



Management for Ruptured Liver Hydatid Cysts in the Chest: Experience of a Moroccan Center

Sani Rabiou^{1*}, Hicham Harmouchi¹, Layla Belliraj¹, Fatima Z Ammor¹, Ibrahim Issoufou¹, Kassim Sidibé², Baladougou Sylla³, Marouane Lakranbi¹, Dafarallah Benajah^{3,4}, Yassine Ouadnoui^{1,4} and Mohamed Smahi^{1,4}

¹Department of Thoracic Surgery, CHU Hassan II, Fez, Morocco

²Department of Imagery, CHU Hassan II, Fez, Morocco

³Department of Hepato-gastrology, CHU Hassan II, Fez, Morocco

⁴Department of Medicine and Pharmacy, Sidi-Mohamed-Ben-Abdellah University, Fez, Morocco

Abstract

Introduction: Hepatic hydatidosis is still a sanitary scourge in Morocco. This benign condition in itself can evolve into serious complications, the rupture of which in the thorax is an example. The management is cumbersome, requiring a perfect collaboration between many specialists.

Material and Method: A retrospective study on the intra-thoracic ruptures of the hydatid cysts of the liver at the CHU Hassan II of Fez over a period of 8 years. Patients were split into 2 groups depending on the nature of the treatment. In the group of patients operated on, the decision was made after a thorough preparation. Preoperative endoscopic sphincterotomy was systematic in the presence of a bilio-bronchial fistula with significant biliptysis. For patients with a contraindication to anesthesia, the treatment was exclusively endoscopic by sphincterotomy.

Results: A total of 31 patients were included in the study (18 women and 13 men), of whom 14 had at least one history of hydatid cyst surgery. Clinical signs were dominated by cough and biliptysis. Twenty eight patients had been operated on and the approach was an exclusive right thoracotomy in all cases. Patients were grouped according to the classification of Mestiri: 13 patients were grade IA, 9 grade IIA, 3 grade IVA, 2 grade IIIC, and a single grade IIIA. Thoracotomy was used to treat lesions in the thoracic, diaphragmatic and abdominal stages in all patients. The operative sequences were marked by the occurrence of a persistent hepatic abscess of the residual cavity in 2 patients controlled by antibiotic therapy and the maintenance of drain in hepato-diaphragmatic intervals during 4 weeks. We have deplored 2 postoperative deaths following a septic shock. In 3 patients with a contraindication to general anesthesia, we opted for an exclusive endoscopic treatment which allowed to completely releasing the bile duct with disappearance of the biliptysis. After an average follow-up of 38 months no case of recurrence was noted.

Conclusion: Through this study, we wanted to highlight the complexity of the management of liver hydatid cysts ruptured in the thorax whose surgery remains the best treatment at the cost of a heavy morbidity and mortality. This morbidity and mortality can be improved by early and accurate diagnosis, but especially by appropriate pre- and postoperative resuscitation measures.

Keywords: Hydatid cyst; Liver; Thorax; Complication; Thoracotomy; Sphincterotomy

Introduction

Intra-thoracic rupture of liver hydatid cysts is a rare clinical entity, since it affects only 0.6% to 16% of cases [1,2]. The association with a bilio-bronchial fistula remains formidable and accounts for the difficulty of management. The treatment of choice remains surgical, and the thoracic approach allows a better approach on the three stages (thoracic, diaphragmatic and abdominal). The introduction of endoscopic sphincterotomy has radically improved the results of this surgery in terms of morbidity and mortality. The aim of this study was to clarify the contribution of endoscopic sphincterotomy while reporting the results of surgical treatment of ruptured liver hydatid cysts in the thorax by exclusive thoracotomy.

Materials and Methods

This was a retrospective study carried out in the department of thoracic surgery of the CHU

OPEN ACCESS

*Correspondence:

Sani Rabiou, Department of thoracic surgery C1, CHU Hassan II, Fès, Morocco, Tel: +212680772220; E-mail: rabiou sani2@icloud.com

Received Date: 02 Sep 2017

Accepted Date: 15 Nov 2017

Published Date: 23 Nov 2017

Citation:

Rabiou S, Harmouchi H, Belliraj L, Ammor FZ, Issoufou I, Sidibé K, et al. Management for Ruptured Liver Hydatid Cysts in the Chest: Experience of a Moroccan Center. *Clin Surg*. 2017; 2: 1757.

Copyright © 2017 Sani Rabiou. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.



Figure 1: Biliptysis collected from a patient.



Figure 2: Thoracic tomodensitometry showing a multi-partitioned hydatid cyst in the ruptured liver in the thorax.



Figure 3: Thoracic tomodensitometry: Lower right lobular pulmonary lesion, rounded, surrounded by a hyperdense wall containing some air bubbles, associated with an encysted right pleural effusion. Note also the presence of a cyst in the liver level seeming to continue with the lung lesion.



Figure 4: CT scan of a ruptured liver hydatid cyst in the thorax. Note the presence of a pleural effusion with significant pneumopathy of the right lower lobe.

Hassan II of Fez from January 2009 to March 2016 that is an 8 years' duration. We included 31 patients, supported for ruptured liver hydatid cyst in the thorax. For each patient, once hospitalized, the

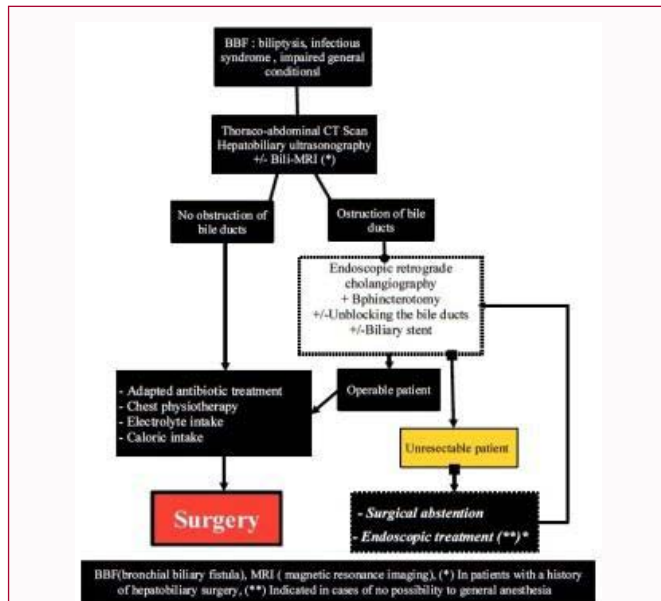


Figure 5: Conduct in front of a ruptured hydatid cyst in the thorax with biliobronchial fistula.

clinical examination sought essentially the history of surgery for liver or pulmonary hydatid cysts, or the combination of two. The diagnosis was based on clinical examination (presence of biliptysis) supplemented by the results of the thoracic and abdominal CT scans showing images in favor of a liver hydatid cyst appearing to continue with intra-thoracic lesions. In the presence of biliptysis, we carried out its daily quantification and then a hepatobiliary ultrasound scan supplemented the balance by looking for the presence of an obstruction of the bile duct by hydatid material. Thus, in case of obstruction of the biliary tract, an endoscopic sphincterotomy was indicated preoperatively with the objective of restoring normal biliary flow. It was only after proper preparation by correction of hydro-electrolyte, nutritional and infectious disorders that the surgery had been proposed to the patients. In case of contraindication to general anesthesia, we opted for exclusively endoscopic treatment. Thus, for each patient, an information sheet was used, including the epidemiological data (age, sex), clinical (underlying terrain or surgical history) and para-clinical (imaging). The data was entered and analyzed using the Excel software.

Results

There were 18 women and 13 men, with an average age of 40 years, with outsiders between 17 and 81 years. Fourteen patients had at least one history of hydatid disease surgery. Clinical symptomatology was dominated by a cough in 50% of cases, whereas biliptysis was only found in 13 patients (Figure 1). We noted a bilio-cutaneous fistula in one case. One patient had been admitted to a table of febrile jaundice. Clinical symptomatology was associated with biological cholestasis in 9 cases. The thoracoabdominal computed tomography (CT) performed in all patients had consistently shown a hepatic cystic lesion appearing to continue with a thoracic lesion that varies according to patients (Figures 2-4). Bronchial fibroscopy performed in only 11 patients showed biliptysis in 4 cases, with evidence of a hydatid membrane in a patient. Preoperative retrograde endoscopic cholangiography was reported in only 5 patients, all of whom had significant biliptysis at admission. It allowed to completely releasing the biliary tract by sphincterotomy with hydatid membrane extraction

in 3 patients and placement of a biliary prosthesis in one case.

Surgical treatment

Surgical treatment involved 28 patients. The first approach was an exclusive posterolateral right thoracotomy in all cases. Table 1 gives the grouping of patients according to the classification of Mestiri. This approach initially made it possible initially to perform the phreno-pulmonary and hepato-diaphragmatic disconnection and then the treatment of lung lesions and through the diaphragmatic breccia lesions of the liver. The gestures performed at the pulmonary and hepatic levels are summarized in Table 2. Diaphragmatic closure was achieved without the need for prosthesis in all patients. This was a direct suture of the diaphragmatic breccia after mobilization of the holy diaphragm at the level of the costo-diaphragmatic sac. The operative sequences were marked by the occurrence of a persistent hepatic abscess of the residual cavity in 2 patients controlled by antibiotic therapy and the maintenance of drain in hepato-diaphragmatic intervals during 4 weeks. We deplored 2 cases of postoperative death following a septic shock. In other cases the surgical follow-up was simple and no case of recurrence was noted after a follow-up of an average duration of 38 months.

Endoscopic treatment

There were 3 patients with a contraindication to general anesthesia. The clinical picture involved a biliptysis associated with an alteration of the general state in all these patients. Ultrasound and cholangiography were in favor of dilatation of the bile ducts in these cases. The management was exclusively endoscopic by sphincterotomy with extraction of hydatid membranes thus releasing the bile duct (Figure 5). Post-endoscopic surveillance consisted of the quantification of biliptysis which had decreased by more than half in the first 48 hr in all patients. An antibiotic therapy with a respiratory physiotherapy had been instituted throughout the duration of hospitalization with a good evolution. Patients were reviewed once a week for the first month and then once a month for the first semester. With an average follow-up of 10 months, there were no complications or deaths in this group.

Discussion

The hydatid cyst of the ruptured liver in the thoracic cavity is a rarely reported clinical entity whose frequency appears to decline from 16% [3] to 2.5% [4]. The better knowledge of hydatidosis and the availability of more and more precise means of investigation such as abdominal ultrasound seem to have a role in the regression of this frequency by allowing an early management of hydatid cysts of the liver, avoiding its evolution at the stage of complications. It is a serious complication of the hydatid cysts of the liver due to the multiplicity of lesions which at the same time concern the abdominal and thoracic stages via the diaphragm [5-7]. These are, in most cases, very old hydatid cysts located in the hepatic dome. The association with lesions of the bile duct is a determining factor in the evolution of this disease. It may be either a simple lesion of the fine canaliculi or the open and wide rupture of the hydatid cyst of the liver in the main bile duct. It follows then the migration of the daughter vesicles and the debris of hydatid membranes in these biliary tracts causing the obstruction of the latter with appearance of the clinical signs like the jaundice. The occurrence of infection of the hepatic cyst, the formation of the intracystic putrid porridge containing bladed hydatid vesicles, fragments of membranes, pus and bile, will lead to a peri-cystic inflammatory state responsible

Table 1: Distribution of the 28 operated patients according to the Mestiri classification.

Classification of Mestiri	Number of patients
Grade IA	13
Grade IIA	9
Grade IVA	3
Grade IIIC	2
Grade IIIA	1

Table 2: The different gestures performed at the pulmonary and hepatic level.

Treatment of pulmonary lesion
<ul style="list-style-type: none"> Lobectomy (2) Segmentectomy (3) Sous segmentectomy (3) Atypique resection (10) Kystectomy (4) Peikystectomy (1) Pulmonary decortication only* (5) (*Pulmonary decortication is associated with all pulmonary gestures)
Treatment of hepatic lesion
<ul style="list-style-type: none"> Perikystectomy+capitonnage of résiduel cavity (1) Resection of Dome protruding+capitonnage of résiduel cavity (5) Resection of Dome protruding+Épiploplasty (1) Resection of Dome protruding (11) Kystectomy (6) Perikystectomy (4)

Table 3: Classification of Mestiri.

Classification of Mestiri and al. [6]
Type I: direct fistulization of the cyst in the bronchi
◦IA: small-sized bronchial fistula
◦IB: large-caliber bronchial fistula
Type II: intrapulmonary cavern
◦IIA: without bronchial fistula or bronchiolar fistula
◦IIB: with large bronchial fistula
Type III: encysted intrapleural intermediate poucket
◦IIIA: without bronchial fistula
◦IIIB: with bronchial fistula
◦IIIC: with fistula on the wall
Type IV: rupture in the large pleural cavity
◦IVA: acute rupture: biliohydatidic pleurisy
◦IVB: secondary pleural hydatidosis

for the formation of adhesion between the liver and the diaphragm with constitution of a lobar hepatitis or segmental hydatid. The intimate contact of this hepatic dome with the diaphragm favors the crossing of the diaphragmatic barrier by the hydatid cysts of the liver sometimes resulting in the rupture of these cysts in the pleura or in a lobe lung. Thus, the intrathoracic evolution of these lesions will depend on several factors [2,8,9]: the intimate contact of the hepatic dome with the diaphragmatic dome; The thoracoabdominal pressure gradient which tends to aspirate the contents of the hydatid cyst of the liver towards the thoracic cavity; The erosion of the diaphragm by ischemia-necrosis caused by the eventual superinfection of the cyst and the inflammatory phenomena; The corrosion of all the tissues in contact with the bile [3]. The cyst, after having passed through the diaphragm, can be fistulized in the bronchi, generally of the lower and/or middle lobe, rarely of the upper lobe or of the lingula [9]. The bile will cause by caustic action on the lung or the bronchial tree, lesions ranging from simple hydatid pneumonia to the constitution of a cave, or even to the total destruction of the pulmonary parenchyma [5,9]. These lesional aspects made it possible to define several classifications, including that of Mestiri et al. (Table 3) [6]. This classification emphasizes the importance of small bronchial fistulas without neglecting biliary fistulas [4,6]. Indeed, these biliary lesions are at the origin of the super infection of the cyst of the liver, which

through the intermediary of the bilio-bronchial fistulas will spread to the level of the lung. In addition, there is no parallelism between bronchial and biliary lesions [10]. Our retrospective study, carried out over a period of 8 years, is marked by the improvement of the diagnostic means, the evolution towards the choice of a first thoracic approach and the reduction of the mortality of the hydatid cysts of the liver ruptured in the thorax. Several approaches have been proposed for this surgery: thoracotomy alone, thoraco-phrenolaparotomy, laparotomy alone or associated with thoracotomy [4,8,11]. For some authors, the abdominal route is often sufficient to control all lesions and reserve the thoracotomy for pleuropulmonary lesions which do not regress after disconnection of the liver to the diaphragm by the abdominal route alone [7]. The choice of the approach is always a matter of controversy. In fact the maximum comfort and safety is obtained with the thoracotomy because of the important lesions of the pulmonary parenchyma which we have observed, often requiring a regulated pulmonary resection such as lobectomy or segmentectomy [7,12]. In our study, the thoracotomy made it possible to perform a phreno-pulmonary disconnection and then to treat pulmonary lesions. After the hepato-diaphragmatic disconnection, treatment of hepatic lesions, hepato-diaphragmatic drainage is carried out through the diaphragmatic breccia. Phrenoplasty is made possible after resection of all the sclerotic parts and release of the costo-diaphragmatic sac asses to mobilize the rest of the diaphragm thus giving a suture without tension. We did not use the interposition of prosthesis to fill the diaphragmatic breccia. In the study carried out by S. Msaad et al. [13], the first approach was a right subcostal laparotomy in 3 patients, an exclusive thoracotomy in 1 case, and a thoracoabdominal approach in another case. As in the case of our 3 patients, we believe that exclusive endoscopic sphincterotomy may be an alternative in some non-operable patients (Figure 5). It allows unclogging the bile duct in order to establish a normal biliary flow [14].

Conclusion

Despite improved resuscitation and surgical techniques, intra-thoracic rupture of hydatid cysts retains a severe prognosis. The introduction of more and more per-endoscopic sphincterotomy in preoperative surgery has improved the prognosis and even treated exclusively some non-operable patients. Thoracotomy is an excellent first-line treatment for thoracic, abdominal and diaphragmatic lesions. It reduces not only the duration of hospitalization but also the cost of overall care.

Authors' Contribution

All authors contributed to the study design. Sani Rabiou, Hicham Harmouchi and Marouane Lakranbi performed data collection and data analysis. Ibrahim Issoufou, Layla Bellirej, Balladougou Sylla and Fatiam Z Ammor performed the cost analysis. All authors

critically interpreted all data analysis. Yassine Ouadnoui, Dafarallah Benajah and Mohamed Smahi composed the manuscript, and all the remaining authors provided critical edits to the final draft. All authors read and approved the final manuscript.

References

- Gomez R, Moreno E, Loinaz C, De la Calle A, Castellon C, Manzanera M, et al. Diaphragmatic or transdiaphragmatic thoracic involvement in hepatic hydatid disease: surgical trends and classification. *World J Surg.* 1995;19(5):714-9.
- Kilani T, Daoues A, Horchani H, Sellami M. Place of thoracotomy in thoracic complications of liver hydatid cysts. *Ann Chir Thorac Cardiovasc* 1991;45:705-10.
- Goinard P, Pelissier G. Bilio-bronchial fistula of hydatid origin. *Mem Acad Chir.* 1965;91(10):383-6.
- Abi F, el Fares F, Khaiz D, Bouzidi A. Localisations inhabituelles des kystes hydatiques. Unusual localizations of hydatid cysts. Apropos of 40 cases. *J Chir.* 1989;126(5):307-12.
- Moumen M, El Fares F. Bilio-bronchial fistula of hydatid origin. Apropos of 8 cases. *J Chir.* 1991;128(4):188-92.
- Mestiri S, Kilani T, Thameur H. Thoracic migrations of hydatid cysts of the liver: proposal for a classification. *Lyon Chir.* 1987;83:12-6.
- Sakhri J, Benali A, Letaief R, Derbel F, Dahmen Y, Ben Hadj Hmida R.. Les kystes hydatiques du foie rompus dans le thorax : aspects diagnostiques et thérapeutiques. *J Chir.* 1996;133(9-10):437-41.
- Karydakos P, Pierrakakis S, Economou N, Ninos A, Raitsiou B, Bobotis E, et al. Traitement chirurgical des ruptures des kystes hydatiques du foie. *J Chir.* 1994;131(8-9):363-70.
- Chehab F, Khaiz D, Lakhoulfi A. Bilio-bronchial fistula of hydatid origin in 9 cases. *Sem Hôp.* 1997;73:800-4.
- Kilani T, El Hammami S, Horchani H, Ben Miled-Mrad K, Hantous S, Mestiri I, et al. Hydatid disease of the liver with thoracic involvement. *World J Surg.* 2001;25(1):40-5.
- Bouzidi A, Chehab F. Surgical treatment of biliary cystic fistula caused by hydatid cysts. Apropos of 83 cases. *J Chir.* 1997;134(3):114-8.
- Msougar Y, Lakranbi M, Bouchikh Y, Ouadnoui Y, Maida M, Fenan H, et al. Outcomes after thoracotomy for intra-abdominal hydatid cysts ruptured into the thorax (a 160 case series). *Rev Mal Respir.* 2010;27(5):417-20.
- Msaad S, Yangui I, Ketata W, Abid N, Feki W, Abid H, et al. Hydatid cysts of the liver ruptured into the thorax (about five cases). *Rev pneumol clin.* 2015;(75)5:255-63.
- Rabiou S, Lakranbi M, Ouadnoui Y, Panaro F, Smahi M. Surgical management of hydatid Bilio-bronchial fistula by exclusive thoracotomy. *Int J Surg.* 2017;(41):112-18.