Govaresh/ Vol. 27, No.2, Summer 2022; 118-122.

# An Unusual Presentation of Massive Hydatid Cyst of Liver in a 39-year-old Female Patient

**Amir Mohammad Salehi [1], Hossain Salehi[2], Amin Akhlaghi[1] ,Faeze Foroughi-Parvar[3,*]**

[1] Medical student, School of Medicine, Hamadan University of Medical Sciences, Hamadan, Iran
[2] Gastroenterology Ward, Baharloo Hospital, Tehran University of Medical Sciences, Tehran, Iran
[3] Department of Medical Parasitology and Mycology, School of Medicine, Hamadan University of Medical Sciences, Iran.

Case Report

## ABSTRACT

Hydatidosis, a zoonotic infection of the larval stage of Echinococcus granulosus, has a worldwide distribution. Due to the late onset of clinical manifestation, the cyst can involve the greater part of the tissue. This case study was performed on a massive hydatid cyst (HC) (85×115mm), reported in a 39-year-old woman. She was referred to the clinic with right upper quadrant pain and gastric pressure. Magnetic resonance imaging (MRI) revealed that the vast space of the liver had been occupied by the massive cyst in the right lobe of the liver. Routine liver assays were normal. IgG enzyme-linked immunoassay (ELISA) test using antibody was negative (0.19 mg/dL). Resection laparoscopic surgery was scheduled for the patient due to severe pain. The massive HC was confirmed in microscopic examination of the cyst.

**Keywords:** Echinococcosis; Surgery; Hydatid cyst

*please cite this paper as:*
Salehi AM, Salehi H, Akhlaghi A, Foroughi-Parvar F. An Unusual Presentation of Massive Hydatid Cyst in Liver of a 39-Year-Old Female Patient. *Govaresh* 2022;27:118-122.

## INTRODUCTION

Hydatidosis in humans as an accidental intermediate host occurs as a result of the localization of the larval (metacestode) stage of the Echinococcus granolusus(1). Although E. granolusus is regarded as a worldwide distributed infection, Eastern Europe, the Mediterranean region, Africa, Australia, and South America report a high frequency of hydatidosis(2). Hydatid cyst (HC) predominantly involves the liver (70%) followed by the lung (20%), while 10% of cysts can develop in other organs such as the brain, body musculature, heart wall, kidney, eye, and marrow cavity of bones(3). Depending on the site of the cyst, the clinical manifestation may vary. Diagnosis is mainly implemented through clinical symptoms, imaging (ultrasonography, computed tomography (CT) scan, and magnetic resonance imaging (MRI)), and serological tests(4,5). The disease is usually asymptomatic, but cyst expansion when a large portion of the organ is affected leads to serious complications. So the delay between the infection and the onset of symptoms leads to giant cysts(6). The cysts of 10 or greater than 10 cm in diameter are considered giant cysts. Regarding the rare emersion of giant cysts as atypical cases, the treatment and surgery are complicated(7,8).

***Corresponding author:***
Faeze Foroughi-Parvar, MD
Department of Medical Parasitology and Mycology, School of Medicine, Hamadan University of Medical Sciences, Hamadan, Iran.
Tele: + 98 38380462
Fax: + 98 38380208
Email: f.foroughi@umsha.ac.ir
Orchid ID: 0000-0001-9554-3099

Received: 24 Nov. 2021
Revised: 03 Apr. 2022
Accepted: 04 Apr. 2022

## CASE REPORT

A 39-year-old Iranian woman was referred to the Emergency Medicine Department of our hospital complaining of the right upper quadrant pain and gastric pressure. The patient was a teacher who mentioned visiting villages near Rasht (north of Iran) approximately a month prior (August 2021), staying for

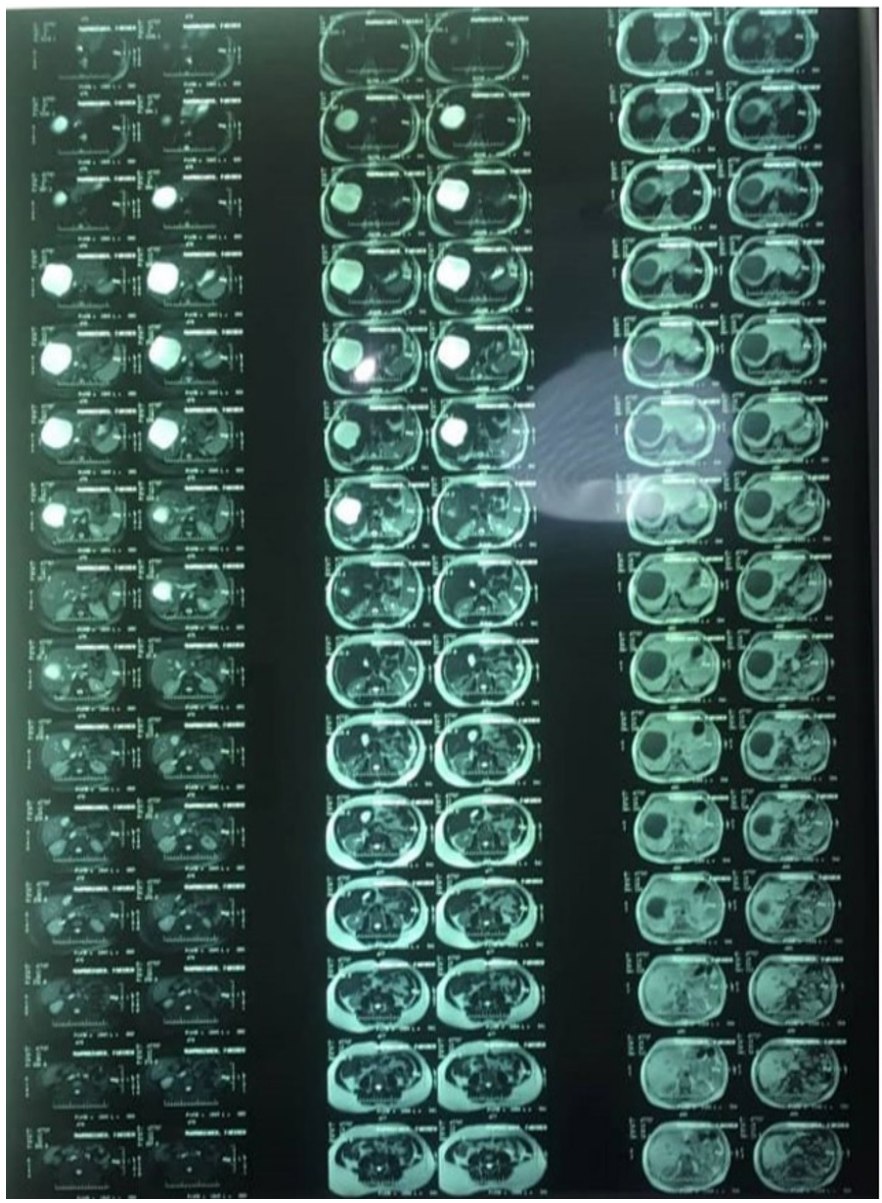

**Fig.1: MRI shows thin wall lesion with low signal on T1 and bright signal on T2**

10 days, and consuming the local cuisine. The patient was conscious during the physical examination. Physical examination showed an enlarged liver with a palpable mass in touch. Weight loss, normal level of liver enzymes, cell blood count (CBC), and erythrocyte sedimentation rate (ESR) were confirmed in a routine examination. Other suspected hepatic problems were ruled out during hospital admission to the Gastroenterology ward. The IgG (enzyme-linked immunoassay) ELISA test using native antigen B (AgB) on the sera of the patient revealed a negative result even after two repetitions (0.19 mg/dL). MRI revealed a huge single 85×115 mm cystic mass (figure1) as craniocaudal view and transverse diameter within the sub-capsular region of the right hepatic lobe while bile ducts and porta hepatis left unaffected. The patient had no history of liver disease or abdominal trauma. However, the routine liver assays were normal. Regarding the massive size of the cyst and acute abdominal pain, resection

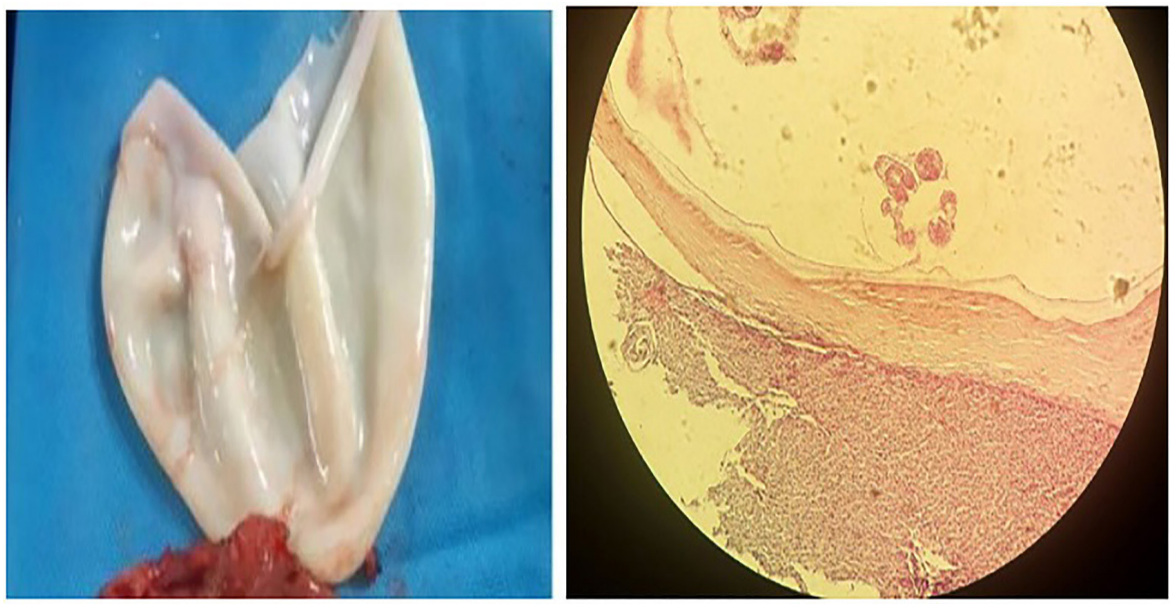

**Fig.2: Cystic echinococcosis removed from a 39-year-old woman (a). The pathological specimen of the removed cyst included the chitinous layer and the germinal layer region in the incision (b).**

surgery with the suspicion of a benign simple cyst was scheduled for the patient before any pre-surgery treatment. Protoscolices observation through the postoperative examination on cyst fluid confirmed the hydatid cysts, as well as the cyst wall identification in histopathologic analysis (Figure 2a, b). Albendazole was prescribed to the patient over the 3 months after surgery.

## DISCUSSION

According to the World Health Organization-Informal Working Group on Echinococcosis (WHO-IWGE), cystic Echinococcosis (CE) is classified into active (CE1 and CE2), transitional (CE3), and inactive (CE4 and CE5) stages based on biological activity, which is supported by clinical manifestations(9,10).

HC may survive latent for several years. Symptoms arise when cysts become large and palpable with abdominal enlargement and pressure. Imaging techniques are frequently used in the detection of HCs. Developing, dying, and dead cysts can be properly detected by CT scans and MRI. Radiography, ultrasonography, and scintigraphy are useful methods for the diagnosis of HC(11).

The present case reported abdominal pain as the first complaint. Abdominal pain is considered the most common symptom, along with hepatomegaly and abdominal enlargement(12,13). In a Chinese female patient, the giant liver cyst was misdiagnosed as hepatic hemangioma(14). The feeling of abdominal pressure was due to the increasing cyst dimension, which raises the risk of intraperitoneal rupture of HC(15). CEs (single cysts) < 5cm that have not responded well to medical therapy and cysts> 5cm are indicated for PAIR (puncture, aspiration, injection, re-aspiration) procedure(16). Enzyme-linked immunosorbent assay IgG was negative in two repeated examinations. False-negative results are reported in some surgically confirmed CE cases(17). It can be explained by the lack or weak antibody production, the presence of circulating immune complexes, and in small quantity antigen release in CE1, CE4, or CE5 stages(18). Immunodiagnosis of CE is still remained debating. Questionable performance of serological tests, low sensitivity (up to 30% false negativity), and also low specificity (up to 25% false positivity), made difficulties in interpretation of CE. Antigen preparation, cyst size, stage, and location of the cyst, as well as the patient immune system, could be accounted for in differences in test results. Nevertheless, serological assays as complementary techniques of imaging are effective in the diagnosis of hydatidosis(17).

According to a retrospective study by Sarkari

and colleagues, women are more likely to develop echinococcosis cysts than men(19), and the same results have been confirmed in studies by Aboudaya et al. in Libya and O`leary et al. in Kenya(20,21).

In the present study, the patient did not receive any pre-surgery medicine. Due to the severe abdominal pain, she was referred for resection. According to WHO guidelines, albendazole or mebendazole therapy should be applied three or four days before the surgery. It should be followed for 3 months after resection(22). Albendazole is constantly used as a pre-surgery treatment twice daily as 400 mg. It has an effective protosclicide activity in vitro, and in the report by Morris and colleagues, the germinal layer has been found damaged following the pre-surgical albendazole therapy(23).

In conclusion, although albendazole therapy reduces the incidence of surgery, surgery is the best choice in the case of large cysts, which involves a huge part of the organ. Due to the low sensitivity and specificity of serological tests, it seems that the diagnosis of HC should be made by a combination of imaging and immunodiagnosis methods. Given that most statistics and information on cystic echinococcosis are from hospital operations and case reports, further research on the prevalence of this parasite in dogs, other definitive hosts, and intermediate hosts, as well as humans should be explored in this area.

## ETHICS APPROVAL:

The patient signed the informed consent related to hospital care and for academic purposes. The Ethics Committee of the Hamadan University of Medical Science approved the publication of this report.

## CONFLICT OF INTEREST

The authors have no conflicts of interest.

### Human and Animal Guidelines:

Helsinki Declaration has been followed for involving human subjects in the study.

### Source of Support:

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

### Availability of data:

Access to data is permitted with the author's permission.

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