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Research Note

Intrahepatic Paragonimiasis – A Case Report –

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Paragonimus species is principally a parasite of the lung causing pulmonary paragonimiasis in the final host including human beings. However, because of the complexity of its migration route in the final host, this parasite often causes an ectopic infection in various sites such as skin, peritoneal cavity, diaphragm, liver, brain, etc. As the sites of ectopic infection with mature/immature *Paragonimus* worms, brain and skin are well known (Yokogawa, 1960). However, the reports of ectopic infection in the peritoneal cavity or in the abdominal viscera are rare, probably because infections in these sites are often asymptomatic and difficult to diagnose. Here we report a case of intrahepatic paragonimiasis which was proven by the postoperative histopathological examination of the calcified nodular lesions in the liver.

Case Report

History

The patient is a 62 years-old female who was born and grow up in Miyazaki Prefecture. She has no experience of travelling overseas. Earlier in 1980 when she was admitted to a regional hospital because of the control of hypertension, cholelithiasis was unexpectedly noted. However, because it was asymptomatic at that time, it had been left untreated. On 1990, she began to complain occasional right low back pain. When she was admitted to a regional hospital, calcified nodular lesions were, in addition to the presence of the stones in the gall bladder, found in the right lobe of the liver by plain abdominal rentogenography (Fig. 1), ultrasonography (Fig. 2), and also by computed tomography (Fig. 3). Since intracystic and intrahepatic cholelithiasis was suspected, she was admitted to the 2nd Department of Surgery, Miyazaki Medical College, on the 5th Dec. 1990 for workup.

Laboratory Data and Clinical Course

Her laboratory data at the time of admission were as follows; RBC 4.2×10^6/mm^3, WBC 5.1×10^3/mm^3 (Eo 3.0%), Ht 36.5%, Hb 12.5 g/dl, Total protein 5.68 g/dl, Albumin 3.68 g/dl, A/G ratio 1.84, Na 146 mEq/L, K 4.3 mEq/L, Cl 111 mEq/L, GOT 18 IU/L, GPT 16 IU/L, LDH 279 IU/L, γ-GTP 31 IU/L, Al-Pase 118 IU/L. Chest rentogenogram appeared to be normal.

Partial hepatectomy of the right lobe including calcified nodular lesions was carried out together with cholecystectomy on the 14th Jan. 1991.

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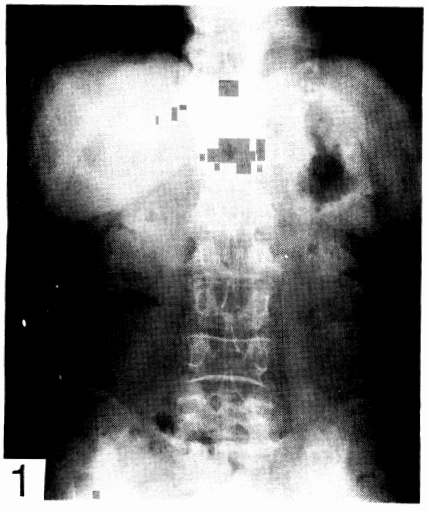

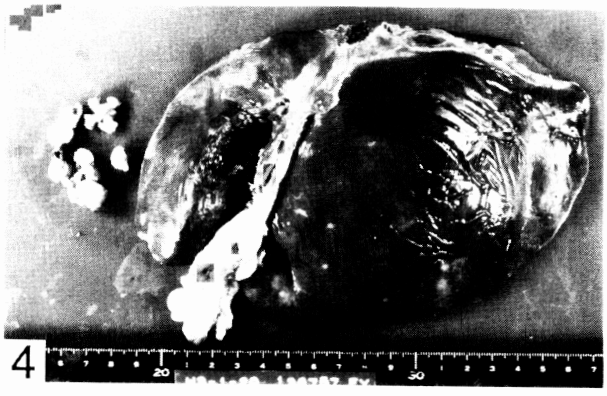

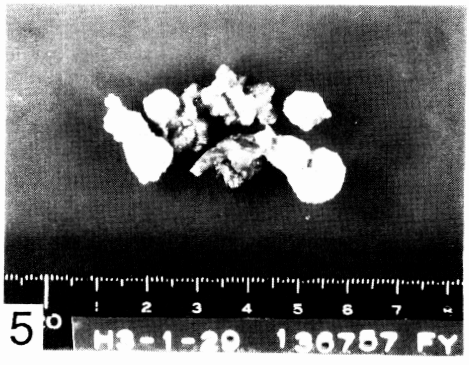

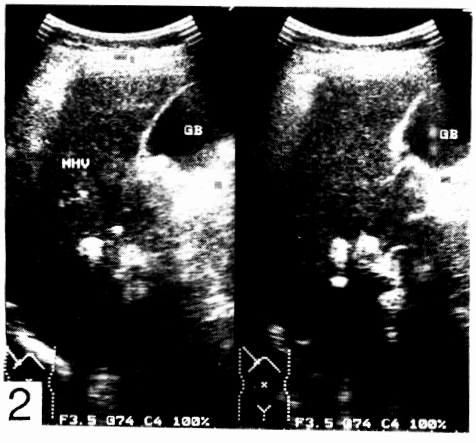

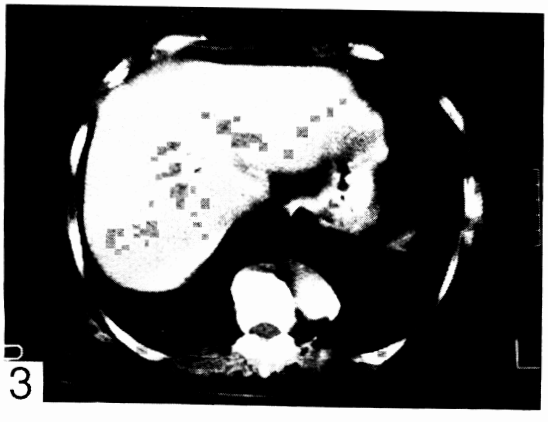

Fig. 1 Plain abdominal rentogenogram showing calcified nodular lesions in the liver.
Fig. 2 Ultrasonogram showing the lesions.
GB: gall bladder, MHV: Middle hepatic vein
Fig. 3 Computed tomograph showing the calcified lesions.
Fig. 4 Surgically removed right lobe of the liver and the dissected nodular lesions.
Fig. 5 Macroscopic appearance of the nodular lesions.

Pathological and Parasitological Findings

The nodular lesion was about 3×4 cm, multilobulated, and was not located in the intrahepatic bile duct but was in the liver parenchyma (Figs. 4 and 5). Histopathological examination revealed that the nodular lesion did not contain calculi. Instead, it was a polycystic lesion containing partially calcified necrotized mass surrounded by thick fibrous connective tissue (Fig. 6). In addition to the scattered cholesterin clefts, numerous parasite eggs were seen especially along with the cyst wall (Figs. 7 and 8). The eggs were thick-shelled, having a flattened operculum at one end (Figs. 9–12). The average size of the biggest 20 eggs was 70.9±5.1 μm by 40.9±4.5 μm. Such morphological characteristics were identical to those of *Paragonimus westermani* eggs. Although immunoserodiagnosis was carried out to confirm histopathological identification, neither the immediate type skin test nor an Ouchterlony's

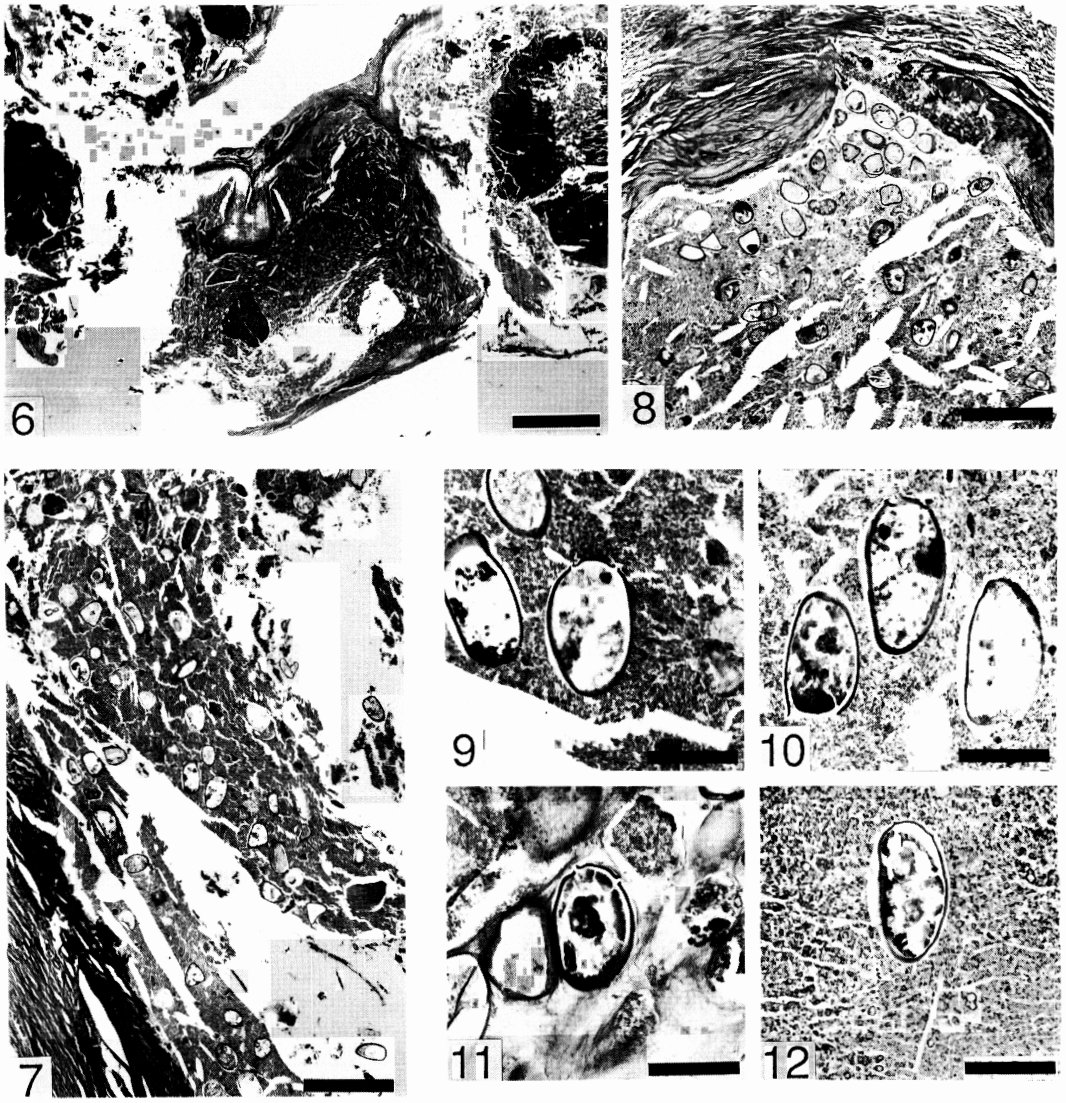

Fig. 6 A cross section of the lesions. Scale bar: 2 mm
Figs. 7 and 8 Necrotised mass containing numerous parasite eggs. Scale bar: 200 μm
Figs. 9–12 High power view of the eggs. Scale bar: 50 μm

double diffusion test using *P. westermani* antigen gave positive results.

In the present study, the bodies of adult *Paragonimus* worms or their fragments were not seen in the lesions. Still, these lesions seem to be formed by infection with adult *Paragonimus* worms and not due to the embolism of parasite eggs because the lesions were the encapsulated multilocular cysts and contained numerous eggs. When metacercariae are ingested by human or other natural final hosts, they excyst in the intestine and penetrate into abdominal cavity. The immature *Paragonimus* worms migrate into abdominal muscles and lodge there for a while and reappear in the abdominal cavity. Then they migrate through the diaphragm and the pleural cavity and finally reach the lung where they become mature adult worms (Yokogawa, 1960). Such a complicated route of migration often causes an ectopic infection along with the migration pathway. In general, ectopically infected helminth parasites are unable to mature into adult stage. However, *Paragonimus* species, especially *P. westermani* is rather exceptional because it can occasionally mature in the ectopic sites. The recovery of *P. westermani* adult worms from ectopic sites such as brain, skin, peritoneal cavity were classically reported in heavily endemic areas (Yokogawa, 1960). Our results provide a further evidence of an ectopic maturation of *Paragonimus* worms.

The parasite eggs found in the present case were identified as *P. westermani* based on the morphometric examinations. Although immunoserological tests are often useful for the diagnosis of paragonimiasis (Imai, 1987) and for the identification of *Paragonimus* species, neither immediate type skin test nor an Ouchterlony's double diffusion test gave positive results in the patient reported here, suggesting that the patient was infected with the parasite very long time ago. Partial calcification and the presence of cholesterin clefts in the necrotized mass also indicate that long time had passed since the granulomatous lesions were formed by the infection.

Miyazaki Prefecture is one of the famous endemic areas of paragonimiasis in Japan and more than 300 cases were found during the late 1950s to the early 1960s. Since 1957 the paragonimiasis project team of the local government conducted the extensive survey, mass treatment, and the campaign to eradicate paragonimiasis, so that the incidence drastically decreased within next 5 years (Hayashi, 1978). However, even nowadays a few sporadic cases are still seen every year in this area (Matsuoka *et al.*, 1986; Ichiki *et al.*, 1989; Nawa, 1991), including cerebral (Kinoshita, 1989) and cutaneous (Ogata, 1989) cases. All these previously reported cases in Miyazaki Prefecture were infected by eating and/or cooking freshwater crabs, *Eriocheir japonicus,* or the flesh of wild boars, *Sus scrofa leukomistax.* The patient reported here has, however, not clear memory as to whether she had eaten or cooked such things. Whatever the source of infection is, discovery of such a case suggest that more undiscovered paragonimiasis cases should be present in Miyazaki Prefecture.

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