CASE REPORTS

CYSTIC ECHINOCOCCOSIS IN THAILAND WITH A SPECIAL NOTE ON DETECTION BY SEROLOGY IN ONE FAMILY

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Abstract. Thailand is a nonendemic area of echinococcosis. We report, herein, 3 cases with a special note on the use of serology in detection of the disease in one family. All cases were Thais. The first case was a man, having a cystic mass in the liver. He was subsequently diagnosed as having echinococcosis following positive serology. The second case, a male, had a renal hydatid cyst revealed by histopathology, and a positive serological test. The third case, a wife of case 2, was positive by serological screening for echinococcosis and subsequently proven to have splenic and liver echinococcal cysts. The present findings support the use of serology in the case detection of echinococcosis in Thailand.

INTRODUCTION

Echinococcosis (hydatidosis) is caused by tapeworm larvae of the genus *Echinococcus*. There are two predominant types: cystic and alveolar echinococcosis (McManus *et al,* 2003; Eckert and DePlazes, 2004). The former is caused by *E. granulosus*, of which domestic and wild dogs are definitive hosts, while herbivores are intermediate hosts. The latter is caused by *E. multilocularis*, of which foxes are a definitive host and microtine rodents, such as voles, muskrats, and deer mice are intermediate hosts.

Human cases of alveolar echinococcosis have limited distribution in the northern hemisphere, while cystic echinococcosis cosmopolitan and more widely distributed (McManus *et al*, 2003; Eckert and Deplazes, 2004). Southeast Asia is generally recognized as a non-endemic area of the parasite and case reports are rare. Two cases were reported from the Philippines (Eduardo, 1991), two from Singapore, which were likely imported cases (Singh *et al*, 1991), and two from Malaysia (Kutty *et al*, 1970). Thailand, with the most reported cases, had 24 cases of echinococcosis from 1932 to 2004 (Waikagul *et al*, 2006). We, report three additional cases of cystic echinococcosis in Chiang Mai, Thailand, detected by serology, two of them were a husband and wife.

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Case 1

A 52-year-old man, who was a resident in Chang Peuk Subdistrict, Mueang District, Chiang Mai Province, Thailand, visited Maharaj Nakhon Chiang Mai Hospital in December 2001 complaining of fever for 5-6 days. Physical examination revealed mild hepatomegaly. Laboratory investigation revealed a hemoglobin level of 12.8 g/dl and a total white cell count of 9.2 x 10^9/l (with a neutrophil level of 4.5 x 10^9/liter, an eosinophil level of 2.9 x 10^9/l,

a basophil level of 0.04 x 10^9 /l, a lymphocyte level of 1.1 x 10^9 /l and a monocyte level of 0.6 x 10^9 /l). A liver function test showed an elevated serum alkaline phosphatase and globulin (157 U/l and 4.1 g/dl, respectively) while liver enzyme and bilirubin levels were normal. An abdominal ultrasonogram (US) showed a 10.8 x 11.5 cm cystic mass in the right lobe of the liver. A high echogenic content was seen in the posterior aspect of the mass. A contrast enhanced liver computed tomogram (CT) revealed a 10 x 11 x 12 cm well-defined cystic mass with partial detachment of the membrane in the right lobe of the liver. The high density content correlated with that seen on the US. The patient's serum was examined for antibodies to echinococcosis by an indirect enzyme-linked immunosorbent assay (ELISA) (Morakote *et al*, 1995) using pig hydatid cyst fluid as an antigen, and it was found to be positive (ELISA value 0.723 compared with 0.178 of pool normal serum, the cut-off value was twice the ELISA value of pool normal serum). The ELISA for amebiasis was negative. Liver unilocular echinococcosis was diagnosed and albendazole given in daily doses of 15 mg/kg of body weight per day. A follow-up at 42 months after treatment by CT revealed a regression of the cyst to 5 x 6 cm size. The patient was born in Nakhon Pathom Province, then moved to Bangkok at 17 years of age. At age 25 he moved to Chiang Mai where he has lived since. He spent 6 months and 9 months in Japan at age 28 and 36 years old, respectively, then visited Australia for one week at age 46.

Case 2

A 43-year-old man who was resident of San Pak Wan Subdistrict, Hang Dong District, Chiang Mai Province, Thailand, visited the hospital in November 1997 complaining of episodes of recurrent fever, right sided abdominal pain, dysuria and hematuria for one month. Physical examination revealed tenderness on compression of the right side of the abdomen. Laboratory investigations revealed a hemoglobin level of 8.3 g/dl, a hematocrit of 25% and a total white cell count of 22.2 x 10^{12} /l (with a neutrophil level of 18.6 x 10^9 /l). Serum blood urea nitrogen and creatinine were 31 mg/dl and 2.7 mg/dl, respectively (normal ranges are 0.6-1.6 mg/dl and 7.24 mg/dl, respectively). Urinalysis showed numerous red blood cells and 8-10 white blood cells per high power field. An abdominal radiograph revealed right renal calculi. A renal scan showed right hydronephrosis with poor function and normal left renal function. The patient underwent right nephrectomy. Histopathology revealed xanthogranulomatous pyelonephritis with a hydatid cyst. Serum ELISA for echinococcosis was positive 4 weeks after surgery at a value of 0.47 (cut-off value 0.212, twice that of normal pooled serum). Albendazole, at 15 mg/kg body weight per day was given. The patient died at home in the year 2000 and an autopsy was not performed.

Case 3

The 43-year-old wife of case 2 was invited with her 25- and 8-year-old daughters and 17-year-old son to have their sera tested for hydatidosis in October 1998. All of them had a history of good health. The wife's serum was the only one positive on ELISA test with a value of 0.428 (cut-off value 0.170, twice that of normal pooled serum). Abdominal US was performed and a 4.5 x 5 cm multiseptate cystic mass was seen in the right lobe of the liver, and another 11 x 12 cm cyst was seen in the spleen. She was also found to be 12-weeks pregnant. The hemoglobin level was 10.2 g/dl, the hematocrit 30% and the total white blood cell count was 8.6 x 10^9/l (with a neutrophil level of 4.5 x 10^9/l, a lymphocyte level of 2.9 x 10^9/l, a monocyte level of 0.7 x 10^9/l, an eosinophil level of 0.3 x 10^9/l and a basophil level of 0.2 10^9/l). The urinalysis was normal. Subsequently, this patient had a spontaneous abortion a few weeks later. She underwent radical resection of the liver and

splenic cysts and made a good recovery. Histopathological examination of the specimens confirmed that both cysts were echinococcal cysts. Albendazole at 15 mg/kg of body weight per day was given.

DISCUSSION

Of 24 cases of echinococcosis reviwed from 1932 to 2004, some cases were foreigners and some imported (Waikagul *et al*, 2006). Two cases had alveolar echinococcosis, while the remainder had cystic echinococcosis. Of the latter, 16 cases were Thais who became ill while in Thailand. Cysts were found mostly in the lungs and liver. Only one case had a cyst found in the kidney, subcutaneous tissues or abdomen. This report added three more cases and noted the use of serodiagnosis in case 1 and 3. In case 1, the presence of hypereosinophilia and hepatic cysts suggested the diagnosis of echinococcosis. Therefore, ELISA for antibodies to echinococcosis was requested and the result confirmed the disease. Although apparently healthy, case 3 was invited to have her serum tested because her husband (case 2) had had echinococcosis. The ELISA was positive and the disease confirmed by ultrasound and histopathological examination. This is the first report of familial echinococcosis in Thailand. Her daughters and son were negative on ELISA. Concerning the sources of infection, case 1 was born in Bangkok and moved to Chiang Mai when he was 25 years old. He had a history of time spent in Australia and Japan at the ages of 46 and 50, respectively. He denied rearing dogs at his home. Thus, it is not clear where he contracted the infection. In contrast, cases 2 and 3 had never gone abroad and they had resided since birth in their village, where they reared a native Thai dog. In addition, there were also stray dogs in the village. The above evidence suggests that the man and his wife were autochtonous cases. Along with a previous report (Thammaprasert, 1993; Rukasem *et al*, 1995), we wonder if the transmission of *E. granulosus* occurred in Chiang Mai and Lamphun.

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