

Research Article

**Single-Stage Surgery of Hepatic and Pulmonary Hydatid Cysts in Children:
A Report of Seven Successful Experiences (2011- 2016)**

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ABSTRACT

Introduction: Cystic echinococcosis (CE) caused by *Echinococcus granulosus* is a major public health problem worldwide for Endemic areas, including Iran. In this paper, we report seven successful treatments of hepatic and pulmonary hydatid cysts in children with single-stage surgery (2011-2016).

Case Presentation: The present retrospective descriptive study recruited seven cases of single-stage surgery of hepatic and pulmonary hydatid cysts in children (2011-2016) in Taleghani Hospital (Gorgan, Iran). A researcher-made checklist was used to collect data, including demographic data such as age, gender, place of residence (urban and rural) and patient's clinical information such as clinical presentations during the visit, ultrasonography report, CT scan report and laboratory findings. After consultation with infectious disease specialist, patients underwent right posterolateral thoracotomy and phreno-laparotomy.

Conclusions: It can be concluded that single-stage surgery has several advantages for the treatment of hepatic and pulmonary hydatid cysts such as avoiding anesthesia-related harm, avoiding the second surgery complications, preventing possible complications of cysts remaining and exacerbation of patient's general health, decreasing recovery time and overall costs of treatment.

Keywords: Hydatid cyst, pediatric surgery, risk factor, Iran

INTRODUCTION

Hydatid disease is caused by the larval stage of *Echinococcus granulosus*, which is responsible for the most common type of hydatid disease in human. (1) Adult worm lives in the intestines of canines. Hydatid cysts develop in human through direct contact with infected dogs and eating food and water contaminated with parasite eggs. Eggs open in the intestine and the larvae penetrate through the intestinal mucosa and enter the blood.

(2) Endemic areas for hydatid disease are countries breeding sheep especially the Mediterranean region, Africa, South America, Middle East, Australia and New Zealand. Iran in the Middle East is also a country with high prevalence of hydatid cyst, in which the disease is considered the most common helminth parasitic disease. (3) Although there are no accurate statistics on the prevalence of infection in Iranian people,

according to studies conducted on patients in hospitals, it is estimated that 1-1.12per 100000 patients suffer from hydatidosis (4) and most cases have been reported in Khorassan, Isfahan and Fars provinces. (5)

Hydatid cyst can infect all organs, but the liver is the most common organ involved in adults (75%), followed by the lungs (15%). (1) In addition, multi-organ involvement has also been reported, such as “liver and spleen” and “liver and lung”. (6) Lungs in children are the most common site of involvement. Hydatid cyst infects the lung 64% and the liver 28% in children. The primary methods of transmission in children include contact with dogs, drinking contaminated water or eating contaminated food. Therefore, it is important to diagnose and treat hydatid cysts in children. (7)

Hydatid cysts have their special features compared to other cysts in other areas. The lungs facilitate the growth of cysts because of the compression and negative pressure in them. As a result, hydatid cysts grow three times as much in the lungs than in the liver (1). At present, surgical removal of cysts is an accepted treatment of choice for pulmonary hydatidosis. Medicinal treatment is used only for those who refuse surgical treatment or those who have contraindications for surgical procedures. (8) It is still controversial to operate extensive bilateral pulmonary hydatidosis. As a result, the selection of surgical technique depends on the location, size and the normal area of hydatid cysts. (7) This study aimed to report the experience of seven successful treatments of hepatic and pulmonary hydatid cysts in children with single-stage surgery (2011-2016).

MATERIALS AND METHODS

The present retrospective descriptive study recruited seven cases of single-stage surgery of hepatic and pulmonary hydatid cysts in children (2011-2016) in Taleghani Hospital (Gorgan, Iran). A researcher-made checklist was used to collect data, including demographic data such as age, gender, place of residence (urban and rural) and

patient's clinical information such as clinical presentations during the visit, ultrasonography report, CT scan report and laboratory findings. The checklist was completed based on the data in patients' medical record (admission sheet, medical history, medication orders, surgical report sheet, and diagnostic and laboratory tests).

Hematology, biochemistry and pulmonary function tests were done on all children attending the center with fever, respiratory distress, abdominal pain, nausea and vomiting. Afterward, abdominal ultrasonography, CT scan of the chest and Ab hydatid cyst serology tests were performed for the diagnosis of hydatid cyst. Clinical examinations of all patients revealed significantly diminished breath sounds. CT scan reports showed large homogeneous cysts with a thin wall both in the lungs and in the liver (Figure 1). Despite numerous large cysts in the liver, liver function tests were normal in this group of patients. After patients were confirmed to have simultaneous hydatid cysts in the lung and the liver, they were selected for one-stage surgery by the physician. Children's fever was controlled prior to surgery and albendazole was initiated for them. After consultation with infectious disease specialist, patients underwent right posterolateral thoracotomy and phreno-laparotomy.

Surgical Technique

After general anesthesia, right posterolateral thoracotomy incision was made in the fifth intercostal space (Figure 2). After excision and capitonnage of pulmonary hydatid cyst, pleural space was rinsed with hypertonic saline. The diaphragm was opened by peripheral incision and hydatid cyst was removed from liver after exposure and cyst cavity was washed with hypertonic saline. The excised hydatid cyst was sent for pathology tests. Then an abdominal drain was placed in the cavity in the liver. Diaphragm was repaired with absorbable suture, a chest tube was placed in the pleural space and thoracic wall was repaired.

The chest tube was removed after the lungs were expanded about the third day after surgery and

when there was no air bubbling. In addition, when the abdominal drainage stopped, the drain was removed on the 5th postoperative day. The patients were discharged in good general condition and without fever, with an albendazole prescription. In addition, all patients were visited every two months for six months to control the disease and ensure complete treatment through laboratory tests and ultrasonography. The collected data were analyzed by descriptive statistics in SPSS.

RESULTS

From a total of seven children with hydatid cysts, three were girls (42.8%) and four were boys (57.14%). Their mean age was 3.4 and five of them were living in rural areas. The results showed that fever and cough were the most common symptoms of patients with hydatid cyst (Table 2).

affects mostly rural population (11), which is consistent with the results of the present study.

Cysts in humans may be asymptomatic in early stages, but as the cyst enlarges, the patient becomes symptomatic. Liver hydatid cyst can present as a dull pain in the upper abdomen or even a palpable tumor in the upper abdomen. (12) In addition, pulmonary cysts present as dry cough, dyspnea, chest pain and hemoptysis. (13) Gastrointestinal symptoms such as nausea and vomiting associated with fever have been reported in these patients. (14) Most children in the present study came to the hospital with early symptoms of dyspnea, nausea and abdominal pain, which is consistent with the above studies.

Imaging, especially ultrasound, CT scan, and plain radiography are used to diagnose hydatid cyst and the diagnosis is confirmed by finding serum antibodies. (15) The most common location in the

Lung lobe involvement	RLL: Right lower lobe	LmL: Left middle lobe	LLL Left lower lobe.	Liver
Number and percentage	4 (57.4)	3 (42.8)	5 (71.4)	7 (100)

Type of symptoms	Cough	Dyspnea	Fever	Abdominal pain	Nausea	Vomiting
Number and percentage	6 (85.7)	5 (71.4)	6 (85.7)	4 (57.4)	4 (57.4)	3 (42.8)

According to the results, the lower lobe of the right lung (57.4%) was the most common site for hydatid cyst (Table 2).

The mean size of cysts was 29 mm (16-42 mm). All cysts were removed without lung resection. All patients were well controlled and no recurrence and death were found in any of the cases.

DISCUSSION

Hydatid disease is still a national problem in highly endemic countries and epidemiological interventions are needed to eradicate the disease (9). Hydatid disease is known as a rural disease as it is transmitted from animals to humans. (10) The results of various studies indicate that the disease

lungs is lower lobes. More than one cyst exists in 30% of cases and bilateral cysts are found in 20% of cases. (1)

The treatment of hydatid cyst has not changed in recent years and surgery is the treatment of choice. In the study of Bakal et al. (2015) conducted on ten children with hydatid cyst, it was shown that surgical treatment is the treatment of choice for hydatid cysts in children. (16) Two-stage surgical technique is a routine procedure for the treatment of pulmonary hydatid cyst. In this method, patients undergo a two-stage thoracotomy within three to five days, which increases complications and the length of stay. (17) Hasdiraz et al. (2013), in their study on 17 patients, used the two-stage method and reported increased length of stay. (8)

In the present study, hepatic and pulmonary hydatid cysts were treated by a single-stage surgery. Dennis et al. and Galindo et al. used this method on 19 and 24 children, respectively. They believed that single-stage surgery has few complications.(18) It can be concluded that single-stage surgery has several advantages for the treatment of hepatic and pulmonary hydatid cysts such as avoiding anesthesia-related harm, avoiding the second surgery complications, preventing possible complications of cysts remaining and exacerbation of patient's general health, decreasing recovery time and overall costs of treatment.

Several studies show that the majority of large hydatid cysts can be treated with cyclic albendazole, but ultrasound examination is necessary for follow-up (8, 19). However, some studies show that this treatment alone can cause numerous complications and cysts are rarely eradicated. (20) In the above study, patients were treated with albendazole and were followed up by ultrasound after surgery.

Studies show that the mortality rate during surgery is from 1% to 2%. (9) In the present study, no recurrence or death was observed during the follow-up of patients five years after the surgery.

CONCLUSION

The presence of large multi-organ cysts is unusual in young children. The presence of large cysts in children is more harmful than in adults as it leads to compression and progressive destruction of the organ ultimately leading to long-term consequences. The cyst rupture can cause pulmonary fistula in pulmonary cysts and bile duct fistula in hepatic cysts. The present study showed that using one-stage surgery for hepatic and pulmonary hydatid cysts is an appropriate alternative even for children.

REFERENCES

1. Garg, M.K, Sharma, M , Gulati, A, Gorski, U, Aggarwal AN, Agarwal, R, Khandelwal, N.

- (2016) Imaging in pulmonary hydatid cysts. *World J Radiol.* vol -8,issue6,pg 581-587.
2. Yousofi, H, Hashemzadeh, M, Kohansal K, Zabardast, N, Shirzad, H, Shahabi G.A. Survey about Protective Effect of *Echinococcus Granulosus Protoscolices* Surface Antigens in Preventing Secondary Hydatid Cyst. *Armaghane-danesh quarterly journal of yasuj university of medicalsciences* volume 11/no.3/autumn 2006.
3. Goel, M.C, Aggarwal, M.R, Misra, A. (1995) Percutaneous drainage of renal hydatid cyst: early results and follow-up. *Br J Urol*, vol- 75, pg 724-728.
4. Iranian Doctors website. *Echinococcus granulosus*. Available online in August 13 2008 at <http://www.pezeshkan.org>.
5. Rohani, Z, Naroienjad, M. (2007). A Case Report of Hydatid Cyst of Diaphragm. *Iran Univ Med Sci J*, vol- 14, pg 95-99. (Persian).
6. Jahani, M.R, Roohollahi, G.H, Gharavi, M.J. (2004) Splenic Hydatid cyst in a 20 years old soldier. *Mil Med* vol- 169, pg 77-80. (Persian).
7. Ma, J., Wang, X., Mamatimin, X., Ahan, N., Chen, K., Peng, C., & Yang, Y (2016). Therapeutic evaluation of video-assisted thoracoscopic surgery versus open thoracotomy for pediatric pulmonary hydatid disease. *Journal of Cardiothoracic Surgery* vol- 11, pg 129-132.
8. Hasdiraz, L, Onal, O , Oguzkaya F (2013). Bilateral staged thoracotomy for multiple lung hydatidosis. *Journal of Cardiothoracic Surgery*, vol- 8, pg 121-125.
9. Tensaw, I.M.(2010). Hydatid cyst in children, *Ann Ped* vol-6 ,issue2,pg 98-104.
10. Schantz, P.M, Wang, H, Qiu, J, Liu, F.J, Saito, E, Emshoff, A, et al. (2003) *Echinococcosis on the Tibetan plateau: prevalence and risk factors for cystic and alveolar echinococcosis in Tibetan*

- populations in Quinghai Province, China. *Parasitol*.vol-127, pg 109–20.
11. Dopchiz, M.C, Elissondo, M.C, Andresiuk, M.V, Maiorini, E, Gutierrez ,A.M, Muzulin PM,et al. (2009). Pediatric hydatidosis in the south-east of the Buenos Aires province, Argentina. *Rev Argent Microbiol*.vol-41, pg 105–11.
 12. Brunicardi, F, Dana, k, Thmothy, R. (2006).*Schwartz's Principle of Surgery*, 8th ed. New York, The McGraw-Hill Companies.pg 1163-1165.
 13. Bakir, F. (1967).Serious complications of hydatid cyst of the lung. *Am Rev Respir Dis*; vol-96,pg 483-493.
 14. Aydogdu, B , Sander, S, Demirali, O, Guvenc, U, Besik, C, Kuzdan, C, Goya, C, Tireli, G (2015). Treatment of spontaneous rupture of lung hydatid cysts into a bronchus in children. *Journal of Pediatric Surgery*.vol-50,issue9, pg 481–1483.
 15. Erkan, N, Haciyanli, M, Yildirim, M, Yilmaz, C.(2004). A case report of the unusual presence of hydatid disease in the pancreas and breast. *J Pancreas (JOP)*,vol-5,pg 368-72.
 16. Bakal, U, Simsek, S, Kazez, A. (2015).Surgical and Molecular Evaluation of Pediatric Hydatid Cyst Cases in Eastern Turkey. *Korean J Parasitol* Vol- 53, issue 6,pg 785-788.
 17. Shehatha, J, FRACS, Alizzi J, (2008). Thoracic hydatid disease; a review of 763 cases. *Heart Lung Circ*. vol- 17, pg 502–504.
 18. Galindo, R, Cherkaoui, O, Abdelaoui, A, Bennis, A, Biaz, A, Laraki, A.(1981). Le traitement du kuste hydatique pulmonaire ches l'enfant. *Ann Chir*. vol-35, issue3, pg 213-215.
 19. Li, T, Ito, A, Pengcuo, R, Sako, Y, Chen, X, Qiu, D (2011). Post-Treatment Follow-Up Study of Abdominal Cystic Echinococcosis in Tibetan Communities of Northwest Sichuan Province, China. *PLoS Negl Trop Dis*.vol- 5,issue10,pg 1364-1367.
 20. Albendazole, J.H. (2000) A review of anthelmintic efficacy and safety in humans. *Parasitology*,vol- 121, pg 113–132.