SURGICAL MANAGEMENT OF PULMONARY HYDATIDOSIS IN CHILDREN

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SURGICAL MANAGEMENT OF PULMONARY HYDATIDOSIS IN CHILDREN (Abstract): Cystic echinococcosis (CE), the most common manifestation of the infestation with the larval form of the tapeworm Echinococcus granulosus, is a public health problem in endemic areas such as Moldavia. Aim: To retrospectively analyze pediatric patients who underwent surgery for pulmonary hydatid cyst over a 5-year period in order to assess the need for extensive lung resection. Material and methods: In the interval January 2007 – January 2012 30 pediatric patients with pulmonary hydatid cyst were surgically treated in the Department of Pediatric Surgery at the Iasi “St. Maria” Children’s Hospital. We retrospectively reviewed the medical records of these patients. Results: Patient age ranged from 3 to 14 years, and 21 patients (70%) came from rural areas. Respiratory symptoms (cough, chest pain, hemoptysis, fatigue, and dyspnea) were the main complaints. Chest radiography, computed tomography and serological tests made the diagnosis in all cases. The most common location of cystic lesions was the left lower lobe (40%). All patients were treated by enucleation and capitonnage of the residual cavity. Postoperative complications occurred in 5 cases (16.67%) and in one case (3.33%) a new hydatid cyst was diagnosed in the right lung after 2 years. Conclusions: The surgical treatment of choice was cyst enucleation with capitonnage, thus sparing the pulmonary parenchyma. Major lung resections are to be avoided in children when no severe lung destruction is detected. Keywords: PEDIATRIC PATIENTS, PULMONARY HYDATID DISEASE, ECHINOCOCCUS GRANULOSUS, ENUCLEATION, CAPITONNAGE

Cystic echinococcosis caused by \textit{Echinococcus granulosus} is a very serious social, medical and public health problem in Romania with a yearly incidence of more than 3 cases/100,000 population in some geographical areas (1).

Echinococcosis is an anthropozoonosis with worldwide distribution caused by a small tapeworm of genus Echinococcus that can infect humans in its larval state. By passing through the intestinal wall into the portal circulation, the larva can locate itself into various organs of the intermediate host, the humans. Four types of \textit{Echinococcus} (\textit{E. Granulosus}, \textit{E. Multilocularis}, \textit{E. Vogeli}, and \textit{E. Oligarthus}) cause infection in humans. \textit{Echinococcus granulosus} and \textit{Echinococcus multilocularis} are the most common, causing cystic echinococcosis and alveolar echinococcosis, respec-
Echinococcus Vogeli and E. oligogarthrus cause polycystic echinococcosis and are less frequently associated with human infection (2, 3). Liver is the most commonly involved organ in more than 60% of cases, followed by lungs (10-40%), muscles, bones, peritoneum, kidneys, spleen and brain. Unlike in adults, in children the pulmonary tissue is the most commonly involved site and, unfortunately, does not have specific symptoms (4, 5). During the cystic stage the disease may remain asymptomatic for a long period of time. When symptoms occur, they are related to parasite size, location and load and caused by compression and complications (rupture, infection).

Preoperative diagnosis is based on a combination of methods such as history, physical examination, medical imaging procedures, laboratory and serological tests. Pulmonary hydatidosis is usually managed surgically or by combining surgery with administration of antiparasitic drugs. Medical treatment alone is rarely used and controversial (6, 7).

The aim of our paper was to retrospectively analyze the pediatric patients who underwent surgery for pulmonary hydatid disease over a 5-year period in order to assess the need for extensive lung resection surgery.

MATERIAL AND METHODS

The clinical records of 30 patients who underwent surgery for pulmonary hydatid disease in the interval January 2007 – January 2012 at the Department of Pediatric Surgery at St. Maria Children’s Hospital, Iasi, Romania were analyzed. Hydatidosis was diagnosed based on physical findings, clinical symptoms, radiological examinations (chest X-ray and computed tomography), ultrasound, laboratory and serological test results (eosinophilia, enzyme-linked immunosorbent assay) and histopathological examination of surgical resection specimens.

Clinical, epidemiological, and laboratory data were analyzed together with surgical protocol details. IgG antibodies against Echinococcus granulosus were measured using commercially available enzyme-linked immunosorbent assay (ELISA) kit RIDASCREEN® Echinococcus IgG (produced by R-Biopharm AG, Landwehrstr. 54, D-64293 Darmstadt, Germany) that uses purified antigens bonded to a microwell plate. Antibodies in patient serum attach themselves to the antigens and are determined during the second phase of the test by using enzyme-labelled anti-human antibodies. The enzyme converts the colourless substrate to a blue end product, the final measurement being performed in a photometer.

Histopathological examination was performed on fragments collected during surgery, fixed by immersion in a 10% formaldehyde solution and embedded in paraffin blocks. Routine Hematoxylin and Eosin (H&E) staining was performed on sections cut from these blocks.

Surgical intervention was performed through posterolateral thoracotomy in most cases, and staged thoracotomy was the preferred surgical approach in bilateral cases. The cyst was surrounded by iodine-soaked gauze packs to prevent spillage and dissemination to surrounding structures. After neutralizing the cyst and aspirating its content, the intact germinative membrane was removed. The laminated layers were removed and then capitonnage of the residual cavity was performed. All patients received prophylaxis with Albendazole (15
mg/kg/day given orally in 2 divided doses) for 3–6 months postoperatively, in cycles consisting of 45 days of treatment alternating with 14 drug-free days. Patients were regularly followed-up postoperatively at 1 month after discharge and every 3 months thereafter for a period of 6–18 months. These visits included clinical examination, abdominal US, chest X-ray / CT if necessary, laboratory and serological tests in order to evaluate disease recurrence during the follow-up period.

RESULTS
The current study was performed in order to evaluate the clinical characteristics and surgical management of children with pulmonary hydatidosis. Of the 30 patients, 16 (53.33%) were males, 14 (46.67%) were females, and age ranged from 3 to 14 years (mean age 9.3 years). Most patients (21 cases - 70%) lived in rural areas and interacted daily with domestic animals, mainly dogs. Epidemiological investigation revealed that hydatidosis was previously diagnosed among family members of 8 (26.67%) patients. The patients presented for respiratory symptoms in 20 (66.67%) cases: cough (12 patients), chest pain (8 patients), hemoptysis (3 patients), fatigue (11 patients), and dyspnea (6 patients). A palpable abdominal mass was present in 2 (6.67%) patients having concomitant hepatic hydatid cysts. Lung hydatidosis was correctly identified on chest X-ray in 27 (90%) patients, 3 patients undergoing a computed tomography (CT) examination (fig. 1).

Laboratory test results showed eosinophilia in 21 (70%) patients, and erythrocyte sedimentation rate was elevated in 23 (76.67%) patients. Serological tests (ELISA kit RIDASCREEN® Echinococcus IgG) were performed in 12 patients and 10 (83.33%) of them were found positive, thus allowing an accurate preoperative diagnosis. The 2 patients with negative tests had uncomplicated cysts of 5.5 and 7.8 cm, respectively.

Fig. 1. Hydatid cyst located in the right lung

Cysts were located in the left lung in 14 (46.67%) patients, right lung in 12 (40%) patients, and 4 (13.33%) patients had bilateral cysts. The most frequent location was in the lower lobes, 21 (70%) patients (fig. 2).

Preoperative abdominal ultrasound was performed in all cases as part of the standard preoperative evaluation protocol, and identified concomitant liver hydatidosis in 6 (20%) patients, 2 of them with palpable masses. In 3 patients with hepato-pulmonary hydatidosis, the two locations were treated in a single operative time.

In 21 (70%) patients surgery was performed through an axillary thoracotomy; in 5 (16.67%) patients through an anterolateral thoracotomy; in 3 (10%) patients through a posterior thoracotomy; and only in one (3.33%) patient through an anterior thoracotomy. The cyst was intact in 18 (60%) patients. Bronchial fistulas were identified
in 13 (43.33%) patients and were repaired with absorbable material (Vicryl). In all cases the surgical technique consisted in enucleation and capitonnage of the residual cavity after previous inactivation with saline solution (30% NaCl) for 5-10 minutes.

![Number of cases](image)

**Fig. 2.** Location of diagnosed hydatid cysts

Postoperative complications occurred in 5 (16.67%) patients: infection (abscess) of the residual cavity in 2 patients, treated conservatively with antibiotics; pneumothorax due to reopening of bronchial fistula in 2 (40%) patients, and atelectasis of the left lung in 1 patient. No patient required reoperation, but bronchoscopy with mucus plug aspiration was required in the patient with left lung atelectasis. Hydatidosis recurrence was recorded in one patient 2 years after the first intervention; the new hydatid cyst was located in the same lung (right), and reoperation was needed despite medical treatment with Albendazole one year after the first surgery.

Mean hospital stay was 12.7 days (range 10-25 days) with a 0% mortality rate. Mean follow-up period was 2.9 years (2 months to 5 years) by chest X-ray at a 6 months interval in the first postoperative year and then annually.

**DISCUSSION**

As previous studies stated, 10-20% of pulmonary hydatidosis cases are diagnosed during childhood, the statement that in children the lung is the most commonly affected organ by cystic echinococcosis being controversial (8, 9). As in other locations, pulmonary hydatid cyst may be asymptomatic for a long period of time and diagnosed incidentally, or manifest itself only when voluminous or complicated. Pulmonary hydatidosis in children becomes symptomatic more easily compared to liver hydatidosis, due to the compressible nature of lung parenchyma, its higher vascularity and lower negative pressure (10). In endemic areas, general practitioners as well as specialist doctors (pediatrician, pediatric surgeon, general or thoracic surgeon), should be aware and suspect hydatidosis in all cases at risk (rural area, animal contact, eosinophilia, non-specific respiratory or digestive symptoms, palpable abdominal mass, acute abdomen) and rule out this disease. Chest radiography is a simple yet essential method in diagnosing lung hydatidosis as well as an ideal tool for localization and surgical planning (11). In the present study, pulmonary involve-
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...ment appears to be slightly more prevalent in boys (53.33%), possibly because they get into contact with infected materials more often than girls. As in order studies, pulmonary infestation is encountered more frequently in children older than 9 years (mean 9.3 years) (12).

The most frequently encountered symptoms in patients with pulmonary hydatidosis are determined by the mass effect exerted on surrounding tissues, rupture or supra-infection of the cyst causing pneumothorax, pleural effusion, or empyema. In the present study, patients were admitted with nonspecific respiratory symptoms in 2/3 of cases (20 patients – 66.67%). Serological tests like IgG ELISA can be used for diagnosing hydatidosis, but they should always be doubled by other methods as false negative results occur in uncomplicated lung cysts as it was noted in our study. In the present study, hydatid cysts were most frequently located in the lower lobes (21 patients – 70%), similar results with the ones reported in the literature (2, 5, 6).

Currently, there is no standardized treatment for pulmonary hydatidosis, the aim of any treatment being parasite eradication and recurrence prevention, while the additional aims of surgical treatment are prevention of intraoperative rupture and obliteration of the residual cavity (capitonnage). Surgical options in pulmonary hydatidosis are represented by enucleation with or without capitonnage, wedge resection, lobectomy and pneumectomy.

The aim of the surgical treatment is complete excision of the cyst with maximum preservation of the lung tissue. In pediatric patients there is no need to perform major lung resections like lobectomy or pneumectomy as adjacent lung parenchyma destruction is rare. In peripherally located cysts, wedge resection may be performed with minor or no sequels. In our study group, all patients were treated by enucleation with capitonnage of residual cavity. The low rate of complications, conservatively manageable, supports this type of surgical approach. Large pulmonary resections may be more deleterious to the patient and cause future morbidity.

Initially, concomitant pulmonary and hepatic hydatidosis was treated in two-stages (3 patients), enucleation of the pulmonary cyst being the first intervention due to the high risk of rupture during abdominal surgery. With the progress of anesthetic methods and better postoperative management, which allows early intensive physical therapy, hepatic and pulmonary hydatidosis have been addressed in a single surgical time (3 cases).

Axillary thoracotomy was the preferred surgical approach in our study group as it allows entering the chest only by sectioning intercostal muscles, thus producing less postoperative pain and facilitating an early recovery, crucial in pediatric surgery. At the same time, aesthetic results are better, as the scar is located in an inconspicuous area and there are no long-term sequels like scoliosis. Thoracoscopic procedures and percutaneous drainage have been tried by several authors but they are not worldwide accepted due to the high risk of dissemination and anaphylactic reaction (13).

In our study postoperative recovery was favored by an early started respiratory physiotherapy.

The use of antiparasitic drugs like Mebendazole and Albendazole has been shown in several studies to be helpful in preventing relapses (14). Their efficacy when used as a single primary treatment is debatable, as the drug has to reach the germinall layer, a process achievable only in
small cysts with thin walls. In our study, Albendazole was prescribed in 3 high-risk patients (bronchial fistulas), one of them being re-operated after two years for a new hydatid cyst in the same lung. However, due to insufficient availability of this resource and limited compliance to treatment in rural areas, a complete therapy cannot be ensured in all cases.

CONCLUSIONS
The preferred surgical treatment of pulmonary hydatidosis in our study group was cyst enucleation and capitonnage of the residual cavity, thus sparing pulmonary parenchyma. Major lung resections were avoided, as severe lung destruction is rare in children. Surgical treatment should be completed by administration of antiparasitic drugs in high-risk patients, when hydatidosis may disseminate by fistulas or secondary to perioperative complications. Pulmonary hydatidosis should be carefully considered in the differential diagnosis of respiratory or digestive symptoms in endemic areas.

REFERENCES