Evaluation of the effect of pulmonary hydatid cyst location on the surgical technique approaches

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INTRODUCTION

Hydatid disease had been recognized since the Galen and Hipocrates, and in 1808 Rudolphi used the term hydatid cyst to describe echinococcosis in humans. A hydatid cyst is a parasitic infestation caused most commonly by echinococcal granulosus. The most common location of a hydatid cyst is liver and lungs at the second place (10-40%).[1] Generally, a pulmonary hydatid cyst is diagnosed by radiological imaging, physical examination and history findings.

The preferable treatment for pulmonary hydatid cysts is operation. Various surgical procedures are described in the literature. The surgical procedures may be conservative (cystostomy, enucleation of intact cysts, removal of the cyst after needle aspiration with or without pericystectomy, with maximal preservation of lung parenchyma) or radical (lung parenchyma resection with cystotomy and pericystectomy, wedge resection, segmentectomy and lobectomy).[2] However, the selection of surgical technique depends on the conditions throughout surgery. While hydatid disease is endemic in Iran and the highest rate is reported for Khorasan Province and few studies have evaluated the relation between location of the cysts and type of surgical technique, we aimed to perform this study to assess surgical treatment of pulmonary hydatid cysts and evaluate whether the location of a cyst affects the choice of surgical technique.

MATERIALS AND METHODS

In this study, the patients with a pulmonary hydatid cyst who had been referred to thorax surgery section of Qaem Hospital related to Mashhad University of Medical Sciences from 2010 to 2012 were evaluated. Among 99 patients, 12 cases were excluded because 6 patients...
had multiple bilateral lung hydatid cysts and they treated medically, 4 patients were inoperable due to other medical problem and 2 patients rejected surgical treatment.

Sampling method was non-probable and simple sampling. Initially, all conditions of the study were completely described for the patients and in case of their assent; they would sign a written informed consent. Moreover, the study was approved by the Regional Ethics Committee of Mashhad University of Medical Sciences (project number: 900415).

The studied variables included age, gender, clinical symptoms, type of surgery, postoperative complications, number of hydatid cysts in lungs and their size and location, hospitalization, bilateral lung hydatid cyst and mortality. A questionnaire was prepared and the information was recorded.

The patients were admitted as elective or emergency. Diagnosis was definite in the patients referred as elective. For the patients who had been referred as emergency, after obtaining history and physical examination, chest x-ray and CT scan were performed. For the cases with unsure diagnosis, indirect hemaglutinin serology test was done. The surgical technique approach was based on the location, size and number of cysts.

Preoperative evaluation was performed. Among 87 patients, for 14 cases with over 40 years old we did an electrocardiography and in 13 patients with history of chronic lung disease, lung function tests were performed. According to the results of these tests and having no contraindication for surgery, the patients were candidates for surgery.

Surgical technique
Before the operation, the anesthetist was asked to start corticosteroids to prevent anaphylactic shock. All the cases were put in decubitus lateral position and posterolateral thoracotomy was performed through the fifth, sixth or seventh (depending on the location of the cyst) intercostal spaces. To prevent contamination of thoracotomy site and the surrounding tissue with hydatid cyst fluid, the surrounding of cyst was trapped with gas containing hypertonic sodium chloride solution 20%. During the operation, two suctions were ready; one inserted into trocar and the other in terminal suction.

According to the location, number and size of cysts, we selected each of the following operation techniques: Cystotomy and bronchial closure and capitonnage, cystotomy and bronchial closure, segmentectomy, enucleation, and lobectomy. Then, the patients were evaluated for the relation between the location of a cyst and the choice of surgery. The patients were followed up for 1 month in terms of postoperative complications.

To describe data, statistical charts and tables were used and data analysis was performed by SPSS version 19. The Chi-square test was used to compare treatment outcome and morbidity in the different methods of surgery and variance analysis was used to compare the duration of hospitalization in the different methods of surgery. P ≤ 0.05 was considered statistically significant.

RESULTS
A total of 87 patients with diagnosis of a pulmonary hydatid cyst were evaluated. Among them, 46 cases (52.9%) were males and 41 (47.1%) were females. The most affected age group was patients in their third decade with a mean age of 29.61 ± 14.27 (minimum 7 years and maximum 70 years). Clinical symptoms of the patients are shown in Table 1.

The incidence of hepatic involvement was 19.5% (17 cases) and the hepatic localization was not complicated in the chest in 16 patients. We operated a lung hydatid cyst at first and then a liver cyst operated in the second stage (6 weeks later). One patient was referred due to bilioptysis and he was treated for both lung and liver cysts in first stage. The surgical technique approach for the patient was right posterolateral thoracotomy incision. We found a cyst in the middle lobe and fistula between diaphragm and lower lobe. We evacuated a lung cyst and capitonized. Then diaphragm was opened and the hepatic cyst was drained and was capitonized. Sub-diaphragmatic drain and chest tube were inserted.

The rate of bilateral hydatid cyst was 22 cases (25.3%) and unilateral hydatid cyst was 65 cases (74.7%).

Among 87 patients, 61 cases (70.1%) had a cyst with size of <10 cm and 26 (29.9%) had cyst with size of ≥10 cm. In terms of the number of cysts, 5 patients (5.7%) had more than three cysts and 67 cases (77%) one cyst, 9 (10.3%) two cysts, and 6 (6.9%) three cysts.

About the location of the cysts, in 19 cases (21.8%), the location of cyst was right lower lobe, in 12 (13.8%) right upper lobe, in 26 (29.9%) left lower lobe, in 11 (12.6%) left upper lobe, in 3 (3.4%) middle lobe, in 5 (5.7%) we observed pleural involvement and multi-lobe involvement in 11 (12.6%).

Table 1: Symptoms of a pulmonary hydatid cyst in the studied groups

| Symptoms                          | N (%) |
|----------------------------------|-------|
| Cough                            | 57 (65.5) |
| Chest pain                       | 31 (35.5) |
| Blooding expectoration           | 29 (33.3) |
| Fever                            | 29 (33.3) |
| Vomica                           | 18 (20.7) |
| Hemoptisis                       | 4 (4.6) |
| feeling of pressure in the thorax| 27 (31) |
| Asymptomatic                     | 14 (16.1) |
| Anaphylactic phenomena           | 5 (5.7) |
| Growth disorder                  | 2 (2.3) |
| Weight loss                      | 18 (20.7) |
| Asymmetry of the chest           | 1 (1.15) |
The surgical technique approach according to the location of the cysts in the patients with a pulmonary hydatid cyst is shown in Table 2. There was significant relation between the existence of the cyst in the middle lobe and performing lobectomy ($P = 0.016$).

The choice of operation technique according to the size of the cysts in the patients with a pulmonary hydatid cyst is shown in Table 3. No significant relation was found between the size of cyst and lobectomy ($P = 0.682$).

Selection of operation technique according to the number of the cysts in the patients with a pulmonary hydatid cyst is shown in Table 4. No significant relation was observed between the number of cysts and lobectomy ($P = 0.344$).

In terms of hospitalization, nine cases had hospital stay for more than 9 days and five of them had undergone lobectomy.

The most common postoperative complication in the studied patients was prolonged air leak, which occurred in 12 cases. Other postoperative complications were empyema in 1 case, hemoptysis in 5 patients and bile secretion from drain in 1 case. In the last patient we performed conservative treatment because the patient was severely ill. After 2 months bile leakage subsided. We observed no complication in 68 patients. The rate of mortality was 1.15% (one case). He had lung and mediastinal cysts. The mediastinal cyst was contagious to great vessels. After opening the cyst, the patient had severe bleeding. We managed bleeding process during operation but the patient expired 2 days later in ICU due to multiorgan failure.

The mean follow-up time was 1 month. Among the referred patients, 96.8% had no severe problem and 3.2% had complication and 25 patients didn't refer for follow-up.

**DISCUSSION**

A hydatid cyst is a parasitic infestation caused mostly by echinococcal granulosus. It is an important health problem in regions where people earn the living by agriculture and livestock raising, while veterinary services, public health and preventive policies are poorly offered. Common complaints of patients with lung hydatid cysts are dry cough, hemoptysis and feeling pressure in the thorax. Symptoms are related to size, location and eventual rupture of the cysts. Hydatid disease when is located in lungs can reach a certain size without causing any symptom. A cyst is identifiable in chest x-ray and computed tomography (CT) scanning as a round or oval homogenous opacity that can be differentiated from pulmonary parenchyma. Serological investigations have a limited value in diagnosis of lung hydatid cysts. Some studies reported that in patients with pulmonary hydatid disease, the lung parenchyma should be preserved as much

**Table 2: Type of operation according to the location of cysts**

| Type of operation Location of cysts | Segmentectomy | Lobectomy | Enucleation | Cystotomy+bronchial closure | Cystotomy+bronchial closure+capitonnage | Total N (%) |
|-------------------------------------|---------------|-----------|-------------|-----------------------------|----------------------------------------|-------------|
| Right lower lobe                    | 1             | 2         | 1           | 6                           | 9                                      | 19 (21.8)   |
| Right upper lobe                    | 1             | 1         | 0           | 0                           | 10                                     | 12 (13.8)   |
| Left lower lobe                     | 4             | 0         | 1           | 4                           | 17                                     | 26 (29.9)   |
| Left upper lobe                     | 0             | 0         | 0           | 4                           | 7                                      | 11 (12.6)   |
| Middle lobe                         | 0             | 2         | 1           | 0                           | 3                                      | 3 (3.4)     |
| Pleural involvement                 | 1             | 1         | 0           | 3                           | 0                                      | 5 (5.7)     |
| Multi-lobe involvement              | 1             | 1         | 0           | 2                           | 7                                      | 11 (12.6)   |
| Total                               | 8             | 7         | 3           | 19                          | 50                                     | 87 (100)    |

**Table 3: Type of operation according to the size of cysts**

| Type of operation Size of cysts | Segmentectomy | Lobectomy | Enucleation | Cystotomy and bronchial closure | Cystotomy and bronchial closure and capitonnage | Total number (%) |
|---------------------------------|---------------|-----------|-------------|-------------------------------|-----------------------------------------------|------------------|
| <5 cm                           | 4             | 1         | 0           | 0                             | 12                                            | 17 (19.5)        |
| 5-9 cm                          | 2             | 3         | 3           | 11                            | 13                                            | 25 (50.6)        |
| ≥10 cm                          | 2             | 3         | 0           | 8                             | 13                                            | 26 (29.9)        |
| Total                           | 8             | 7         | 3           | 19                            | 50                                            | 87 (100)         |

**Table 4: Type of operation according to the number of cysts**

| Type of operation Number of cysts | Segmentectomy | Lobectomy | Enucleation | Cystotomy and bronchial closure | Cystotomy and bronchial closure and capitonnage | Total number (%) |
|-----------------------------------|---------------|-----------|-------------|-------------------------------|-----------------------------------------------|------------------|
| 1                                 | 6             | 4         | 2           | 18                            | 37                                            | 67 (77)          |
| 2                                 | 1             | 1         | 0           | 1                             | 6                                             | 9 (10.3)         |
| 3                                 | 1             | 1         | 0           | 0                             | 5                                             | 6 (6.9)          |
| >3                                | 0             | 2         | 1           | 0                             | 2                                             | 5 (5.7)          |
| Total                             | 8             | 7         | 3           | 19                            | 50                                            | 87 (100)         |
as possible and radical procedures must be avoided.\textsuperscript{[12]} Karavdic \textit{et al.} reported that surgical treatment is related to size, localization, clinical manifestation and eventual complication of the cysts.\textsuperscript{[9]}

The most common localization of a hydatid cyst is the liver (50-60%) and secondly the lungs (10-30%).\textsuperscript{[7]} A hydatid cyst should be treated as soon as diagnosis is made, since it may cause serious complications by means of rupture into bronchi and pleural cavity or vital organ compression.\textsuperscript{[8]} Surgical treatment is preferred in pulmonary hydatid cysts.\textsuperscript{[9]} Medical treatment is considered for small cysts without complication, in patients who are high risk for surgery and those who reject the surgery.\textsuperscript{[10]} The choice of surgical technique is based on the location, size and the number of cysts. Since, hydatid disease is endemic in some parts of the world including Iran, we aimed to perform this study to evaluate whether the location of cyst affects the choice of surgical technique or not.

In this study, the most affected age group was 21-30 years with a mean age of 30 years and when compared with other studies, the same age group was involved.\textsuperscript{[11]} Although it may occur in any age group, it is more common in the third and fourth decades and is more prevalent in men.\textsuperscript{[12]} In this study, the rate of incidence of a pulmonary hydatid cyst was 52.9% in male and 48.1% in female. In a study by Bulent \textit{et al.} no significant differences were found between the incidence rates by the gender.\textsuperscript{[13]} However, our result about the gender-related incidence rate was similar to those reported in the study performed by Bilgin \textit{et al.}\textsuperscript{[14]} But, the study of Ghaffarifar reported that the incidence rate is more common in females.\textsuperscript{[15]}

In the referred patients of this study, 16.1% were asymptomatic and 83.9% were symptomatic. Common complaints were dry cough and chest pain. This was similar to the study of Bagheri \textit{et al.} which reported that in the patients with a pulmonary hydatid cyst, cough and chest pain were the most common complaints (62%, 56%, respectively).\textsuperscript{[12]} Sehitogullari \textit{et al.} in the study with 102 patients, 62 males and 40 females, reported that there is a relation between symptoms of patients and localization and size of the hydatid lung cysts.\textsuperscript{[16]}

In this study, the incidence of hepatic involvement was 19.5% and the rate of bilateral hydatid cyst was 25.3%. Various studies have reported multifocal involvement rates of 8% to 30%. Dogan \textit{et al.} in the study involving a series of 1055 patients reported that the incidence rate of concomitant hepatic and pulmonary involvement was less than 10%.\textsuperscript{[17]} In the study of Karavdic \textit{et al.} cyst localization was unilateral in 87.5% and bilateral in 12.5%.\textsuperscript{[8]}

In the present study, 70.1% of the patients had a cyst with size of <10 cm and 29.9% had a cyst with size of ≥10 cm. No significant relation was found between the size of cyst and lobectomy ($P = 0.682$). Ushler \textit{et al.} performed a study on two groups of the patients with a hydatid cyst: Those smaller than 10 cm (group A) and those 10 cm or greater (group B). There were 462 patients (86%) in group A and 75 patients (14%) in group B. Lobectomy rates were significantly higher in group B compared with group A ($P = 0.038$).\textsuperscript{[18]} Also, Isitmangil \textit{et al.} reported that one of the indications for lobectomy is large cysts involving more than 50% of the lobe.\textsuperscript{[19]} Moreover, in this study, we didn't find any significant relation between the number of cysts and performing lobectomy ($P = 0.344$). However, the literature review demonstrated one of the major indications of lobectomy, the number and not the size of the cysts.\textsuperscript{[11]}

In our study, the left lower lobe was the most frequently affected area of the lung (29.9%) and the right lower lobe was affected as 21.8%. Ulu\textit{k} \textit{et al.} concluded that the right lower lobe was the most frequently attacked area of the lung.\textsuperscript{[16]} The study of Sehitogullari showed that the right lower lobe was the most affected area of the lung.\textsuperscript{[17]} Their finding is not similar to our study. Also, in the present study, there was a significant relation between the existence of a cyst in the middle lobe and lobectomy ($P = 0.016$). Rochan \textit{et al.} stated that lobectomy should be performed in cases where more than half of the lobe is involved.\textsuperscript{[7]} Of course, no study has been performed so far to evaluate the relation between the location of cysts and lobectomy.

Postoperative morbidity rates have varied between 0% to 17%, with air leak as the most common complication.\textsuperscript{[17]} In our study also, the most common postoperative complication was air leak in 12 patients of which 25% of cases of air leak was observed in the patients who underwent lobectomy. There was a significant relation between postoperative air leak and lobectomy ($P = 0.052$).

In the medical literature, mortality rates ranged from 0% to 2%.\textsuperscript{[20]} In our study, the rate of mortality was 1.15%.

\textbf{CONCLUSION}

In spite of having not enough cases (89 patients) and cases with a middle lobe cyst were few (7 patients), our data indicate if a hydatid cyst is located in the middle lobe, it may need lobectomy. Although size and number of cyst are determining factors for the selection of operation, but the location and the ratio of destruction of lung are important factors too.

Also, in patients with underlying pulmonary diseases if the lesion is completely removable with lobectomy, it is better not to insist on conservative surgical therapy, because it leads to incomplete tissue removal and therefore more complications would be expected. Moreover, since, pulmonary hydatid disease is endemic in the area of Khorasan, registry of the cases for better organizing the health, medical and statistical burden and prevalence of this disease is recommended. This will help to reduce the incidence of this disease. However, more studies with larger sample size are required to obtain more accurate results.
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REFERENCES

1. Guska S, Cerimagić Z, Pilav I. Conservative surgical treatment of pulmonary hydatid disease in children. Med Arh 2007;61:11-5.
2. Bagheri R, Haghi SZ, Amini M, Fattahi AS, Noorshafiee S. Pulmonary hydatid cyst: Analysis of 1024 cases. Gen Thorac Cardiovasc Surg 2011;59:305-9.
3. Blanton R. Echinococcus. In: Behrman RE, Kliegman RM, Jenson HB, editors. Nelson Textbook of Pediatrics. 16th ed. Philadelphia: WB Saunders; 2000. p. 1079-81.
4. Köseoglu B, Bakan V, Onem O, Bilici S, Demirtas I. Conservative surgical treatment of pulmonary hydatid disease in children: An analysis of 35 cases. Surg Today 2002;32:779-83.
5. Mirshemirani AR, Razavi S, Sadeghian S. Surgical treatment of pulmonary hydatid cyst in 72 children. Tansaffos 2009;8:56-61.
6. Karavdic K, Guska S. Surgical treatment of pulmonary hydatid disease in children: A retrospective study. Med Arh 2011;65:16-9.
7. Rochan RB, Rice CL, Carrico CL. Hydatid disease of the lung. In: Shields TW, editor. General Thoracic Surgery. 4th ed. Philadelphia: Lippincott Williams and Wilkins; 2000. p. 1021-38.
8. Ramos G, Orduña A, García-Yuste M. Hydatid cyst of the lung: Diagnosis and treatment. World J Surg 2001;25:46-57.
9. Shaw JM, Bormann PC, Kriege JE. Hydatid disease of the liver. S Afr J Surg 2006;44:70-2, 74-7.
10. Ulku R, Yilmaz HG, Onat S, Ozçelik C. Surgical treatment of pulmonary hydatid cysts: Report of 139 cases. IntSurg 2006;91:77-81.
11. Harlafts NN, Aletras HA, Symbas PN. Hydatid disease of the lung. In: Shields TW, LoCicero J, Ponny RB, Rusch VW, editors. General Thoracic Surgery. 6th ed. Philadelphia: Lippincott Williams and Wilkins; 2009. p. 1187-95.
12. Harlafts NN, Aletras HA, Symbas PN. Hydatid disease of the lung. In: Shields TW, LoCicero J, Ponn RB, editors. General Thoracic Surgery. 6th ed. Philadelphia: Lippincott Williams and Wilkins; 2005. p. 1298-308.
13. Kocer B, Gulbahar G, Han S, Ilhan MN, Dural K, Sakinci U. Analysis of pulmonary hydatidosis according to their segmentary locations. Clin Pal Med 2008;15:8-12.
14. Bilgin M, Oguzkaya F, Akaçi Y. Is capitonnage unnecessary in the surgery of intact pulmonary hydatid cyst? ANZ J Surg 2004;74:40-2.
15. Ghaffarifar F. Echinococcusgranulosus and evaluation of patients with hydatid cyst. Peyvand Mehri, Tehran: 2009.
16. Sayir F, Cobanoğlu U, Şehitoğulları A, Bilici S. Our eight-year surgical experience in patients with pulmonary cyst hydatid. Int J ClinExp Med 2012;5:64-71.
17. Şehitoğulları A. Our results in surgical treatment of hydatid cyst of the lungs. Eur J Gen Med 2007;4:5-8.
18. Usluer O, Ceylan KC, Kaya S, Sevinc S, Gursoy S. Surgical management of pulmonary hydatid cysts: Is size an important prognostic indicator? Tex Heart Inst J 2010;37:429-34.
19. Isitmangil T, Sebita S, Tunca H, Gorura R, Erdika O, Pocanb S, et al. Clinical Experience of Surgical Therapy in 207 Patients with Thoracic Hydatidosis Over a 12-year Period. Swiss Med Weekly 2002;132:548-52.
20. Hasdiraz L, Oğuzkaya F, Bilgin M. Is lobectomy necessary in the treatment of pulmonary hydatid cysts? ANZ J Surg 2006;76:488-90.

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