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Case Report

A Case Report of Serologically Diagnosed Pulmonary Gnathostomiasis

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Since Ogata and his colleagues discovered the first (Nawa *et al.*, 1989) and subsequent two confirmed human cases (Ogata *et al.*, 1988) of gnathostomiasis doloresi, over 20 cases including suspected ones were found in Miyazaki Prefecture, Japan, for recent 10 years. All these previously discovered cases were the form of cutaneous larva migrans showing creeping eruption and/or mobile erythema (Ogata *et al.*, 1992; Nawa *et al.*, 1993; and unpublished data). According to the accumulated records of human gnathostomiasis caused by *G. spinigerum* infection in Thailand (Daengsvang, 1980) and Japan (Miyazaki, 1960), the larvae, or sometimes adults, can emerge almost everywhere of human body to cause non-cutaneous gnathostomiasis with extreme diagnostic difficulties. Here we describe a case of serologically diagnosed pulmonary gnathostomiasis of which causative agent was suspected to be *Gnathostoma doloresi* by circumstantial evidences.

Case Report

The patient was a 38-year-old male living in Saito-City, Miyazaki Prefecture, Japan, where is located in the center of the endemic area of *G. doloresi*. He often eats flesh of various wild animals and freshwater fishes including brook trout (common Japanese name: "Yamame"). On March 1994, he complained of right chest pain and high fever and visited a general physician. Because moderate eosinophilia (WBC 7200 with 16.9% eosinophils) was noted by hematological examinations, he was advised to visit a general hospital for further work-up. In addition to eosinophilia (WBC 8700, 18% eosinophils) elevation of total IgE (2125 IU/ml) was noted. Plain chest radiogram (Fig. 1A) and computed tomogram (Fig. 1B) revealed a nodular lesion on r-S_4. Because parasitic infection of unknown etiology was suspected, he was transferred to the 1st Department of Internal Medicine, Miyazaki Medical College. By a multiple dot enzyme-linked immunosorbent assay (ELISA), positive reaction was detected against *G. doloresi* antigen (Fig. 2). Reliability of the diagnosis by a dot-ELISA test was confirmed further by an Ouchterlony's double diffusion in agarose gel in that the patient's serum produced a precipitin band against *G. doloresi* antigen but not against other parasite antigens (Fig. 3). After admission, the respiratory symptoms of the patient was gradually improved by treating with anti-inflammatory drugs and antibiotics. Without specific treatment by antihelminthic drugs, the lung lesion and eosinophilia gradually reduced along with the reduction of anti-*G. doloresi* antibody titer in the serum, though total IgE remained high level (Fig. 4). The nodular lesion in the r-lung was diminished almost completely by

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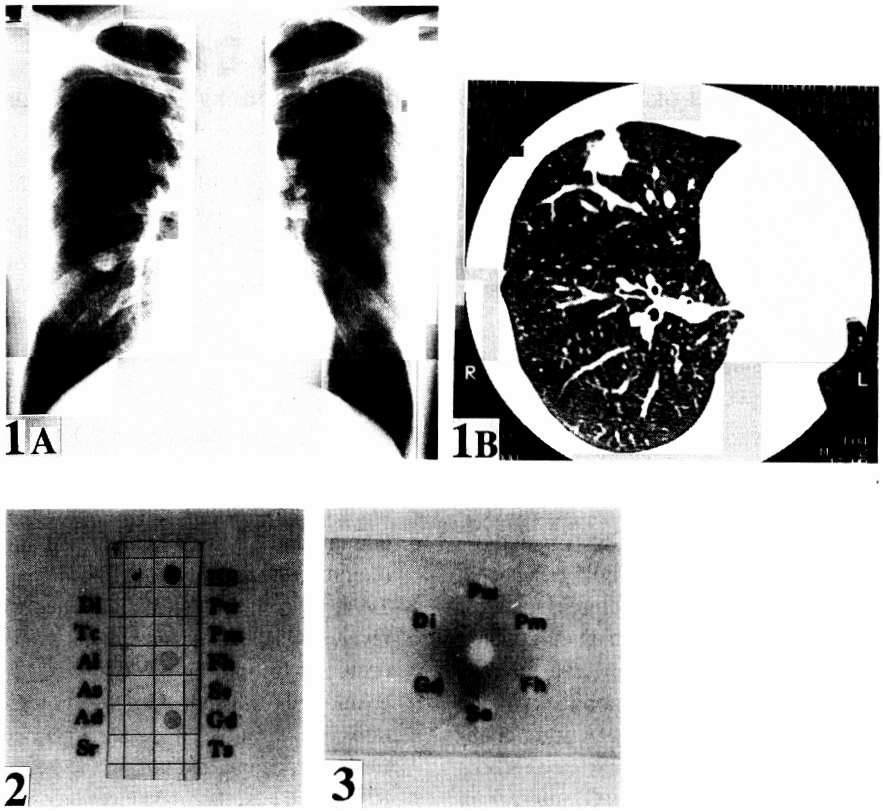

Fig. 1 Plain chest radiogram (A) and computed tomogram (B) of the patient showing a nodular lesion in the right S_4 at the time of admission.

Fig. 2 Multiple-dot ELISA.
HS: Normal human serum, Di: *Dirofilaria immitis*, Pw: *Paragonimus westermani*, Tc: *Toxocara canis*, Pm: *Paragonimus miyazakii*, Al: *Ascaris lumbricoides*, Fh: *Fasciola hepatica*, As: *Anisakis simplex*, Se: *Spirometra erinacei*, Ad: *Ancylostoma duodenale*, Gd: *Gnathostoma doloresi*, Sr: *Strongyloides ratti*, Ts: *Trichinella spiralis*.

Fig. 3 Ouchterlony's double diffusion test in agarose gel.
Center well: the patient's serum (5-fold concentrated).
Abbreviations used were the same as those appeared in Fig. 2.

the end of May 1994. During admission and subsequent following-up study, cutaneous lesion such as creeping eruption or mobile erythema was never observed in the patient.

Discussion

Gnathostomiasis is one of the important food-borne parasitic zoonoses (Nawa, 1991). Among twelve *Gnathostoma* spp. (Daengsvang, 1980), *G. spinigerum* had long been considered as the only causative species of human gnathostomiasis. Recently, however, confirmed human cases of infection with *G. hispidum* (Tsushima, 1980; Araki, 1986), *G. doloresi* (Ogata *et al.*, 1988; Nawa *et al.*, 1989) and *G. nipponicum* (Ando *et al.*, 1988) were reported from Japan. Probably due to well-preserved traditional ways of cooking, eating habits, and natural resources, Miyazaki Prefecture is known as the almost only endemic area of human gnathos-

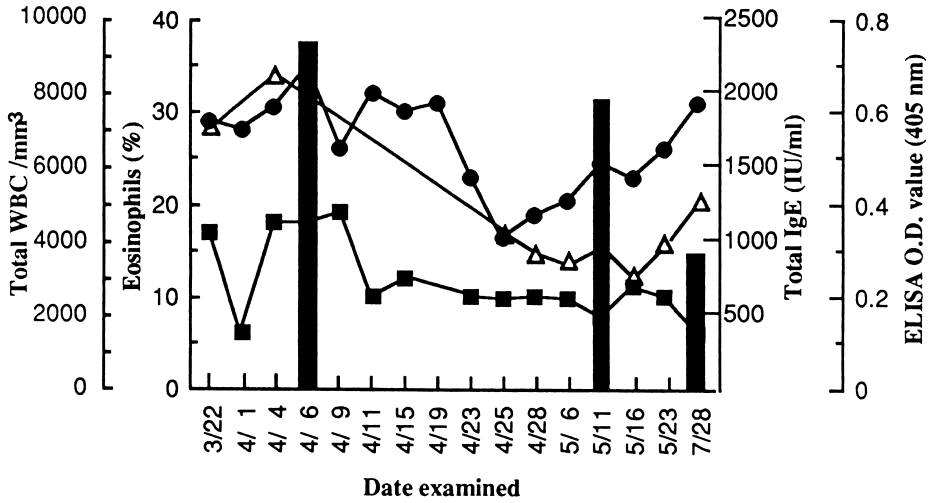

Fig. 4 Kinetics of total white blood cell count (●), eosinophilia (■), total IgE (Δ), and ELISA values (closed column) of the patient.

tomiasis doloresi, though the natural life cycle of *G. doloresi* is spreaded widely in the mountaineous area of central to south-western Japan (Miyazaki, 1960). The present patient is living in the middle of the endemic area and often eats flesh of various wild animals including brook trout, "Yamame". Almost all wild boars caught in the endemic area were infected with *G. doloresi* (Nawa and Imai, 1989). Furthermore, we have found *G. doloresi* larvae from snakes (Imai *et al.*, 1988) and fishes (Nawa *et al.*, 1993) captured in the endemic area. Over 90% of the gnathostomiasis patients found in Miyazaki Prefecture have common past history of eating flesh of brook trout, "Yamame". Therefore, the present patient is likely to be infected with *G. doloresi*.

Although gnathostomiasis is primarily a disease of the skin, the larvae sometimes appear in an unexpected site causing serious diseases. Among such non-cutaneous gnathostomiasis, only few cases of pulmonary gnathostomiasis caused by *G. spinigerum* has been reported from Thailand (Caslens, 1935; Prijyanonda *et al.*, 1955) and Japan (Okamura *et al.*, 1955). Matsuoka *et al.* (1981) reported a gnathostomiasis case with small amount of pleural effusion which was eventually confirmed by detecting the parasite in histopathological sections of the biopsied skin, though the species was not identified. In their report, the patient had eaten flesh of a snake, *Agkistrodon halys*, which was proven as the important paratenic host of *G. doloresi* (Imai *et al.*, 1988). Therefore, together with the present case, pulmonary gnathostomiasis seems to be caused also by *G. doloresi*. In addition, recently Matsushita *et al.* (1993) reported a case of pulmonary gnathostomiasis caused by *G. hispidum*, in that the worm eventually emerged in the anterior chest skin and specified by histopathological examinations. Different from those reported by Matsuoka *et al.* (1981) or by Matsushita *et al.* (1993), the patient reported in the present study never elicit a cutaneous lesion during admission and over 4 months follow-up study, indicating that the parasite was entrapped in the granuloma in the lung. The gradual decrease in antibody titers by the microplate-ELISA supports this idea.

In addition to the present case, very recently we encountered an ileus case due to eosinophilic nodular lesion formed around migrated *G. doloresi* larvae in the colonic subserosa (Seguchi *et al.*, manuscript in preparation). Therefore, in conclusion, the presence of such cases indicates that *G. doloresi* larvae can, like other *Gnathostoma* spp., migrate into various tissues and organs of human body to

cause visceral gnathostomiasis with unexpected clinical manifestations.

(This case was reported orally at the 226th Annual Meeting of the Japanese Society of Internal Medicine, Kyushu Branch held in the University of Occupational and Environmental Health, Japan, Kitakyushu, Fukuoka on Sept. 1994)

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