Pulmonary Hydatidosis - An Abandoned Surgical Approach

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Rezumat

Hidatidoza pulmonară - procedeu chirurgical abandonat

De-a lungul anilor, hidatidoza, ca entitate patologică medico-chirurgicală, a fost analizată de mulți chirurghi ce au încercat să identifice metoda optimă de tratament prin care să limiteze cât mai mult leziunile parenchimului pulmonar, aducând cât mai multe beneficii posibile pacienților. În acest articol, autorii încearcă să aducă în centrul atenției un procedeu chirurgical abandonat datorită posibilelor complicații dar care a fost adaptat perfect în cazul descris mai jos. Prezentăm cazul unui pacient în vârstă de 59 de ani, fără antecedente patologice semnificative, care avea o simptomatologie minimă nespecifică și la care s-a utilizat cu succes procedeu chirurgical Perez-Fontana în tratamentul hidatidozei pulmonare.

Cuvinte cheie: echinococcus, chist hidatic pulmonar, Perez-Fontana

Abstract

Over the years, hydatidosis as a medical-surgical problem, has been researched by many surgeons, trying to find an optimal treatment method that produces as few as possible lung parenchyma lesions and that has as many benefits as possible for the patient. In this article, the authors are bringing to attention, an abandoned surgical method due to its possible complications but which has adapted perfectly to the case describe bellow. We present the case of a 59-year-old patient, with no significant personal pathological history for the underlying disease, who

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presented with minimal nonspecific symptomatology and with whom the surgical treatment performed was successful practicing the Perez-Fontana technique.

Key words: echinococcus, pulmonary hydatid cyst, Perez-Fontana

Introduction

In a 1941, Jose Arcé, a reputed South American surgeon, a surgical professor and director of the Surgery Institute at the University of Buenos Aires, highlights the affinity of the hydatid disease for the lung, describing its appearance, localisation, clinical manifestations (1).

Human echinococcosis is a parasitic disease that has remained to date a major, worldwide spread, medical and surgical problem. The World Health Organization's 2015 Foodborne Disease Burden Epidemiology Reference Group (FERG) estimated that echinococcosis is responsible for 19,300 global deaths each year. A recent (March 2017) World Health Organization report shows that over 1 million people are affected at the same time and there are projects for validating effective strategies for controlling cystic echinococcosis by 2020 (2). Echinococcosis prevention and control activities and programs focus on deworming of dogs and sheep, which are the definitive hosts.

In most of the cases, this human parasitic disease is caused by taenia echinococcus granulosus. This etiological agent must be distinguished from taenia echinococcus multilocularis, which produces multilocular lesions (alveolar type) and which has an increased potential for local invasion (3). In the evolutionary cycle of this disease, man is considered to be the casual intermediate host, acquiring the infection as well as the other intermediate hosts (dog, sheep, and camel), but are not involved in transmitting the infection to the definitive host. The most important pulmonary forms in humans are cystic echinococcosis (CE) produced by taenia echinococcus granulosus and alveolar echinococcosis (AE) caused by taenia echinococcus multilocularis (4).

Treatment in hydatid disease is a topic debated since 1804 when Laenec explained that these hydatid cysts are only a phase of the developmental cycle of a tapeworm and do not represent abnormal human tissue growth as stated before. Later, in 1835, Recamier surgically approached leaver hydatid cyst in a two-stage procedure, which was completed in one stage in 1877 by Lindemann. Thomas practiced the incision of pulmonary parenchyma and the removal of the hydatid cyst in 1884, pouching the remaining cavity to the chest wall and leaving the wound open. The enucleation process of the hydatid larva cyst was proposed in 1964 by Ugon (5) and a year later Allende proposed simple enucleation of the cyst but without padding the residual cavity (6).

In 1948 Perez-Fontana described a new technique presenting it under the name of pericystectomy - surgical removal of the cyst together with the fibrous capsule (7,8).

As with any surgery, Perez-Fontana technique has both advantages (it's a radical operation, conservative, with reduced chance of relapse) but also disadvantages (increased risk of bleeding, risk of bronchopleural fistula, with air leak and the appearance of pneumothorax), that is why it was abandoned, other surgical techniques taking its place.

The authors are presenting the case of a 59 years old patient, to whom this surgical method has been successfully practiced.

Material and Method

This paper presents the case of a 59-year-old patient, known for 1st degree hypertension in drug-controlled which reports that the illness had started a month before, with diffuse pain in the left hemithorax, dyspnea, asthenia, lack

Figure 1. Thoracic radiography: pulmonary opacity in the upper left lobe

of dynamics, hemoptysis in small quantities.

Upon admission to our clinic, the objective clinical examination did not provide us with conclusive data for the diagnosis.

Thoracic radiography (*Fig. 1*) highlights a pulmonary opacity, localized in the upper left lobe, with well-defined margins, 7/4 cm dimension, possibly in direct contact with the parietal pleura.

The functional tests performed in our service were within normal limits. BK sputum exam - direct and culture - negative. Laboratory analyses with eosinophils in normal range.

Computed tomography examination (*Fig. 2, 3, 4*) reveals a space replacement, with liquid content (UH between -11 and 37) in intimate contact with the parietal pleura, without invasion of the chest wall, with a maximum diameter of 75 / 43.2 mm, well defined, ellipsoidal shape; without the presence of mediastinal lymphadenopathies.

After a general preoperative preparation, surgical intervention was decided. We entered the pleural cavity by incision on the medial axillary line and left thoracotomy was practiced through 4th intercostal space. We found a whitish cystic formation located and exteriorized in the left upper lobe of the lung parenchyma, having macroscopic signs of uncomplicated hydatid cyst (*Fig. 5*).

The cystic formation was isolated with

Figure 2. CT - mediastinal window pulmonary cystic lesion

Figure 3. Chest CT - parenchymal window intrapulmonary cystic mass

Figure 4. Chest CT sagittal reconstruction: cystic lesion of the upper pulmonary left lobe

sterile fields and surgery intervention was decided.

As surgical technique the authors opted for

Figure 5. Left upper lobe pulmonary hydatid cyst - intraoperative aspect

Figure 6. Pericyst and hydatid membrane excised

ideal chisto-perichistectomy - Perez-Fontana procedure.

The objective of surgery is to remove the parasite along with pericystic cavity with preservation of functional pulmonary parenchyma. This surgery intervention was performed in general anaesthesia with a selective right bronchial intubation.

The release of the cystic formation goes to the near-close release of the pericystic formation together with the cyst, with great care to avoid its intraoperative breakage of the cyst, at the junction between the lung parenchyma and pericystic cavity. We used both dissection scissors and finger dissection where adhesions are "more lax" to allow this manoeuvre. At the time of "finger dissection", the left lung was partially ventilated at the surgeon's request for an attempt of release in better viewing conditions of the air lost.

There is a relatively large possibility of vascular intrusion, therefore the dissection was performed with great care and at the lowest bleeding the dissection was stopped and haemostasis was performed.

Scissor dissection was performed with great care, step by step, to avoid any important vascular intrusion that floods the operator field and make the maneuver difficult.

The removal of the hydatid membrane and the pericyst was entirely accomplished (Fig. 6, 7, 8) and bronchial microfistulas and bleedings did not represent a danger that would require abandoning this technique

Figure 7. Pericyst and hydatid membrane sectioned

Figure 8. Hydatid membrane and pericyst separated

and the use of another one. Subsequently, the aerostasis control was done, by introducing physiological serum into the remaining cavity (Fig. 9) and suture with 4.0 slowly resorbable

Figure 9. Aerostasis control with physiological serum

Figure 10. Pulmonary micro fistulas aspect

thread was proceeded, where remaining microfistles have been observed (*Fig. 10*).

Perilesional destruction of pulmonary parenchyma determines its modification and transformation into a thin foil tissue.

Next surgical step was the resection of the so-called "lung flap" with its marginal suture (*Fig. 11, 12*).

The sectioning of the "flap" and the 4-0 resorbable suture of the resection edges to seal the suture trench are shown in the figures below (*Fig. 13, 14*).

Although there was no air loss, preventively, a second, superficial, safe layer of suture was performed without damaging the architectural pathway of the lung parenchyma. Intra-

Figure 11. The margins of the "flap" caught in forceps

Figure 12. The beginning of pulmonary "flap" resection

Figure 13. The resection of the remaining pulmonary parenchyma

Figure 14. Pulmonary parenchyma sutured

Figure 15. Second suture layer of pulmonary parenchyma. Intrathoracic drainage tube

operatively, at the end of surgery, there was a complete expansion of the lung, without air leak, final stage - the drainage of the pleural cavity (*Fig. 15*).

Pulmonary radiological aspect, 24 hours after surgery, with completely expanded lung and the presence of the two drain tubes (*Fig. 16*).

Postoperative recovery was favourable and fast, with patient discharge 5 days after the surgery. The patient was then sent to a parasitology service for complementary parasiticide treatment.

Discussions

Positive and differential diagnosis of hydatid cyst is based on clinical, laboratory and imagistic findings and varies depending on location. The table below shows some of the diseases suitable for the differential diagnosis, both the uncomplicated and complicated hydatid cyst (*Table 1*).

Pulmonary hydatid disease, although pathologically similar to hepatic hydatid disease, have a different medical-surgical management.

Medical therapy with benzimidazole medication class is valuable in smaller hydatid lesions, but also in disseminated disease, secondary lung or pleural hydatidosis, as well in poor surgical risk patients (9,10). Studies of the different types of treatment

Figure 16. Postoperative radiography

reveal, however, that the gold standard in the treatment of this disease remains surgical treatment, regardless of the method chosen (11,12).

As it is well known, there are several types of surgical approach to echinococcosis, each intervention having its advantages and disadvantages. Lamas Mondino surgery (13), although of historical interest, is indicated in patients who are biologically unable to withstand thoracotomy (70-80 years, comorbidities) with altered respiratory tests.

Arcé procedure consists in the complete

Table 1. Differential diagnosis of uncomplicated and complicated hydatid cyst (3)

Uncomplicated pulmonary hydatid cyst	Complicated pulmonary hydatid cyst
Pulmonary tuberculosis (pseudotumoral forms)	Pulmonary tuberculosis cavern with an air-fluid level
Primitive malignant lung tumours	Pulmonary abscess
Pulmonary metastases	Infected pulmonary cyst
Benign lung tumours	Pulmonary bulla with an air-fluid level
Congenital cystic pulmonary malformations	Pulmonary excavated infarction
Mediastinal cystic lesions	Spontaneous pneumothorax
Loculated pleural effusions	Parapneumonic pleural effusion
Parietal thoracic lesions	Neoplastic pleural effusion
Aortic aneurysms	
Diaphragmatic deformities (image "en brioche")	
Hydatid cyst of the liver dome	

and relatively slow evacuation of the hydatid fluid, followed by a minimal pneumonotomy at the level of the maximal cyst corticalisation area (1,13,14).

By Finochietto procedure, the hydatid fluid is suddenly and completely aspirated by a thick needle, this manoeuvre reducing the risk of secondary sowing. It is followed by pneumonotomy and membrane extraction (13,14).

Within the so-called "ideal cystectomies", are included the Ugon (5,15), Dubau (15,16) and Perez-Fontana procedures (3,7,8), through which the hydatid is completely extirpated without puncture and without opening it, presenting the great advantage of never putting into contact the surgical wound with the hydatid liquid, therefore the risk of allergic accidents such as anaphylactic shock and secondary sowing is virtually excluded.

Most authors recommend removal of the hydatid membrane after punching, partial or total discharge of the hydatid fluid, inactivation of the cyst with absolute alcohol solution or hypertonic saline solution (3,17).

For the treatment of the remaining pericystic cavities, several procedures have been developed, including Juvara (13), Posadas-Carpinisan (14), Guedj-Melnikov (13,14,18), Gerulanos (3,13,14), Horvat (the "valley" principle) (3,15) and Dor (19). The last one represents the method of upholstering by successive, overlapping stumps and answers to all the requirements regarding the

remaining cavity treatment, prevention of infection, but also avoidance of remaining cavity ballooning, as well as quick functional pulmonary recovery.

Most surgical interventions are based either on evacuation of the cyst and of its content, the closure of the bronchial fistulas inside the pericystic cavity to prevent the occurrence of the balloon cavity with superinfection and the occurrence of the secondary lung abscess, or on the complete excision of the lesion. Preserving as much as possible from lung parenchyma is a major objective for the surgeon, especially in the case of children (20).

All intraoperative precautions are necessary to prevent accidental rupture of the cyst. In the case of a rupture, all measures should be taken to prevent and combat anaphylactic shock (21). It is crucial to have a lung separation and one lung ventilation during general anaesthesia. Double lumen endobronchial tubes are currently the most widely used by the anaesthesiologists for this purpose in order to prevent broncho-pulmonary contralateral hydatid dissemination in a case of accidental rupture of the cyst (22,23). However, in part of the pediatric patients (weighting < 30 kg), single lumen tube was advanced into the main contralateral bronchus under the guidance of bronchoscope.

Monitors attached for ECG, continuous invasive arterial blood pressure, end-tidal CO₂, and (SpO₂).

Intravenous access secured with large bore

cannulas (22,23). Without these measures, mechanical ventilation or even surgical pulmonary manipulations can mobilize solid fragments of parasitic

membrane or small daughter cysts into the bronchial tree resulting in acute obstruction of the airways (22,23). Before induction of anaesthesia intravenous corticoids administration (hydrocortisone 100 mg) were always indicated to prevent anaphylactic reactions (21,24). Adrenaline and Theophylline kept in standby for any emergencies.

In rare situations, surgical treatment can reach the pulmonary exeresis, from atypical resections to pneumonectomy (extremely rare cases), when we encounter a complicated pulmonary festered cyst complicated with piosclerosis, major intraoperative vascular accidents, giant pulmonary hydatid cyst with pulmonary parenchyma destruction, significant bronchial stenosis with incomplete lung expansion after pericystic cavity treatment (3,13,14).

We consider that hydatid cysts over 2 cm have to be surgically approached before antiparasitic treatment because we observed a possible evolution to pulmonary abscess along with parasite's death by local infection of the pulmonary cyst. Unlike the liver hydatidosis, the risk of infection is greater in lungs because of bronchial micro fistulae which are opened at the level of the pericystic cavity (3,10).

Postoperatively, antiparasitic treatment enhance long-term favourable evolution of the disease and to stop recurrence.

Our experience suggests that the most effective therapy in treating multiple pulmonary hydatidosis is the combination of surgery with chemotherapy in case of at least one hydatid cysts 2 cm over or only antiparasitic chemotherapy in case of all cysts 2 cm under (10).

In situation of multiple visceral uncomplicated hydatidosis with pulmonary and liver cysts we recommend pulmonary surgical approach first because the major risk of pulmonary cyst rupture during mechanical ventilation (general anaesthesia for liver surgery) with broncho-pulmonary or pleural

hydatid dissemination. Simultaneous approach of both pulmonary and hepatic hydatid cysts could be possible by thoracotomy in a situation of liver dome cysts or through combined thoracic and abdominal approaches in other hepatic cyst's localisations.

Albendazole given as described together with surgery is considered to have the highest success rate in treating cystic hydatidosis (10).

Perez-Fontana surgery is an ideal cysto-perichistectomy, which consists in removing intact the hydatid cyst along with the pericyst. In 1948, when this method was published, the authors grounded it theoretically through the existence of a space between the adventitia and the pulmonary parenchyma. In the case of large, centrally located hydatid cysts, there is the risk of lesions of large vessels, even the pulmonary artery, even leading to pneumonectomy (3,7,8,13,14,15).

This procedure has been abandoned precisely because of the possibility of massive haemorrhage through the injury of large intraparenchymal vessels and bronchial fistulas which can raise both intra and post-operative problems if one does not intervene through a suitable attitude.

Despite the latest evolution of thoracoscopic surgery, many thoracic surgeons are still adopting the classical open thoracotomy for approaching on pulmonary hydatid cysts (24). They believe of not being able to control the contents of the cyst during the thoracoscopic procedure which could contaminate the pleural cavity or causing an anaphylactic response. However, some recent publications on thoracoscopic management of pulmonary hydatid cysts have been initiated in adults and children (24-29). Lungs isolation is crucial to conduct video-assisted thoracic surgery (VATS) operations and prevent hydatid dissemination or anaphylactic reactions. The carefully exploring of the lung's surface and the pleural cavity for any ruptured cyst or free pleural membranes is facilitate by 10 the presence of the high-definition thoracoscopic lens inside the thoracic cavity which provides a very clear and accurate images by focusing the image on the area of operative interest.

Additional advantage is that the all members of the surgical team, even the anaesthesiologist can follow the details of the surgery through the screen, which may help them to observe and take the immediate and appropriate action in any moment of the operation. Identification of the small deep intraparenchymal cyst is more challenging and requires intraoperative ultrasound examination. After identifying the cyst, several pads soaked with hypertonic saline (23.4%) are placed around the cyst (to prevent contamination in case of rupture) and the cyst wall is punctured using 21 gauge needle (23).

The fluid contents is carefully aspirated until the cyst becomes collapsed and hypertonic saline “23.4% concentration” is injected through the same needle for parasitic inactivation. After waiting for 10–15 minutes, the fluid is re-aspirated and the cyst is opened. The cystic membrane is removed avoiding any contact with the patient’s tissue. After the cyst exploring for additional membranes, ventilation is done to check air-leaks through cyst-bronchial connections. The bronchial openings are closed one by one using 3-0 Vicryl sutures “4-0 in pediatrics” (20,29). The remaining pulmonary cavity is treated by capitonnage and by plicating the cyst walls. The authors highlights the limited amount of surgical trauma and pain provided by VATS which was the reason of performing the single-stage bilateral VATS operations (for bilateral pulmonary cysts) or combined thoracoscopic and laparoscopic procedures in cases of multi-visceral echinococcosis (concomitant pulmonary and abdominal cysts). These authors concluded that the surgical treatment of pulmonary hydatid cysts via VATS techniques is feasible to be performed safely, under intraoperative ultrasound guidance for localizing small and deep lesions and has many advantages over to the conventional open thoracotomy approach (24-29).

Conclusions

The recommended treatment of the hydatid cyst is the surgical one and the main objectives are to evacuate the hydatid content

with the extirpation of the hydatid membrane and to treat the pericystic cavity with preserving the bronchial and vascular structure and sparing the pulmonary parenchyma.

We consider that the Perez-Fontana procedure can be performed relatively safely with carefully dissection in a cases of peripheral pulmonary cystic localization because of the minimal risk of severe bleeding or air losses (little risk of vascular or bronchial fistulas), which can be easily resolved intraoperatively, without altering the architectural design of pulmonary parenchyma.

It is recommended that the surgical treatment to be followed by the medical one, in order to avoid pleural and pulmonary parasitic recurrence.

Conflict of Interest

The authors declare no conflicts of interests.

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