

A case on multiorgan echinococcal involvement in Tibet and its reflection

Case Report

Siwei Chen¹, Haiqiang Jin¹, Yu Hao², Yuhua Zhao², Chunyu Liu³, Feiqi Zhu⁴ and Yongan Sun^{1*}

¹Department of Neurology, Peking University First Hospital, Beijing, China

²Department of Neurology, People's Hospital of Tibet Autonomous Region, China

³Peking University Third Hospital, Beijing, China

⁴Cognitive impairment ward of neurology department, The Third Affiliated Hospital of Shenzhen University, China

Received: Jan 27, 2020; **Accepted:** Feb 08, 2020; **Published:** Feb 11, 2020

***Corresponding author:** Dr. Yongan Sun, Department of Neurology, Peking University First Hospital, Beijing, China

Copyright: © 2020 Dr. Yongan Sun. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Abstract

Echinococcosis, or hydatidosis, is a chronic zoonosis caused by adult or larval stages of tapeworms (cestodes) belonging to the genus *Echinococcus*. Because of the special geographic environment and lifestyle in Tibet, the incidence of the echinococcosis is pretty high there. This essay is to introduce a typical case on multiorgan echinococcal involvement including liver, lung and brain in Tibet and some reflection from this case.

Keywords

echinococcosis, multiorgan involvement, Tibet population.

Introduction

Echinococcosis is a near-cosmopolitan zoonosis caused by adult or larval stages of tapeworms (cestodes) belonging to the genus *Echinococcus* (family Taeniidae). The two main forms are *Echinococcus granulosus* and *Echinococcus multilocularis*, which can cause cystic echinococcosis and alveolar echinococcosis dividedly [1].

Echinococcosis has a worldwide distribution, especially in temperate zone [2,3] and communities where agriculture is dominant [4]. China is the only country where there are four species of *Echinococcus* [5]. In Tibet, two major types of echinococcosis are in high prevalence. The incidence rate of alveolar echinococcosis is relatively lower than cystic echinococcosis in Tibet Autonomous Region (TAR), but the prevalence rates of both types are among the highest around the world [6]. Besides, in recent years, a

new type of echinococcus-*E. shiquicus*-has been confirmed from Tibetan fox and pika on the Qinghai-Tibet plateau [5,7], which arose great attention for the echinococcosis in Tibet.

Echinococcosis mainly develops in liver and lung, while it rarely invades heart, brain, spine or ovary [8]. Among all the cases infected, brain invasion only occupied 0.4-1% [9,10].

Case

A middle-aged married Tibetan male working as a herdsman had the complaint of the right limb weakness with numbness for seven months and later he was admitted to the People's Hospital of Tibet Autonomous Region (TAR). During the course, he started with paroxysmal right lower limb weakness with numbness and gradually

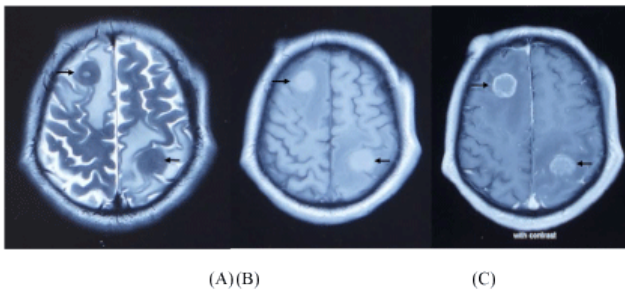


Figure 1: (A) Axial T2-weighted image reveals two heterogeneous intensity masses with peripheral high signal intensity consistent with oedema in the right frontal lobe and left parietal lobe. (B) Axial T1-weighted image. (C) Irregular rim enhancement can be seen after contrast material.

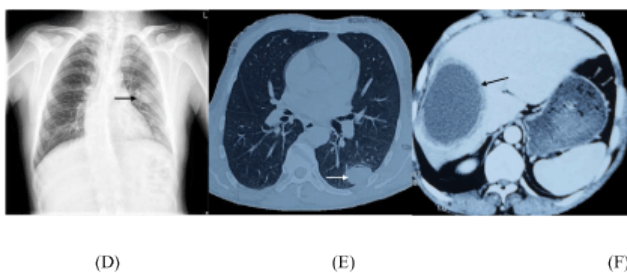


Figure 2: (D) Chest X-ray. (E) The lesion can be seen more clearly from the chest CT scan and air-fluid level exists in the cyst-like lesion. (F) A large cystic lesion with a rough rim can be figured out in the right liver lobe.

developed sudden loss of consciousness accompanied with right limbs twitching for 4 episodes. Each episode lasted for almost 2 minutes. For the past history, he had a liver hydatid resection in the hospital 5 years ago, which confirmed that he had liver hydatid according to the post-surgery pathology. For neurological physical examination, there were no positive signs. The brain Magnetic Resonance Imaging (MRI) showed that the right frontal lobe and left parietal lobe presented to have abnormal intensity nodular lesions which can be enhanced (Figure 1). His chest CT (Figure 2) discovered a high-density round lesion in the left, given the high possibility of pulmonary hydatid. In the follow-up for 14 months, his brain MRIs showed that the oedema around the lesions was significantly less but the number and the size of the lesions remained the same. Besides, his chest X-ray (Figure 2) still indicated a round high-density shadow in the left. Meanwhile, his abdominal CT scan in contrast (Figure 2) also had a liver cyst. There was no obvious abnormal index for auxiliary examinations including blood test and cerebral spinal fluid.

During the medical process, the patient took albendazole as the only therapy but the effect of anti-echinococcosis

drug seemed to be plain and the lesions remained as large as before according to the imaging results. Given the risk of surgery in brain and recurrence rate, the patient gave refusal to surgery in brain and lung but only had a liver surgery again. The patient was finally diagnosed with liver, lung, and brain echinococcosis clinically this time. Later follow-up for the patient is on-going.

Discussion

Echinococcosis can be asymptomatic to lethal [9]. Early infection may be asymptomatic. However, as the course develops, the clinical symptoms may occur and deteriorate. The extent of harm is closely related to the location, size, quantity, allergic reactions after cystic rupture and secondary infection [1,2]. For the brain echinococcosis infection, it's important to differentiate echinococcosis from cerebral malignancies (metastasis, high-grade gliomas) and other infective cerebral diseases like tuberculosis, bacterial abscess, fungal infection and toxoplasmosis etc [11]. Besides, there may be multiorgan involvements after the echinococcosis infection. The liver is the most likely affected organ. Then, it disseminates usually to the lung and brain but rarely to the pericardium, skin and bone [12]. Therefore, comprehensive inspections are in need clinically.

Diagnosis depends mainly on typical imaging findings, as well as serum-specific antibodies, pathology and nucleic acid examination. Nevertheless, serology has only limited use in the diagnostic process because of its low diagnostic sensitivity, while cross-sectional imaging is of utmost importance [13].

The best choice for treatment is total cystectomy. The more radical the surgery is, the lower risk of recurrence there will be. But it is also accompanied by more complications (1). Medical treatment (mainly albendazole) and percutaneous drainage can also be used (2). Conservative therapy is suitable for patients with multiple echinococcosis or high surgical risks [14].

Tibet is a place with special geographic environment, where there is a high prevalence rate of echinococcosis. The vast forest in Tibet may cause more incidence of alveolar echinococcosis than other countries [15-17].

What's more, Tibetans have special living and eating

habits. A large number of Tibetans are herdsmen and they usually eat raw meat, especially yak meat. An epidemiological survey supported that high prevalence of hydatid infection has been reported in sheep and yaks (99%), cattle (88%) and pigs (70%) [18]. Also, drinking unboiled water can also lead to the occurrence of the disease. As a nomadic or seminomadic ethnic group, Tibetans have many opportunities to contact cattle, sheep and dogs. They usually use animal's faeces as fuel for everyday life. Contacting with livestock's excrement is also a way to be contaminated.

Besides, Tibetans have special rite of funerals that are different from other ethnic groups. For example, they revere the celestial burial. In addition to the vultures, the corpses would also be swallowed by wolves and dogs. If he/she is a patient carrying larva or eggs of echinococcus, these carnivores can be the definitive hosts following the human beings. Some poor people in Tibet may have water burial after they pass away, leading to the disease spreading through water sources. Other evidence supporting this idea is that hydropathic pathology frequently exchanges in Morocco and the Mediterranean region and most cases were reported by countries around the Mediterranean such as Turkey and Spain [19]. Therefore, human may not be the definitive host during the disease transmission process. Under the unique circumstances of animals eating corpses or drinking water contaminated by corpses, human beings can act as intermediate hosts. These special rites of funerals make it difficult to control the disease in Tibet.

Tibet is a vast region with a low density of population distribution. Because of the poor medical conditions in some areas, most patients cannot receive treatment timely, resulting to the proliferation and metastasis of hydatid cysts [20]. To identify the disease in an early stage, ultrasound is an effective and economic friendly screening method for populations in the regions where both cystic echinococcosis and alveolar echinococcosis are endemic.

In conclusion, the education and prevention about the echinococcosis disease in Tibetan areas are still in urgent need. Meanwhile, effective screening and early treatment should be applied as early as possible.

Author contributions

SC, HJ, and YS collected data and wrote the manuscript.

YH, YZ, CL and FZ provided clinical data and helped write the manuscript.

Consent

The patient has given the written informed consent for publication.

Conflict of Interest Statement

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

References

- McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *The Lancet*. 2003;362(9392):1295-304.
- Arminanzas C, Gutierrez-Cuadra M, Farinas MC. Hydatidosis: epidemiological, clinical, diagnostic and therapeutic aspects. *Revista espanola de quimioterapia: publicacion oficial de la Sociedad Espanola de Quimioterapia*. 2015;28(3):116-24.
- Fasina O, Ogun OG. Hydatid cyst of the orbit in a young Nigerian female: a case report. *Ghana medical journal*. 2017;51(4):204-6.
- Baradan Bagheri A, Zibaei M, Tayebi Arasteh M. Cystic Echinococcosis: A Rare Case of Brain Localization. *Iranian journal of parasitology*. 2017;12(1):152-5.
- Ito A, Budke CM. The echinococcoses in Asia: The present situation. *Acta Trop*. 2017;176:11-21.
- Feng X, Qi X, Yang L, Duan X, Fang B, Gongsang Q, et al. Human cystic and alveolar echinococcosis in the Tibet Autonomous Region (TAR), China. *J Helminthol*. 2015;89(6):671-9.
- Xiao N, Qiu J, Nakao M, Li T, Yang W, Chen X, et al. Echinococcus shiquicus n. sp., a taeniid cestode from Tibetan fox and plateau pika in China. *Int J Parasitol*. 2005;35(6):693-701.
- Ogul H, Kantarci M. Primary Alveolar Echinococcosis Presenting with Simultaneous Cerebral and Craniofacial Involvement. *J Craniofac Surg*. 2018.
- Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev*. 2004;17(1):107-35.
- Salamone G, Licari L, Randisi B, Falco N, Tutino R, Vaglica A, et al. Uncommon localizations of hydatid cyst. Review of the literature. *Il Giornale di chirurgia*. 2016;37(4):180-5.
- Kvascevicus R, Lapteva O, Awar OA, Audronyte E, Neverauskiene L, Kvascevicene E, et al. Fatal Liver and Lung Alveolar Echinococcosis with Newly Developed Neurologic Symptoms due to the Brain Involvement. *Surg J (N Y)*. 2016;2(3):e83-e8.
- Kayacan SM, Vatansever S, Temiz S, Uslu B, Kayacan D, Akkaya V, et al. Alveolar echinococcosis localized in the liver, lung and brain. *Chin Med J (Engl)*. 2008;121(1):90-2.

13. El Saqui A, Aggouri M, Benzagmout M, Chakour K, El Faiz Chaoui M. Cerebral hydatid cysts in children: about 15 cases. *Pan Afr Med J.* 2017;26:205.
14. Adel F, Ramia JM, Gijon L, de la Plaza-Llamas R, Arteaga-Peralta V, Ramiro-Perez C. Extrahepatic and extrapulmonary hydatidosis. *Cir Cir.* 2017;85(2):121-6.
15. Cai H, Guan Y, Ma X, Wang L, Wang H, Su G, et al. Epidemiology of Echinococcosis Among Schoolchildren in Golog Tibetan Autonomous Prefecture, Qinghai, China. *Am J Trop Med Hyg.* 2017;96(3):674-9.
16. Ito A, Urbani C, Jiamin Q, Vuitton DA, Dongchuan Q, Heath DD, et al. Control of echinococcosis and cysticercosis: a public health challenge to international cooperation in China. *Acta Trop.* 2003;86(1):3-17.
17. Wang Q, Qiu J, Yang W, Schantz PM, Raoul F, Craig PS, et al. Socioeconomic and behavior risk factors of human alveolar echinococcosis in Tibetan communities in Sichuan, People's Republic of China. *Am J Trop Med Hyg.* 2006;74(5):856-62.
18. Grosso G, Gruttadauria S, Biondi A, Marventano S, Mistretta A. Worldwide epidemiology of liver hydatidosis including the Mediterranean area. *World J Gastroenterol.* 2012;18(13):1425-37.
19. Saqui AE, Aggouri M, Benzagmout M, Chakour K, Faizchaoui ME. [Hydatid cyst of the posterior fossa]. *Pan Afr Med J.* 2017;26:133.
20. Tefera E, Knapp J, Teodori M. Hydatid cyst of the interventricular septum. *Glob Cardiol Sci Pract.* 2017;2017(1):e201709.