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Research Note

Two Cases of Paragonimiasis Westermani with Pleural Effusion in Young Girls Living in the Southern Part of Miyazaki Prefecture, Japan

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Paragonimiasis westermani is one of common parasitic diseases in Japan, especially in the south-western parts. In Miyazaki Prefecture, the endemic areas of this disease are widely spread along with the rivers. Recently, however, like other endemic areas of Japan, the prevalence of this disease in Miyazaki Pref. drastically decreased mainly because of the improvement of public health and also of the changes in food habit of the people. Yet, a few sporadic cases are occasionally found in Miyazaki Pref. Although classically known form of paragonimiasis westermani is characterized by persisting cough, bloody sputum, and nodular or ring shadows in the lung field by chest X-ray (Yokogawa 1965), atypical form of paragonimiasis westermani cases showing pleural effusion and/or pneumothorax, resembling clinically to paragonimiasis miyazakii, were recently reported from various places (Kanzaki *et al.*, 1983; Tashiro *et al.*, 1984; Nishida *et al.*, 1986; Matsuoka *et al.*, 1986; Ooga *et al.*, 1988). Here we report two cases of paragonimiasis westermani in young girls with massive pleural effusion.

Case No. 1: 9-year-old girl. She was born in Nichinan-City and occasionally eats raw, sliced meat of wild boars and also fresh-water crab (*Eriocheir japonicus*) soup. These materials were locally obtained and she often helps cooking them. On Jan. 1988, she had suffered from dry cough for two weeks, but it disappeared spontaneously without treatment. From the middle of March 1988, she complained of dyspnea at the exercise. At this stage emaciation was noticed by her mother. On the 7th April 1988, she consulted a physician with water-like diarrhea. By auscultation, respiratory sound of the right lung field was weak. Massive retention of pleural effusion was noted in the right lung by chest X-ray. She admitted to the Department of Pediatrics, Miyazaki Medical College for work-up. Physical examination revealed a skinny, emaciated child [height: 122.7 cm (−2.1 S.D.), weight: 21.0 kg (−1.8 S.D.)]. By chest X-ray, pneumothorax with massive pleural effusion was noted in the right lung (Fig. 1). Laboratory findings related to parasitic diseases were as follows; WBC 7,500 (Eosino. 8.0%); IgE 247.2 IU/ml. Pleural effusion was yellowish white in colour, turbid with numerous inflammatory cells (Eosino. >80%) and fibrin clots. By an Ouchterlony's double diffusion test in agarose, the patient's serum and also pleural effusion produced strong precipitin band against crude extract antigen preparations of *P. westermani* and *P. miyazakii* with the predominance of the reaction against the former (Fig. 2). She was treated successfully with bithionol (30 mg/kg/day for 10 alternative days).

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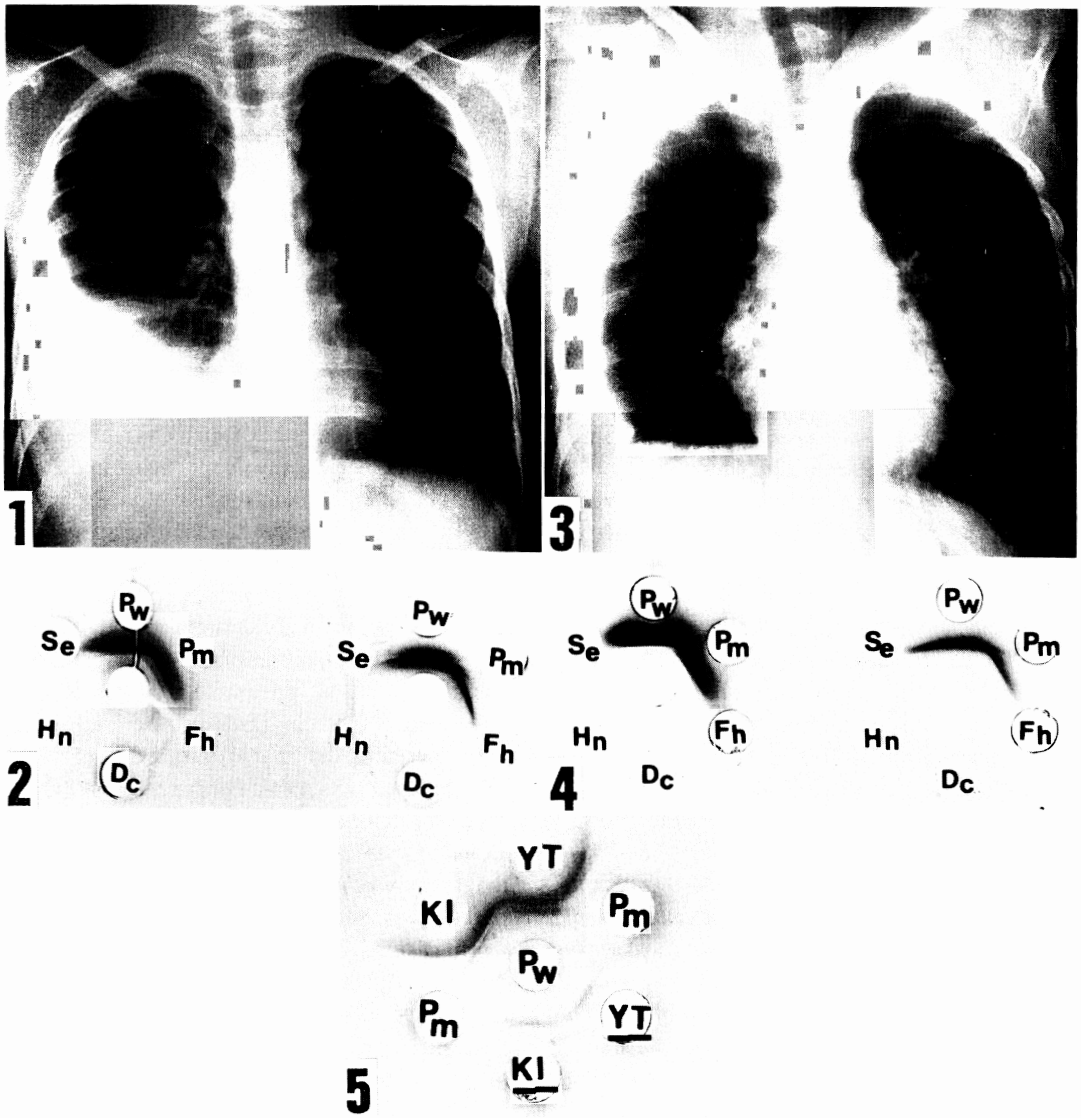

Figs. 1 and 3 Chest roentogenogram on admission
Fig. 1: case No. 1
Fig. 3: case No. 2

Figs. 2 and 4 Ouchterlony's double diffusion test in agarose
Fig. 2: case No. 1
Fig. 4: case No. 2
In both cases, left center well contains patient's serum, whereas right center well contains pleural effusion.
Pw: *Paragonimus westermani* Pm: *Paragonimus miyazakii*
Fh: *Fasciola hepatica* Dc: *Dipylidium caninum*
Hn: *Hymenolepis nana* Se: *Spirometra erinacei*

Fig. 5 Absorption test
Pw: *Paragonimus westermani* Pm: *Paragonimus miyazakii*
KI: serum of case No. 1 YT: serum of case No. 2
Patients' initials with underline indicate that the sera were adsorbed with *P. miyazakii* antigen.

Case No. 2: 11-year-old girl. She was born in Kitago-Cho, a neighbouring town to Nichinan-City. Like case No. 1, this patient also has a past history of cooking and eating locally obtained fresh-water crab, *E. japonicus*. On Sept. 1988, she consulted a local dermatologist with purplish swelling of the right ankle which was successfully treated with anti-allergic drugs for 2–3 weeks. A similar purplish swelling with induration reappeared on the right forearm on Jan. 1989 and she presented to a regional hospital. By laboratory examinations, eosinophilia (WBC 21,000, Eosino. 21%) and elevated serum total IgE level (11,000 IU/ml) was noted. Subsequently on the 10th Feb. 1989, pleural effusion was noted in the right lung by chest X-ray. On the 13th Feb. 1989 she was admitted to the Department of Pediatrics, Miyazaki Medical College, for work-up. Massive pleural effusion in the right lung was confirmed by chest X-ray at the time of admission (Fig. 3). The pleural effusion was yellowish-white in color, slightly sticky and turbid with fibrin clots and containing numerous inflammatory cells (Eosino. 60%). Immediate type intradermal skin test using *P. westermani* antigen was positive (wheel: ϕ 13 mm, erythema: ϕ 140 mm). Also, by an Ouchterlony's double diffusion in agarose, her serum and pleural effusion gave positive results similar to those observed in case No. 1 (Fig. 4). At first she was treated with praziquantel but this treatment was ineffective. Then she was treated successfully with bithionol.

In both cases, inflammatory lesions were not seen in the lung parenchyma, and the parasite eggs were not detected in their sputum, stool, nor in pleural effusion. When their sera were absorbed with *P. miyazakii* antigen and tested by an Ouchterlony's double diffusion in agarose, precipitin reaction against *P. westermani* antigen was observed, whereas that against *P. miyazakii* was completely abolished (Fig. 5). These two patients have a common past history of eating fresh-water crab, *E. japonicus*, but have never eaten *Geothelphusa dehaani*. Since Miyazaki (1982) reported that in Miyazaki Prefecture the metacercariae of *P. westermani* found in *E. japonicus* were exclusively of triploid type, the pathogen for these two cases are assumed to be *P. westermani* triploid type.

These two patients are living in (Case No. 1) or adjacent to (Case No. 2) Nichinan-City where has long been known as one of famous endemic area of *P. westermani* in Miyazaki Prefecture (Hirose, 1974). A sporadic occurrence of these and other cases (unpublished observation) in Miyazaki Pref. indicates that the natural life cycle of *P. westermani* is still preserved well in this area. The patients reported here sometimes help cooking fresh-water crabs and eat them. A high risk of ingestion of metacercariae of *P. westermani* during cooking was reported by Yokogawa (1952). The occurrence of such children cases indicates that recently the people living in this area payed little attention to the risk of infection with this parasite by cooking and eating fresh-water crabs.

In the present study, massive pleural effusion accompanied with eosinophilia and elevated serum IgE level was commonly observed in both cases. In addition, pneumothorax was noted in case No. 1. Such findings are rather the characteristics of paragonimiasis miyazakii (Yokogawa *et al.*, 1974; Hibiya *et al.*, 1984). Increase in number of such atypical cases may be explained as such that the confirmation of diagnosis was done earlier enough before the parasite reaching the lung parenchyma to form worm cyst. Alternatively, these patients might be infected with a single worm. Yokogawa *et al.* (1960) reported that, after an experimental infection with a single metacercaria of *P. westermani*, the worm could reach the pleural cavity but failed to form worm cyst. Whatever is the explanation, our results together with others show that immunodiagnosis is helpful for the differential diagnosis and identification of the causative species of paragonimiasis.

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