

## Case Report

# An Iatrogenic Hepato-Peritoneal Hydatidosis Revealed at the Stage of Abdominal Cutaneous Fistula: A Case Report

Othmane Bourouail<sup>\*</sup>; Anwar Rahali; Elmustapha Halim; Abdelkarim Laalou; Aziz Elfadili; Njoumi Nouredine; Abderrahman Elhjouji; Aziz Zentar; Abdelmounaim Aitali

Visceral Surgery Service II, Military Teaching Hospital Mohamed V, Ibn Sina University Hospital Centre, Rabat, Morocco

**\*Corresponding author: Othmane Bourouail**

Resident of General Surgery, Visceral Surgery Department, Military Hospital of Instruction Mohammed V, Rabat, Morocco.

Tel: +2120666784726

Email: Th.bourouail@gmail.com

**Received:** July 03, 2023

**Accepted:** August 05, 2023

**Published:** August 15, 2023

## Abstract

Hydatidosis is a parasitic disease caused by the development of the larval form of *Echinococcus granulosus*, it is a significant public health problem in Morocco due to its frequency and the severity of some of its complications. Hepato-peritoneal hydatidosis is a rare serious condition, with a high risk of recurrence and significant morbidity and mortality. The peritoneal localization is most frequently iatrogenic caused by a previous hydatidosis surgery; the revelation by external abdomino-parietal fistulization makes it more uncommon. The large laparotomy surgery excision is the chosen treatment. Furthermore, to prevent possible recurrence a long-term antiparasitic therapy should be implemented.

In our case, it was an intra-abdominal hydatid contamination due to the iatrogenic intraperitoneal rupture of the hepatic hydatidosis. This condition was revealed at the stage of cutaneous abdominal fistulization. The abdominal CT scan showed both positive and topographic diagnosis. our therapeutic strategy associated a large laparotomy surgery and antiparasitic medical treatment.

The secondary peritoneal hydatidosis caused by iatrogenic trauma is rare but constitute a serious problem with short free recurrence disease and a high risk of morbi-mortality. This case illustrates the potential complication of hydatid cyst surgery, and the latency of the peritoneal localization.

**Keywords:** Hepato-peritoneal; Iatrogenic hydatidosis; Abdomino-cutaneous fistula; Surgery; Antiparasitic.

## Introduction

Hydatidosis is a parasitic disease caused by the development of the cestode *Echinococcus Granulosus* (EG) larva especially in the liver or lungs. The food's ingestion contaminated by the excrement of a carrier animal or the contact with infected animals are the most common modes of transmission [1]. The diagnosis is based on epidemiological, clinical, radiological, and biological characteristics [2]. The diagnosis generally made at stage of complication, in which the cyst rupture is the main complication and affects 15 to 40% of hepatic Hydatid Cysts (HC) [1,2]. This opening can occur in the bile ducts (40 to 60%) as well as in other abdominal or thoracic organs [3]. The HC rupture on the peritoneal cavity is uncommon (2-7%), mostly caused by a previous surgical intervention of the hydatidosis [1-3]. This condition constitutes a pejorative turning of the disease, immediately by the risk of anaphylactic choc and secondary, by the intra-peritoneal dissemination. In this case, we report an iatrogenic hepato-peritoneal hydatidosis (HPH), widely disseminated and revealed at the stage of the Abdomino-Cutaneous Fistula (ACF).

## Materials and Methods

A 62-year-old male patient presented with a cutaneous fistula in the abdomen. The patient had a history of surgical intervention for liver hydatid cyst 8 years prior with protruding dome resection.

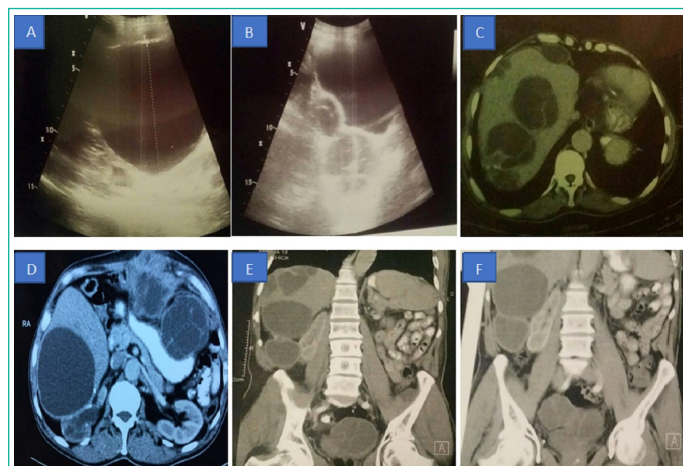
Upon physical examination, the patient had an abdominal fistula that was discharging a clear fluid. His vital signs were stable, and there were no signs of systemic infection.

Imaging examinations were conducted, including an abdominal ultrasound and Computed Tomography (CT) scan. The ultrasound revealed the presence of a large hepatic mass with multiple cysts, while the CT scan confirmed the hepatic HCs and revealed multiple peritoneal cysts (Figure 1). The laboratory investigations showed elevated levels of eosinophils and IgG antibodies to *Echinococcus granulosus*.

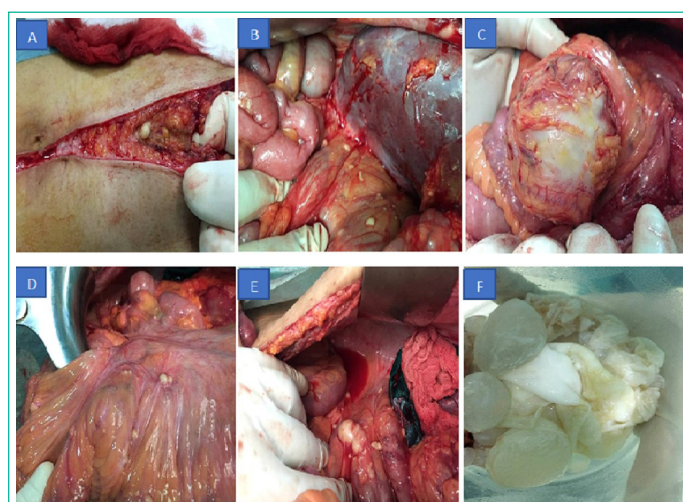
Based on these findings, the diagnosis of HPH was made. The therapeutic strategy associated the perioperative antiparasitic

drugs, including albendazole and praziquantel, and the surgery with a large laparotomy.

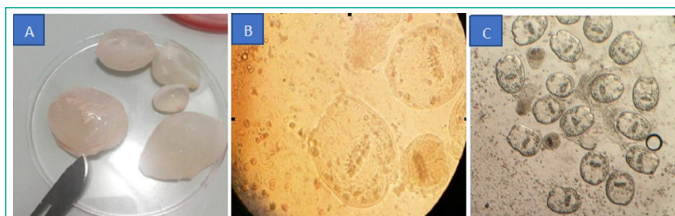
This latter was performed under general anesthesia with a xyphopubic laparotomy, after a large abdominal exploration, the entire peritoneal cavity was invaded, it was a veritable intraperitoneal hydatid metastasis. After appropriate measures to avoid the spillage of cystic fluid and prevent the spread of the disease, the majority of hydatid cysts were excised with radical and conservative method depending on location and relationship with adjacent organs and vessels (Figure 2). The patient tolerated the procedure well and was discharged from the hospital with a prescription for continued antiparasitic therapy. The anatomopathological and parasitological examination of the specimen confirmed the diagnosis of the hydatidosis (Figure 3).



**Figure 1:** HPH revealed by ultrasound and an injected abdominopelvic CT imaging. (A) Abdominal ultrasonography showing an HC in the hepatic dome. (B) A pelvic ultrasound revealing two retrovesical cystic lesions. (C) Multiple hepatic HCs are visible on a CT scan's axial slice. (D) CT image with axial section displaying liver HC at segment VI and two cystic lesions affixed to the big stomach curvature, one of which is fistulized to the abdominal wall. (E) The right transverse mesocolon, the liver, and the pre-vesical region all have cystic lesions as seen on the CT image. (F) Cystic lesions of the Right transverse mesocolon, rectovesical space are visible on a frontal section CT scan.



**Figure 2:** Preoperative image of various-sized and-located intra-abdominal hydatid cysts. (A) Abdominal wall fistula. (B) Right transverse mesocolon and liver HC. (C) A right colon-adhering hydatid mass. (D) Multiple Hydatid lesions of mesentery. (E) Pelvic cysts in close proximity to the retroperitoneal and pre-vesical spaces as well as the right iliac arteries. (F) Extirpated cysts with proligerous membranes and daughter vesicles.



**Figure 3:** Hydatid cysts: (A) Vesicles daughter. (B) Scolex of EG. (C) Protoscolex.

Follow-up imaging and laboratory investigations were conducted to monitor the patient's progress. After six months of antiparasitic therapy, the patient's IgG antibody levels decreased significantly, and there was no evidence of disease recurrence on a radiological imaging study. The patient was advised to continue antiparasitic therapy for a total of one year, and follow-up visits were scheduled to monitor his condition.

## Discussion

The intraperitoneal rupture of the hepatic hydatid cyst is an uncommon complication, that presents a serious problem with a primary risk of anaphylactic choc and a large dissemination after an evolution's long period [2,3]. This condition is mostly caused by an iatrogenic trauma during a surgical procedure of hydatidosis, leading to the intra-abdominal development of *Echinococcus granulosus* larvae and the formation of HCs in different organs [4]. This issue can be latent for a long time and as they increase in size, they compress adjacent tissues, organs and/or vessels, thus causing symptoms, which are site specific. Otherwise, the intraperitoneal hydatid cysts can undergo the same complication as that of hepatic localization such as the sur-infection, the cyst rupture and fistula [3,4]. In this case, the patient presented with the Abdominal Cutaneous Fistula (ACF), rare eventuality, caused by mechanic and inflammatory factor [5]. Its revealing phase in this instance, reflects the latency and the disseminated form of the disease.

Imaging examinations such as ultrasound and Computed Tomography (CT) scan are useful to confirm the diagnosis [6]. Ultrasound is a simple and non-invasive imaging technique that can detect the presence of hydatid cysts with high accuracy [7]. CT scan is more sensitive in detecting small cysts and can provide the topographic diagnosis of the disease [6-8]. The reference's treatment for HPH is the antiparasitic drugs combined to the surgery [9]. Antiparasitic drugs such as albendazole and praziquantel are effective in killing the larvae and preventing the growth of cysts [1-9]. Surgical intervention by large laparotomy using a radical and a conservative method is necessary to achieve the resection of all HCs and prevent further dissemination of the disease. The surgical team must take appropriate measures to avoid cystic fluid spillage and prevent the spread of the disease [3-9].

The management of hydatidosis depends on various factors, including the cyst's localization, the size and the presence of complications [9]. In this case, the patient had multiple hepatic and peritoneal cystic mass, which required a large exploratory laparotomy to ensure as much hydatid mass resection as possible by radical and conservative method [10].

Using the antiparasitic drugs in the hydatidosis's management is still controversial. Some studies have shown that the preoperative antiparasitic treatment can facilitate the surgery by reducing the size. After surgery, employing the medical therapy for a long period prevent the recurrence's disease. However, other studies have reported no significant benefits

of antiparasitic therapy. In this case, the patient underwent a perioperatively combination of albendazole and praziquantel, that have been continued for one year. The IgG antibody levels decreased significantly, and there was no evidence of disease recurrence on imaging studies [11].

The prognosis of hydatidosis depends on the recurrence's potential of the disease, the relationship with a neighboring organs and vascular structures, reflecting on its removability, and the presence of complications. In general, the prognosis is good if the disease is diagnosed and treated early. However, the presence of the intra-abdominal hydatid mass's complications such as the infections or the rupturing of the cyst can make the prognostic poor [12]. In this instance of disease, the morbidity related to surgery may reach 16% while the mortality can vary between 0 to 20% [13].

This case presents a veritable intra-abdominal iatrogenic hydatid metastasis, controlled both by large laparotomy surgery and a long-term antiparasitic therapy. The prevention of this condition involves judicious blood transfusion and rigorous sterilization of surgical instruments. It is important for healthcare providers to be aware of the potential risks of iatrogenic hydatidosis and take appropriate measures to prevent its occurrence. Early diagnosis and treatment are crucial to prevent the progressing of the disease and causing serious complications [14].

### Conclusion

Hepato-peritoneal hydatidosis is one of the rarest conditions caused by previous hydatidosis surgery. This instance illustrates the importance of considering it in the differential diagnosis of patients with a history of hydatidosis surgery and its potential complications. The revelation by the ACF reflects the latency and dissemination of the disease. Healthcare providers should be aware of the potential risks of iatrogenic hydatidosis and take appropriate measures to prevent its occurrence.

### Author Statements

#### Acknowledgement

We appreciate the contribution of all surgeons, co-workers and friends of radiology and parasitological involved in this case of report and thanks the editor and the reviewers for their help with this manuscript.

#### Authors' Contributions

All authors contributed significantly to this work and agree to be accountable for all aspects of the work. OB, AR and EH Contributed to the study design, art drawing, acquisition of data, drafting and revising the manuscript. AL, AE contributed to the study design and art drawing. NN, AE, AA participated by revising the manuscript, giving a critical analysis. AZ performed the surgical procedure and according publication after revising the manuscript.

#### Funding

There are no resources of funding to be reported or declared.

#### Availability of Data and Materials

All data is contained within the manuscript file.

#### Ethics Approval and Consent to Participate

This case of report is only involving objective retrospective

description therefore not applicable to ethics approval. The written informed consent was obtained from the patient to participate to this case report and any accompanying images.

#### Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

#### Competing Interests

The authors declare no competing interests.

#### References

- Koç C, Akbulut S, Şahin TT, Tuncer A, Yılmaz S. Intraperitoneal rupture of the hydatid cyst disease: single-center experience and literature review. *Ulus Travma Acil Cerrahi Derg.* 2020; 26: 789-97.
- Akbulut S, Ozdemir F. Intraperitoneal rupture of the hydatid cyst: four case reports and literature review. *World J Hepatol.* 2019; 11: 318-29.
- Akbulut S. Parietal complication of the hydatid disease: comprehensive literature review. *Med (Baltim).* 2018; 97: e10671.
- Toumi O, Noomen F, Salem R, Rabeh H, Jabra SB, Korbi I, et al. Intraperitoneal rupture of hydatid cysts. *Eur J Trauma Emerg Surg.* 2017; 43: 387-91.
- En-Nafaa I, Moujahid M, Alahyane A, Amil T, Hanine A, et al. Hydatid cyst of the liver ruptured into the abdominal wall and the psoas muscle: report of a rare observation.
- Belli S, Akbulut S, Erbay G, Koçer NE. Spontaneous giant splenic hydatid cyst rupture causing fatal anaphylactic shock: a case report and brief literature review. *Turk J Gastroenterol.* 2014; 25: 88-91.
- Sarkar D, Ray S, Saha M. Peritoneal hydatidosis: A rare form of a common disease. *Trop Parasitol.* 2011; 1: 123-5.
- Ran B, Shao Y, Yimitei Y, Aji T, Shayiding P, Jiang T, et al. Spleen-preserving surgery is effective for the treatment of spleen cystic echinococcosis. *Int J Infect Dis.* 2014; 29: 181-3.
- Sozuer E, Akyuz M, Akbulut S. Open surgery for hepatic hydatid disease. *Int Surg.* 2014; 99: 764-9.
- Shalayiadang P, Jiang T, Yimitei Y, Ran B, Aini A, Zhang R, et al. Double versus single T-tube drainage for frank cysto-biliary communication in patients with hepatic cystic echinococcosis: a retrospective cohort study with median 11 years follow-up. *BMC Surg.* 2021; 21: 12.
- Mejri A, Arfaoui K. Ruptured hydatid cyst into the peritoneal cavity: an ultimate multidisciplinary emergency. *J Emerg Trauma Shock.* 2021; 14: 117.
- Sheikhy K, Rouhani R, Pejhan S, Sanei Motlagh A, Sheikhy A. Evaluation of clinical status, diagnosis, treatment and radiological findings of pulmonary hydatid cyst: 5-years' experience at tertiary lung center. *Caspian J Intern Med.* 2022; 13: 44-50.
- Benamr S, Mohammadine E, Essadel A, Lahlou K, Taghy A, Chad B, et al. L'hydatidose péritonéale secondaire. Mise au point à propos d'une série de 50 cas. *Med Maghreb.* 2000; 82: 15-20.
- Adhikari S, Bhattarai M, Gyawali S, Subedi S, Bhattarai A, Awale L et al. Acute abdomen due to rupture of a hydatid cyst of the liver: a rare complication - a case report. *Ann Med Surg (Lond).* 2023; 85: 1172-6.