



Tehran University of Medical
Sciences Publication
<http://tums.ac.ir>

Iran J Parasitol

Open access Journal at
<http://ijpa.tums.ac.ir>



Iranian Society of Parasitology
<http://isp.tums.ac.ir>

Case Report

Co-Occurrence of Cystic and Alveolar Echinococcosis in the Liver: A Case Report

Qiang WANG^{1,2}, Shunyun ZHAO^{1,2}, Jide A^{1,2}, Yamin GUO¹, Jinyu YANG¹,
Ashraf NAVEED², *Wei GAO^{1,2}

1. General Surgery Department of Qinghai Provincial People's Hospital, Xining, Qinghai 810000, P.R. China
2. Diagnosis and Treatment Center for Echinococcosis in Qinghai Province, Xining, Qinghai 810000, P.R. China

Received 12 Sep 2020
Accepted 18 Dec 2020

Keywords:
Echinococcosis;
Echinococcus;
Case report;
China

***Correspondence
Email:**
644740875@qq.com

Abstract

Morbidity of mixed cystic and alveolar echinococcosis (CE and AE) is exceptionally rare. Less literature retrieved from a database on the internet detailed the content, including radiography, pathology, and therapeutics data. Such a case of co-occurrence of the different *Echinococcus* species was diagnosed and treated at our hospital center from Nov 2019 to Feb 2020. A 30 yr old female from the pastoral area in Qinghai Province, China, was diagnosed with a case of echinococcosis and diagnosis was confirmed after image studies, immunoassaying of hydatid enzymes, life history and pathology result. The patient underwent hepatectomy along with excision of the internal capsule. Post-operative pathology was done, and it confirmed a mixed infection of both CE and AE. The patient recovered well without complications after liver-protecting and tissue repair treatment for 15 days. Knowing about infective mode and immune method of the case might be vital for research on variation for *Echinococcus* infection.

Introduction

Echinococcosis is a zoonosis, which is distributed all over the world, especially in the west of China, like Qinghai and Xinjiang Province. *Echinococcus* is a patho-

gen that can curiously divide into two categories: *E. granulosus* and *E. multilocularis*, leading to cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively. Although



Copyright © 2021 Wang et al. Published by Tehran University of Medical Sciences.
This work is licensed under a Creative Commons Attribution-NonCommercial 4.0 International license
(<https://creativecommons.org/licenses/by-nc/4.0/>). Non-commercial uses of the work are permitted, provided the original work is properly cited.

these two species of echinococcosis are most commonly present in the Tibetan, the Kazakh and the Mongol ethnic minorities (1), there are extremely few patients infected with CE and AE at the same time (2).

In 1992, the research team for echinococcosis in Xinjiang Medical University reported the first case of multiple infections (3) and then after that four more cases were reported (4). Recently imaging findings of such a complicated case of mixed infection of CE and AE were described (2). However, there are no integral details to the treatment process of such patients so far.

We aimed to provide data for the treatment of mixed echinococcosis patients, including imaging and surgery procedure. A patient from the countryside was admitted at the center for the echinococcosis in Qinghai Province who was later confirmed as a case of co-occurrence of CE and AE.

Case Presentation

The informed written consent was taken from the patient as well as approved by the Ethics Committee of Qinghai Provincial People's Hospital.

A 30 yr old patient from the pastoral area presented with a history of contact with a sheepdog. The patient had pain in the right epigastrium and took Tibetan medicine for more than two years. During this period, the pain was not relieved. In Nov 2019, she was admitted to our center because of growing distension. Blood count, coagulation, liver, and kidney function were within normal limits. Hydatid enzyme immunoassay (target Em2 antigen) was positive. Color Doppler Ultrasound examination revealed $76 \times 63\text{mm}$ heterogeneous boundary echo located in S4-5 of the liver and $48 \times 41\text{mm}$ solid echo in S6-7 near the portal vein with a small strong photoelectric reflection inside. On plain computed tomography (CT) scan of the upper abdomen, the focus in S6-7 was located between the first

porta and inferior vena cava (Fig. 1). The T2 signal of S4-5 lesions was significantly higher than that of the other one (Fig. 2). Two lesions (arrow) expressing different HU values did not seem to be adjoint on plain computed tomography.



Fig. 1: Plain phase on computed tomography

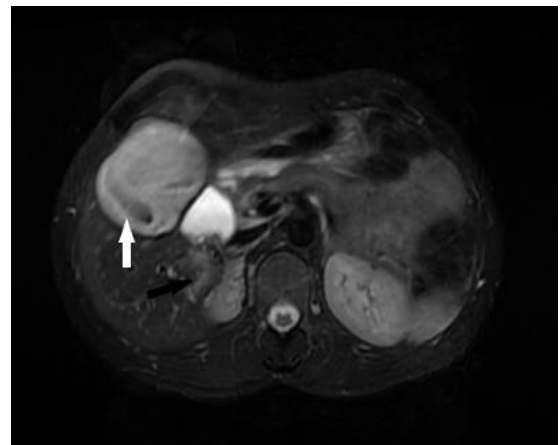


Fig. 2: Magnetic resonance image (MRI) of the lesions

Two lesions (arrow) were revealed in T2 signal on MRI. Cyst with a complete capsule containing several daughter cysts in the T2 signal was adjoined to the gallbladder wall.

Under general anesthesia, two lesions were seen on the surface of the liver when the abdominal cavity was opened, the one in the S4-5 segment was elastic, while the other in the

S6 -7 segment was uneven and hard. We resected the right half of the liver and separated the lesions. Daughter cysts and yellowish-

white matter were filled within each lesion (Figs. 3-5). Daughter cyst and necrosis material could be spotted when incising tissue.

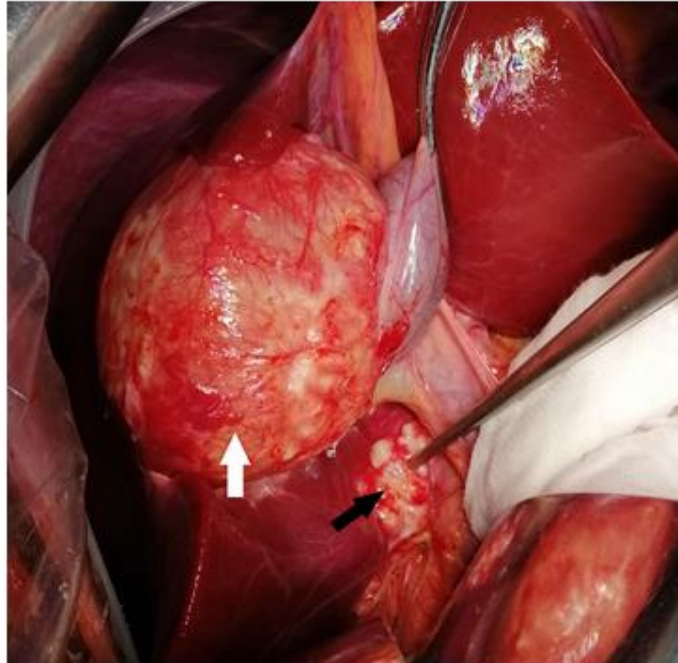


Fig. 3: Lesions location (arrow)



Fig. 4: Specimen and its content

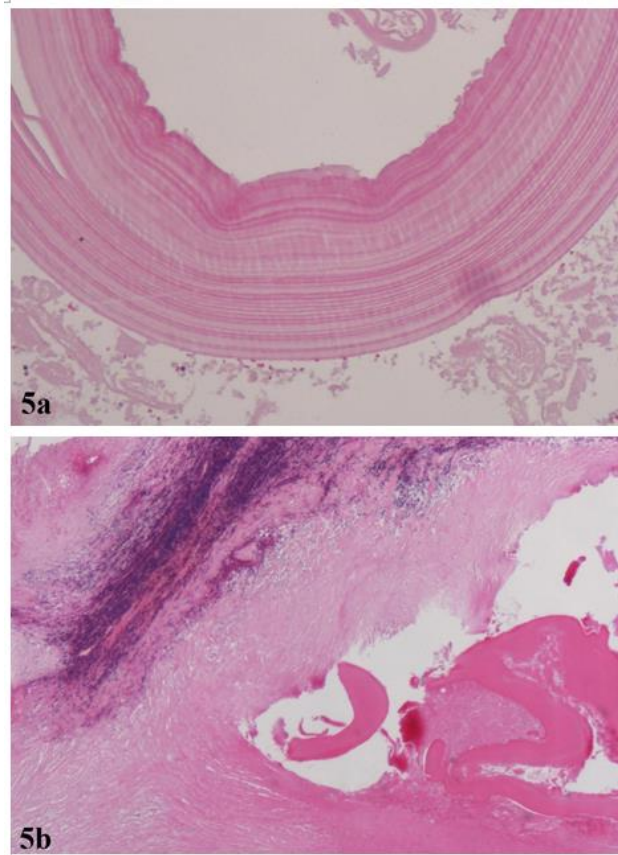


Fig. 5: Demonstration of the pathological result

Lamellar structure (a, x200) and infiltration of inflammatory cells (b, x200) are typical in cystic and alveolar echinococcosis respectively.

Discussion

The eggs of echinococcosis pass through the portal vein after entering the digestive tract and then reach the hepatic sinuses. The focus of CE was composed of boundary outer capsule wall, and the contents were daughter cysts or necrotic substance. AE is significantly different from the CE in growth pattern and whether or not with a complete capsule and clear infiltration zone (2). The lesions similar to the tumors, can invade the structure of intrahepatic vessels, bile duct and metastasize to distant organs, including lung, brain, heart, spleen, and so on (5).

Hydatid fluid lipoproteins antigen B and antigen five are used in an immunoassay for CE, while Em 2, Em 492, EM 10 are more specific for *Em* infection (6-8). Nevertheless, the serology test may lead to false positive or false negative results (6). Combining the results of imaging findings and serology tests is necessary for the confirmed diagnosis of CE and AE's co-occurrence.

In general, there are very few patients presenting with both CE and AE at the same time because of the diseases' natural history. The successive reports of such cases may indicate that the infection of echinococcosis is subtly changing according to the local environment and its treatment means (9). Surface hydatid antigen may vary for immunity in the wake of a more used antiparasitic agent. Retrospective analysis of these reported cases

from different centers needs to be researched for the etiology and epidemiology.

We should be aware of the significance of increasing these cases, which indicate other changes, e.g., communicability and ecosystems.

Conclusion

The diagnosis and treatment of mixed hydatid disease are of great significance to the study of this kind of patient.

Financial source

None.

Conflict of interest

All authors declared that there was no conflict.

References

1. Craig P. Epidemiology of echinococcosis in China. *Southeast Asian J Trop Med Public Health*. 2004;35:158-169.
2. Du Q, Hu C, Li Y. Mixed infection with two types of echinococcosis misdiagnosed as hepatic alveolar echinococcosis. *Rev Soc Bras Med Trop*. 2019;52:e20190259.
3. Wen H, Tian WL, Zou PF, et al. A rare case of mixed cystic and alveolar hydatidosis. *Trans R Soc Trop Med Hyg*. 1992;86(3):290-291.
4. Yang YR, Liu XZ, Vuitton DA, et al. Simultaneous alveolar and cystic echinococcosis of the liver. *Trans R Soc Trop Med Hyg*. 2006;100(6):597-600.
5. Brunetti E, Kern P, Vuitton DA. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop*. 2010;114(1):1-16.
6. Wen H, Vuitton L, Tuxun T, et al. Echinococcosis: Advances in the 21st Century. *Clin Microbiol Rev*. 2019;32(2):e00075-18.
7. Vuitton DA, Gottstein B. *Echinococcus multilocularis* and its intermediate host: a model of parasite-host interplay. *J Biomed Biotechnol*. 2010;2010:923193.
8. Tappe D, Frosch M, Sako Y, et al. Close relationship between clinical regression and specific serology in the follow-up of patients with alveolar echinococcosis in different clinical stages. *Am J Trop Med Hyg*. 2009;80(5):792-797.
9. Ran B, Wang M, Jian W, et al. Simultaneous occurrence of hepatic alveolar and cystic echinococcosis. *J Helminthol*. 2019;94:e80.