Case Report: Surgical Treatment of Multiple Hydatid Cysts in the Liver of a Pediatric Patient

Hailong Lv,[†] Yufeng Jiang,^{*†} Guisheng Liu, Shijie Zhang, and Xinyu Peng^{*}

Department of Hepatobiliary Surgery, First Affiliated Hospital, School of Medicine, Shihezi University, Shihezi, Xinjiang Province, China;
Department of Histology and Embryology, Medical College, Shihezi University, Shihezi, Xinjiang Province, China

Abstract. Multiple hydatid cysts in the liver rarely occur in the pediatric population. Here, we present the case of a 16-year-old girl who presented with six hydatid cysts in the liver. The cysts were surgically removed and all found to be infertile. Interestingly, the patient had post-operative eosinophilia. From this experience, we conclude that individualized treatment is necessary for patients with multiple hydatid cysts.

INTRODUCTION

Human echinococcosis is a zoonotic infection caused by larval forms (metacestodes) of tapeworms of the genus *Echinococcus* found in the small intestine of carnivores. Although there are different species of *Echinococcus* described, only five of them—*E. granulosus*, *E. multilocularis*, *E. oligarthrus*, *E. vogeli*, and *E. shiquicus*—are formally recognized currently as taxonomically relevant. To distinguish the diseases caused by these different species, the World Health Organization (WHO) proposed the designation cystic echinococcosis (CE) for the disease caused by *E. granulosus*, alveolar echinococcosis (AE) for the disease caused by *E. multilocularis*, and polycystic echinococcosis (PE) for the disease caused by either *E. vogeli* or *E. oligarthrus*. No human cases caused by *E. shiquicus* have ever been observed, and only CE and AE are of public health significance. CE is the more prevalent form, and it occurs in countries in the Mediterranean, Middle East, eastern Europe, and Africa as well as Argentina, Chile, western China, Australia, and New Zealand.¹ The most frequently infected organ is the liver (55–70%) followed by the lungs (18–35%), and around one-fourth of all cases show multiple affected organs.^{2,3} Surgery involving removal of all cysts remains the main method for managing hydatid disease. Although the lesions generally appear grossly benign, establishing a definitive treatment plan is difficult because of vast differences in clinical manifestations among patients. Here, we report a case of multiple hydatid cysts in the liver that was successfully managed in our hospital.

CASE REPORT

A 16-year-old girl was referred to our unit in Xinjiang, China on January 24, 2014 because of a multiple cystic mass in the liver, which was identified on abdominal ultrasound during a routine check-up. She had no history of past illness and denied any contact history with pets or livestock. On physical examination, no icterus or tenderness was recognized in the right upper abdomen, and no mass was palpable. The patient had no allergic manifestations, such as urticarial rash and rhinitis. Laboratory examination on admission showed a

white blood cell (WBC) count of $6.5 \times 10^9/L$, eosinophilia of 0.1%, and normal values for hemoglobin, electrolyte profiles, and biochemical parameters, including liver enzymes and bilirubin. Urinary albumin was positive. Her tumor markers were within the normal limits. Upper abdominal computed tomography (CT) showed that the liver had multiple low-density lesions: the largest was a unilocular hepatic cyst with an 11-cm diameter in segments VI and VII, and five unilocular cysts were found in segments I, III, IV, VI, and VIII, suggesting liver hydatid cysts (Figure 1). The Casoni test yielded a positive reaction. The patient was scheduled for surgery and administered $15 \text{ mg kg}^{-1} \text{ d}^{-1}$ albendazole for 3 days pre-operatively.

With the patient under general anesthesia, a syringe was inserted into the largest unilocular hepatic cyst after liver exposure, and intracystic fluid was aspirated. The hydatid cyst fluid was faint yellow. Next, the cyst was opened, the internal capsule was removed, and 20% hypertonic saline was injected for 15 minutes (Figure 2A and B). Subtotal cystectomy was performed, because the cyst wall was adhered to the first hepatic portal vein.⁴ Intraoperatively, the remnant cyst wall was inspected for evidence of bile leaks, and visible biliary openings were sutured individually in healthy tissue. The cyst in the caudate lobe was found to communicate with the largest cyst, and its contents were removed completely. The remaining four hydatid cysts were completely resected—total cystectomy—through the subadventitial space. (Figure 2C–F).⁴ The hepatic hydatid cysts along the virtual space between the adventitia and the outer membrane were completely stripped. Intraoperative blood loss was 150 mL. Microscopically, no protoscoleces were observed in any cyst. On post-operative days 1, 3, and 8, the peripheral blood eosinophilic leukocyte counts were 29.5%, 35.4%, and 50.53%, respectively (Figure 3A). At 1 and 3 months post-operatively, the serum eosinophil count had decreased to 25.3% and 12.3%, respectively. Hydatid disease was confirmed by pathological examination of the cyst tissue (Figure 3B). The patient recovered well from the operation, and the post-operative course was uneventful. Ultrasonographic follow-up was conducted for 3 months, and no evidence of recurrence was found.

DISCUSSION

Hydatid disease is a serious health problem worldwide, and clinical presentation varies according to the anatomic location of the cyst. Although most hydatid infections are acquired in childhood, they can remain latent for 5–20 years. Furthermore, hydatid cysts are usually asymptomatic and diagnosed incidentally or when various complications occur. In such cases, patients experience no discomfort before diagnosis.

^{*} Address correspondence to Yufeng Jiang, Department of Histology and Embryology, Medical College, Shihezi University, Shihezi, Xinjiang Province 832008, China; or Xinyu Peng, Department of Hepatobiliary Surgery, First Affiliated Hospital, School of Medicine, Shihezi University, Shihezi, Xinjiang Province 832008, China. E-mails: yufengjiang03@126.com or pengxinyu2000@sina.com

[†] These authors contributed equally.

FIGURE 1. Computerized tomographic scan showing multiple liver cysts. (A) Hydatid cysts in liver segments VII, VIII. (B-C) Hydatid cysts in liver segments I, III, VI, VII. (D) Hydatid cysts in liver segments IV.

Most hydatid cysts found in adults are hepatic, although some are seen in the lungs.⁵ Studies on hydatid disease in children have recorded varying incidences of cysts in the lungs and liver.⁶⁻⁸ However, multiple liver cysts are relatively rare in the pediatric population.

The optimal treatment of liver hydatid cysts remains controversial^{9,10} because of the various treatment options available: namely, surgery, percutaneous aspiration, injection and

reaspiration, drug treatment, and the watch-and-wait approach. Nonetheless, surgical excision is the only potentially curative treatment of hydatid cysts. The surgical techniques can be categorized as radical or conservative. Radical methods involve total excision of the cyst, whereas conservative methods involve removal of the cyst contents and inactivation of protoscolices or removal of the parasite only (hydatidectomy) and partial cystectomy. Some surgeons favor subadventitial cystectomy,

FIGURE 2. (A and B) Subadventitial subtotal cystectomy was performed for the hydatid cyst in liver segments VI and VII. (C-F) The hydatid cysts in liver segments III, IV, VI, and VIII were completely resected.

FIGURE 3. (A) Eosinophils in a peripheral blood smear examined on post-operative day 8 (arrows). (B) Photomicrograph of a histopathologic section showing liver tissue and a laminated layer of the hydatid cyst (hematoxylin and eosin).

pericystectomy, or even hepatectomy, if required, whereas others, especially those in endemic areas, prefer conservative surgery. Treatment of liver hydatid cysts is very difficult for many reasons, but mainly because the disease characteristics and clinical manifestations differ widely among patients. For complex diseases like this one, a one size fits all approach is not suitable, and clinicians must adopt a patient-specific approach to treatment.¹¹

Because of the threat posed by hydatid disease on human health, it is crucial to devise methods for early diagnosis and promote individualized treatment plans. Treatment should be administered according to the specific conditions presented by the patient. In this case, we selected different forms of surgical excision of the liver hydatid cysts. We have performed subadventitial exocystectomy procedures for most cases of liver hydatid disease in our centers. This procedure consists of two approaches: subadventitial total (radical procedure) and subtotal (conservative procedure) exocystectomy. In subadventitial total exocystectomy, sectioning of the liver parenchyma began at the border between the liver parenchyma and the cyst surface. During dissection, careful identification of the potential space between the pericyst and adventitia is important, and the hydatid cysts can be completely separated from the liver. In subadventitial subtotal exocystectomy treatment, a small portion of the ectocyst may remain adjacent to the junction of the vena cava or upper hepatic vein.⁴ To reduce post-operative recurrence and residual cavity complications, radical surgery was conducted for four of six unilocular cysts, and the largest hepatic cyst, which was in segments VI and VII, was treated with conservative surgery because of its proximity to the first hepatic portal vein. The last cyst in the caudate lobe was also treated conservatively. The treatment option selected should be based on cyst stage, size and location, observed complications, clinical experience, and comorbidities.

It is noteworthy that cysts in this case were infertile and that the patient had eosinophilia. Fertile hydatid cysts are formed in intermediate hosts and produce protoscoleces. Bovines generally have infertile cysts, whereas infected sheep generally have fertile ones.¹² To our knowledge, no literature is available on the fertility rates of hydatid cysts in humans. In

this case, no protoscoleces were observed microscopically in any cyst. For unknown reasons, some cysts are infertile and do not produce protoscoleces. Some researchers have hypothesized that, when the oxidative damage to DNA in the germinal layers exceeds the capability of DNA repair mechanisms, apoptosis is triggered, and hydatid cysts become infertile.^{13,14} As one of the interfaces between the host and the parasite, the proteins present in the hydatid cyst fluid are likely to reflect differences between fertile and infertile cysts.¹⁵ Cyst infertility may also be the result of the gradual build-up of the patient's immune function against the hydatid infection.

Eosinophilic granulocytes normally represent a small percentage of the leukocytes in blood, although they are much more abundant in the tissues. These cells can destroy parasites and also have immune regulatory functions.¹⁶ Observation of eosinophilia in peripheral blood smears may support a diagnosis of parasitic infection, especially in case of diagnostic dilemma. However, eosinophilia is not usually observed in patients with hydatid disease. In most cases, it is either mild (< 15%) or absent, although in patients with ruptured cysts in the biliary tree, eosinophilia is often marked and shows transient elevation (up to 60%).¹⁷ In this study, eosinophil count was normal before the surgery, but it increased prominently after the surgery. A reason for this could be that some rupture occurred during surgical removal of the hydatid cysts.

In conclusion, this case report validates the diversity and complexity of the clinical manifestations of liver hydatid cysts. We adopted radical surgery whenever possible, although a conservative approach was required in some cases. Although surgical resection is the best curative procedure, hydatid cyst management must be individualized for each patient. The most important factor for successful management is intact excision of the cysts.

Received July 16, 2014. Accepted for publication November 19, 2014.

Published online January 5, 2015.

Financial support: This case report was supported by National Natural Science Foundation of China Grants U1303121 and 81360410.

Authors' addresses: Hailong Lv, Guisheng Liu, Shijie Zhang, and Xinyu Peng, Department of Hepatobiliary Surgery, First Affiliated

Hospital, School of Medicine, Shihezi University, Shihezi, Xinjiang, China, E-mails: lvhailong0993@126.com, woweiyikuang@sina.cn, zhangshijie@126.com, and pengxinyu2000@sina.com. Yufeng Jiang, Department of Histology and Embryology, Medical College, Shihezi University, Shihezi, Xinjiang, China, E-mail: yufengjiang03@126.com.

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