

## Complicated Hydatid Cyst of the Liver: A Case of Hepato-Pulmonary Fistula

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### ABSTRACT

Cystic echinococcosis is the term commonly used name for the larval stage of the tapeworm in a zoonotic parasitic infection, caused by species of *Echinococcus* [1].

Infection can potentially occur in any organs and tissues of the human body but predominantly it can affect the liver (75%) and the lung (15%) [1]. Hydatid disease (HD) can remain asymptomatic, and many hydatid cysts (HCs) represent incidental clinical or radiological findings [2].

Presence of symptoms mostly depend on the site of involvement, stage of growth, mass effect, complications or hematogenous spread [3]; reason why different scenarios may occur.

Even if typical features of HD, especially hepatic no-complicated manifestations, are nowadays well known, imaging still plays a key-role not only to reach the fast and correct diagnosis, but also in the visualization of any complications, that are sometimes life-threatening conditions [4].

Ultrasound (US) is almost always the first approach for diagnosis, staging, differential diagnosis, leading any interventional management and usually the first choice during the follow-up [1].

Computed tomography (CT) best demonstrates cyst wall calcification and cyst infection [5].

CT and magnetic resonance (MRI) are furthermore indicated to detect cyst wall defects and also to identify the passage of contents through any fissures or transdiaphragmatic migration [5].

MRI remains superior in demonstrating any neural involvement [5]. The aim of this case report is to show one of the not-so-uncommon complications of this infection, meaning a complex hepatic cyst complicated by hepato-pleural-pulmonary fistula.

### Keywords

Cystic echinococcosis, Hepato-pulmonary fistula.

### Introduction

Cystic echinococcosis (CE), is a conventional name for the tapeworm larval stage of a zoonotic parasitic infection, caused by species of *Echinococcus* [1].

Commonly the name hydatidosis refers to the larval phase of *E. granulosus*; in fact, regarding human infection, the most relevant species are *Echinococcus granulosus*, causing cystic echinococcosis and *Echinococcus multilocularis*, causing alveolar echinococcosis (AE) [1].

CE is the most frequent type, highly endemic in many livestock-rising areas worldwide (1) such as cattle farming areas of South America, the Mediterranean region, the Middle East, Africa, Asia and Australia [3]. After ingestion of food or water firstly contaminated with *Echinococcus* eggs by animals, humans can be infected secondarily.

In human infection, the liver and portal venous circulation are the first stop and line of defense against the tapeworm, reason why the liver is the most common site of involvement (75%); the right hepatic lobe is involved in 80% of cases and the left lobe in 20% [2].

Less commonly, HD can involve lungs (15%), spleen, kidney, bones, brain and other although rare anatomic sites and tissues [3].

Regarding the thoracic district infection, lung parenchyma is the most common involved, but any extrapulmonary region including the pleural cavity, fissures, mediastinum, heart, vascular structures, chest wall and diaphragm, can be involved as well [4]. As nowadays widely known, the typical HC has a multi-layered structure. HD can remain asymptomatic, and many HCs represent incidental clinical or radiological findings [2]. Presence of symptoms mostly depends on the site of involvement, stage of growth, mass effect, complications or hematogenous spread [3]. Most symptomatic HCs are complicated with secondary bacterial infection, rupture or because of their mass-effect in large size cyst, causing symptoms like right upper abdominal pain, swelling and general discomfort [2].

Complications are not-so-uncommon and sometimes they can represent life-threatening conditions without an early diagnosis and treatment. One of the most common complications is the HC rupture; it can be intrabiliary, intra-peritoneal, intra-thoracic through the diaphragm, or toward organs of the gastrointestinal tract [2]. Reason why different scenarios may occur, the role of imaging is pivotal not only for the diagnosis of HC, but also in its staging, extension and complications. Ultrasound (US) is almost always the first approach for diagnosis, staging, differential diagnosis, leading any interventional management and usually the first choice during the follow-up [1].

US of the upper abdomen can detect HCs located in the diaphragm or in the peripheral segments of the lungs and transthoracic echocardiography can identify those in the cardiac chambers [3]; however, US alone is not the best method for assessing the precise extent of the HC, while CT and MRI can demonstrate it in detail. CT is particularly effective in demonstrating cyst wall calcification and cyst infection [4], while MRI, along with CT, is indicated for detecting cyst wall defects, identifying the passage of contents through fissures or transdiaphragmatic migration [5] and, ultimately, evaluating treatment efficacy by assessing changes in the size, number and shape of HCs [4].

MRI is still superior in demonstrating any neural involvement [5]. Imaging findings depend on the organ involved, host reaction

and stage of the cyst, ranging from purely cystic lesions to solid masses-like aspect [3]. Cysts can be solitary/multiple, unilocular/multivesicular, with/without calcification; furthermore, daughter vesicles or membranes into the cyst, peripheral cyst wall or calcification may be important for differential diagnosis of HD [3]. Even though different classifications exist, HCs are most commonly classified into four types based on their imaging appearance.

Type I: simple cyst with no internal architecture.

Type II: cyst with daughter cysts and/or matrix.

Type III: calcified cyst.

Type IV: complicated HC.

The aim of this case report is to show one of the not-so-uncommon complications of HD infection, meaning a complex HC complicated by right hepato-pleural-pulmonary fistula (Type IV).

### Case Presentation and Clinical History

An 82 years-old man presented to the emergency room with a history of recent fatigue, fever, almost 39°C, productive cough with purulent sputum and right upper quadrant pain.

He was alert and collaborating and at triage findings were reduced vesicular murmur at the right lung base, regular cardiac activity and non-peritonitic abdomen. He had a previous positive history of a non-complicated big hepatic HC, HCV-related liver disease, type 2 diabetes mellitus, asthma and arterial hypertension.

### Imaging Findings

In the emergency room, US of the abdomen and basal chest plus superior abdomen CT were performed.

Abdominal ultrasound revealed the already known big hepatic HC, with hypo-anechoic content and partially calcified wall, sited subcapsular in the right liver lobe between VII and VIII segments, measuring almost 6cm (Figure 1).

Furthermore, an heterogeneous echogenicity between the liver capsule and the ipsilateral pleural cavity was shown close to the cyst wall (Figure 1a, 1b).

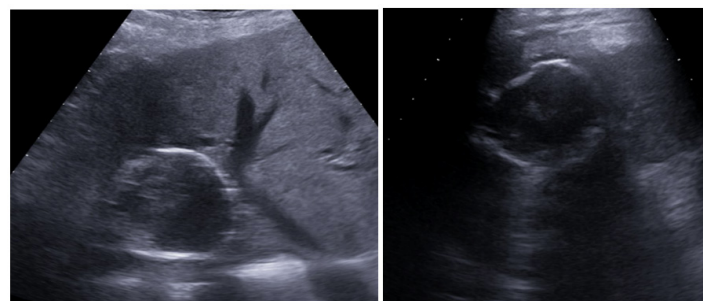


Figure 1a-b

Basal chest plus superior abdomen CT was then performed. The CT scan revealed parenchymal lung consolidation into posterior and lateral segments of the right lower lobe (Figure 3a-b).

Furthermore, a right-basal pleural thickening was shown, contiguous and not cleavable from the hepatic HC and from the liver capsule (Figure 4).

The CT scan subsequently demonstrated the full extent of the hepatic HC and its partially calcified wall (Figure 4a-b-c). Air bubbles inside the HC were also shown, suggesting a strongly suspected hepato-pulmonary fistula (Figure 4b). The patient was then hospitalized, and an MRI was soon scheduled.

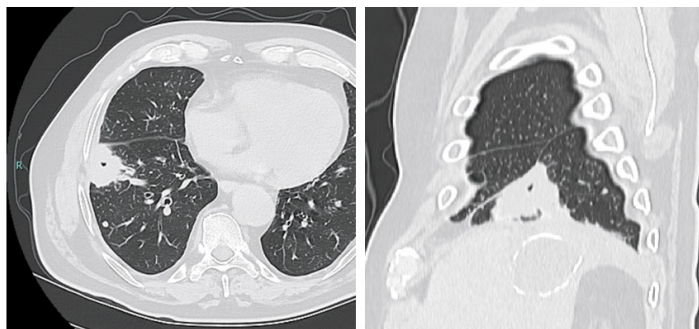


Figure 2a-b

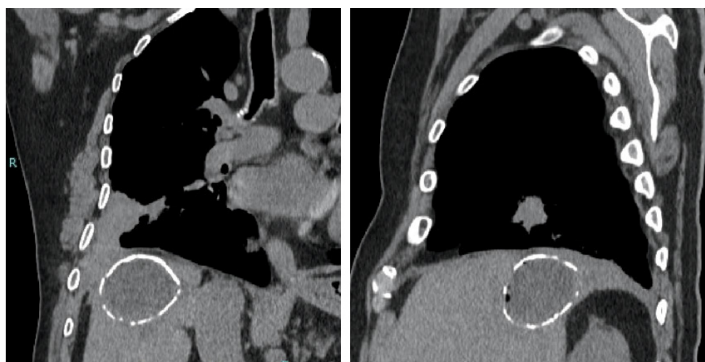


Figure 3a-b

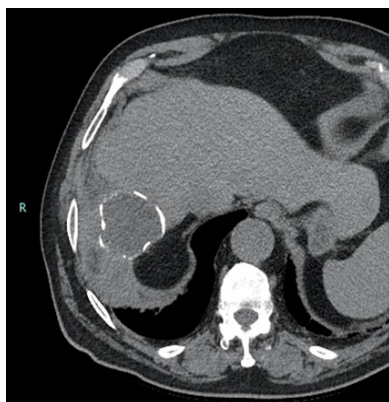


Figure 3c

A few days later, MRI with and without contrast injection was performed, demonstrating the previously known findings related to the hepatic HC and confirming the new diagnostic suspicion.

In fact, an interruption of both the postero-lateral wall of the right diaphragm and of the wall of the HC was shown. Especially after-contrast injection, MRI best identified a fistulous tract growing between the hepatic HC and the thoracic cavity, reaching the right lung parenchyma through the ipsilateral pleura (Figure 7a-b, 8a-b).

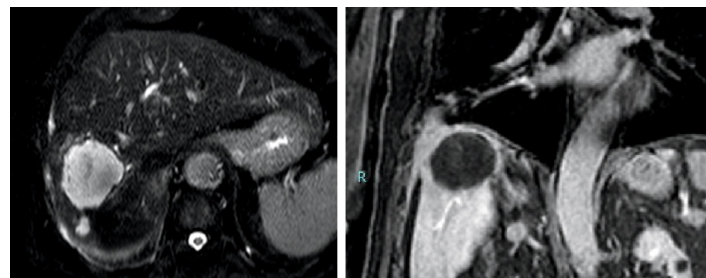


Figure 4a-b

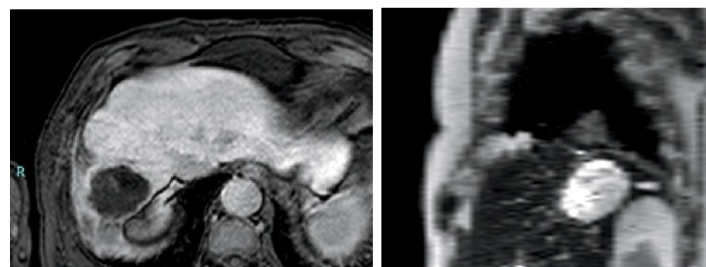


Figure 5a-b

Antifungal, antibiotic and anthelmintic therapy were then started before any further pneumological or surgical evaluation.

## Discussion

The diagnosis of HD and HC is based on clinical findings, serologic values, and imaging features [6]. HC mostly affects the liver (75%), followed by the lungs and, less commonly in the spleen, kidneys, brain and musculoskeletal system [1].

HC has three layers: the outer fibrous layer is called pericyst and consists of modified host cells including fibroblasts, giant cells and eosinophils [1]; the middle laminated acellular membrane and the thin inner germinal layer form the endocyst, that is the true wall of the HC [1].

The infectious embryonic tapeworm known as “scolex” develop from an outpouching of the germinal layer [1]. Complications of HC are generally rupture, infection, exophytic growth and portal venous system involvement [6]. Rupture of the hydatid cyst is the most frequent complication (20-50%), mostly depending on degeneration of the parasitic membranes, aging of the hydatid cyst or trauma [2]. US, CT and MRI help to reach the correct diagnosis, to show their complications and to manage the follow-up as well. While CT scan is sometimes preferred for its speed and costs and in preoperative management [2], MRI is particularly effective in showing the presence of fistulous ducts in communication with other viscera, as in case presented.



In literature, three types of complication have been described:

### Contained Rupture

The endocyst ruptures and the hydatid fluid escapes into the space between the pericyst and the endocyst, leading to the collapse of the endocyst [2].

In this kind of collapse, CT scan may show the diagnostic sign called the “water lily sign” or “snake or serpent sign”, consisting of detached and undulating membranes inside the hydatid cyst without reduction of its size [2].

### Communicating Rupture, with involvement of the biliary tree.

This is the most common type of rupture (44-64%); in this condition the content of hydatid cyst evacuates into the biliary radicles involved by the pericyst [2]. Depending on the size of communication, small fissures in small biliary radicles or wide perforation into a main biliary radicle may be shown.

**Direct Rupture** into peritoneal cavity, pleural cavity, abdominal wall and hollow viscera. It occurs when both the endocyst and pericyst are torn and the content of the cyst escapes into the peritoneal cavity, the pleural/thoracic cavity through the diaphragm, the mediastinum or hollow organs.

Although rare, this condition can be fatal without the appropriate management; allergic shock, peritonitis or implantation of scolices in several organs, known as “metastatic hydatidosis”, may occur. Sometimes, direct rupture of hepatic hydatid cyst can be delimited by Glisson’s capsule, self-limiting into the hepatic subcapsular space [2].

As the presented case, transdiaphragmatic-intrathoracic rupture of a hepatic HC is a complication due to direct rupture, occurring in almost 0.6-16% of patients with hepatic HD [2].

Because of the lack of peritoneal covering, the posterior segments of the right hepatic lobe, also known as “bare area” of the liver, is the most common site of transdiaphragmatic migration of hydatid material. When CT scan reveals the presence of air inside the HC, communication with the bronchial tree must be strongly suspected [2]. Due to chemical reactions from the migration of hydatid material into the lungs, CT scans commonly show pleural effusions, pleural thickening, lung consolidation and atelectasis, similar to the described case as well.

### Conclusion

HD predominantly affects the liver, followed by the lungs, but it can potentially affect almost any organ in the human body with different sizes and shapes [1]; commonly it demonstrates characteristic imaging findings but sometimes it can create diagnostic challenges miming benign or malignant neoplasms [1].

As symptoms are non-specific, imaging plays a crucial role in the diagnosis of hydatid cyst in common/uncommon locations and its staging or complications [4].

Complications of HD occur in about one third of patients [7] and may potentially be life-threatening if not promptly diagnosed.

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