater fish and vegetables. She had no history of cutaneous migratory swelling before admission to the hospital.

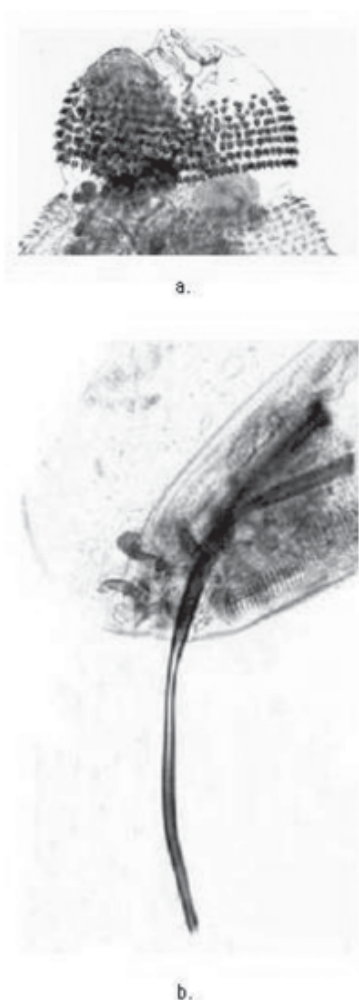


Fig 1- Anterior end (a) showing rows of hooklets and posterior end (b) showing two unequal spicules of adult male *Gnathostoma spinigerum* removed from the stomach of a patient by gastroendoscopy.

Worm's description

The worm is cylindrical, covered with spines throughout the whole body. Cephalic bulb armed with eight rows of hooklets (Fig 1a). The posterior end provided with two unequal spicules (Fig 1b). No spermatozoa were found in the sperm duct.

Case 2

A 61-year-old Thai male lived in Nong Khai Province, Thailand, presented with the symptom of paraplegia seven days before admission to Bung Kan Hospital. He had a full feeling, painful sensation and itching on the chest region and abdomen. The painful, itching sensation migrated around in the chest for a few days. Four days before admission, he could not raise his thighs, with weakness of the left side more than the right, and he felt numbness in some parts of his legs. He could not void or defecate. The weakness and sensation of both lower extremities became progressively worse. He was referred to Nong Khai Hospital for further investigation.

Physical examination. He was well conscious, looked strong, the upper extremities were powerful, the motor tone of the right lower extremity was grade I, and left lower extremity was grade II, (grade V is strongest); sensation was impaired below the umbilicus to the groin region and anterior aspect of both thighs.

Laboratory findings. Hemoglobin 11.2 g/dl, hematocrit 30.5%, WBC 14,000 cell/mm³, neutrophil 39%, lymphocyte 16%, eosinophil 44%, monocyte 1%, platelet adequate. Electrolyte was normal, K⁺ 4.11 mEq/l, BUN 22 mg%, creatinine 1.3 mg%. Urine finding was few WBC, few red blood cells (RBC) and others within normal limit.

Film KUB revealed that there was staghorn renal stone on the left side.

By history taking. He had a habit of eating raw fish and half-cooked frogs, even ingesting pila snails sometimes. Since the patient had headache symptom but no stiff neck, the history and symptoms indicated that a cerebrospinal fluid finding was necessary to differentiate between gnathostomiasis and angiostrongyliasis. Lumbar puncture was performed and routine cerebrospinal fluid (CSF) examination was carried out. The cerebrospinal fluid and plasma samples were sent to the Department of Helminthology, Faculty Tropical Medicine, Mahidol University in Bangkok, for serologic test for both gnathostomiasis and angiostrongyliasis. Initial CSF pressure was 22 cmH₂O and slightly turbid, protein 273 mg/dl (normal 5-45 mg/dl), glucose 35 mg/dl (normal 40-50mg/dl);

cell count: WBC 170 cells/mm³, neutrophil 3%, lymphocyte 41 %, monocyte 2%, eosinophil 59%. Indian ink was negative. Serologic tests for *Gnathostoma spinigerum* were positive both in plasma and CSF, but serologic tests for *Angiostrongylus cantonensis* were all negative.

MRI of T-spine showed abnormal intramedullary signal intensities along the thoracic cord with hypersignal on T2 w and prominent size of the thoracic cord. The differential diagnosis was thoracic myelitis or transverse myelopathy, and clinical profile made the most likely diagnosis of gnathostomiasis. The patient was treated with albendazole 400 mg bid for 21 days, the weakness of the lower extremities was not worse but did not recover; he could not walk and was retained on a urinary catheter.

DISCUSSION

These two cases represent the problem of food-borne disease caused by *Gnathostoma*, which is still an important parasite, causing morbidity and even death to people in endemic areas, especially people ingesting uncooked or raw fish, frog, and other animals. Emphasis on the need for preventive education about food-borne parasitic zoonosis at the community level is required; treatment of infections with this group of parasites, in particular gnathostomiasis, require multiple doses of drug (albendazole), and some cases could not be cured.

The first case showed that gastroscopy is an effective method for investigating a patient with epigastric pain. It had been used to remove an *Anisakis* nematode from the stomach wall of a patient effectively (Kagei *et al*, 1978). This is the first time that morphologic adult gnathostome was recovered in the stomach wall of a human, which leads to the question of whether humans can be a definitive host of this nematode. In the animal definitive host, several male and female gnathostomes lived together in a tumor of the stomach wall, and became sexually mature egg-laying females (Daengsvang, 1980). Gnathostomes recovered from humans varied in development, from advanced third-stage larvae, immature males and females, to mature males (Prommas and Daengsvang, 1934; Chitchang *et al*, 1981; Radomyos and Daengsvang, 1987). If a mature male and female met in the stomach of a human, could they form a tumor in the stomach wall and become sexually mature worms? If this is possible, then humans can also act as a definitive gnathostome host which would change the theory of the gnathostome life-cycle. The chance is

small, nevertheless there is a possibility. Physicians are encouraged to investigate all cases of acute epigastric pain by gastroscopy.

Case number 2, although we could not remove the parasite, had a clinical profile that was highly suggestive of migratory parasite manifestation. The patient had habit of eating raw fish and uncooked freshwater animals. He had the symptom of migratory radicular pain at the abdominal and chest region. This type of pain is quite typical of *Gnathostoma* manifestation (Vejjajiva, 1979) and cannot be explained by an other etiology. WBC was hypereosinophilic, suggesting that it was a tissue-manifesting parasite. CSF findings showed eosinophilic and serologic test both in serum and in CSF were positive for gnathostomiasis by immunoblot but were negative for *Angiostrongylus cantonensis*. Immunoblot gave a more specific result than ELISA, and it is a reliable test for gnathostomiasis (Lertanekwattana *et al*, 1994; Dekumyoy *et al*, 2002).

The disability of this case remained after receiving albendazole 400mg bid for three weeks, the dose that could eliminate migratory swellings within one week with no recurrence (Kraivichian *et al*, 1992). After albendazole, worms at the periphery tended to move outward to the dermis and were readily removed (Suntharasamai *et al*, 1992) and symptom gradually disappeared. Deep infection may need a long time for recovery, since it depends on how much the organ was damaged. Further investigation is needed into the relationship between organ involvement and duration of cure/recovery. In addition, the incubation period, the duration from ingesting the parasite to the time symptoms develop, is still not clearly established since it was difficult to pinpoint the actual time of ingestion that initiated the infection and it depended on the organs of parasite involvement as well.

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