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Management of Thoracic Hydatid Disease and Its Complications

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Introduction

Hydatid disease of the lung or lung echinococcosis is caused due to an infection with the cestode *Echinococcus granulosus* predominantly. The three types of echinococcosis are cystic echinococcosis caused by *E. granulosus*, alveolar echinococcosis caused by *E. multilocularis*, and polycystic echinococcosis caused by *E. vogeli* or *E. oligathrus*, being the first one the predominant, by far [1].

The term hydatid recognizes its first use from the French *hydate* around 1680, which comes from the Greek *hydatis/hydatos*, meaning vesicle with serous liquid, and this last, from *hydor/hydatos*, which means water or liquid.

This parasite has been known from the times of Hippocrates and Galen. Magnúson described the presence of the *E. granulosus* in German dogs circa 1200, but the first description belongs to the German anatomist Adam Christian Thebesius (1686–1732) in the seventeenth century. Zabert and Van Deinsen considered this disease to have originated in Iceland and introduced to Europe by the dogs of whale hunting ships [2]. In 1808 Karl Asmund Rudolphi (1771–1832), known as the "father of helminthology," was the first one to coin the term "hydatid cyst" to refer to the disease in humans [3].

In 1889, John Davies Thomas (1844–1893), an Australian surgeon based in Adelaide, was the first to propose the surgical treatment of pulmonary hydatidosis [4, 5]. Some years later, in 1899, Alejandro Posadas (1870–1902) performed in Buenos Aires, Argentina, a thoracic procedure for the treatment of hydatid disease in what is considered the first recorded surgical procedure in world surgery [6]. His technique included the performance of a thoracoplasty with the "harpooning" or "anchorage" of the lung to prevent its collapse and the aperture of the pericystic layer to remove the germinative layer and the cyst content [7].

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G. Tsoulfas et al. (eds.), *The Surgical Management of Parasitic Diseases*, https://doi.org/10.1007/978-3-030-47948-0_10

Parasite Biology and Cycle

The life cycle of *E. granulosus* requires two host types, an intermediate and a definitive one. The first are represented most commonly by sheep, however cattle such as horses, goats, pigs, and camels may be potential intermediate hosts. Humans are intermediate but accidental or aberrant intermediate hosts due to infestation or contact with diseased dogs, but do not play a role in the biological cycle.

The definitive hosts are dogs or other canines, such as foxes or wolves. The adult worm inhabits the small intestine of the definitive host; the size ranges from 2 to 7 mm long and gets attached to the intestinal mucosa by a double row of hooklets placed in its scolex and has at least three proglotides with several eggs. These eggs pass out in the stool and stick to the fur or fall to the grass. They can survive for at least 1 year in the outside world, time during which they are widely dispersed, even with the help of the wind. When intermediate or accidental hosts become infected, either by contact with infected dogs or by ingestion of eggs from contaminated food, water, or soil, the embryos are released after hatching in the intestinal mucosa and then enter the portal circulation making their first station the liver, or otherwise the lung [8]. The parasite then grows to form a cyst filled with fluid. Since two mammal species are required for completion of the life cycle of the parasite, direct transmission from human to human is not possible.

The fully developed cyst is organized in three layers, composed of both host and parasite tissue:

- (a) The outer layer, or pericyst, is the adventitia and is formed by inflammatory fibrous tissue from the host as a reaction to a foreign body (parasite).
- (b) The middle layer is known as the exocyst or laminated layer and is an acellular membrane composed by mucopolysaccharides.
- (c) The inner layer is the endocyst, which is the germinal layer of the parasite and gives rise to larvae scolices which bud internally. The fluid contained inside is antigenic and contains hooklets and scolices and is known as "hydatid sand." There is an average 5 cc of this hydatid sand which contains half a million hooklets and scolices, which indicates its huge aggressiveness.

The cyst exists in different forms: intact or ruptured, single or multiple, unilateral or bilateral, exclusively located in the lung or in other locations (the liver, pleura, pericardium, rib cage, spine, etc.). The hydatid cysts are able to grow faster and larger in the lung due to the structure of these organs, which is easily collapsible, in comparison to the liver parenchyma.

The hydatid cyst can be classified according to its morphology:

- Type I: simple cyst
- Type II: cyst with daughter cysts and matrix
 - Daughter cysts at the periphery.
 - Larger daughter cysts which fill most of the mother cyst.
 - Cyst with calcifications and daughter cysts. Calcifications in the lung are partial and in spots, quite different from the calcification of liver cysts.
- Type III: calcified cyst, which means that the cyst is not viable, and cannot infect the host
- Type IV: complicated cyst, e.g., ruptured cyst

Epidemiology

Cystic echinococcosis can be seen worldwide, while endemic areas are represented by South and Central America, the Middle East, North and sub-Saharan Africa, Eurasia, Australia and New Zealand, China, and Russia [9].

A huge distinction needs to be made with alveolar echinococcosis caused by *E. multilocularis*, which is significantly less frequent than hydatid disease by *E. granulosus*. Definitive hosts include small rodents and the borders of endemic areas of alveolar echinococcosis never cease to expand [10].

Regarding the geographical distribution in Argentina, 30% of our territory carries risk of infection (1.271.912 km2) and 8% of our entire population (about 2.850.000 inhabitants) are at risk for acquiring the disease [11].

In children, the lung is the most common location for the cystic hydatid disease [12]; 20 to 40% of patients with thoracic involvement also present with liver cysts [13]. According to a multicenter Argentinian experience, 30% are multiple pulmonary cysts, 20% are bilateral, and 60% are located in lower lobes [14].

Clinical Presentation and Diagnosis

Pulmonary hydatidosis is the second most frequent location of cystic hydatid disease caused by *E. granulosus*. The lower lobes are more frequently affected than the upper ones, and one in five cases are bilateral. The size ranges from 1 to 20 cm and is the organ where hydatid cyst can become the largest due to the easy compressibility of lung tissue. Calcification of lung hydatid cysts is very rare, contrary to the liver ones.

Pulmonary hydatid cysts can present themselves in a variety of ways, making the diagnosis difficult and requiring high suspicion from the physician. The background of the patient in reference to the birthplace and residence in rural regions is very important and should be particularly targeted. Lung cysts may remain asymptomatic and thus silent for years, appearing when chest X-rays are requested for screening or due to other reasons. Initial symptoms include cough, discomfort, chest pain, fever, and hemoptysis. The development of an anaphylactic reaction and the appearance of vomiting are indicative of rupture of the cyst. In this latter case, the sputum direct study may reveal the presence of echinococcus granulosus' hooklets. When confronted with an initial thoracic hydatid cyst, it is mandatory to preclude the presence of a liver location [15].

A simple chest X-ray may provide the first suspicion and let the treating team know if this is a complicated or non-complicated cyst. Next diagnostic steps include serology together with imaging modalities, such as ultrasound and CT scans not only of the thorax but also of the abdominal and pelvic cavity.

Serological tests include the following modalities:

- Casoni intradermal reaction and complement fixation test have been abandoned in clinical practice.
- Indirect hemagglutination test: it is positive in just 50% of patients with pulmonary hydatidosis and in 90% of those with liver cysts. False-positive reactions with other helminthic diseases should be taken into account.

- Arc 5 test: this test measures antibodies against antigen 5, which is a major parasite antigen found in the inner aspect of the germinal layer, in daughter vesicles (brood capsule) and protoscolices. It achieves low sensibility but high specificity, and the subunit 8 kDa (located in the protoscolices) offers greater specificity [16]. Nonetheless the sensibility for lung hydatid cysts is in the range of 35%.
- ELISA (enzyme-linked immunosorbent assay): the immunoglobulin G enzymelinked immunosorbent assay test represents the most sensitive technique. When this test shows a positive result, it should be confirmed by a western blot determination.

The diagnosis is usually performed by combining the clinical background, serology, and imaging modalities. It is important to highlight the role of screening X-ray examinations among the population in endemic places [17].

Differential diagnosis includes tuberculoma or tuberculous cavern, malignant tumor (primary or metastatic), lung abscess, bronchopulmonary infections, Wegener's granulomatosis, bronchiectasis, pneumothorax, and empyema, among others.

Puncture is not recommended due to the risks of cyst rupture, anaphylactic reaction, and risk of seeding the pleural cavity. Bronchoscopy is usually not necessary; besides the location of the cysts tends to be peripheral and not central.

Plain chest radiograph is helpful and may provide useful information regarding the status of a pulmonary hydatid cyst. When the cyst is intact, the typical image is that of a round or oval mass with clean borders surrounded by normal lung tissue (Figs. 10.1 and 10.2). The size may be variable and sometimes is not single. Sometimes the cyst shows an air-fluid level (Fig. 10.3). When the cyst is ruptured or infected, the typical image achieves the "water lily" or "hyacinth" sign. Due to the rupture or if bronchial erosion has occurred, an air fluid level can be found or the thin crescent or meniscus sign. From a clinical point of view, the patient will present with vomiting, with expulsion of cyst content (scolices and daughter vesicles).

Rupture or perforation of a pulmonary cyst represents its most frequent complication, with an incidence close to 50% of the cases. In 90% of the cases, the rupture is to the bronchial tree, and in the rest of the cases, the opening is toward the pleural cavity. The CT can provide further details as can be seen in Figs. 10.4 and 10.5 ("water lily sign" and presence of parasites). Two conditions should be considered regarding the hydatid cyst rupture: (a) the adventitial layer of lung cysts does not develop as well as in the liver and is more prone to rupture, and (b) the increase in the size of the cyst causes an increase in the intrathoracic pressure and can also favor the rupture. Rupture also conveys the risk of subsequent infection, with a similar course as that of a pulmonary abscess.

Simultaneous lung and liver locations for hydatid cysts are observed in less than 10% of the cases [18]; nonetheless when the patient initially presents with a thoracic location, liver involvement must be ruled out by ultrasound and CT scan.

Manterola introduced the concept of the thoracic involvement of hepatic echinococcosis, TIHE [19]. The hydatid cyst of the ruptured liver in the thoracic cavity is a rarely reported clinical entity whose frequency appears to be declining [20].



Compromise of both the liver and the thoracic cavity by hydatid cysts can be seen in Figs. 10.6, 10.7, 10.8, 10.9, and 10.10.

According to the degree of evolution of the diaphragmatic and/or thoracic involvement, the following five stages or grades have been described [13]:



- 1. Adhered cyst, firm adherences between the cyst surface and the diaphragm but no perforation.
- 2. Hydatid transit, with perforation of the diaphragm, but little or no invasion of the thoracic cavity.
- 3. Pleurothoracic vesiculation, the cyst perforates the muscle and grows inside the thoracic cavity, and daughter vesicles are established in the pleura.



Fig. 10.4 Ruptured hydatid cyst, chest X radiograph, and CT scan (water lily sign). Parasites can be seen in the CT scan



Fig. 10.5 Ruptured hydatid cyst, chest X radiograph, and CT scan (water lily sign)



Fig. 10.6 Liver cyst with thoracic evolution (chest X-ray)

Fig. 10.7 Liver cyst with thoracic evolution (chest X-ray)



- 4. Disease of the pulmonary parenchyma, the cyst connects to the bronchi, or there may be compression and/or atelectasis of the pulmonary parenchyma.
- 5. Chronic bronchial fistula, either postoperative or usually as a result of the spontaneous evolution of the abdominal hydatid disease.

Fig. 10.8 Liver cyst with thoracic evolution (CT scan)







A condition that favors the upward migration is the negative pressure in the thoracic cavity and the obstruction of the bile duct system by parasites. Sometimes, the final evolution is the establishment of an abdominothoracic fistula, which is an uncommon but severe complication of hydatid disease. Biliptysis is pathognomonic of a biliary-bronchial fistula communication. The fistula usually is organized through trans-diaphragmatic penetration, which leads to a large cyst's rupture into the lung lower lobe. Though uncommon, the underlying cause is the perforation



Fig. 10.10 Liver cyst with pleural and bronchial communication (CT scan)

| Table 10.1 | Classification of |
|--------------|-------------------|
| the hydatid | liver cysts |
| migrating to | the lung (after |
| Mestiri, 22) | |

| 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 pocket | |
| 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: biliohydatic pleurisy | |
| IVB: secondary pleural hydatidosis | |
| | |

from the right subphrenic space into the posterior basal segment of the right lower lobe [21]. Mestiri et al. proposed a classification for the migration of liver cysts to the lung emphasizing the importance of small bronchial fistulas over the biliary fistulas [22], as seen in Table 10.1.

Although calcification occurs frequently in liver cysts, it requires a lapse between 5 and 10 years at least, whereas it rarely happens in pulmonary cysts.

Other thoracic locations include the pleural cavity, the pericardium, the spine, and the rib cage. Pleural compromise can be achieved by different mechanisms: (a) spread of the cyst's (either hepatic or thoracic) content after its rupture which allows the nesting of daughter vesicles in the pleural space, (b) direct infestation with the

parasite, (c) hematogenous or lymphatic route, and (d) postoperative sequelae. Primary pleural hydatidosis is neglected by most authors [23]. The surgical treatment mandates thoracotomy and pleural decortication.

Location in the mediastinum is very rare but should be suspected in patients with a mediastinal mass coming from or living in an endemic area [24]. Surgery is mandatory due to the need to confirm the diagnosis and rule out malignancy if the images are not definitive and also due to the presence of vital structures. According to the location of the mediastinal hydatid cyst, the approach may be by right or left thoracotomy or thoracoscopy or even through a median sternotomy [25].

Pericardial hydatid involvement may be due to migration (secondary hydatidosis) or due to the rupture of a hydatid cyst located in the left heart (cardiac muscle). The role of magnetic nuclear resonance is very useful to show cardiac involvement (Fig. 10.11a, b). Pulmonary arteries are rare locations for hydatid cysts, with the most frequent cause arising from embolism originating in a primary cardiac location; another possibility is that the embryos of *Echinococcus granulosus* pass through the liver and then into the inferior vena cava and from there to the pulmonary arteries through the right cardiac chambers.

Hydatid compromise of the spine is very seldom seen [26]. CT and MRI usually show a lobulated lesion with some compromise of the epidural space of one or several thoracic vertebrae (Fig. 10.12a, b). The course is usually aggressive, with compromise of the spinal cord and thus neurological symptoms, recurrence is not infrequent, and surgery usually involves resection and vertebral instrumental stabilization [27, 28].



Fig. 10.11 (a) Pericardial hydatid disease (CT scan, sagital view). (b) Pericardial hydatid disease (CT scan, lateral view)



Fig.10.12 (a) Hydatid disease of the spine. (b) Hydatid disease of the spine

The chest wall is a rare location of secondary hydatidosis, but it may occur after the rupture of a lung cyst, from a liver cyst invading the diaphragm into the pleural cavity, following previous hydatid thoracic surgery, or by hematogenous spread of embryos with a diameter of less than 0.3 mm, which may escape from the liver or from the lung capillaries to involve any organ via the systemic circulation [29].

The rib location of hydatid cysts (Fig. 10.13) mandates differential diagnosis with the following entities, among others: fibrous dysplasia, tuberculosis, simple bone cysts, plasmacytoma, osteo- or chondrosarcoma, and other primary and meta-static bone tumors.



Fig. 10.13 Hydatid disease affecting the rib cage, posterior arch

Surgical Treatment

Initial treatment with albendazole before scheduled surgery is always recommended [30]. The dosage is 10 mg/kg/day twice a day, and the recommendation is after lunch and dinner rich in lipids, so as to improve absorption of the drug, with a maximum dosage of 800 mg/day [31]. If possible, preoperative prophylaxis is recommended for at least 15 days and for up to 3 months postoperatively [32].

Surgery should be considered for every thoracic hydatid cyst; initially, the surgical treatment consisted of simply performing the marsupialization of the cyst [33]. Currently, the goal of the surgical treatment for hydatid cysts of the lung is the complete excision of the disease process together with the maximum preservation of lung tissue, together with the elimination of the parasite and the prevention of recurrences and/or seedings [3]. The WHO guidelines mandate surgical intervention in the following cases: impending cyst rupture, compromise of vital organs due to mass effect, hemoptysis, secondarily infected cysts, infection due to obstruction, and unmanageable pain [34].

It is essential to prevent the contamination of the operative field due to rupture and/or spillage of the cyst's content, as its result may be the implant of scolices and the production of secondary cysts [35]. The approach maybe through a conventional or vertical thoracotomy, whereas a median sternotomy is reserved for those infrequent cases of bilateral cysts. The minimally invasive treatment of pulmonary hydatid disease was introduced by Becmeur et al. for the treatment of children [36], and though many surgeons do not feel comfortable with this access due to safety issues, the feasibility of this access has been proven by others [37, 38]. Alpay considers that video-assisted thoracic surgery treatment of pulmonary hydatid cysts is superior to conventional thoracotomy due to shorter operative time, lower chest drainage volume, shorter duration of tube placement, and less postoperative pain [39]. The minimally invasive or thoracoscopic approach is reserved for small and peripheral cysts, which after puncture and drainage are resected "en bloc" with the aid of mechanical sutures. Contraindications to this approach include larger pulmonary cysts (larger than 5/6 cm), multiple cysts and those with a hilar location. It is important to bear in mind that the total enucleation of an unruptured pulmonary hydatid cyst, which can be safely performed in open surgery, cannot always be safely performed via a thoracoscopic approach since the small size of the incisions would prevent the safe removal of the cyst.

The conservative procedures include the following:

- Enucleation (intact endocystectomy): known as the "delivery" or the Armand Ugón technique [40], it may be used when the cyst is relatively small and not complicated, and the risk of rupture is low. The hydatid delivery requests the expertise and the attention of the whole team, including the anesthesiologist who must not ventilate the lung while the surgeon makes an opening in the adventitial layer, and introduces the finger to dissect the adherences between the cyst and the wall. After the parasite is fully freed from the surrounding tissues, the insufflation of the lungs will aid in the delivery, pushing the parasite through the air expelled through the bronchi (Fig. 10.14). Irrigation with saline solution during the whole procedure is recommended in order to prevent desiccation and a leak on the membrane [41].
- Removal after insertion of a trocar-suction device (R. Finochietto), for adequate parasite and cyst's content aspiration (Fig. 10.15). The puncture plus aspiration is a precision maneuver which necessitates avoidance of injury to the lung parenchyma, as well as spillage of the cyst's content. Care should be taken to prevent membranes falling into the pleural space.
- Cystotomy (marsupialization): is a conservative technique with usually very good results, frequently with an added capitonnage.
- Capitonnage: consists of the obliteration of the cavity with purse string sutures. There is no clear agreement regarding its use, and one of its main consequences is the distortion of the pulmonary parenchyma, especially when large cysts have been removed [42].

All the abovementioned techniques leave the adventitial layer is left in site.

Fig. 10.14 Delivery of hydatid cyst



Fig. 10.15 Finochietto trocar-suction aspirator



Fig. 10.16 Final result of a capsule resection (pericystectomy)



• Pericystectomy (capsule resection): known as the Pérez-Fontana method, consists in the removal of the cyst with the perycystic layer and the adventitia and the posterior obliteration of the residual cavity, Fig. 10.16 [43].

The use of the PAIR (puncture, aspiration, injection, and reaspiration) technique, originally described for the treatment of abdominal hydatid cysts, is not favored for thoracic locations since several contraindications have been reported [44].

The radical procedures, which include lung parenchymal resections, should be reserved for specific situations. They should be considered in the following cases:

• When the hydatid cysts have produced simultaneous and irreversible alterations in the surrounding lung parenchyma

- Multiple cysts in a lung segment or lobe
- When a large cyst replaces a small lobe (lingula or medium) The options include the following:
- Pulmonary resection (e.g., "wedge"), when the size of the cyst is not large and there is no peripheral impact on the lung parenchyma.
- Segmentectomy, characterized by a high morbidity.
- Lobectomy: most of the times it is considered as the technique of choice. The surgical team should rely on this approach when confronted with large cysts involving more than 50% of the lobe, cases with severe pulmonary suppuration, multiple unilobar cysts and sequelae such as pulmonary fibrosis, bronchiectasis, and hemorrhage.
- Pneumonectomy: strictly only in cases of necessity.
 Some general principles that should be taken into consideration are:
- Double lumen intubation is recommended to keep ipsilateral lung collapse during the procedure and in this way facilitate the surgical maneuvers.
- Protection of the pleura and the adjacent tissues may be achieved with towels soaked in 20% hypertonic saline solution.
- Avoidance of spillage of hydatid liquid to the bronchi.
- Injection of scolicide agents in the cyst: usually some fluid from the cyst is aspirated and replaced with hypertonic saline solution for about 15 minutes before the cyst's contents are evacuated with a powerful and wide aspirator.
- Treatment of the adventitial layer: according to the local conditions, the resection will include only the protruding or emergent part or its entirety. If the complete excision is prone to excessive risk, the remaining adventitial layer should be left on sit with a meticulous hemostasis of the borders.
- Closure of small bronchi: after the partial or full resection of the pericystic areas, and since the bronchi are opened to the cyst cavity in a tangential fashion, it is imperative to achieve a safe closure of the opening in a very thorough and detailed fashion with simultaneous irrigation and ventilation to observe bubbles from the opened bronchi. The folds of the remaining adventitial layer should be checked to prevent any bronchi from being closed. A hydrostatic test should be performed to check the closure.
- Management of the residual cavity: by means of a capitonnage or leaving it open so as to be resolved by the pulmonary expansion [45].

Intraoperative complications are represented mainly by the rupture of the hydatid cyst and bleeding. The first one is, by large, the most frequent and the cause of severe sequelae in the long-term follow-up (Fig. 10.17). All the protective measures should be taken to prevent the perforation and, if it happens, the spillage of its contents.

Postoperative complications may be *immediate* or *delayed*. Among the first ones, the bronchopleural fistula, hemopneumothorax, empyema, and atelectasis should be considered and will be briefly addressed.

• Bronchopleural fistula: usually due to an inadvertent bronchial opening that was not sutured during the operation. Usually, it tends to be self-limited and resolves

Fig. 10.17 Intraoperative spillage



with the pleural drainage favoring the parenchymal expansion of the lung, adhering itself to the parietal pleura.

- Hemo- and hemo-pneumo-thorax, their treatment does not differ from other situations.
- Empyema: usually develops in patients with long-standing and infected cysts, with compromise of the lung parenchyma.
- Atelectasis: is the most frequent postoperative complication. Usually due to secretions, the presence of hydatid membranes occluding the bronchial tree should be ruled out. Fiberoptic bronchoscopy is the treatment of choice and is both diagnostic and therapeutic.

Delayed postoperative complications are represented by:

- Residual cavity, usually treated as simple lung air cysts: no treatment is needed, but the risk is bacterial colonization and posterior pulmonary abscess (Fig. 10.18).
- Seeding: it is secondary to the intraoperative rupture of a hydatid cyst. The pleura are not a very fertile location for the nesting of the scolices, but when it is compromised, the prognosis is severe (Fig. 10.19). Bronchial seeding is by far a more serious complication and is the consequence of the filling of the bronchial tree with hydatid liquid and sand. The immediate consequence is the development of an anaphylactic shock, frequently during the operation and the patient under general anesthesia. It may be followed by the quick development, between 2 to 3 months, of multiple intrapulmonary cysts in the alveoli (Fig. 10.20).
- Local recurrence: due to the persistence of membrane remainders in the residual cavity and the consequence will be the development of a new hydatid cyst.

The management of biliary bronchial fistulas has not changed much with the advent of years. The premises of the surgical treatment include the drainage of the pleural space and the treatment of the pleural and lung lesions and the interruption



Fig. 10.18 Residual cavities





of the communication between the liver, the diaphragm, the pleura, and the lung, the so-called pleura-peritoneal "divorce" or "splitting" [46, 47]. The approach tends to be simultaneous and the approach is through a thoraco-phreno-laparotomy with simultaneous treatment of the structures: the liver, diaphragm, pleura, and lungs.



Key Points

- Diagnosis based on background, epidemiology, chest X-ray.
- Serology (arc 5 test, ELISA, and western blot).
- Surgery remains the primary choice of treatment in cystic pulmonary hydatid echinococcosis.
- Prevention should be the goal for the cestode infestation.
- A better understanding of hydatid immunoregulation may pave the way to rational immunotherapy and a future vaccine development.

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