

An imported case of hepatic unilocular hydatid disease

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Abstract: A 38-year old man visited a private clinic complaining of epigastric discomfort for 2 months. A huge hepatic cyst was found by sonography and computerized tomography. An exploratory laparotomy was performed under the impression of hydatid disease. The cyst was successfully removed. A lot of living protoscolices of *Echinococcus granulosus* were found from the cystic fluid under light microscopy. During the operation, however, the cyst was accidentally ruptured and the cystic fluid spilled out. The patient was medicated with albendazole, and had been well without any signs of anaphylaxis or recurrence for 1 year follow-up period. He had been in Saudi Arabia for 3 years. This is the 16th case of hydatid disease reported in Korea and a case without immediate complication in spite of rupture of the cyst.

Key words: *Echinococcus granulosus*, man, unilocular hydatid cyst, imported case, albendazole

INTRODUCTION

In Korea, there were a few reports on the occurrence of hydatid cysts among the cattle reared in Cheju-do (Jang and Oh, 1974; Seo *et al.*, 1975). As for human infections, however, only one out of total 15 cases reported since 1983 was suspected as indigenous infection and other 14 were regarded as imported ones especially from the Middle East (Chung *et al.*, 1983; Park *et al.*, 1985; Lee *et al.*, 1986; Kim *et al.*, 1986; Kim and Chang, 1987; Im *et al.*, 1987; Suh *et al.*, 1987; Jeon *et al.*, 1988, Lee *et al.*, 1988; Kwon *et al.*, 1988; Huh *et al.*, 1988; Suh *et al.*, 1989; Kim *et al.*, 1993).

The prognosis of the unilocular hydatid cyst is fair if the cyst is at an operable site and can be removed without spillage of the cystic fluid, which may cause anaphylactic reactions. Recurrence of the disease, due to development

of a secondary cyst at the time of operation, may also be anticipated in 50% of the cases with unilocular cysts lodged in critical centers, and in such cases the prognosis is usually grave (Beaver *et al.*, 1984). In this paper we describe the 16th case of unilocular hydatid disease in Korea, of which the cyst was ruptured during operation but without anaphylaxis or any evidence of recurrence for 1 year.

CASE DESCRIPTION

In February, 1994, a 38-year old man residing in a suburban area of Kyonggi-do visited a private clinic because of indigestion lasting for 2 months. Ultrasonography was done, and a cystic mass suspected as the hydatid cyst was detected in the right lobe of the liver. He was transferred to Seoul National University Hospital (SNUH) for management of the cyst. The sonography done in SNUH revealed that near the upper portion of the cyst located a daughter cyst (Fig. 1), and multiple

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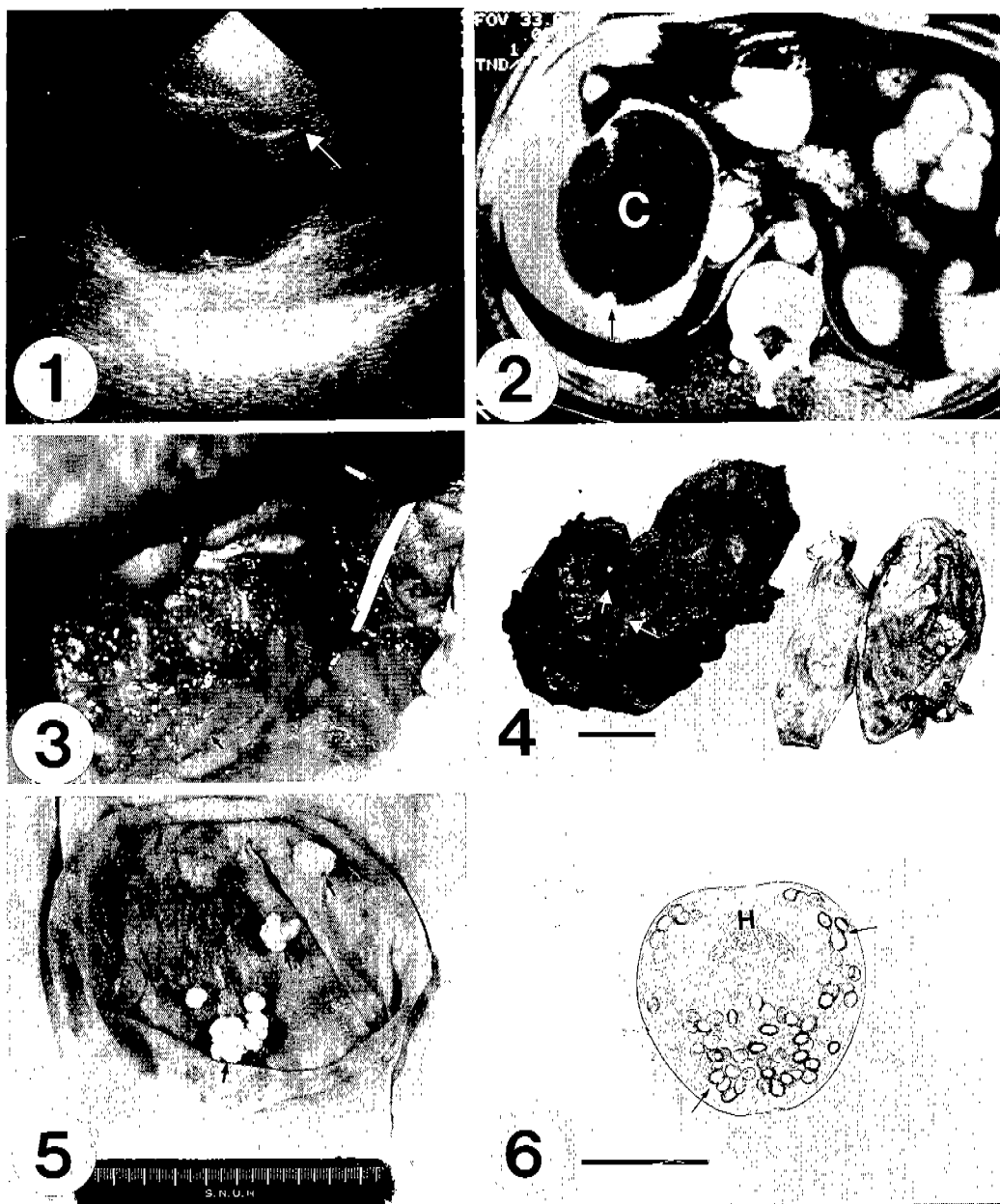


Fig. 1. Ultrasonographic finding of the liver showing a huge cystic mass. Near the upper portion located a daughter cyst (arrow). Before operation. **Fig. 2.** Abdominal CT showing a well demarcated cystic mass (C) in the right lobe of the liver. A small portion of the wall has been calcified (arrow). T₁₂ level. **Fig. 3.** Operative finding of the hydatid cyst of the liver which shows whitish, glistening, and membranous external surface (arrows). **Fig. 4.** Resected hydatid cyst (11 cm × 11 cm × 10 cm). Daughter cysts are also seen (arrows). Scale bar = 5 cm. **Fig. 5.** Inner surface of a cyst showing cauliflower-like calcified protuberances (arrows). **Fig. 6.** A protoscolex recovered from the cystic fluid having many hooklets (H) and calcareous corpuscles (arrows). Scale bar = 50 m.

small mural nodules appeared along the cyst wall. Gallbladder stones were also detected. In

computerized tomography (CT), a huge hepatic cyst was discovered. The outer margin was

Table 1. Results of serologic test for parasite-specific IgG antibodies by micro-ELISA^{a)}

Antigen	Absorbance of serum sample No. ^{b)}			Positive criteria
	1	2	3	
<i>Taenia solium</i> metacestode	0.11	0.25	0.11	0.18
<i>Paragonimus westermani</i>	0.01	0.01	0.04	0.25
Sparganum	0.02	0.04	0.05	0.25
<i>Clonorchis sinensis</i>	0.03	0.05	0.07	0.25
Hydatid cyst	0.19	0.24	0.30	0.18

^{a)}The test was done by the Department of Clinical Pathology, Yongsan Hospital, Chung-Ang University College of Medicine. ^{b)}Three times of sera collection were done.

encapsulated with a thick wall, and a small portion of the wall was calcified (Fig. 2). He had the history of working in Saudi Arabia for 3 years, from 1982 to 1984, and reared a dog near his residence.

On physical examination, the liver was not palpable and others unremarkable. The blood picture was within normal range, and the eosinophil count 0.9%. Serologic tests by ELISA showed that the patient's serum was reactive to both hydatid cyst and *Taenia solium* metacestode (Table 1). Endoscopic retrograde cholangiopancreatogram (ERCP) for the evaluation of biliary communication proved that there was no communication.

Exploratory laparotomy was done under the impression of hepatic echinococcosis. The cyst was adhered to the duodenum, omentum, and colon (Fig. 3). Cyst excision was performed with cholecystectomy and segmentectomy (Fig. 4). However, the cyst was ruptured accidentally during the operation. Fortunately, there was no anaphylactic reaction due to the spillage of the cystic fluid. After the operation, he was treated with albendazole 200 mg/day for two weeks. Three months after the operation, he had to be reoperated due to postoperative stricture of the common bile duct and hepatic duct. After then, he was discharged and has been well without any sign of clinical or radiological recurrence for one-year follow-up period.

Grossly, the hydatid cyst was 11 cm in diameter, and encapsulated with a thick cyst wall of 0.7 cm in thickness, containing 5-6 large daughter cysts. The outer surface of the cyst was pinkish gray, smooth and glistening and the inner surface was yellowish gray.

Focal hemorrhage was also noted. There were several whitish, rubbery, firm polypoid lesions growing from the inner surface of a large daughter cyst (Fig. 5). Many living protoscolices were found from the sediment of the cystic fluid (Fig. 6). They were armed with hooklets. The above findings were consistent with the characteristics of *E. granulosus* metacestode.

DISCUSSION

The unilocular hydatid cyst may go for 5-20 years before it is diagnosed (Beaver *et al.*, 1984). Hence, the present case can be regarded to have been infected 10 years ago when he was in Saudi Arabia. Yet a possibility of indigenous infection can not be completely ruled out, since Seo *et al.* (1975) found hydatid cysts from the cattle in Cheju-do, and before that time many authors reported prevalence of the enzootic cycle of echinococcosis in Korea (Issiki, 1944; Kim *et al.*, 1969; Jang and Oh, 1974). Possible occurrence of indigenous cases in Korea should be further clarified.

The 15 cases previously reported in Korea are summarized in Table 2. Among them, 13 cases were imported from the Middle East, and one from Pakistan. The first case of Chung *et al.* (1983) had never been abroad. The age of patients were distributed from twenties through fifties. Although the liver is the well known predilection site, the lung also showed a similar frequency among the Korean cases. The chief complaints were mostly chest pain in case of pulmonary echinococcosis and epigastric discomfort in case of hepatic ones. The duration between exposure to the infection

Table 2. Brief summary of 15 reported cases of hydatid disease in Korea

Case No.	Age & Sex	Chief complaint	History of abroad travel	Incubation period ^{a)}	Involved organ	Reference
1	27/F	Chest pain	Absent	?	Lung	Chung <i>et al.</i> (1983), Yim <i>et al.</i> (1985) ^{b)}
2	25/M	Chest pain	Kuwait	3 years	Lung	Chung <i>et al.</i> , (1983), Yim <i>et al.</i> (1985) ^{b)}
3	31/M	Asymptomatic	Middle East	4 years	Lung	Park <i>et al.</i> (1985), Suh <i>et al.</i> (1989) ^{b)}
4	30/M	Chest pain	Middle East	6 months	Lung	Lee <i>et al.</i> (1986)
5	32/M	Epigastric discomfort	Saudi Arabia	5 years	Liver	Kim <i>et al.</i> (1986)
6	39/M	General malaise & epigastric discomfort	Saudi Arabia	3 years	Liver	Kim and Chang (1987)
7	49/M	Pleuritis symptoms	Saudi Arabia	21 months	Lung	Im <i>et al.</i> (1987)
8	25/M	Epigastric pain & weight loss	Pakistan	2 years	Liver	Suh <i>et al.</i> (1987), Kim <i>et al.</i> (1993) ^{b)}
9	39/M	Epigastric discomfort	Saudi Arabia	11 years	Liver	Jeon <i>et al.</i> (1988)
10	33/M	Lower abdominal discomfort	Saudi Arabia	6 years	Peritoneal cavity	Lee <i>et al.</i> (1988)
11	26/M	Asymptomatic	Libya	2 years	Lung	Kwon <i>et al.</i> (1988), Park <i>et al.</i> (1988) ^{b)}
12	39/M	General malaise	Saudi Arabia	11 years	Liver	Huh <i>et al.</i> (1988)
13	44/M	Asymptomatic	Saudi Arabia	7 years	Lung	Suh <i>et al.</i> (1989)
14	55/M	Epigastric discomfort	Middle East	10 years	Liver	Kim <i>et al.</i> (1993)
15	43/M	Epigastric discomfort	Saudi Arabia	?	Liver	Kim <i>et al.</i> (1993)

^{a)}means the duration between exposure to the infection source and onset of symptoms. ^{b)}Original case record was published in the above reference respectively.

source and onset of symptoms ranged from 6 months to 11 years.

The clinical diagnosis of the hydatid disease is usually based on the history, clinical findings and serology results. The cystic material provides the definite diagnosis. Beggs (1985) reported that most of the cysts are acquired in the childhood, although they are not diagnosed until the third or fourth decade. It may cause little symptom, and present as a mass or asymptomatic calcifications (Noah *et al.*, 1986). Since hydatid cysts usually elicit symptoms after growth to appropriate sizes, the method of early diagnosis should be established. The lesion by hydatid disease can now be detected by ultrasonography, CT, and radioisotope scan, but in view of availability and price, sonography seems the most appropriate one.

The possibility of *E. multilocularis* for the present case was easily ruled out. In the cyst caused by *E. multilocularis*, a host-produced adventitial layer encapsulating the invasive

mass is often lacking, and protoscolices are produced only rarely. The parasite is multivesicular, forming irregular spaces that consist of folded and collapsed hyaline membranes and germinal epithelium (Beaver *et al.*, 1984). All of these features were different from the present case.

It is well acknowledged that surgical removal is the best way to manage the hydatid cyst, but it may result in a little consistent rate of recurrence (Mottaghian and Saidi, 1978). Also in the present case cysts may recur remotely in the future, although he was treated with albendazole for 2 weeks as a prophylactic measure. A long term follow-up observation is strongly recommended.

Medical treatment of the hepatic hydatid disease with benzimidazole compounds was advocated by some authors on the basis of clinical improvement and cyst shrinkage as evidenced by CT imaging (Kern, 1983). However, limitations due to side effects (Woodtli *et al.*, 1985) and inconvenience of

long-term administration (Morris *et al.*, 1985) also emerged. A more effective, convenient, and safe drug for hydatid disease should be searched for in the future.

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=초록=

해외에서 유입된 간 포충 낭종증 1례

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소화불량을 주소로 내원한 38세 남자의 초음파 촬영과 전산화단층촬영 소견에서 간의 낭종이 발견되어 낭종적출술을 시행하였다. 낭종은 성공적으로 적출되었으며, 현미경 하에서 낭액 내에 살아 움직이는 단방조충(*Echinococcus granulosus*)의 protoscolex가 다수 관찰되었다. 그러나, 수술중 낭종이 터져 낭액의 유출이 있었다. 술후 환자는 albendazole을 투약받았으며, 아나필락시스 등 합병증은 관찰되지 않았다. 환자는 1982년부터 3년간 사우디아라비아에서 근무한 적이 있었다. 이 환자는 국내에서 확진된 단방포충 낭종증의 제16례로, 수술 중 낭종이 터졌으나 아나필락시스를 일으키지 않은 1례이었다.

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