Hydatid Disease of the Liver: the First Indigenous Case in Thailand and Review of the Literature

Pattaya Riengchan MD*, Chusana Suankratay MD, PhD*,
Henry Wilde MD**, Voranuch Thanakit MD***

** Department of Medicine, Faculty of Medicine, Chulalongkorn University*
*** Queen Saovabha Memorial Institute, Thailand*
**** Department of Pathology, Faculty of Medicine, Chulalongkorn University*

The authors describe the first indigenous case of hepatic hydatid disease in Thailand. A 58-year-old female presented with progressive right upper quadrant abdominal discomfort over a 6-month period. Ultrasonography and computed tomography showed a solitary cystic lesion 11x12x13 centimeter in size at the left lobe of the liver. She had never been abroad and had no livestock exposure. The first operation was complicated by spillage of the parasite which required a combination of albendazole and praziquantel and a second operation for intracystic instillation of hypertonic saline solution. Unfortunately, uncontrolled generalized seizures developed due to severe hypernatremia. She never regained consciousness and expired 3 weeks after admission. In addition, the authors also reviewed previous reports of hydatid disease in Thailand. To date, only 9 cases have been reported since 1932.

Keywords : *Hydatid disease, Echinococcosis, Echinococcus granulosus, Itraoperative spillage, Intraperitoneal instillation, Albendazole, Praziquantel*

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Echinococcosis (hydatid disease) is a zoonosis caused by the larval tapeworm of *Echinococcus granulosus, E multilocularis, E vogeli* or *E oligarthrus*[1-6]. The definitive hosts are dogs that pass eggs in their feces. The intermediate hosts include sheep, cattle, goats, camels, horses and humans in *E granulosus*, and rodents in *E multilocularis*. After ingestion of eggs, embryos are released and penetrate the intestinal mucosa, enter the portal circulation, and disseminate throughout various organs particularly the liver and lungs. *E granulosus*, which causes unilocular cystic lesions, is prevalent in areas where livestock is raised in association with dogs including Australia, latin America, eastern Europe, Africa and the middle East. *E multilocularis*, which causes multilocular alveolar lesions, is prevalent in alpine, sub-arctic or arctic regions. It is now spreading over western Europe where the fox (*Vulpes vulpes*) is the definitive host[7]. *E vogeli* and *E oligarthrus*, which cause polycystic lesions, are prevalent only in central and latin America. Wild canines are their definitive hosts. Human hydatid disease is extremely rare in Thailand. To date, only 9 cases have been reported since 1932[8-16]. The authors report a fatal case of hepatic hydatid disease complicated by intraoperative spillage from the cyst, treated postoperatively with hypertonic saline by intraperitoneal instillation and a combination of albendazole and praziquantel. Previous reports of hydatid disease in Thailand were also reviewed.

Correspondence to : Suankratay C. Department of Medicine, Faculty of Medicine,Chulalongkorn University, Bangkok 10330, Thailand. Phone: 0-2256-4249, Fax: 0-2256-4578, E-mail: schusana@hotmail.com

Case Report

Four weeks prior to admission, a 58-year-old Thai female presented to the surgical department complaining of progressive right upper quadrant abdominal discomfort over a 6-month period. Ultrasonography and computed tomography were performed, and demonstrated a solitary cystic lesion 11 x 12 x 13 centimeter in size at the left lobe of the liver (Fig. 1). She was born in Nakorn Srithammarat, in southern Thailand, and worked in a supermarket at

Samutprakarn, near Bangkok. She denied having been abroad and had no livestock exposure. She had no pets and no history of fever, chills or weight loss. Her other medical history was unremarkable. She was admitted for elective cystectomy. Physical examination revealed hepatomegaly (7 centimeter below the right costal margin at the mid-clavicular line) and no other abnormalities. She had a normal total white blood cell count of 4.5 x 10^9 cells/liter, a hematocrit of 35.5 percent, a platelet count of 169 x 10^9 cells/liter and no eosinophilia. During laparoscopy, there was a cyst at the left lobe of the liver. It had a thin wall, and contained colorless clear fluid. The cyst wall was partially removed. Histopathological examination showed laminated eosinophilic membranes. Parts of protoscolices of *Echinococcus* were seen (Fig. 2). A

Fig. 1 The computed tomography of the abdomen showed a non-enhancing solitary cystic lesion 11x12x13 centimeter in size at the left lobe of the liver

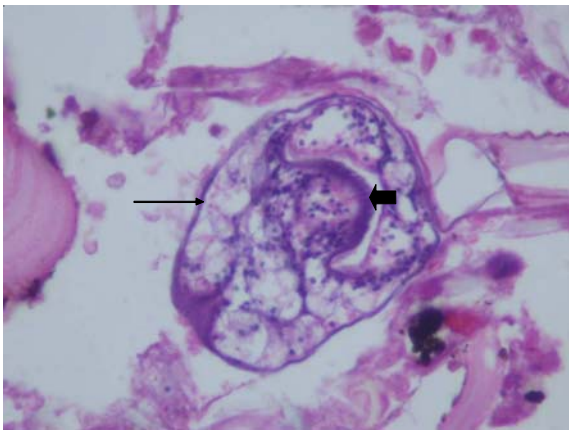

Fig 2. The hydatid cyst revealed brood capsule (⟶) with scolex (⬅). The scolex was ovoid and contained suckers, acid-fast hooklets

combination of praziquantel and albendazole was administered postoperatively. After 3 days of medical treatment, she was re-explored for total cystectomy and visceral peritoneal protoscolicidal seeding was observed. Eight liters of 15 percent saline solution as a protoscolicidal agent was instilled into her peritoneal cavity, left for 15 minutes and then removed. Six hours after operation, she developed generalized tonic-clonic seizures. Her blood sodium level was found to be 203 mEq/L. Seizures were controlled with anticonvulsants, muscle relaxant and ventilatory support. Hypernatremia was corrected by immediate continuous venovenous hemofiltration (CVVH). However, she never regained consciousness and expired 3 weeks after admission.

Discussion and Literature Review

Human hydatid disease can involve the liver (50-70 percent), lung (20-30 percent), kidney (4 percent), muscle (4 percent) and bone (1-4 percent)[1-6]. The silent period of the disease may be more than 10 years. The most frequent clinical presentations include pain, swelling or pressure effect from enlarging cysts.

Awareness is needed for the diagnosis of hydatid disease, particularly in non-endemic regions. Patients usually present with a solid or cystic lesion(s) demonstrated by radiography, ultrasonography (USG), computed tomography (CT) or magnetic resonance imaging (MRI)[1-6,17]. A definite diagnosis is made by microscopic examination of the cyst wall, comprising three layers and containing scolices and/or protoscolices of *Echinococcus*. Failure of preoperative diagnosis increases the risk of accidental intraoperative rupture and spillage leading to dissemination of infectious scolices, secondary bacterial infection or hypersensitivity reactions to the cyst's content. These can range from urticaria to life-threatening anaphylaxis. Hydatid disease was not suspected preoperatively in the present patient. She had no hypersensitivity reaction but there was peritoneal dissemination of parasites. Unfortunately, there was a fatal outcome from severe hypernatremia after incautious hypertonic saline instillation into the peritoneal cavity. This complication has been reported previously following intracystic hypertonic saline instillation, either alone or as adjunctive therapy before operation to minimize dissemination of infectious scolices[18-21].

The incidence of hydatid disease from *Echinococcus granulosus* varies among different

geographical areas. It is highest in Australia, Latin America, Eastern Europe, the middle East and Africa. In Thailand, human hydatid disease is extremely rare. To date, a total of 9 case reports of hydatid disease have been published (table 1)[8-16]. They included hydatid disease involving the lung (4 cases), liver (2 cases), kidney (1 case), peritoneum (1 case) and soft tissue (1 case). The basis for the difference may in part be due to underestimation of hepatic hydatid disease (which can only be demonstrated by USG, CT or MRI) and which is more likely to be overlooked than pulmonary hydatid disease (which can be easily demonstrated by plain chest radiography).

The male to female ratio was 5:3 (1 absent data). The age of patients varied from 32 to 52 years old. The disease was found in Thai patients without (3 cases) and with (2 cases) a history of living abroad. There are two indigenous cases with unknown travel history. There was only one foreigner (a Nepalese dentist who came to Thailand for medical care). One case report contained no travel data. Among two cases of hepatic hydatid disease, all were imported. The presented patient is the first indigenous case of hepatic hydatid disease in Thailand without a history of traveling abroad or exposure to livestock.

One case of *E multilocularis* hydatid disease has been reported in Thailand. This parasite is prevalent only in alpine, sub-arctic or arctic regions. Unfortunately, there was no information regarding travel in the report[8].

Nakorn Srithammarat, Krabi, Samutprakarn, Samutsakorn, Rachaburi, and Lumphun have reported cases with this disease. Due to the very long asymptomatic incubation period and the small number of cases, it is difficult to draw many conclusions. However, there was no case from northeastern Thailand. As mentioned above, *E granulosus* is prevalent in areas where livestock is raised in association with dogs. This may imply that in Thailand the parasite may be present in dogs which are the definitive hosts, but the scarcity of the disease may be due to lack of close contact between dogs and livestock. There may also be differences in life style, cooking and eating habits that hinder transmission to humans as intermediate hosts. Furthermore, little is known regarding the prevalence of hydatid disease among potential definite hosts in Thailand. However, it is not uncommon in ruminates, and cestodes of many species are seen in Thai dogs but have not been studied (personal communication, Dr. Suwannee Nithiuthai). Canines are

Table 1. Summary of 9 previous case reports, published from Thailand

Patient, year (reference)	Sex*, age	Birth place, nationality	Where the disease was acquired	Occupation	Site of infection	Species of *Echinococcus*	Eosinophilia (%)	Treatment	Outcome
1936[8]	M, 52	Thailand, Thai	NA (but admitted to Chiang Mai Hospital)	NA**	Peritoneum	*E multilocularis* (autopsy)	NA	Paracentesis	Died 3 years after diagnosis
1952[9]	M, 16	Thailand, Indian	History of travel to India	School boy	Lung	*E granulosus*	27	Pneumonectomy	Survived
1980[10]	F, 56	NA	NA	NA	Lung	NA	NA	Cystectomy	NA
1980[11]	F, 32	Samutsakorn, Thai	Rachaburi	Cattle and sheep farmer	Lung	*E granulosus*	2	Cystectomy	Survived
1989[12]	NA	Thailand, NA	NA	NA	Soft tissue	NA	NA	NA	NA
1992[13]	M, 32	Rachaburi, Thai	Krabi	Agriculture official	Lung	*E granulosus*	18	Albendazole and cystectomy	Survived
1993[14]	F, 39	Thailand, Thai	Lumphun	NA	Kidney	*E granulosus*	NA	Partial removal and marsupialization	Survived
1995[15,16]	M, 36	Nepal, Nepalese	Nepal	Dentist	Liver	*E granulosus*	12	Albendazole and partial left lobectomy and 20% saline intracystic instillation	Survived
1996[17]	M, NA	Ubonrachatani, Thai	The Middle East	NA	Liver	NA	NA	Albendazole and praziquantel	NA

* M: male, F: female; ** NA: not applicable

mobile and the parasite surely is present in this region. The low prevalence of the disease among humans in Thailand, which harbors approximately 10 million dogs, needs more study. Dogs should be surveyed for *E granulosus* ova.

Until recently, surgery was the only option for treatment of hydatid disease. However, it has been associated with frequent recurrences and a high rate of perioperative morbidity and mortality. In the reviewed Thai case reports, the outcome was good and the mortality rate was only 1.4 percent (1 dead, 6 alive and 2 absent data). One case, who presented with progressive ascites, died due to unrecognized *E multilocularis* peritoneal infestation. Praziquantel and benzimidazole compounds such as albendazole have been associated with high treatment failures[22]. The current recommendation by the World Health Organization is percutaneous puncture under sonographic guidance, aspiration of cystic fluid, injection of a protoscolicidal agent (saline or alcohol) and reaspiration of cyst content (PAIR)[23]. This procedure needs to be further evaluated in larger studies[24]. In the Thai case reports, the treatment was conservative (1 case), surgery alone (4 cases), surgery with medication (2 cases), medication alone (1 case) and surgery with medication and saline instillation (1 case).

To the best of our knowledge, this is the first case report of hepatic hydatid disease in an indigenous Thai patient without travel abroad. Hydatid disease should be included in the differential diagnosis of solid or cystic lesion(s) of various organs even in indigenous Thai patients without a travel history. Failure to recognize it may increase the risk of complications related to dissemination of scolices.

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โรคถุงน้ำ *hydatid* ของตับ: รายงานแรกในผู้ป่วยคนไทยที่ไม่ได้เดินทางไปต่างประเทศ และการทบทวนวารสาร

พัทธยา เรียงจันทร์, ชุษณา สวนกระต่าย, เฮ็นรี่ ไวลด์, วรนุช ธนากิจ

ได้รายงานผู้ป่วยรายแรกของประเทศไทยที่เป็น hydatid disease ของตับ และไม่ได้เดินทางไปต่างประเทศ โดยเป็นผู้ป่วยหญิงอายุ 58 ปี มาโรงพยาบาลด้วยเรื่องปวดท้องด้านขวาบนมานานประมาณ 6 เดือน โดยไม่มีอาการทางระบบอื่น การตรวจคลื่นเสียงความถี่สูงและเอ็กซเรย์คอมพิวเตอร์พบถุงน้ำขนาด 11 x 12 x 13 เซนติเมตร ที่กลีบซ้ายของตับ ผู้ป่วยไม่เคยมีประวัติเดินทางไปต่างประเทศ และไม่เคยเลี้ยงสัตว์ใด ๆ การผ่าตัดครั้งที่ 1 มีภาวะแทรกซ้อนเกิดขึ้น โดยการแตกของถุงน้ำและมี protoscolex ของ Echinococcus แพร่กระจายไปทั่วช่องท้อง หลังผ่าตัดได้ให้การรักษาโดยให้ albendazole และ praziquantel หลังจากนั้นจึงผ่าตัดครั้งที่ 2 เพื่อใส่ hypertonic saline solution ในช่องท้องเพื่อทำลาย protoscolex หลังผ่าตัดผู้ป่วยมีชักทั้งตัวหลายครั้ง เนื่องจากปัญหา hypernatremia จนหมดสติไป และถึงแก่กรรมต่อมา รวมเวลาอยู่ในโรงพยาบาล 3 สัปดาห์ นอกจากนั้นยังได้ทบทวนรายงานของผู้ป่วยทั้งหมดที่เป็นโรคถุงน้ำ hydatid ในประเทศไทย ซึ่งรายงานครั้งแรกในปี พ.ศ. 2475 จนถึงปัจจุบัน มีจำนวน 9 รายเท่านั้น