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Case Report

A Case Report of Co-infection of Hepatic Cystic and Alveolar Echinococcosis

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Abstract

Pathogenesis of hepatic Echinococcosis is induced by parasitism of Echinococcus species. Generally speaking, there are two types of the disease-- hepatic cystic echinococcosis which is caused by *E. granulosus* and hepatic alveolar echinococcosis which is caused by *E. multilocularis*. Either of the two types is much clinically popular. However, simultaneous appearance of both the two types can be rarely seen. In this study, we reported a case of co-infection of both hepatic cystic and alveolar echinococcosis, aiming to offer experience in diagnosing and treating the co-infection by depicting the clinical characteristics, imaging manifestations and therapies.

Keywords: Hepatic Echinococcosis; Hepatic Cystic Echinococcosis; Hepatic Alveolar Echinococcosis; Co-infection

Introduction

Hydatid disease, also named as echinococcosis, is one of the 17 most easily neglected zoonoses proclaimed by the World Health Organization. It is caused by the larvae of Echinococcus [1]. *E. granulosus* and *E. multilocularis* are the two most important species causing hepatic echinococcosis. The former leads to cystic echinococcosis (CE) and the latter leads to alveolar echinococcosis (AE) [2]. In the epidemic areas, the incidence of CE can reach (1~200)/100 000 each year and the incidence of AE (0.03~1.2)/100 000 [3]. Both CE and AE mainly occur in the liver. Clinically, single illness of cystic echinococcosis or alveolar echinococcosis can be very prevalent, but there are few cases presenting co-infection of the above two. In this study, we reported a case of co-infection of hepatic cystic and alveolar echinococcosis, aiming to offer experience in diagnosing and treating the rare case by depicting the clinical characteristics, imaging manifestations and therapies.

Case Presentation

A 56-year-old Tibetan man complained of chronic epigastric pain for 3 months and jaundice for 2 months. The patient lives in Chenduo County, the area with the highest incidence of echinococcosis in China, and he had been in close contact with cattle, sheep and dogs. He was diagnosed with hypertension 3 years ago. Physical examination revealed jaundice and abdominal tenderness in the right upper quadrant. Laboratory and the liver function were abnormal on March 12th 2021 (Tables 1-3). CT scan suggested a cystic low-density mass in the right lobe of the liver, which was considered to be cystic echinococcosis, surrounding which there was a mixed density mass that was considered to be alveolar echinococcosis (Figure 1). Antibiotics were administered intravenously for the patient, along with liver function-improving medications, adequate nutritional support, symptomatic treatment and percutaneous transhepatic cholangial drainage (PTCD) to relieve intrahepatic cholestasis. Results of

the routine complete blood count (CBC) and liver function tests gradually became normal. On April 9th, we performed extended right hemihepatectomy, choledochotomy and placed a T-tube (Figure 2 and Figure 3). On April 20th, the patient had a sudden loss of consciousness with anuria. A large amount of bile-like fluid (420ml) was drained out through abdominal drainage-tube and no bile could be seen in the T-tube. Results of CBC, liver function, renal function and coagulation function tests became obviously abnormal (Tables 1-3). Bacterial culturing results of the drainage from the abdominal drainage-tube indicated stenotrophomonas maltophilia and enterococcus faecium. The patient was considered to complicate with septic shock, bile leakage, hepatic failure and renal dysfunction, meropenem was applied to strengthen anti-infective treatment, blood transfusion was additionally given along with adequate fluid infusion. The patient's condition gradually improved and results of the laboratory studies finally returned to normal levels. On May 9th 2021, the patient recovered well and was discharged from hospital.

Project name	Result		Unit	Reference range
	March 12th	April 20th		
White-cell count	14.22	109.73	109/L	3.5-9.5
Hemoglobin level	106	77	g/L	119-157
Platelet count	246	222	109/L	70-300
Neutrophils percentage	76.2	75.4	%	40-75

Table 1: Results of routine blood test.

Project name	Result		Unit	Reference range
	March 12th	April 20th		
Alanine aminotransferase	227	23	U/L	7-45
Aspartate aminotransferase	242	22	U/L	13-40
Total bilirubin	379.6	102.3	μmol/L	5.0-21
Direct bilirubin	199.5	63.0	μmol/L	0-3.4
Indirect bilirubin	180.1	39.3	μmol/L	1.3-18.7
Total protein	62.7	49.6	g/L	65.0-85.0
Albumin	25.2	27.3	g/L	40-55
Globuin	37.52	22.3	g/L	20-40
Cholinesterase	1829	944	U/L	5300-11300

Table 2: Results of liver function test.

Project name	Result		Unit	Reference range
	March 12th	April 20th		
prothrombin time	15.3	19.9	Sec	10.0-14.0
Prothrombin time percentage activity	57.7	39.4	%	70-175.7
International normalized ratio	1.29	1.70		0.84-1.40
Activated partial thromboplastin time	43.2	57.2	Sec	22.0-38.0
Activated partial thromboplastin ratio	1.44	1.9		0.73-1.27
Fibrinogen	5.27	1.86	g/L	2.0-4.0
Thrombin time	17.6	18.0	Sec	14.0-21.0
D-dimer	2.41	16.49	μg/mL	0.0-1.5
Fibrinogen degradation products	6.99	39.55	μg/mL	0.0-5.0

Table 3: Results of coagulation function.

Figure 1: The low-density shadow indicated by the pink arrow is cystic echinococcosis with multiple different sizes of subcysts inside, the high-density shadow indicated by the orange arrow is alveolar echinococcosis, which exhibited patchy calcification.

Figure 2: The egg indicated by the black arrow is daughter cyst; the tissue indicated by the orange arrow is alveolar echinococcosis tissue.

Figure 3. a: The pathological examination of surgical specimen manifested that alveolar cysts of different sizes (the site indicated by the arrow), with inflammatory cell infiltration, could be seen in the diseased tissue.

b: The pathological examination of surgical specimen manifested cystic echinococcosis (the site indicated by the arrow).(H&E,10×10).

Discussion

Hydatidosis is widely prevalent all over the world, especially in grazing areas. In China, hydatidosis is mainly distributed in the northwest and southwest of the country and it is reported that the average morbidity is 1.08% in the epidemic areas [4]. Risk factors for hepatic echinococcosis mainly include poor natural environment, close contact with cattle, sheep and dogs, economic underdevelopment in plateau areas, poor hygiene and lack of knowledge of echinococcosis transmission. However, co-infection of hepatic alveolar echinococcosis and hepatic cystic echinococcosis can be rarely seen. Echinococcosis is an animal-borne infectious disease and Echinococcus can complete its life cycle in only two kinds of mammals. Their adults (during which stage eggs can be produced) grow in the small intestine of canines (terminal hosts). The terminal host of *Echinococcus granulosus* is mainly dogs. Dogs are infected by ingesting hydatid cysts from the viscera of intermediate hosts (sheep, cattle, etc.) and the adults generally excrete eggs 45 days later since infection. The terminal hosts of *Echinococcus multilocularis* are mainly foxes and wolves. The intermediate hosts are small mammals such as voles. The adults generally excreted eggs 28 days later since infection. When humans take food contaminated by Echinococcosis eggs, the eggs can finally locate in the liver through the portal system to live on. According to clinical experience, the incidence of hepatic alveolar echinococcosis is higher in the high-altitude areas of Northwest China. That may be a result of the fact that the different terminal hostS--wolves and foxes are more prevalent in high-altitude areas where there are few human residents. Because of the obviously different distribution range of the two kinds of larvae of Echinococcus, the chance of co-infection can be definitely very low. In recent years, the cases of co-infection have gradually increased, that may have three reasons. Firstly, more and more screening work. Secondly, the range of activities of foxes and wolves becomes wider due to the improvement of environment. Thirdly, increased population mobility. Hepatectomy is one of the radical surgical methods for both hepatic cystic echinococcosis and hepatic alveolar echinococcosis. For the case in this study, we performed extended right hemihepatectomy according to the extent of the lesion. However, postoperative complications such as liver failure, septic shock and bile leakage occurred. We analyzed that in addition to hepatitis C, it was also associated with the more complicated condition led by the co-infection. In conclusion, although the co-infection of hepatic echinococcosis rarely occurs, its morbidity is increasing gradually with the improvement of natural environment and the increase of population mobility. Co-infection of hepatic alveolar echinococcosis and hepatic cystic echinococcosis can make the condition more complex and is more prone to induce serious complications, we have to pay attention to it and provide more individualized therapeutic strategy in clinical practice.

Conclusion

The diagnosis and treatment of co-infection of hepatic cystic and alveolar echinococcosis are significant to the study of this kind of patient.

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Conflict of Interest

The authors declare that they have no conflicts of interest.

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