

RESEARCH ARTICLE

DIAPHRAGMATIC HYDATID CYST: DIAGNOSTIC CHALLENGE FOR A RARE PATHOLOGY

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Manuscript Info

Abstract

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..... Hydatid disease mainly affect liver and lung. The incidence of diaphragmatic hydatid cysts, isolated or involving other organs, is very rare, accounting for 1% of the thoracic locations. They may be operative discovery or by their complication. Preoperative diagnosis of hydatid cysts located at the diaphragm is challenging. And surgery remains the only therapeutic approach. The prognosis is generally good apart from the risk of recurrence. We present a rare case of a 44-yearold female, with previous history of liver hydatidosis, who presented to our attention with right hypochondrium pain with fever. The diagnosis was fortuitous during the surgical exploration, while preoperatively, CT scan showed a cyst apparently located on segment VIII of the liver, and announced the hypothesis of fistula between this cyst and the lung. After imagistic investigations we decided for an abdominal approach considering that the cyst was liver related. But after dissection of the adherences with the liver, the cyst remained attached to the diaphragm. The treatment of choice was surgical removal of the cyst. And the patient is now being closely followed up.

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Introduction:-

The hydatid disease, is an endemic disease in many regions worldwide, due to the Echinococcus (E) tapeworm infestation.

Liver and lung are the most common sites of the disease. Hydatid cyst of the diaphragm is rarely reported in the literature with the incidence of around 1%, and generally associated with liver hydatidosis **[1]**.

This case highlights the difficulty of preoperative diagnosis, and the different possible curative approaches.

Case Report:

A 44-year-old female, who hadundergone surgery for a liver hydatid cyst at age of 26 years old.

The second surgery was for Peritoneal Hydatidosis one year ago, revealed by a symptomatology of severe abdominal pain with an infectious syndrome. Surgical procedure was performed, including both radical and conservative treatment for peritoneal and liver hydatid cysts with peritoneal drainage.

And Albendazole treatment was started for 6 months duration.

Currently The patient was admitted in our department for permanent right thoracoabdominal pain for more than 2 months despite the analgesic oral treatment, and recent occasional fever spikes. all evolving in a context of weight loss of 5 kg in two months.

At admission the patient was in good general condition, with low-grade fever and without clinical respiratory manifestations, despite suffering of right thoracoabdominal pain. Two hours after admission and administration of analgesic drugs, the patient was already pain free.

The laboratory tests showed normal results of the serum examinations, except positive hydatid serologies. The white blood cells were normal and no eosinophilia was found.

Chest X-ray performed at admittance showed right pleural effusion. And revealed right-sided subpulmonic opacity with elevation of the right hemidiaphragm. (Fig a)

The CT scan performed the day after, showed cystic lesion of 10cm x 12cm in diameter affecting apparently segment VIII of the liver, extending from the abdominal cavity into the right thoracic cavity, described as hydatid liver cyst fistulized in the right lung. Near to this cyst, there was at the segment III and IV, two smaller ones. (Fig b)

As it was considered that we deal with a liver cyst, in addition to other peritoneal hydatid cysts, we have decided for an abdominal approach.

An exploratory laparotomy was performed, during which time, in the beginning we discovered after a difficult dissection, an infected space between the liver and the right abdominal wall, with pus, also visible on the CT, whose origin was a silk thread used in the second surgery for fixing the drain. Which explains the episodes of recent fever.

This space has been washed and drained. Then we continue the careful dissection, and we discover a hydatid cyst (Fig c) between the muscle fibers of the diaphragm, without liver or lung involvement. During the section of the adherences between the right lobe of the liver and the diaphragm, the hydatid cyst began to separate from the liver and remained attached to the diaphragm (Fig c). So, it was clear that the cyst originated from the diaphragmatic muscle.

The cyst was easily isolated. Then a cystotomy was performed, and the cyst membranes were removed, with using next, of hydrogen peroxide. And we found no communication between the diaphragmatic cavity and the thorax, which eliminates the CT fistula hypothesis in the thoracic cavity.

The surgery also removed the two other liver cysts, well documented at the preoperative CT scan, by using sterilization of hydatid cysts with the hydrogen peroxide after unroofing, and removal of cyst membranes.

Drainage was performed in the three residual cavities.

The other cysts have been spared, and additional treatment is provided by medical treatment for at least 6 months.

The postoperative recovery was uneventful, and chest X-ray did not objectify any pneumothorax.

The patient's tube drainage was removed on postoperative Day 5. There was no bile fistula. The patient was discharged on postoperative Day 7.

Discussion:-

Hydatidosis is still an endemic disease in several regions of the world, because of the close association of human with sheeps and dogs.

The hydatid cyst may evolve in many organs separately [2]. And the localizations in organs other than liver and lung are less frequent [3].

The diaphragmatic localization of hydatid cysts is rare, and has no particular age or gender [4,5,6].

For primitive diaphragmatic localization, it's possible when the embryos reaches that site by arterial or lymphatic circulation [7].

Otherwise, according to Pinna et al., for cases with combined diaphragmatic and liver hydatidosis, the cyst reaches the diaphragm by direct extension from the liver [8]. And more rarely, as in our case, the transplant is secondary to the rupture of a pulmonary or hepatic hydatid cyst. And It can be associated with other intra or extra thoracic localizations.

During its evolution, the diaphragm hydatid cyst can rupture in the pleura, bronchi or in the abdominal cavity, become infected or compress the adjacent organs. Clinically, the uncomplicated forms are asymptomatic and incidentally discovered in 20% of cases. When it's symptomatic, it can manifest as basithoracic pain radiating to the shoulder and sometimes respiratory discomfort or irritative cough **[9,10]**, which was the case with our patient.

For the diagnosis, serological tests are not always helpful [11], but were positif in our case.

The diagnosis is first of all, very challenging for this localization.

The existence of a cyst lying between the thorax and the abdomen represents a great challenge for the radiologist in determining whether it originates from the lung, the diaphragm or the liver.

The difficulty in identifying the exact anatomic location of diaphragmatic hydatid cyst may lead to its misdiagnosis as hydatid cyst of the liver, intrahepatic simple cyst or as other diaphragmatic cystic lesions especially mesothelial cysts and bronchogenic cysts [12].

CT is more accurate in determining the diaphragmatic origin of the cyst **[13].** And It helps detection of other thoraco-abdominal localizations or cyst complications. Which in our case, CT suspected a fistula between the segment VIII cyst and the lower right lobe of the lung.

Otherwise the synchronous trans-diaphragmatic extension of a hepatic hydatid cyst to the right lung is uncommon [14].

MRI and US might be helpful in some cases [13].

But in the end, almost all reported cases of isolated diaphragmatic hydatid cyst were diagnosed as a liver or lung hydatid cyst pre-operatively and found to be diaphragmatic during surgical exploration. [3,15,4], which demonstrate the difficulty of diagnosis.

For diaphragmatic hydatid cyst, Surgery remains the mainstay treatment, although it has to be as conservative as possible [16]. And it remains the treatment of choice for complicated cysts, cysts with multiple daughter cysts, those with biliary communications, or those with calcified walls [17].

Concerning medical treatment with Albendazole, it's used for inoperable patients or for decreasing the recurrence after surgery [18,19]. And there are several controversial studies on the number of phases and the duration of treatment administration [20]. In our department, we have adopted a treatment of at least 6 months.

Finally, the preoperative diagnosis of diaphragmatic localization, despite the fact that it is difficult, remains very useful to adapt the surgical procedure. Indeed, surgery is most often conservative and consists in unroofing especially for large cysts for which radical cure will expose to phrenic repair difficulties [15].

Surgical approach could be most frequently abdominal laparotomy or thoracophreno laparotomy. Laparoscopic approach is possible as well, but rarely used.

For cases with isolated diaphragmatic cyst, resection of the cyst and pericyst with repair of the diaphragm through thoracotomy is the main line of therapy [15]. And this surgical approach could be indicated in the event of the coexistence of liver hydatid disease and pulmonary hydatid involvement in other segments.

The abdominal laparotomy approach was preferred in many studies, for many reasons: 1- abdominal incision provides adequate exposure and the possibility of liberating the liver, allowing more complete removal of the liver cysts. 2- It makes easier the exploration of the bile duct. 3- cyst adherences to the pulmonary parenchyma, if required, can be either dissected or atypically resected through a diaphragmatic aperture. 4- increased morbidity from thoracophrenolaparotomy can be avoided. In the same way, Warrenet al. [21], and MoumenandEl [22], have stated their preference for the abdominal laparotomy approach for resolving bilio bronchial fistular pathology (including pulmonary resection).

in our case, the abdominal laparotomy was preferred, for the reason of presence of multiple abdominal hydatic cysts, the presence of multiple hepatic hydatic cysts, and the suspicion of the fistula between the hepatic cyst and the right lung on CT.

Both radical and conservative surgical treatment can be performed.

In our case, for the diaphragmatic cyst, conservative strategy was maintained, involving the removal of the cyst content and sterilization of the residual cavity. For the cyst, we have chosen a cystotomy instead of partial cyst resection, without closing the diaphragmatic rent, and considering the fibrous hull as a cure to the defect. Which did help us evade a large diaphragmatic dissection, loss of muscle capital in the cyst cavity, and a complex reconstruction. Similar procedure was described in several cases with no negative impact on patients [23].

Conclusion:-

Trans-diaphragmatic hydatid cyst is rarely reported in the literature.

The existence of a hydatid cyst lying between the thorax and the abdomen represents a great challenge in determining whether it originates from the lung, the diaphragm or the liver.

Preoperative careful topographic diagnosis should be made, and it's mandatory to prevent unnecessary excessive incisions.

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.

Conflict of interest:

There is no conflict of interest.

Ethical approval:

Approval has been taken from bioscience center.

Consent: Consent has been taken.

Abbreviations:-

CT: Computed Tomography; MRI: Magnetic Resonance Imaging; US: Ultrasonography. E: Echinococcus. ALB : Albendazole.



Fig A:- Chest X-ray showing right pleural effusion, right-sided subpulmonic opacity and elevation of the right hemidiaphragm.



Fig B:- CT Scan. Anteroposterior view (A), lateral view (B), and transversal section (C), showing the topography of the hydatid cyst in relation to the thoracic, diaphragmatic and intraperitoneal organs.



Fig C:- Hydatid cyst located between the fibers of the diaphragmatic muscle discovered after dissection.

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