A Case of Systemic Paragonimiasis with Ovarian Involvement

Sung-Tae Hong,* Soon-Hyung Lee*, Je G. Chi**, Young-Soo Jin,*** Chung-Soo Kim*** and Yoon-Seok Chang***
Department of Parasitology*, Pathology** and Obstetrics and Gynecology***, College of Medicine, Seoul National University

INTRODUCTION

It is a well known fact that *Paragonimus westermani* causes ectopic parasitism of various sites in human hosts. Major clinical problems of the ectopic paragonimiasis are related to the involved tissue or organs. Accordingly the clinical manifestation is protean and frequently causes serious differential diagnosis of neoplastic disease.

The authors experienced a case of systemic paragonimiasis involving female genital organs. The ovarian involvement in the case caused a tumor mass that was hardly differentiated from ovarian cancer clinically. Exploratory laparotomy was performed and multiple *Paragonimus granulomas* were noted. This case was successfully treated with praziquantel.

CASE REPORT

A 40 year old housewife, an inhabitant of Seoul City, was admitted to Seoul National University Hospital(SNUH) through Emergency Room complaining uncontrollable abdominal pain and a mass in right lower quadrant on May 18, 1981. She had a history of right salpingectomy and appendectomy in 1975.

The patient once ingested fresh water crabs which were bought in a market in Seoul City and soaked for a day in soybean sauce with 3 companions, around June 1979. She remembered episodes of fever, chill, productive cough and hemoptysis in early 1980. She was managed only by symptomatic treatment until June 1980, when the patient was checked by chest X-ray at a private clinic and began antituberculosis therapy for the pleural effusion. The drugs were taken for a year, until the time of this admission. In early 1981, the patient palpated a mass in her right lower abdomen. It was associated with intermittent pricking pain and tenderness which responded well to the analgesics. On May 9, 1981, the patient visited Emergency Room of SNUH complaining severe abdominal pain. The pain did not respond to drugs, and the patient was admitted to Department of Internal Medicine, SNUH on May 18, 1981.

On physical examination, a small fist sized, hard, smooth surfaced and fixed mass was palpable in right lower abdomen, and several subcutaneous masses bean to walnut sized, were also palpated in anterior chest wall, right buttock and mons pubis. Peripheral eosinophil count was 36% and microscopic hematuria was detected. The eggs of *P. westermani* were found in the sputum and feces. The serologic examination (ELISA technique which detected circulating Ig G specific for *Paragonimus* antigen) was strongly positive as O.D. 1.20, positive criterion being O.D. over 0.25. On chest X-ray, hazy mottled peribronchial densities were noted.
in the right lower and middle lung fields and also a nodular lesion was observed in the right upper lobe (Fig. 1). The changes on chest roentgenography were noted after chemotherapy (Fig. 2 and Fig. 3). The abdominal ultrasonography showed an irregular solid echogenic mass with a maximum diameter of 19 cm, posterior to the urinary bladder and also a nodular lesion in right side of uterus. Intravenous pyelography revealed dilated collecting system of the right kidney and the proximal ureter (Fig. 4).

The patient was sent to Department of Gynecology to explore the pelvic mass. On July 1, 1981, the patient was operated under the impression of ovarian neoplasm or ectopic paragonimiasis of pelvis. By surgical observation and frozen section the mass was proved to be the cyst of ectopic paragonimiasis and the patient underwent total hysterectomy, right adnexectomy, omentectomy and adhesiolysis. As for the worm masses, multiple thumb-tip sized nodules with necrotic material were resected from omentum and a small fist sized mass was excised together with uterus and right adnexa. A few thumb-tip sized nodules were found firmly stuck to the posterior wall of urinary bladder, which were inoperable because of excessive bleeding.

The excised masses were examined at the Department of Pathology. Grossly the uterus and right adnexa were the sites of multiple granuloma formations and fibrous adhesion. The parametrium and the right adnexa were studded with greyish white nodules of various size up to several centimeters. Brownish necrotic material was expelled and the eggs of *P. westermani* were seen from those cystic nodules. Microscopically there were two types of lesion; one was characterized by large foci of necrosis with ragged margin and containing amorphous eosinophilic debris in which numerous Charcot-Leyden crystals were included (Fig. 5). The wall of these necrotic lesions consisted of palisading histocytes, fibroblasts and varying number of eosinophils. This lesion was thought to be made by advancing parasite worm. The other type of lesion was widely scattered throughout the affected portions of the removed specimen and was associated with eggs. There was characteristic concentric laminated fibrocollagenous bundles around intact or degenerating ova (Fig. 6). Foreign body giant cells were often seen. Interestingly, sections of ovary showed a deeply situated lesion which was surrounded by parasitic granuloma. The necrotic lesion contained numerous eggs and the wall had ragged margin with palisading histiocytes (Fig. 7). Additionally an adult *P. westermani* was detected from a cystic mass in omentum (Fig. 8).

The patient was treated with praziquantel 25 mg/kg t.i.d. for 3 days one week after the operation. The drug was well tolerated except for a mild headache, itching and mild ocular pain. The number of *Paragonimus* eggs in a gram of feces (EPG) decreased from 1,100 to 30 during the first 5 days after ingestion of the last dose, and only the eggs were detected as EPG 100 on the 17th and 30th day after then, and negative for two months after treatment. As for sputum examination, the eggs counted 2,900/ml of sputum before treatment, and decreased to 400/ml on the second day, and in the range from 14 to 65 until the 9th day. Only on the 18th day after then, 70 eggs were found from 0.5 ml of sputum, and negative in other days. The subcutaneous masses softened remarkably after treatment and diminished in size. One mass near mons pubis was excised on the 10th day after chemotherapy, but only chocolate colored necrotic debris was found without worm. The patient was followed closely for 4 months and no further trouble was noted up to this time of report.

And the 3 companions of her who ingested
the fresh water crabs together with this case all suffered from pulmonary paragonimiasis and treated in other hospitals.

DISCUSSION

Present case could be called as a case of systemic paragonimiasis since the worm involved lung, subcutaneous tissue, omentum and pelvic organs. And the cases of systemic paragonimiasis involving the ovary are not so commonly seen. Only Chung et Kim (1968) reported a case of ectopic paragonimiasis in an infertile woman who had Paragonimus nodules in the uterine wall, ovary, uterine tube and accessory ligaments in Korea. And Hsu et al. (1959) described two cases of ectopic paragonimiasis of female genital organs in Taiwan; one was myometrial invasion and the other was involved of left ovary with abscess formation. The worm migrates through the abdominal cavity normally before it penetrates into the pleural cavity. Ovarian involvement is regarded by direct invasion of the worm through ovarian capsule during the migration period. And this case also suffered from right hydronephrosis and hydrouraeter as complications of ectopic worm cysts. The clinical problems of this case all subsided after surgery and chemotherapy. And the eggs found in feces one month after treatment could be explained as late discharge of remained necrotic debris from the lung.

Although paragonimiasis is one of rare diseases in the gynecology field, it can bring various clinical manifestations. And the worm can involve almost all organs as Musgrave (1907) described. These characters make diagnosis more complicated. However, when a mass is suspected as an ectopic Paragonimus worm cyst, the newly applied technique, ELISA, will be helpful in diagnosing it (Cho et al., 1981). And the drug, praziquantel, which is an excellent chemotherapeutic agent against paragonimiasis (Rim et al., 1981) can promise cure of ectopic paragonimiasis especially the systemic case which is far beyond the surgical measure. With the diagnostic method and the drug, such complicated case of systemic paragonimiasis as present one can be managed more easily.

SUMMARY

The authors reported a case of ectopic paragonimiasis in a 40 year old Korean housewife who came to the hospital because of abdominal mass and pain. She was proved to have multiple parasitic granulomas involving lung, uterus, uterine tube and subcutaneous tissue. The ovary was also the site of parasitism. The past history revealed an episode of ingestion of fresh water crabs soaked in soybean sauce which were obtained in a market of Seoul City 2 years prior to the present illness. The patient was treated for tuberculosis for a year without confirmation of etiologic agent. After surgical removal of all abdominal and most of pelvic granulomas, the patient was treated with praziquantel 25mg/kg t.i.d. for 3 days, and was judged to be cured after 4 months.

REFERENCES


Musgrave, W.E. (1907) Paragonimiasis in the Phili-
EXPLANATIONS ON FIGURES

Fig. 1. Chest P-A film before chemotherapy; white arrow indicates a nodule in right upper lung field.

Fig. 2 & 3. Chest P-A films, taken 2 and 4 months after chemotherapy. White arrows indicate changed nodular density.

Fig. 4. Intravenous pyelography showing right hydronephrosis and hydroureter.

Fig. 5. Low power view (×40) of the omental mass, parasitic worm-associated lesion with ragged margin, containing necrotic debris and amorphous eosinophilic material, H&E staining.

Fig. 6. Egg granuloma found in omental fat. Characteristic concentric lamination around a degenerating egg is seen, H&E staining (×100).

Fig. 7. Low power picture (×40) of ovary section, showing cavitary parasitic lesion in the middle of the ovarian parenchyme. OC represents ovarian capsule and white arrow indicates ovarian ovum and black arrows indicate parasitic ova, H&E staining.

Fig. 8. Adult *P. westermani* isolated from an omental mass, acetocarmine staining.
수술 후 환자는 praziquantel을 25mg/kg의 용량으로 1일 3회, 3일간 복용하였고 부작용으로는 가벼운 두통을 호소하였다. 부약 후 기침, 기액 등의 증상은 7일 이내에 사라졌고 대변과 가래의 증소도 그 수가 급속히 감소하여 한달 후에는 전혀 발견되지 않았다. 하중 및 림프의 크기도 모두 줄고 덜어도 많이 감소하였고, 4개월까지 관찰된 욕부 방사선검사에서는 상부의 결제가 희정한 희미한 혼탁으로 변하였다. 이 환자의 전신의 간 질환에 의한 병리은 수술과 praziquantel 부여에 의해 완화되었다고 판단된다.