



## ***Simultaneous Presence of Hydatid Cysts in the Liver and Spleen: A Case Report with Splenectomy (Running title: Simultaneous Hydatid Cysts of Liver and Spleen)***

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### **ARTICLE INFO**

### **ABSTRACT**

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**Introduction:** Cystic hydatidosis is a major infectious disease associated with severe public health issues in many countries, including Iran. The liver is the most frequently involved organ in this disease. However, cysts may also develop in other organs, such as the spleen. According to a general report, splenic cysts are rare and unusual.

**Case Presentation:** We presented the case of a patient with simultaneous liver and spleen cystic hydatidosis in Maneh and Samalghan city, located in North Khorasan province, Iran. Diagnosis was confirmed by CT-scan and abdominal ultrasonography. The patient underwent successful laparoscopic surgery and splenectomy. Pathological examinations revealed cystic hydatidosis.

**Conclusion:** Due to the possible involvement of the liver and other sensitive organs (e.g., spleen) in endemic and hyperendemic areas, early diagnosis and treatment of cystic hydatidosis could be effective in reducing the incidence of this parasitic infection.

## Introduction

Echinococcosis or hydatid cyst is a common zoonotic disease across the world. Human infection is caused by the cystic larval stage of *Echinococcus granulosus* (*E. granulosus*), which is transmitted to human via contaminated food with the eggs of adult worms in dog feces (1-4). Canines (dogs, wolves, foxes, and jackals) are recognized as the final hosts of this parasite and are infected by contaminated visceral parts, eventually discarding the parasite eggs through their feces (2, 5).

Echinococcosis is prevalent worldwide, while the highest levels of contamination have been reported in the Mediterranean, southern and central regions in Russia, the Middle East, South America, Central Asia, Australia, China, and North and East Africa. The World Health Organization (WHO) has classified echinococcosis as the first global food-borne disease (6, 7). According to the data released by the WHO, Iran is one of the hyperendemic areas of the disease due to the high rate of exposure to livestock (as the intermediate hosts) in dogs. The incidence of the disease in the intermediate host and prevalence of hydatid cyst have been reported to be 11.6-34.6% and 13.1-1.3% in different regions in Iran, respectively (5).

The clinical symptoms and incubation period of echinococcosis may vary depending on the involved organs, cyst size, and genotype of the parasite (2, 8). Most of the organs in the body, including the liver, lungs, brain, spleen, eyes, and bones, could involve in specific cystic injuries, which cause severe risk and distress in the absence of timely diagnosis and treatment (2, 9).

Liver hydatid cysts lead to liver enlargement, upper abdominal pain, and nausea and vomiting. Larger cysts are often palpable in the abdomen (10). The initial involvement of the spleen mostly occurs after the liver and lungs. Splenic cysts may be solitary or simultaneously affect other organs, such as the liver. In addition, these cysts are usually asymptomatic, and surgery and splenectomy are required in the cases of large cysts (11, 12).

In this paper, we presented a very rare case of simultaneous liver and spleen hydatid cysts, which led to splenectomy.

## Case Presentation

The patient was a 46-year-old housewife who lived in Maneh and Samalghan city, located in North Khorasan province in the northeast of Iran (Figure 1). She referred to Imam Ali Hospital in Bojnourd, North Khorasan, with the symptoms of abdominal pain and nausea and vomiting, which has started in the past 24 hours.

Epigastric pain was evident in the preliminary examination. Abdominal ultrasonography showed a cyst (diameters: 39×48 mm) in the right lobe of the liver. The volume and thickness of the gallbladder and bile ducts were normal. Spiral CT-scan was performed on the patient, and two cysts were detected in the right lobe of the liver, as well as one cyst in the spleen (Figure 2). No free fluid was observed in the abdominal cavity.

In the laboratory investigation, hemoglobin level was 9.93 g/dL, and white blood cell count was 9,500 / $\mu$ L (76% neutrophil, 15% lymphocyte, 4% monocyte, and 5% eosinophil). Liver function tests (alanine aminotransferase, aspartate aminotransferase, amylase, and alkaline phosphatase) were within the normal

ranges. Further evaluation was carried out via abdominal ultrasonography and CT-scan, which revealed simultaneous hydatid cysts in the liver and spleen of the patient.

For the diagnosis of the hydatid cysts, the patient was subjected to laparotomy using the chevron incision (double-sided cutting below the ribs). During the surgery, we observed a rupture of the liver hydatid cyst with severe adhesion to the right diaphragm (Figure 3A). After embedding hypertonic saline gases around the cysts, the adhesions were freed from the diaphragm, and the cyst containing a large number of daughter cysts, hydatid sand, and its typical membranes was removed completely. Finally, a Penrose drain was inserted into the removed cysts, and the cavity was completely closed.

Since the patient was a candidate for splenectomy, after embedding hypertonic saline gases and taking the necessary precautions, the splenocolic, splenophrenic, splenogastric, and splenorenal ligaments were removed, and the spleen was directed upwards. After the ligation of the short gastric arteries and splenic arteries, the cystic spleen was removed completely (13) (Figure 3B).

After controlling the homeostasis and insertion of the drain into the abdomen, the operation was completed. Recovery was satisfactory within the prescribed time frame, and the patient was discharged.

It is notable that before the surgery, the patient received a dose of pneumococcal vaccine and prophylaxis. Moreover, a sample was sent to the laboratory for final confirmation through pathology examination, and the diagnosis of hydatid cyst was confirmed in both the organs (Figure 4).

Due to rupture of the cyst before surgery and in order to prevent infection recurrence, the patient was immediately prescribed with albendazole (400 mg/kg/day) for six months. Eight months after the infection, the physical examination for hydatid cyst, medical laboratory investigations, and CT-scan were repeated. The patient was normal and well with no evidence of hydatid cyst involvement. Written consent was obtained from the patient.

### **Discussion**

In the present study, we described a rare case of simultaneous hydatid cysts in the liver and spleen. Due to the space-occupying cyst, the spleen was completely removed. After the surgery, the patient was thoroughly treated, and despite the rupture of the cyst before and during the operation, relapse of the parasitic infection was not observed.

Hydatidosis is a zoonotic parasitic infection and a significant health issue caused by the larval stage of the *E. granulosus*. This infection is widespread globally and highly common in farming livestock areas, such as the Middle East (Iran), North Africa, New Zealand, Australia, and South America (14-16). The incubation period of the disease may vary, ranging from a few months to a few years; the growth rate also varies depending on individuals and regions (2). Since the adult worm lives in the intestinal tract of dogs, its eggs are excreted through the feces and released into the environment. In endemic areas where infected and untreated dogs live close to humans, it is highly likely that the eggs cause human infection through food (e.g., vegetables). After causing infection in humans, the eggs in the larval stage become the hydatid

cysts that cause various symptoms and complications (2).

Hydatid cysts are diagnosed by imaging modalities, especially ultrasound, CT-scan, magnetic resonance imaging (MRI), and simple radiology. Diagnosis is confirmed by finding special antibodies in the blood serum. Evidently, the screening and diagnosis of hydatid cysts in human are practical and convenient by serological tests; unfortunately, these methods are not viable in some endemic areas, such as Iran (17).

The presence of hydatid cysts may vary in different organs. More than 90% of hydatid cysts are detected in the liver, lungs or both, and the spleen is the third most common location with the frequency of 2.5-5.5% (11, 14, 18). Splenic cysts are often asymptomatic, and patients may have no complaints; these cysts are accidentally identified in radiological examinations (11). Combination of laparotomy and total splenectomy is a selective method for the treatment of splenic cysts (14, 15). Differentiation from other cysts is the major issue in the diagnosis of splenic hydatid cysts. However, the disease should be considered in endemic areas, and serological and pathological tests should be performed (15).

In this report, the patient referred to the hospital with epigastric pain and became a candidate for surgery since cysts were detected in the imaging of the spleen and liver. Through laparotomy, the observation of a large hydatid cyst in the spleen made total splenectomy obligatory. Due to the risk of cyst rupture, postoperative complications, and excessive hemorrhage, removal of cysts or partial splenectomy are not recommended for very large cysts. In the present study, since the

patient had a very large cyst in the umbilical region of the spleen, we avoided cyst removal or partial splenectomy and completely removed the spleen as the optimal therapeutic choice (19) (Figure 3).

During the surgery, the liver cyst had a mild rupture due to adhesion to the diaphragm (Figure 3), which could have increased the risk of anaphylactic shock in the patient. Therefore, all the internal components of the cysts were removed immediately and completely. No anaphylactic shock occurred in the patient.

Reports in this regard have indicated that the rupture of the cyst could increase the risk of the distribution of parasites at the larval stage to different areas of the body (20). As such, in order to prevent the release of the cyst with mild rupture in the patient, anti-parasitic treatment with albendazole was initiated immediately. Recurrence or relapse of infection with a new cyst was not reported in the patients six months after the surgery.

According to the results of this case report, hyperendemic diseases are characterized by a cycle of infection between livestock, dogs, and humans, especially in countries such as Iran. Patients presenting with epigastric symptoms should refer to health centers, and the hydatid cyst should be examined meticulously. Early diagnosis of hydatid cysts by simple techniques, such as the combination of routine clinical history and CT scan, is vital to preventing the severe complications of the infection.

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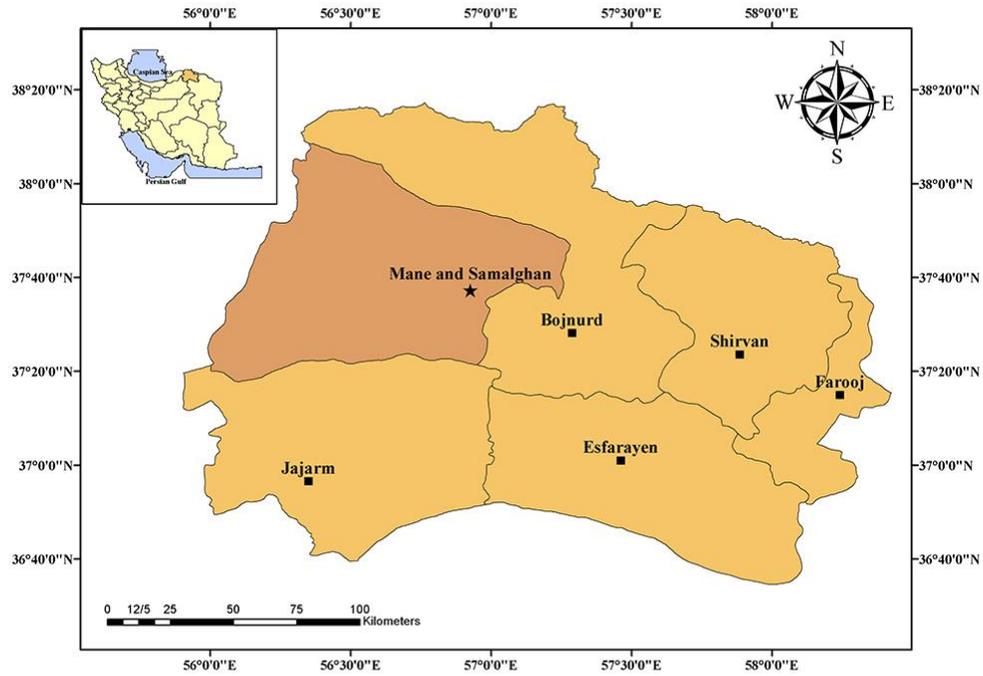
**Conflicts of interest:** None declared.

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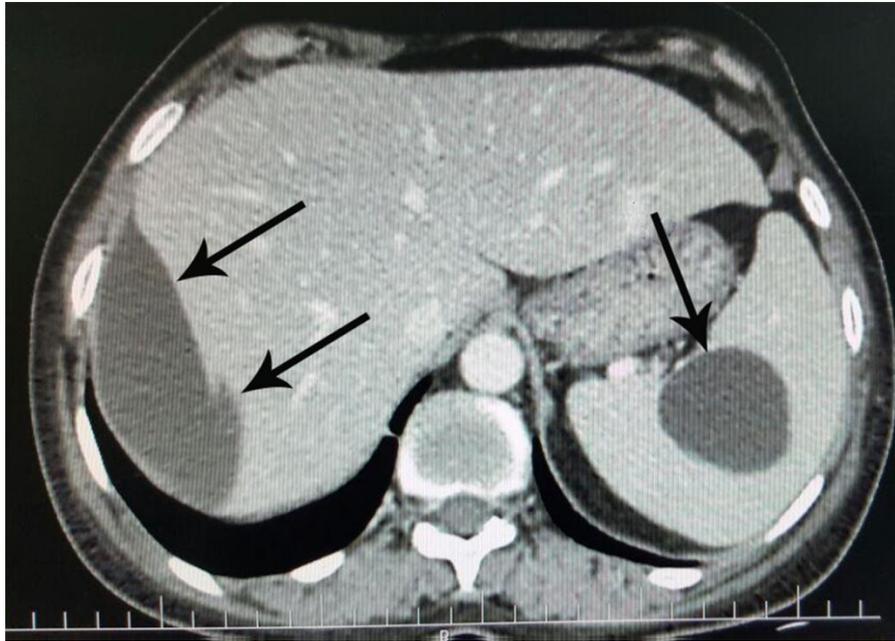
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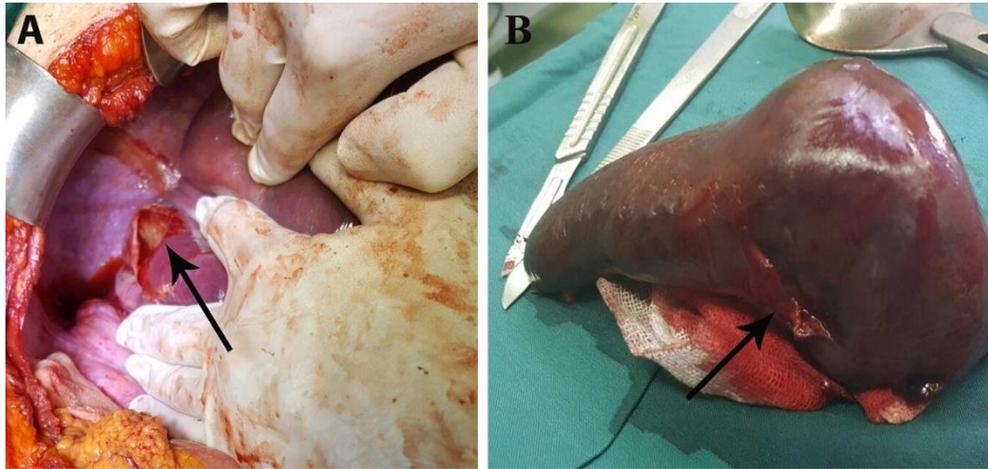
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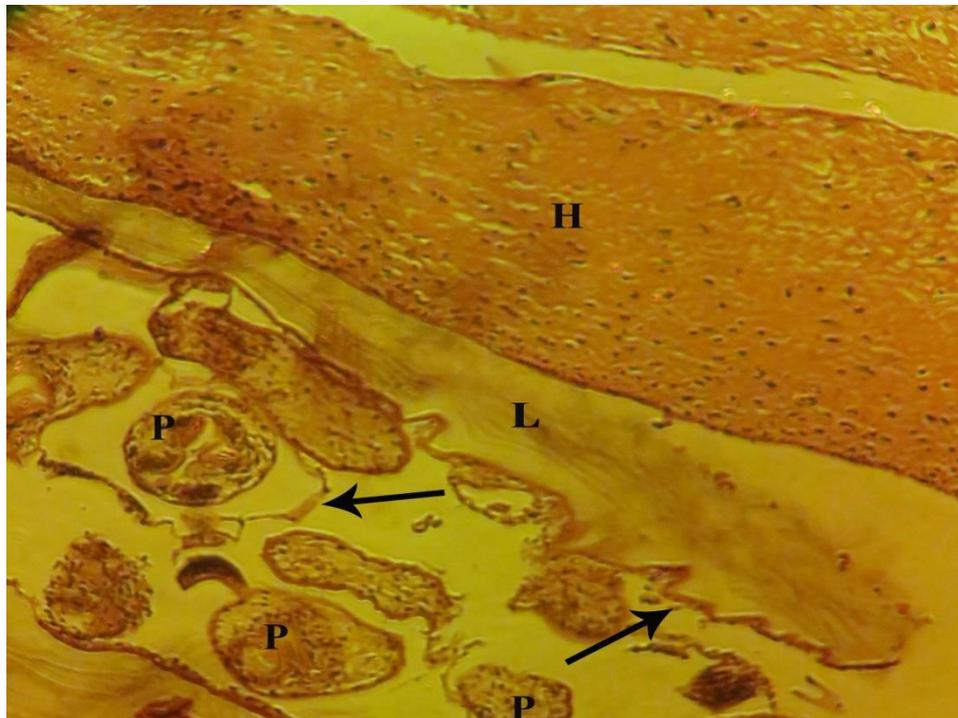
**Figure 1.** Map of Iran Showing Location of Maneh and Samalghan City in North Khorasan Province, Northeast of Iran (created by Arc GIS version 10.2).



**Figure 2.** Abdominal Spiral CT-scan Findings (two cysts were detected in the right lobe of the liver and one cyst was detected in the spleen [black arrows] with no free fluid in the abdominal cavity)



**Figure 3.** Adhesion of Liver (A) and Spleen (B) Cysts to Diaphragm (showing location of cyst in umbilical region of spleen)



**Figure 4.** Microscopic Section of Hydatid Cyst in Liver (H: host hepatic cells, L: laminated layers of cyst wall, P: protoscolices located inside brood capsule, arrows: germinal layers of cyst wall [hematoxylin and eosin stain, 40X])