



Case report

Hydatid cyst: Introducing a new surgical approach towards hepato-pulmonary fistula, a case report

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ABSTRACT

Introduction and importance: Hydatid disease, caused by the zoonotic parasite *Echinococcus granulosus*, presents as cysts most commonly in the liver and the lungs. A hepato-pleural fistula is considered one of its complications and surgery remains the mainstay of treatment. A simultaneous invasive thoracic and abdominal access is routinely needed which is accompanied by increased morbidity and mortality.

Case presentation: In this case report, we present an innovative, less invasive, non-classical approach.

Clinical discussion: We introduce a successful transabdominal transdiaphragmatic laparoscopic technique for the management of a hepato-pulmonary fistula secondary to hydatid disease of the liver.

Conclusion: Compared to the classical method, this new surgical approach towards hepato-pulmonary fistula paves the way for minimally invasive surgeries to manage complicated hydatid disease with a lower mortality and morbidity, faster recovery and shorter hospital stay. The work has been reported in line with the SCARE 2020 criteria.

1. Introduction

Hydatid disease is considered a remarkable health problem worldwide, mostly endemic in developing and underdeveloped countries such as the Middle East and Africa. It is caused by the zoonotic parasite *Echinococcus granulosus* [1] presenting as cysts most commonly in the liver (50–80%) and lungs (10–40%) [2]. Several complications of hydatid disease of the liver may develop, one of which is formation of a hepato-pleural fistula. Hepato-pleural fistula is considered one of the reported complications and includes the formation of a fistula between the liver parenchyma and the pleural space [3]. Surgery remains the mainstay of treatment of hydatid disease, especially in established cases of hepato-pleural fistula [4]. It has been traditionally done through a trans-thoracic approach in addition to an abdominal access. However, it was associated with high morbidity and mortality especially in developing countries that have modest expertise in this field [1]. Recently, less invasive surgical approaches have been advocated for management of hepato-pleural fistula [5]. Here, we report successful transabdominal

transdiaphragmatic laparoscopic approach to manage liver hydatid cyst with hepato-pleural fistula yielding satisfactory outcomes. The work has been reported in line with the SCARE 2020 criteria [17].

2. Case presentation

We report a case of a 51-year-old female housewife who was referred from an outside physician to our clinic at the American University of Beirut Medical Center for intermittent episodes of dyspnea and nonproductive cough of 2 months duration. Patient reported no fever, chills, night sweats, weight loss, chest pain, palpitations or hemoptysis. She also denied any abdominal pain, change in bowel habits, dysphagia, vomiting, jaundice, or tea colored urine/clay colored stools. Patient is also known to have hypothyroidism and diabetes mellitus type II and is a former smoker. She does not have any past surgical history. To mention, this patient is an inhabitant of a rural area close to a farm in Lebanon.

On presentation, the patient was awake, alert and oriented to person, place and time. Physical exam revealed stable vital signs with decreased

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breathing sounds on the right lower lung field with no wheezing. Her abdomen was soft, non-distended with no rebound tenderness or any peritoneal signs, and murphy sign was negative. Baseline investigation was unremarkable with normal complete blood count levels except for mild eosinophilia (5%), normal basic metabolic panel, liver function tests and coagulation profile. A plain antero-posterior chest radiogram (CXR) was performed showing blunting at the Right costophrenic angle (Fig. 1). Further evaluation with a computed tomography (CT) scan of the chest and abdomen/pelvis was done. CT revealed 2 large cystic lesions (measuring 7×8 cm and 10×7.5 cm) in the right lobe of the liver, communicating, and containing several irregularly shaped daughter cysts within the mother cyst, with no calcifications. Communication with the pleural space was noted with a right lower lobe consolidation. A suspicion of hydatid cyst was confirmed by a hydatid indirect hemagglutination test with a value $>1:32768$.

Patient was started on albendazole and was scheduled for laparoscopic evacuation of liver hydatid cyst with transabdominal transdiaphragmatic laparoscopic approach of the hepato-pleural fistula.

Intraoperatively, general abdominal laparoscopy was performed by a general surgeon specialized Liver Transplantation and Hepatopancreaticobiliary Surgery, through a 10 mm umbilical port. Respectively, a 5 mm trocar was inserted in the epigastrium, 12 mm trocar in the mid right sub-costal area (retrieval bag and scope if needed) and a 5 mm trocar in the lateral right sub costal area (Fig. 2). During inspection, two large hydatid peri-cysts were identified arising from the hepatic dome adhering to the supra-hepatic diaphragm. The cyst contents were managed laparoscopically via closed circuit of tubing system by aspirating 100 cc of clear intra-cystic fluid and replaced by similar volume of hypertonic saline as a scolicial agent. Using electrocautery, a small opening was done in the wall of the cyst that allowed introduction of the suction where the cyst was aspirated till dryness and repeatedly irrigated with hypertonic saline (Fig. 3). The peri-cysts were divided completely from the diaphragmatic surfaces. This allowed management of the hepatic hydatid by excising the fibrotic component of the pericysts, retrieving the membranes and thoroughly inspecting the potential existence of bile leak or cysto-biliary fistula (Fig. 4). Then, the intra-thoracic component of the hydatid was managed via the diaphragmatic communication through the same abdominal approach, where the membranes were retrieved, and cyst cavity was irrigated and washed. There was definite pleural communication and erosion to the lower surface of the lower right pulmonary lobe with tiny air leak but no

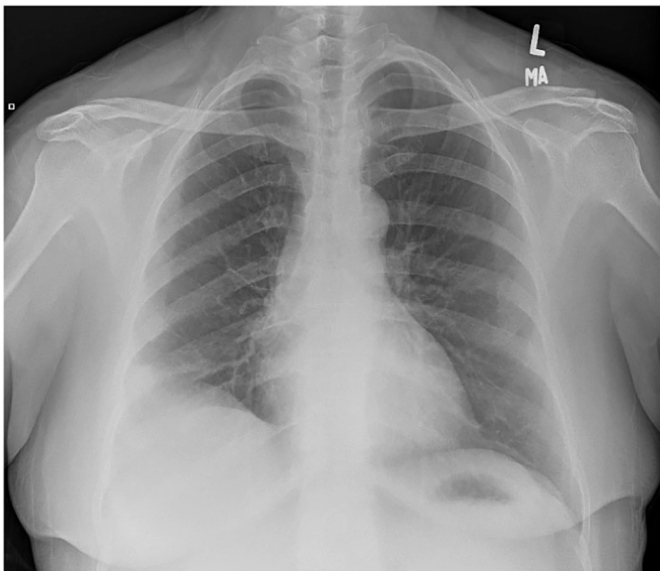


Fig. 1. Chest X-ray preoperatively with blunting at the Right costophrenic angle.

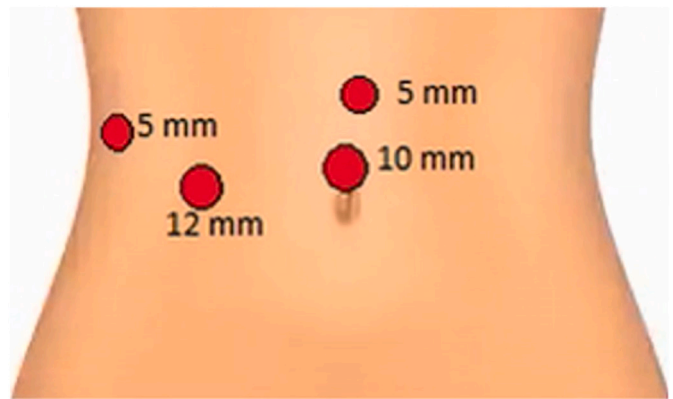


Fig. 2. Surgical planning with laparoscopic transabdominal approach.

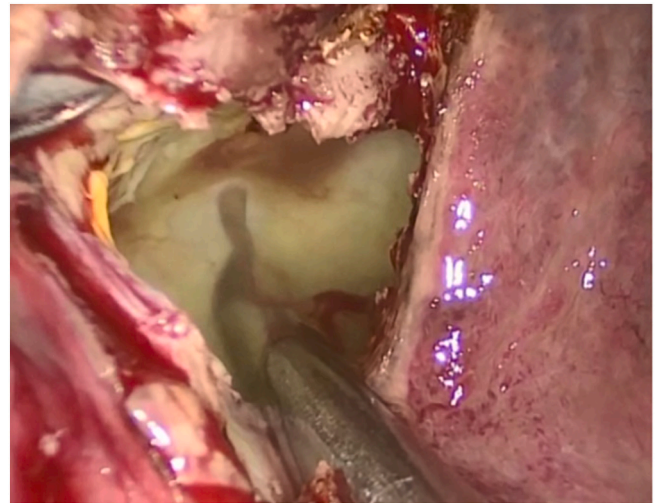


Fig. 3. Evacuation of the daughter cysts and protoscolices.



Fig. 4. Drainage of the hydatid liver cyst with its pleural component.

communication to major bronchioles as it was identified by air water tests. And an intra-abdominal drain was inserted to drain both the chest and hepatic cavities (Fig. 5). There was no need for right chest tube insertion, since the lower surface of the diaphragm was inseparable from

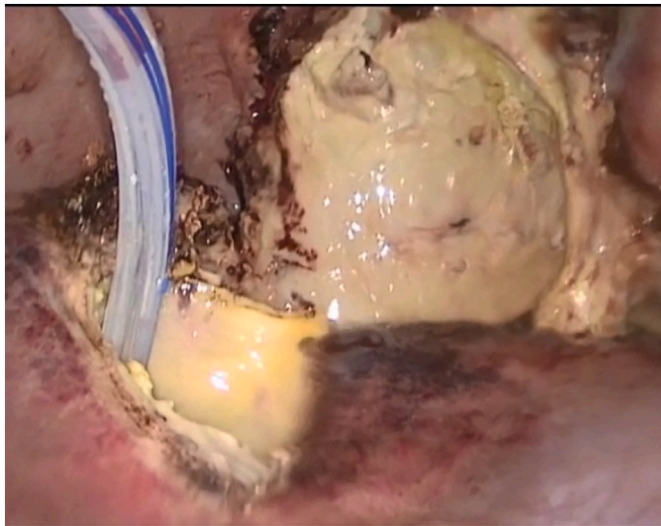


Fig. 5. Photographic demonstration of the pleuro-biliary fistula after evacuation and drainage.

the lung due to the inflammatory process. Drained specimen and tissue were sent out for pathology and the report could be seen in Fig. 6.

Postoperatively, the patient was doing well. Biliious drainage was noted on postop day 2, and a biliary leak was suspected. ERCP with sphincterotomy was performed to decrease biliary pressure, and a 10 Fr x 10 cm plastic stent was inserted into the biliary tree. Drain output was followed up. At postoperative day 8, patient was doing well, and she was discharged home on albendazole, and the drain was removed.

3. Discussion

Hydatid disease is endemic in agricultural and animal raising societies especially in the Middle East. This disease carries significant burden on public health and economy [6]. For all cysts that are larger than 15 cm, those that are complicated or those that are not suitable for

interventional radiology (IR), surgical approach is the mainstay treatment [6].

One of the major complications of the hydatid cyst of the liver is its rupture into the pleura and bronchial tree. This happens either due to the pressure from the expanding hydatid cyst leading to erosion into the diaphragm or from the destructive effect of a superimposed infection [7]. The most prominent sign is biliopneumothorax. However, often patients may have nonspecific respiratory symptoms for weeks or months, as the case of our patient. In the past years, different invasive surgical approaches have been used for hydatid cysts with a pneumo-biliary fistula. They comprise methods of gaining access either through the abdomen or through the lung, or both [8–10]. Transthoracic approach is the surgical method of choice via posterolateral thoracotomy; however, sometimes thoraco-laparotomy or thoraco-phreno-laparotomy is performed. On the other hand, reports of minimally invasive approaches have been published like performing ERCP with micro-coils and ERCP with fibrin sealing [11,12].

Indeed, advancement of laparoscopic surgery provides a technical basis for minimally invasive surgery for hydatid cysts with less morbidity, rapid recovery and a shorter hospital stay compared to conventional open procedures [13,14]. There are no reported case reports in the literature for the management of hepato-pleural fistula through a laparoscopic approach [15,16]. Here, we introduce a successful transabdominal laparoscopic approach for the management of hepato-pleural fistula.

It is crucial to highlight that the understanding of the pathophysiology of the disease allows for the feasibility of our presented surgical approach. This is due to the fact that when the hydatid cyst in liver erodes from the dome of the liver through the diaphragm, then the pleura and reaches the lung, it forms a fibrotic tract with no communication with the surrounding structures. This allows surgeons to reach the lung through an abdominal approach without injuring the biliary structures around it. Moreover, such an approach offers several advantages. First, there is no need for posterolateral thoracotomy with this approach. Second, the use of laparoscopy will allow reduced length of incision and minimal abdominal trauma and abdominal adhesions. Moreover, during the surgery, the magnified view of laparoscopy and the use of trocars will reduce the complications such as bleeding and bile leakage and maximize the preservation of normal liver tissue [13]. In addition, patients can resume diet earlier after surgery and have a shorter hospital stay [13]. To note, verbal consent was provided by the patient for write-up of this case report.

Provenance and peer review

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Ethical approval

The write up of this case report was approved by the institutional review board (IRB).

Consent

Verbal consent was provided by the patient for write-up of this case report.

Author contributions

All authors contributed to: Conception of the work, Design of the work, Drafting the work and Revising the work critically for important intellectual content, Final approval of the version to be published, and

Tissue Exam:	Order:
Collected	Status: Final result
Component	
Diagnosis	
<p>LIVER, PARTIAL HEPATECTOMY: – HYDATID CYSTS AND SCOLICES. – FIBROTIC TISSUE WITH NECROSIS AND INFLAMMATION.</p>	
Electronically signed by	
Gross Description	
<p>Intrahepatic hydatid cysts. Formalin. Received are multiple flat pieces of cystic tan-white translucent tissue measuring 0.1 in thickness and 7.8 x 6.5 x 2 cm in aggregate. Also received are pieces of congested and focally fibrotic tissue measuring 4.2 x 3 x 1.4 cm in aggregate. Partially submitted.</p>	
Summary of sections	
1A fibrotic tissue	
1B cystic tissue	

Fig. 6. Intra-operative tissue exam pathology report.

Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Research registration

NA.

Guarantor

The corresponding author is the guarantor of submission.

Declaration of competing interest

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