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Original Article

Cystotomy with Non-Capitonnage in Treating Children with Pulmonary Hydatid Disease

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Purpose: Pulmonary parenchyma saving method (cystotomy and enucleation) has been globally accepted in lung hydatidosis. However, whether capitonnage is performed or not after cystotomy is still controversial. This study aims to improve the diagnosis and treatment of patients.

Methods: We retrospectively analyzed the data of 12 pediatric patients with pulmonary hydatid cysts. These 12 patients (10 males and 2 females), with an average age of 8.7 years, underwent cystotomy without capitonnage. The mean follow-up period was 36 months.

Results: Among the 12 patients, 10 underwent thoracotomy cystotomy and 2 underwent thoracoscopic surgery with excellent outcomes except one case of postoperative broncho-pleura fistula, which was treated through thoracoscopic surgery. The mean hospital stay was 8 days. No death or recurrence occurred during the follow-up period.

Conclusion: Good therapeutic effect can be expected by combining cystotomy of pulmonary hydatid cysts with postoperative anti-hydatid drug therapy. For those unruptured (uncomplicated) hydatid lung cysts, cystotomy with the non-capitonnage method seems to be the best option, which needs to be verified by well-designed studies.

Keywords: cystotomy, non-capitonnage, children, lung, hydatid disease

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Introduction

Echinococcosis is a common zoonotic parasitic disease with global existence, mainly in regions with animal husbandry. It is prevalent in some Mediterranean and Middle Eastern countries such as New Zealand, Australia, South Africa, and South America.[1–5] In China, it mainly occurs in Xinjiang, Qinghai, Gansu, and Tibetan areas in western Sichuan and Inner Mongolia. It usually infects human organs separately or in groups, especially the liver and lung. Lung is the most commonly involved organ in children.[3–5] Children with pulmonary echinococcosis are mainly diagnosed by respiratory symptoms such as cough, expectoration, chest pain, shortness of breath, fever, and so forth. Lung hydatid cysts are more prone to rupture than those in the liver.[6] Once ruptured, it easily causes allergic reactions or suffocation, and anaphylactic shock or even death in severe

cases. To the best of our knowledge, there are rare studies[6–11] that exclusively concentrate on children with pulmonary hydatid cysts, which are partially listed in **Table 1**. This study retrospectively analyzed the data of 12 children with pulmonary echinococcosis admitted and treated in our department; we aim to accumulate experiences and thus, improve the diagnosis and treatment of patients.

Materials and Methods

The study was approved by the ethics committee of the institution (Sichuan Academy of Medical Sciences and Sichuan Provincial People's Hospital, Chengdu, China) and written informed consent was obtained from the patients for publication of this report and any accompanying images. From July 2010 to September 2018, 12 patients with pulmonary hydatid cysts were admitted and treated in our department, all of whom came from the Tibetan areas in western Sichuan (Ganzi and Aba prefecture). Among them, there were 10 males and 2 females, aged from 4.4 to 14.8 years (the average age was 8.7 years), including 8 cases of left lung, 3 cases of right lung, and 1 case of bilateral lung. The mean diameter of the hydatid lung cysts was 12.8cm (range from 4.7 to 15.6 cm). There were 8 out of 13 (one case with two cysts from bilateral lung) pulmonary cysts located at the lower lobe of the lungs (the most common site involved). The liver was concomitantly involved in 5 patients. Chief complaints included dyspnea, recurrent cough, hemoptysis, fever, and chest pain. In addition, some of the patients were accidentally found to be abnormal through physical examination. Chest radiography (**Fig. 1A**) and chest CT scan (**Fig. 1B**) suggest that the lung cyst was a single or multiple round or quasi-circular shadow, with uniform lesion density, clear boundaries, ring calcification, and "band sign." Serological tests were routinely arranged and half of the patients tested positive. Diagnosis of pulmonary echinococcosis was established from the above conditions. All the 12 patients with pulmonary hydatidosis underwent cystotomy and removal of the inner capsule (**Fig. 2A**); there were 10 cases of conventional thoracotomy and 2 cases of thoracoscopic surgery. If the bronchial cyst fistula (**Fig. 2B**) was found during the operation, it was repaired at the same time. All patients underwent prophylaxis with albendazole for 3–5 months postoperatively (15 mg/kg/day of albendazole given twice a day orally, in cycles consisting of 3 weeks of treatment alternating with 7 drug-free days). During the anti-parasitic treatment phase, liver function was monitored routinely because of drug toxicity.

Under satisfactory general anesthesia, the surgeon took the lateral position (for multiple cysts on both sides, the side where the cyst was more complicated was disposed of and then, the opposite side was tackled in the secondary operation), made the 5th or 6th intercostal incision of about 5–8 cm, and exposed the yellow and

Table 1 Results of various studies of pediatric pulmonary hydatid diseases

Authors	HE*	Kabiri[6]	Amine[7]	Haberal[8]	Aydin[9]	Cevik[10]	Karavdic[11]
Country, year	China, 2020	Morocco, 2019	Tunisia, 2014	Turkey, 2018	Turkey, 2013	Turkey, 2014	Bosnia and Herzegovina, 2011
Patients No.	12	19	25	25	42	120	72
Age (yr)	8.7	9.4	8	10.5	2-7	10.15± 3.93	10.36 ± 4.28
Mean cyst diameter (cm)	12.8	-	23/25< 5, 2/25 >8	< 7	6.2	3.89	-
Mean operative time (min)	50	-	75	-	-	-	-
Mean duration of hospital stay (d)	8	13.5	5	7	7.2	7.27	-
Complication (%)	8.3	10.5	4	16	4.76	15.1	5.56
Mean period of follow up (mo)	36	24	48	12	39	11.3	72 or more
Mortality and recurrence (%)	0	0	0	0	0	0	2/72
Capitonnage or not (yes or no)	No	Yes	Yes	Yes	Yes	Yes	No

"*" means authors from present study, "-" means unmentioned.

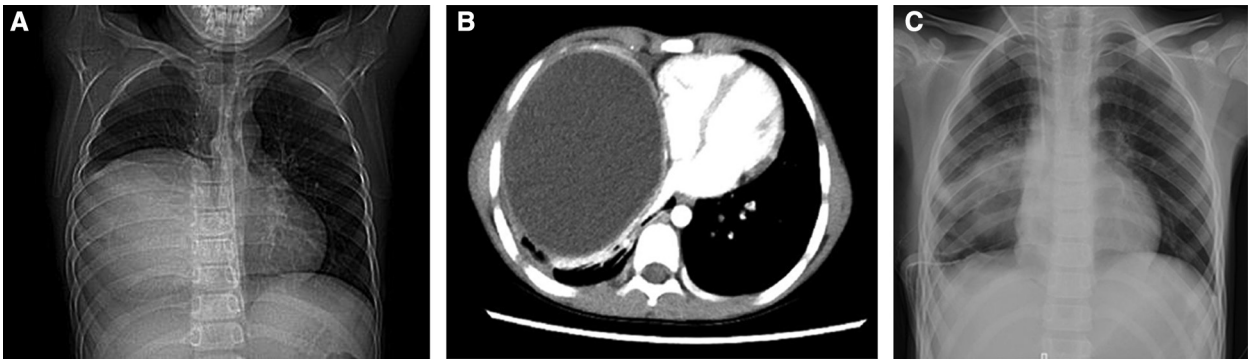

Fig. 1 (**A**) A giant spherical cystic mass was displayed in the lower part of the right thorax, resulting in atelectasis of the right lung in the chest X-ray. (**B**) A CT scan image showing a fluid-filled thick-wall cyst (nearly occupying the whole right thoracic cavity), pushing the right lung to the back and the heart to the left of the thorax. (**C**) On day 1 postoperative, a chest X-ray demonstrated that the cystic mass disappeared and the right lung partially re-expanded, leaving a cloudlike opaque where the cyst originally was. CT: computed tomography

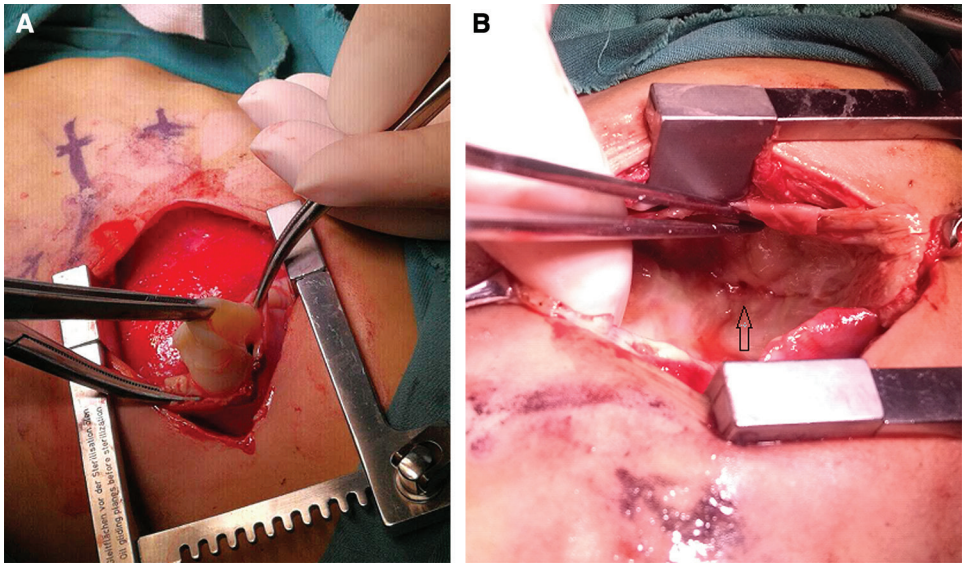

Fig. 2 (**A**) A surgical operation showing the ruptured endocyst of the pulmonary hydatid cyst being removed after cystotomy. (**B**) The bronchus connected with the pericyst of the hydatid lung cyst, forming a broncho-cyst fistula (see arrow).

white hydatid cyst lesions. The tissue around the lesion was protected with a gauze full of 10% hypertonic saline to avoid any allergy caused due to the overflow of intracystic fluid and subsequent contact with the extracystic tissue. The cystic fluid was removed through puncture and aspiration with a 50-mL syringe, the needle was fixed and the syringe was removed; sufficient amount of 10% hypertonic saline was drawn, and was finally injected into the capsule through the needle. The hypertonic saline was removed after being kept for 10 min. After cutting open the wall of the outer cyst, the collapsed cyst in the yellow-white hydatid, which separated itself from the outer cyst, could be observed. After removing the inner capsule, the residual cavity was soaked with hypertonic saline to kill the scolex. Normal saline was injected into the residual cavity for the air leakage test. If there was a bronchial fistula between the outer capsule and the bronchus, it was repaired with an absorbable suture like the figure "8."

The indications of thoracoscopy in this group were peripheral pulmonary hydatid, a cyst diameter of less than 5 cm, a single cyst, and no obvious adhesion to the chest. For selection of incision, the 6th intercostal incision in the midaxillary line was taken as the observation hole, and the 4th intercostal incision in the midaxillary line or the 7th intercostal incision in the posterior

axillary line was taken as the operation hole according to the location of the lesion. The procedure was the same as open surgery.

Results

All 12 cases were treated and discharged from hospital after 11–28 days. Among the patients co-infected with hepatic hydatid disease, two patients underwent one-stage surgery, three patients underwent pulmonary cystotomy 2 months prior to hepatic surgery, and patients with bilateral pulmonary lesions underwent staging surgery, with an interval of 2 months. All the 12 patients successfully underwent the surgery, and there were no serious complications such as intraoperative massive hemorrhage and thoracic organ injury. Typical chest X-ray image of hydatid lung cyst on day 1 postoperative was presented in **Fig. 1C**. One patient had a postoperative bronchopleural fistula, which was not healed after 2 weeks of conservative treatment and was repaired using (figure "8" suture) thoracoscopic surgery. The operation time was 65–130 min, with an average of 90 min. The blood loss was 30–110 mL, with an average of 42 mL. No wound or intrathoracic infection occurred postoperatively, and the patient was discharged 5–24 days after surgery, with an average of 6.6 days. In all of the patients, the pathological diagnoses indicated the echinococcus granulosus disease. All the children were followed-up from 15 to 82 months, with an average of 36 months. All patients were followed-up without recurrence.

Discussion

Cystic hydatid lung diseases can be diagnosed according to their history, typical symptoms, and radiological methods (such as chest X-ray and CT scan). A cyst is identified in a chest X-ray and CT scan as a round or oval homogenous opacity that can be differentiated from pulmonary parenchyma. Abdominal ultrasonography should be listed as routine to detect concomitant hepatic cysts. The Casoni experiment and hydatid complement binding experiment have been eliminated due to low sensitivity and specificity and false-negative results.[12] Among the 12 cases in this group, 11 cases were diagnosed preoperatively because of the typical chest X-ray and chest CT manifestations, and only one case was diagnosed intraoperatively. The child was admitted to the hospital in emergency complaining about "chest pain and cough for 3 days, and fever for 1 day." After admission, the chest X-ray and CT findings were different from those of a typical pulmonary hydatid cyst, and pulmonary cystic lesions could not be completely differentiated from pulmonary bullae or a cystic adenomatoid deformity or pulmonary abscess. So, thoracotomy was performed in emergency. The rupture of the pulmonary hydatid cyst was confirmed intraoperatively, and it was speculated that the child had developed an allergic reaction before admission. Fortunately, no more serious complications occurred.

It is believed that children with hydatid disease have a higher incidence of anaphylactic shock than adults, and pulmonary hydatid disease is more prone to cause anaphylactic shock in patients than hepatic hydatid disease.[10,13] In this group, no anaphylactic reaction occurred during the operations.

There may be several risk factors such as size and location of the cysts, diameter of the bronchial fistula,[14] and so on, which could lead to the rupture of the hydatid lung cysts. By analyzing the data (diameter of the bronchial fistula) of two groups (ruptured cyst versus intact one) of patients with pulmonary hydatid cyst, Akgul Ozmen C et al.[14] concluded that rupture of the hydatid lung cyst was closely related to the diameter of the bronchial fistula. Furthermore, a bronchial fistula with diameter greater than 3 mm was considered to be associated with 100% cyst rupture. That is to say, the operators could observe the size of the diameter of the bronchial fistula by means of bronchoscopy, and decide whether it was the best time or not to choose surgery for the patients based on the research. It is theoretically feasible to adopt this policy. However, the patients being examined by means of bronchoscopy do not benefit more than those who do not do bronchoscopy. When it comes to size of the pulmonary hydatid cyst, Dakak et al.[15] and Usluer et al.[16] have suggested that the large diameter of a cyst increases its propensity to rupture. In contrast, Kuzucu et al.[17] did not find statistical differences between small cyst group and giant cyst group with respect to the overall frequency of complicated (ruptured) lesions. Maybe a multi-center research has to be initiated to obtain more data to evaluate the relationship between cyst size and cyst rupture. Reasonably, if the lung cyst is located at the outermost part of the lung parenchyma with the cyst wall adjacent to the visceral pleura, it is more prone to rupture into the thoracic cavity than into the bronchus. Since rupture of the hydatid lung cyst can lead to severe complications such as dyspnea, anaphylactic shock, tension

pneumothorax, empyema, or pyopneumothorax, the best measure is to remove the inner capsule (including the contents) before it ruptures. Based on this, pulmonary hydatid cysts should be treated immediately once the diagnosis is established. As long as the patients are able to tolerate the anesthesia and operation, it is feasible to complete cystotomy of pulmonary and hepatic hydatid cysts successively in one operation, which does not only reduce the risk of reoperation and reanesthesia for patients but also benefits the patients' family in terms of cost.

Generally, the principle of surgery is to remove the lesion and retain as much lung tissue as possible. The mainstream treatment is cystotomy and enucleation of the internal capsule of the hydatid. The advantages include (1) the operation is simple and easy to be mastered; (2) it is a one-stage operation for lesions from single lung or bilateral lungs; (3) it can be used for deep cyst or burst cyst; and (4) most patients can tolerate the operation. Ayed et al.[18] and Hasdıraz et al.[19] reported single (64 cases) and bilateral (17 cases) cystotomy of pulmonary echinococcosis, with satisfactory results. In this group of patients, 10 out of 12 underwent thoracotomy cystotomy and enucleation of hydatid lung cyst (non-capitonnage); the effect was satisfactory. Traditional thoracotomy surgery can obtain satisfactory treatment effect, but it is more traumatic and the postoperative recovery of children is slower. With the introduction of the concept of minimally invasive surgery and the development of minimally invasive technology, the advantages of thoracoscopic surgery have become more and more prominent. That is, small trauma and slight pain, which is conducive for postoperative cough and respiratory function exercise, but reduces the incidence of pulmonary infection, atelectasis, and other complications. However, satisfactory results can only be achieved by accurately grasping the surgical indications and excellent operating techniques mastered by the operator. Ma et al.[20] compared thoracoscopic and open-thoracic pulmonary hydatid cystotomy, and concluded that the former method had the advantages of less trauma, less bleeding, less postoperative drainage flow, less pain, low incidence of complications, and low hospitalization cost.

Lobectomy and segmental wedge resection, as an alternative of enucleation of internal capsule, involve the removal of the inner and outer cyst along with the surrounding lung tissue to achieve the goal of eradicating the lesion, which, meanwhile, may cause lung parenchyma loss to a certain extent.

The necessity of capitonnage is still a controversial issue. Karavdic and colleagues[21] considered that the non-capitonnage surgical method resulted in a significantly shorter duration of surgery, smaller drainage of secretions, atelectasis and pleural empyema events, shorter time required for re-expansion of the surgically intervened lungs, and better overall outcome. Amine et al.[7] concluded that capitonnage appears to prevent pneumothorax, emphysema formation, and a remaining residual cavity in the long term with a significant difference. Actually, obliterating the residue cavity after cystotomy is real. However, how to perfectly carry out the measures (capitonnage) while reducing the relevant complications as much as possible is another issue. Probably, the surgeon's experiences and preference play a part in making the decision whether or not to perform capitonnage. Karavdic and Guska[11] reported that among 72 pediatric patients treated with the non-capitonnage method, there were only four minor postoperative complications (two had infection of operative wound and two had prolonged parenchymal air leakage), and air leak was managed by continuous negative aspiration.

The World Health Organization's guidelines (1996) recommend that drug treatment should only be performed in patients with primary hepatic or pulmonary echinococcosis who cannot undergo surgery, and in patients with a recurrent disease involving two or more organs. According to the WHO-IWGE,[22] the medical treatment of hydatid cysts is more effective for multiple cysts less than 5 cm in diameter compared to those greater than 10 cm in diameter. Cevik et al.[10] and Aydogdu et al.[23] hold similar views. Dogru et al.[24] reported that 30–34% of lung hydatid cyst patients treated with albendazole achieved full recovery, with another 34% of the patients showing some treatment benefit.

If the hydatid cysts do not rupture, however, Todorov et al.[25] and Cevik et al.[10] consider that anti-hydatid treatment makes the walls of the hydatid cyst brittle, thus increasing the risk of cyst rupture. The question is how to balance medical therapy and surgical intervention while avoiding cyst rupture as much as possible. Keramidas et al.[26] recommended that patients with cystic echinococcosis who receive medical therapy are followed closely for at least 2 months as most complications occurred approximately 2 months from the start of treatment. Echinococcosis is a systemic disease, and pulmonary hydatidosis is only a manifestation of systemic hydatid cysts in the lungs. Therefore, in terms of treatment, comprehensive methods combining internal and

external factors should be adopted.[10,27] Currently, anthelmintic drugs such as albendazole or mebendazole have been used globally for decreasing the risk of recurrence in patients undergoing surgery for cystic echinococcosis.

All the children in this group received conventional anti-hydatid drug treatment for 3 months (5 months for those who underwent staging surgery) postoperatively.

Conclusion

The analysis of the 12 cases of children with pulmonary echinococcosis in this group shows that good to excellent therapeutic effect can be expected by combining cystotomy of the pulmonary hydatid cysts with postoperative anti-hydatid drug therapy. According to the indications, it is feasible to remove the inner capsule of pulmonary hydatid cyst using thoracoscopic surgery. For the unruptured (uncomplicated) hydatid lung cysts, cystotomy with the non-capitonnage method seems to be the best option, which needs to be verified by well-designed studies.

The shortcomings of this study lie in the fact that it was a retrospective study and the sample size was small, thus not forming a strong clinical evidence. Prospective studies need to be designed and more data need to be collected and analyzed, to form convincing evidence. In this study, the average follow-up time was 3 years, which could only indicate the medium-term effect, and the patients need to be followed-up for a longer time to evaluate the long-term effect.

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Disclosure Statement

The authors declare that they have no conflicts of interest to declare.

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