

## Case Report

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# The Trans-Diaphragmatic Hydatid Cyst: An Unconventional Surgical Strategy

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### ABSTRACT

**Introduction:** The hydatid disease is a zoonotic infection due to the tapeworm echinococcus granulosus (TEG). In 50-70% of the cases, the hydatid cyst is observed with a hepatic localization. The trans-diaphragmatic extension of a liver hydatid cyst is rarely reported in the literature. Here, we report the singularity of our observation which focuses on an abdominal approach rather than a thoracotomy and the way we handle the diaphragmatic defect.

**Observation:** A 34-year-old male patient presented with a right hypochondriac pain evolving since 2 years. The abdominal examination found a bulging just below the right costal margin. The ultrasound and computed tomography (CT) scan images show an enormous liver hydatid cyst covering the entire posterior right section and extending beyond the diaphragm to the right hemi-thorax. Our therapeutic strategy consisted of a resection of the protruding dome with aspiration and evacuation of all the hydatid material. We did not close the diaphragmatic defect because there was no communication with the thorax contents. Our management had no negative impact on the patient in 2 years of follow-up.

**Conclusion:** Being rarely reported in the literature, the trans-diaphragmatic hydatid cyst is an uncommon situation. The surgical intervention is the main stay treatment. Our management of the diaphragmatic defect was unconventional. The singularity of our approach is to not close the diaphragmatic defect since we considered the remaining fibrous capsule as a closure, avoiding a laborious dissection and a complex diaphragmatic reconstruction.

**KEY WORDS:** Surgery; Trans-diaphragmatic; Hydatid cyst; Zoonotic infection; Tapeworm; Echinococcus granulosus.

**ABBREVIATIONS:** TEG: Tapeworm Echinococcus Granulosus; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; E: Echinococcus; WBC: White Blood Cell; ELISA: Enzyme-linked immunosorbent assay.

### INTRODUCTION

The hydatid disease or the echinococcosis is a parasitic infection due to the Echinococcus (E) tapeworm infestation. It is an endemic disease in many regions worldwide. Mainly, it is a zoonotic infection caused by the adult E-worm beared by the definitive host. Humans are infected accidentally by ingesting the worm's eggs. Many organs can be observed to be the sites of localized hydatid cyst especially during the primary echinococcosis. In adults, the liver is the main site in 50-70% of the cases followed by the lungs in 10-30%.<sup>1,2</sup> The trans-diaphragmatic extension of a hepatic hydatid cyst is an uncommon situation, rarely reported in the literature. Herein, we present the case of a voluminous hydatid cyst of the hepatic right posterior section extended to the inferior pulmonary right lobe and our surgical management for the same.

### CASE REPORT

A 34-years-old male, with no previous medical history, consulted our department for a right

upper quadrant pain evolving since 2 years without radiation associated to asthenia and weight loss. The patient had no history of fever, neither digestive nor respiratory symptoms. During the abdominal examination, we found a bulging just below the right costal margin with no tenderness during palpation. The respiratory system examination showed dullness instead of resonance at the percussion of the inferior part of the right hemi-thorax. Biologic investigations revealed hemoglobin 11 g/ml, white blood cell (WBC) 8400/mm<sup>3</sup> and eosinophils were not elevated. An abdominal ultrasound confirmed the diagnosis of a voluminous hepatic hydatid cyst partially calcified which extended through the diaphragmatic dome. An additional thoraco-abdominal CT scan showed a large hydatid cyst covering almost the totality of the right posterior section of the liver and extending beyond the diaphragm in the right hemi-thorax. This cyst was calcified in its inferior part and contained multiple daughter vesicles (Figure 1). The indirect immunofluorescence and the enzyme-linked immunosorbent assay (ELISA) test for the *E. granulosus* were significantly positive. A pre-operative Albendazol therapy (400 mg per day) for 3 months was indicated. The surgery was done through a midline laparotomy; no hepatectomy was done instead of that a protruding dome resection was performed just above the calcified part with aspiration and evacuation of its contents (liquid, membranes and the daughter cysts), taking the pre-caution to confine the operation site with H<sub>2</sub>O<sub>2</sub> soaked compresses to avoid any spillage. There was no biliary fistula identified. The

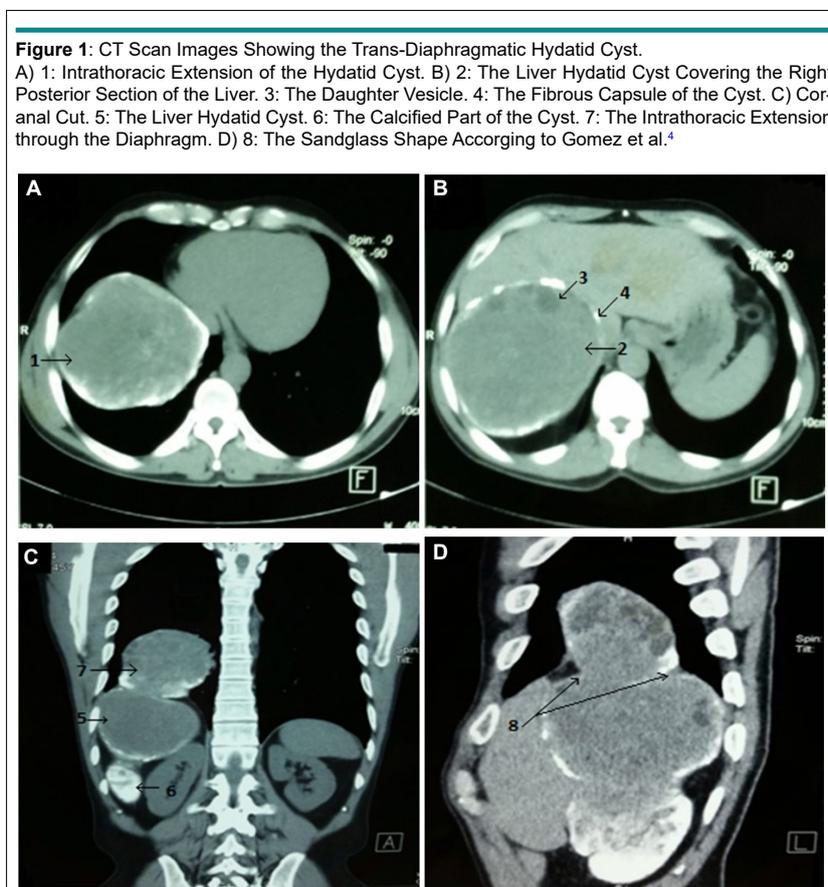
rent in the diaphragm was about 7 cm, made essentially from the fibrous hull or capsule of the superior part of the hydatid cyst without any bronchial fistula. We also found that the superior part of the fibrous capsule of the cyst was continuous with the diaphragm, preventing any communication between the thoracic and the abdominal contents. Our decision was to end the surgery just by a drainage of the remnant cavity (inter hepato-diaphragmatic space) without closing the diaphragmatic rent. We show you our surgical strategy by 4 diagrams (Sketches).

The post-operative course was uneventful. The patient remained asymptomatic after 24 months of follow-up; the recent CT scan showed no recurrences, also there was no negative impact of our decision to consider the fibrous capsule as a diaphragmatic closure (Figure 2).

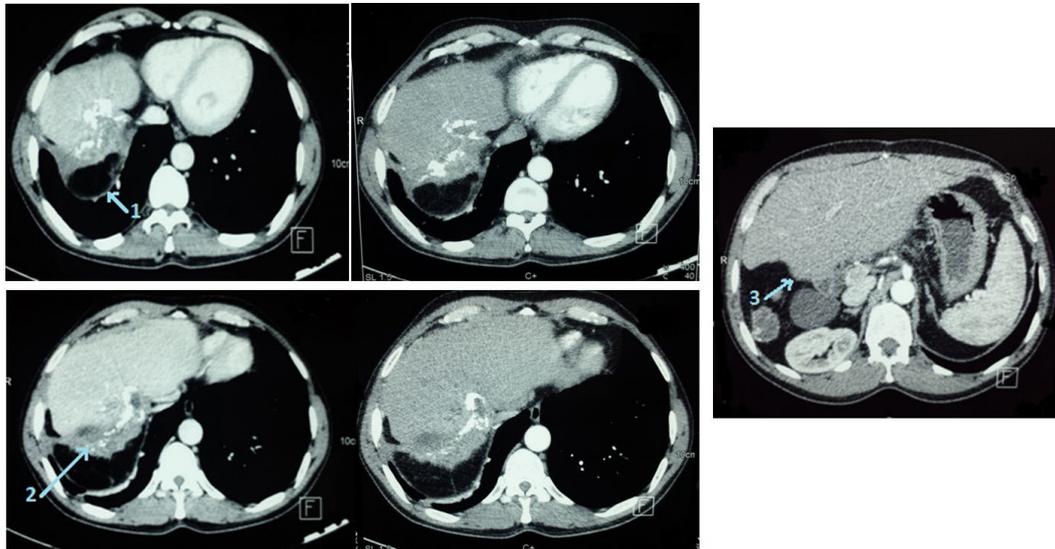
## DISCUSSION

The hydatid disease is a parasitic infection cause by the *Echinococcus* tapeworm. It has worldwide prevalence, especially in countries like Morocco where sheep rearing is carried out on a large scale. Although, the liver and the lungs are the most frequently affected viscera by the hydatid cyst, other organs such as the spleen, the kidneys, and the brain could also be affected.

The trans-diaphragmatic presentation where the he-

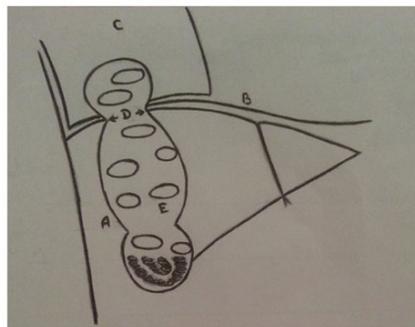


**Figure 2:** Post-operative CT Scan Images Showing No Recurrences and No Negative Impact of our Decision of not Closing the Diaphragmatic Rent.

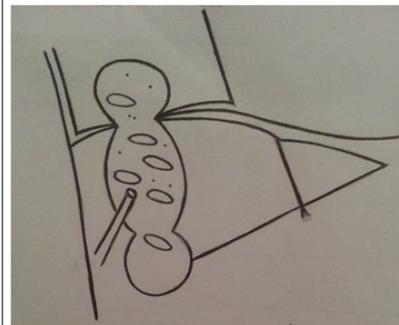


1. The fibrous capsule is continuous with the diaphragm.
2. Absence of any recurrences of the the hydatid cyst.
3. A liver compensatory hypertrophy.

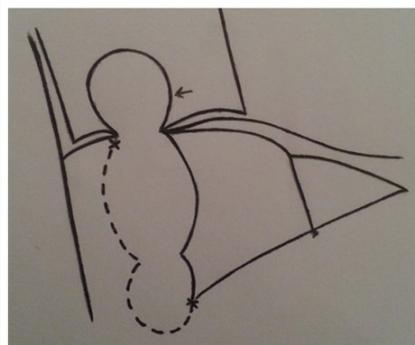
**Sketch 1:** A) The Trans Diaphragmatic Hydatid Cyst. B) The Diaphragm. C) The Right Lung. D) The Diaphragm Rent. E) Daughter Vesicles.



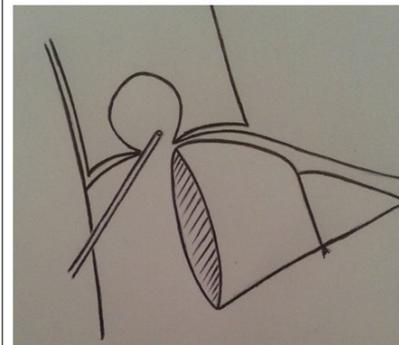
**Sketch 2:** Aspiration and Evacuation of the Hydatid Cyst Content.



**Sketch 3:** Resection of the Protruding Dome without Performing any Hepatectomy.



**Sketch 4:** Drainage of the Remnant Cavity without Closing the Diaphragmatic Rent Defect.



hepatic hydatid cyst invades the lung is rarely reported; it represents only 2% in a study with more than 1150 patients with liver hydatid cyst.<sup>3</sup> According to the classification that evaluates the diaphragmatic and trans-diaphragmatic extension of a hepatic hydatid cyst by Gomez et al<sup>4</sup> our patient had a Grade 3 trans-diaphragmatic involvement: a “sandglass” cyst perforating the diaphragm and growing inside the thoracic cavity without any connection with the bronchi.

The diagnosis is easily confirmed by ultrasound, CT scan or even magnetic resonance imaging (MRI),<sup>2,5</sup> offering a number of information such as the cyst measurement, its localization and impact on the adjacent anatomical structures (vascular and biliary structures), the WHO classification, and the presence of a diaphragmatic rent, its dimensions or a communication with the bronchial tubes.

The treatment of hydatid cysts is well established by the WHO: an anthelmintic treatment (Albendazol), pre- and post-operatively combined with surgery, which remains the mainstay treatment although it has to be as conservative as possible for the healthy parenchyma.<sup>6</sup> Recently, a percutaneous treatment has been developed based on echo-guided puncture, aspiration of the hydatid material, injection of protoscolicidal agents and finally re-aspiration (PAIR).<sup>2</sup> The percutaneous treatment isn't indicated in communicating hydatid cysts.

The hydatid cyst may evolve in many organs separately.<sup>7</sup> The synchronous trans-diaphragmatic extension of a hepatic hydatid cyst to the right lung is uncommon.<sup>8</sup> In the literature, the one stage trans-diaphragmatic approach for the liver cyst through a right thoracotomy (7<sup>th</sup>-8<sup>th</sup> intercostal space) is highly recommended, because it can cure the lung and the hepatic cysts, simultaneously eliminating the requirement of a second surgical intervention.<sup>8,9</sup> On the contrary, the trans-diaphragmatic approach for a right lung cyst through a laparotomy is only reported in 1 patient from the 50 patients evaluated by Aydin et al<sup>10</sup> series. In fact put 4 conditions to adopt this specific approach:

- An absolute indication for laparotomy;
- The pulmonary cyst isn't voluminous or complicated;
- Absence of pleural adhesions;
- The pulmonary cyst is reachable by a trans-diaphragmatic approach through laparotomy (the right lower lung lobe).

Whatever the approach is (thoracotomy or laparotomy), the surgical procedure must contain the following steps: protection by soaked compresses with proto-scolicidal agents, aspiration of the cyst liquid through a needle, opening the cyst to evacuate the remnant material, fistulae obliteration (bronchial or biliary), opening the diaphragm to reach the other cyst and re-do the same steps and finally drainage and closure of the diaphragmatic defect.

The particularity of our observation is the second case reported

to treat a trans-diaphragmatic extension of a liver hydatid cyst through a laparotomy. This approach was adopted for multiple reasons:

- The patient had no respiratory symptoms which means there was no communication between the cyst and the bronchi; a condition that will be difficult technically to cure through a laparotomy;
- The cyst was Grade 3 of Gomez classification and invading the inferior pulmonary lobe making it reachable *via* a laparotomy;
- The liver hydatid cyst was large and covering the entire right posterior section. The probability of biliary fistulae is very high. We thought that the laparotomy approach would give us the global view for an optimal management of these biliary fistulae.

Our patient had a 24 month follow-up. There were no recurrences or complications directly related to our decision of not resecting the fibrous capsule or approaching the diaphragm's margins (Figure 2).

## CONCLUSION

Trans-diaphragmatic hydatid cyst is rarely reported in the literature. The surgical treatment is generally performed through a thoracotomy. The laparotomic approach may be used only if the hydatid cyst is reachable and affecting the inferior part of the right lung. Our strategy of not closing the diaphragmatic rent and considering the fibrous hull as a cure to the defect did help us evade a large diaphragmatic dissection and a complex reconstruction with no negative impact on our patient.

## AUTHORS' CONTRIBUTIONS

HH drafted the manuscript. AM, MA, MR, FS, AH critically revised the manuscript. All authors read and approved the final manuscript.

## CONSENT

The patients gave a signed statement, which authorizes the use of her personal and/or medical information in the publication of this study.

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None. Compliance with ethical guidelines.

## CONFLICTS OF INTEREST

The authors declare that they have no competing interests. First and corresponding author Dr. Hajar Hachim. 5<sup>th</sup> year resident in General surgery.

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