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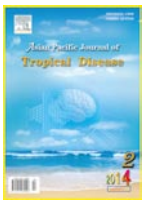

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# A case of disseminated hydatid disease by surgery involving multiple organs

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ABSTRACT

Hydatid disease is the most common parasitic infection in the world, and is caused by the parasite *Echinococcus granulosus*. The most common site of this disease is the liver (75%), followed by the lungs, kidney, bones, and brain. Multiple abdominal organ and peritoneal involvement can also be seen in some cases. The dissemination of hydatid cyst disease can develop spontaneously or secondary to trauma or surgery. Here, we present the case of a 69-year-old man with multiple cyst hydatidosis, who underwent surgery for acute appendicitis approximately 20 years previously. Computed tomography of the abdomen shows the multiple active and inactive cystic lesions in the liver, spleen, right kidney, and mesentery. This patient required surgery several times, as well as medical treatment, after the rupture of a mesenteric hydatid cyst during the appendectomy. Combined anthelmintic treatment was recommended to the patient who refused further surgical treatment.

## 1. Introduction

Echinococcosis is especially endemic to the Mediterranean Region, Australia, the Middle East, Turkey, Africa, and South America, and is the most common zoonotic infection in the world. Echinococcosis in humans is caused by the larval stages of cestode species of the genus *Echinococcus*, predominantly by *Echinococcus granulosus*[1]. These cysts are often seen as a single cyst in humans, although multiple cysts or multiple organ involvement has been observed. Echinococcosis can involve any organ in the human body[2]. The liver (50%-70%) is the most common site of hydatid cysts, followed by the lungs (20%-30%)[3], kidney (2%-3%), brain (1%-2%), spine (1%), and eyes (0.2%)[4]. Occasionally, heart, thyroid, spleen, pancreas, and muscle involvement may be seen[3,5]. Secondary peritoneal involvement can occur due to the spontaneous rupture of a hydatid cyst in the liver or spleen, or accidental spillage during surgery. Primary peritoneal involvement is very rare, and constitutes less than 0.5% of all intra-abdominal hydatidosis[1]. Here, we present an unusual case of multiple cystic hydatidosis with radiological findings.

## 2. Case report

A 69-year-old man presented to the hospital with abdominal pain and tenderness, and had a known medical history of cystic hydatidosis. Abdominal distention and tenderness were noted upon physical examination; however, the laboratory tests were normal, with the exception of a high erythrocyte sedimentation rate. He had a medical history of acute appendicitis surgery about 20 years previously. The patient had several surgeries for the multiple cyst hydatidosis. All of the prior surgeries were elective except an emergent surgery due the rupture

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of the mesenteric cyst hydatid during appendectomy. The patient subsequently received long-term treatment of albendazole and praziquantel after the surgeries.

Plain X-ray films of the abdomen revealed calcified foci in the right and left upper and lower quadrants. Abdominopelvic computed tomography (CT) with intravenous and oral contrast agents was performed, and multiple cystic lesions were seen in the liver, spleen, and mesenteric fatty tissue (Figures 1-3). Some of these lesions contained a thin rim of calcification in the walls of the hydatid cyst. Two giant lesions with daughter cysts were seen, with the largest diameters of 124 mm×120 mm in the liver and 120 mm×130 mm in the spleen. Additionally, the CT revealed multilocular cystic lesions (the largest measuring 48 mm×55 mm) adjacent to the aorta and gastric fundus, and distinct imprinting to the gastric cardia and fundus were seen (Figure 4). The presence of rim calcification and daughter cysts primarily showed Gharbi type 3 and 5 hydatid cysts. The thoracic spine CT and all bone structures within the field of view were normal, with the exception of degenerative change.

The result of the serum antibody test for hydatid cysts was positive, and the patient was diagnosed with multiple cystic hydatidosis based on the CT findings and positive serum antibody test. The patient did not accept any new elective surgery, so a prolonged course of albendazole and praziquantel therapy is suggested for the patient. The follow-up time was 1 year. No spontaneous rupture was developed. The patient was advised to continue follow-ups with radiologic examinations at determined intervals in order to screen for possible future complications.

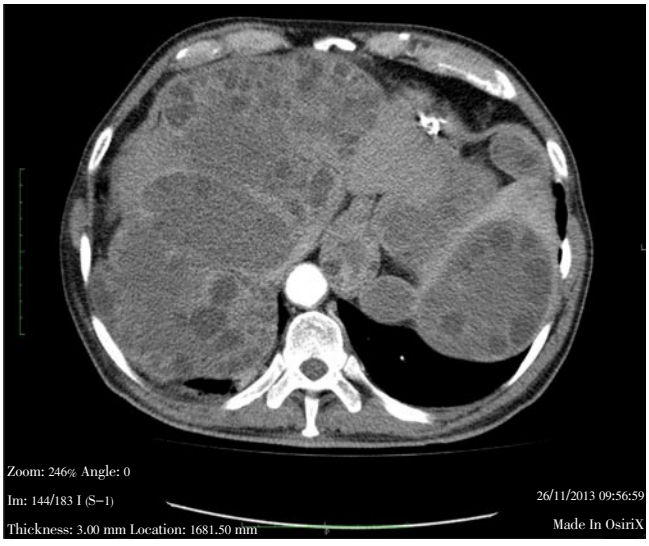


**Figure 1.** Axial contrast enhanced CT of the upper abdomen.
It shows multiple hydatid cysts with multiple daughter cysts (CE3B) in the liver. Also, large hydatid cysts with multiple daughter cysts were demonstrated in the spleen and paraaortic area.

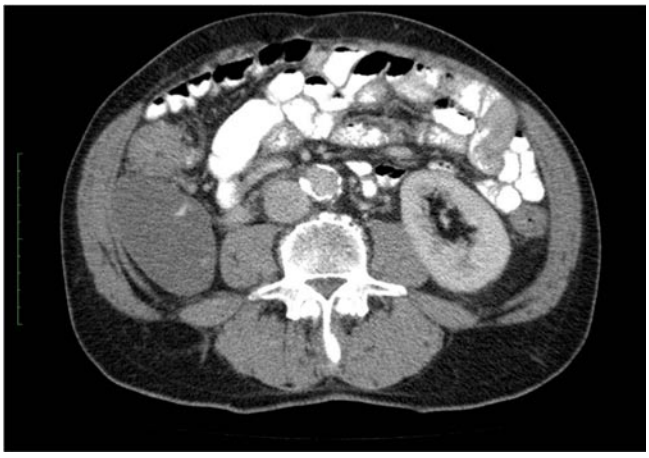

**Figure 2.** Axial contrast enhanced CT of the upper abdomen at the level of the left kidney.
It shows a hydatid cyst (CE1) adjacent to the ascending colon, with the largest diameter of 81 mm×52 mm.

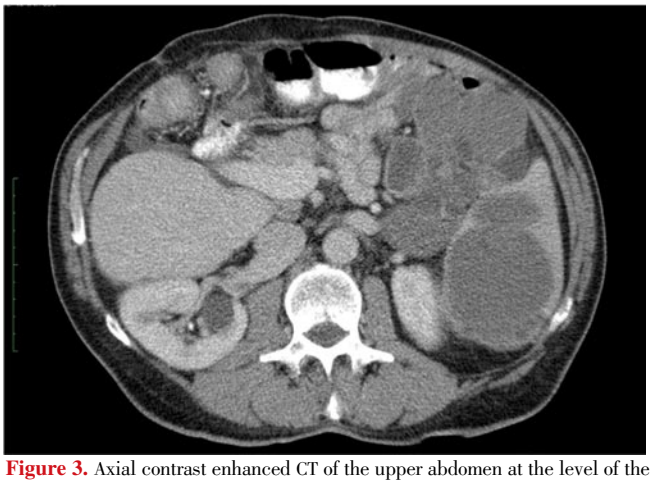

**Figure 3.** Axial contrast enhanced CT of the upper abdomen at the level of the right kidney.
It shows multiple hydatid cysts (CE1) without daughter cysts in the mesenteric fatty tissue. Also, a CE1 cystic lesion was demonstrated, with the largest diameter of 20 mm×21 mm in the cortex of the right kidney.

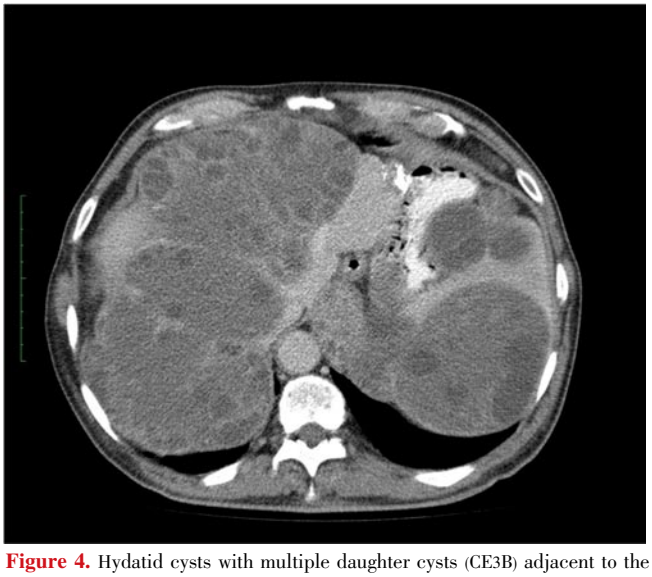

**Figure 4.** Hydatid cysts with multiple daughter cysts (CE3B) adjacent to the aorta and gastric fundus.
Compression and deviation of the gastric fundus by the paraaortic cystic lesions were revealed by CT. Also, compression of the gastric corpus by the CE3B hydatid cyst in the spleen, with the largest diameter of 48 mm×55 mm, was demonstrated by CT.

## 3. Discussion

Hydatid cysts are the most common zoonotic infection in the world, and are caused by the larvae of cestodes from the genus *Echinococcus*. Currently, there are 4 infectious species of this genus known in humans: *Echinococcus granulosus* (cystic echinococcosis), *Echinococcus multilocularis* (alveolar echinococcosis), *Echinococcus vogeli*, and *Echinococcus oligarthrus* (which causes polycystic hydatid disease). Humans become infected through contact with definitive hosts like dogs or other canids, or by consuming contaminated foods containing cestode eggs[1]. After ingestion, liberated eggs travel across the intestinal wall and reach the liver through the portal vein system. Embryos cannot be trapped in the liver and spread to other organs via the systemic circulation[4]. The liver (75%) and lungs (15%) are the most common sites for hydatid cysts.

Disseminated peritoneal hydatidosis has been reported in the literature secondary to the rupture of hepatic or splenic cysts, due to blunt force abdominal trauma or accidental spillage during surgery[6,7]. Although dissemination can develop secondary to trauma or surgery, Thambidurai *et al.* reported spontaneous dissemination of hydatid cyst disease[8]. The spread of hydatid disease can be associated with the lymphatic or systemic circulation[8]. In our case, there was a history of the rupture of a mesenteric cystic lesion during surgery; therefore, it could have been disseminated via surgery to all of the intra-abdominal organs and mesenteric fatty tissue.

Patients may remain asymptomatic for many years. Symptoms of pelvic hydatidosis are usually late onset, and can be associated with the compression of the adjacent organs (rectum, bladder). Rarely, obstructive uropathy, renal failure, allergic reactions, or secondary bacterial infections can be seen[1].

A pancreatic cyst, mesenteric cyst, gastrointestinal duplication cyst, ovarian cyst, lymphangioma, intra-abdominal abscess, or cystic tumor originating from the spleen or liver should be considered for the differential diagnosis of a hydatid cyst. Patients residing in a rural area, and feeding pets with intra-abdominal cystic lesions, should also be kept in mind for the diagnosis of a hydatid cyst[1].

Laboratory findings can be of help in the diagnosis. Eosinophilia can be monitored by a complete blood count test; however, liver and renal function tests are usually normal. Additionally, serological tests like ELISA, indirect hemagglutination, and immunoelectrophoresis can be very useful for the diagnosis. Immunoelectrophoresis is more sensitive than ELISA for anti-hydatid antibodies, but ELISA is more specific. Also, ELISA can be used for follow-up testing for recurrence in the postoperative period[1].

Hydatid cysts have been classified as active cysts (CE1 and 2), transitional cysts (CE3), and inactive cysts (CE4 and 5) by the World Health Organization Informal Working Group (WHO-IWGE)[9]. Multiple cysts or septae are present in the active cysts. WHO type CE1 (active cyst) can be seen as a unilocular anechoic lesion with a double line sign. WHO type CE2 can be seen as multivesicular or multiseptated cysts. The presence of daughter cysts is indicated by a "rosette like" or "honeycomb like" structure upon ultrasonography (US).

The transitional phase of the parent cyst (CE3) includes daughter cysts, hydatid sand, and debris. Cysts with detached membranes (water lily sign) (CE3A) and cysts with daughter cysts in a solid matrix (CE3B) can be shown by US. Cysts can be seen as echogenic in the inactive phase (CE4 and 5), where they appear to be partial or completely collapsed. The CE4 phase cannot include daughter cysts, and can be seen as a heterogeneous hypo/hyperechoic cyst by US. CE5 cysts are characterized by a solid lesion with a calcified wall[8,9].

Most often, Gharbi *et al.*[10] and WHO-IWGE are used for the classification of the hydatid cyst; although, the Gharbi classification is almost the same as the WHO classification. There were two significant additions to the WHO-IWGE in 2001. Gharbi type 2 corresponds to the CE3A in the WHO-IWGE 2001 classification, and according to the WHO-IWGE, Gharbi type 3 is divided into CE2 and CE3B[11]. In our case, the liver, spleen, and mesenteric cysts were defined as CE2 and CE5 according to the WHO-IWGE. Also, CE2 lesions were seen near the aorta, which were compressing the gastric cardia.

US and CT are very helpful for the diagnosis of hydatid cysts. US is the first choice imaging modality for the diagnosis, with a sensitivity of approximately 90%-95%. However, CT is quite sensitive (>95%) when compared to sonography for the identification of hydatid cysts. In contrast enhanced CT examinations, these cysts are observed as well-defined, rounded lesions with low attenuation, without evidence of contrast enhancement[1]. Eggshell calcification of the cyst and diaphragmatic elevation due to the mass effect can be observed with direct X-ray films of the abdomen[1]. In our case, multiple

well-defined, rounded lesions with low attenuation were seen in the liver, spleen, right kidney, mesenteric fatty tissue, and paraaortic locations with contrast enhanced CT, without contrast enhancement. Some of these lesions showed eggshell or rim calcifications. A low attenuation cystic lesion with daughter cysts and rim calcification without contrast enhancement, strongly suggests the diagnosis of a hydatid cyst. In addition, the determination of anti-*Echinococcus* IgG antibodies using ELISA, in a patient with positive radiological findings, supports this diagnosis.

The best method of the treatment is the total excision of the lesion, especially for large cysts and hydatidosis. However, this is not always possible because treatment varies according to the patient[1]. Partial excision of the pericyst with or without omentoplasty is usually the preferred surgery because it has a better clinical response. In fact, total excision is preferred for non-hepatic lesions which are settled in the omentum or free in the peritoneum[5]. Treatment with albendazole is not an alternative to surgery. It is used as an adjuvant for the prevention of recurrence or suppression of the subcentimeter lesions[3].

A multidisciplinary approach is recommended for disseminated echinococcosis. Surgery with albendazole or percutaneous aspiration-injection-reaspiration (puncture, aspiration, injection of scolicidal agent, respiration) with albendazole is preferred for the treatment of disseminated echinococcus[3]. Also, a combination of albendazole/praziquantel treatment can be used as an alternative treatment for disseminated echinococcus or inoperable patients[3,5]. Albendazole is an anthelmintic agent, which can be used for peritoneal hydatid cysts or inoperable patients, and praziquantel is an anthelmintic that is a potent protoscolicidal agent. The treatment of hydatid disease with only albendazole or surgery has shown high recurrence rates. Therefore, continuing medical treatment for at least three months is recommended after surgery, as a multidisciplinary approach[3]. In our case, the patient had already had a several surgeries for cystic hydatidosis without complete curation; therefore, the patient refused additional surgery. So, albendazole with praziquantel was prescribed for the patient, and he was advised to follow-up with radiological examinations to prevent possible complications.

In this case, multiple cystic lesions with or without wall calcification were seen in the liver, spleen, and mesenteric fatty tissue. Primarily, intra-abdominal space-occupying lesions are considered for the differential diagnosis. However, disseminated cystic hydatidosis was determined when the typical radiological findings were correlated with the immunological tests. Early diagnosis and treatment of hydatid cysts is very important for the prevention of severe complications.

## Conflict of interest statement

We declare that we have no conflict of interest.

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