

## Mejora de la calidad de vídeo sobre EDCA en redes locales inalámbricas 802.11e

### The Clinical Finding, Diagnosis and Outcome of Patients with Complicated Lung Hydatid Cysts

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#### RESUMEN

La rotura de un quiste hidatídico puede causar algunos problemas específicos en niños y adultos . La presentación clínica y el diagnóstico preoperatorio y las complicaciones postoperatorias dependen de si el quiste está intacto o roto. El objetivo de este estudio fue examinar los problemas de los quistes hidatídicos pulmonares rotos. Entre 1996 y 2009 , 58 pacientes con un total de 74 ruptura quistes hidatídicos pulmonares fueron operados . Los registros médicos de 58 pacientes de hidatidosis pulmonar se analizaron retrospectivamente las características . Se recogieron datos relativos a síntomas, el diagnóstico preoperatorio y las complicaciones , los procedimientos quirúrgicos , la morbilidad postoperatoria, el tiempo de hospitalización , y la recurrencia del quiste. 42 pacientes ( 72,41 %) eran hombres. la edad media de los pacientes fue de 32 años (rango , de 4 a 69 años ) . Los síntomas clínicos fueron los siguientes : Tos productiva en 47 pacientes ( 81 %), disnea en 42 pacientes ( 72,41 %), dolor en el pecho en 31 pacientes ( 53,44 %) y hemoptisis en 13 pacientes ( 22,41 %). La evaluación de los hallazgos clínicos, radiografías de tórax, tomografía computarizada , se llegó al diagnóstico preoperatorio de la hidatidosis pulmonar en 52 pacientes ( 89,65 %). Lóbulo inferior derecho fue el lugar más común de quistes en los pulmones ( 63 %). En 42 casos ( 72,41 %) se produjo la rotura de un quiste en el árbol traqueobronquial y en 10 pacientes ( 17,24 %) se produjo la rotura de un quiste en el espacio pleural. neumonía recurrente ocurrió en 6 pacientes ( 10,34 %). Se produjo complicación postoperatoria en 20 pacientes ( 34,48 %). Reoperación realiza para FB en 2 pacientes y fístula bilo pleural en 1 paciente. La recurrencia se produjo en 1 paciente que se trata con albendazol . No se produjo mortalidad hospitalaria . La cirugía es la forma primaria de tratamiento para los pacientes con hidatidosis pulmonar. Los casos complicados tienen tasas más altas de complicaciones preoperatorias y postoperatorias y requieren un mayor tiempo de hospitalización y más procedimientos quirúrgicos complejos . El tratamiento inmediato debe realizarse en todo paciente que ha sido diagnosticado con quistes hidatídicos pulmonares.



### ABSTRACT

Rupture of a hydatid cyst may cause some unique problems in children and adult. The clinical presentation and the preoperative diagnosis and postoperative complications depend on whether the cyst is intact or ruptured. The aim of this study was to review the problems of the ruptured pulmonary hydatid cysts. Between 1996 and 2009, 58 patients with a total of 74 ruptured lung hydatid cysts were operated. The medical records for 58 patients of pulmonary hydatidosis were retrospectively investigated. Data related to symptoms, preoperative diagnosis and complications, surgical procedures, postoperative morbidity, hospitalization time, and cyst recurrence were collected. 42 patients (72.41%) were male. the median age of patients was 32 years (range, 4 to 69 years). Clinical symptoms were as follow: Productive cough in 47 patients (81%), Dyspnea in 42 patients (72.41%), chest pain in 31 patients (53.44%) and hemoptysis in 13 patients (22.41%). Assessment of clinical findings, chest roentgenograms, thoracic computed tomography, led to the correct preoperative diagnosis of pulmonary hydatid disease in 52 patients (89.65%). Right lower lobe was the most common location of cyst in the lungs (63%). In 42 cases (72.41%) rupture of cyst occurred into tracheobronchial tree and in 10 patients (17.24%) rupture of cyst occurred into pleural space. recurrent pneumonia occurred in 6 patients (10.34%). Postoperative complication was occurred in 20 patients (34.48%). Reoperation performed for bronchopleural fistula in 2 patients and bilo-pleural fistula in 1 patient. Recurrence occurred in 1 patient which treated with albendazol. Hospital mortality was not occurred. Surgery is the primary mode of treatment for patients with pulmonary hydatid disease. Complicated cases have higher rates of preoperative and postoperative complications and require longer hospitalization time and more extensive surgical procedures. Immediate treatment should be performed in any patient who is diagnosed with pulmonary hydatid cysts.

*Keywords: Echinococcosis, hydatid disease, complicated cysts, intact cysts.*

### Introduction

Human hydatid cyst is still a serious health problem for some countries and remains endemic for many parts of the world, most notably the Mediterranean region, Australia, New Zealand, the Middle East, and South America (1 and 2). In areas where hydatid disease is endemic, it is still a major public health problem. Most individuals who contract this parasite are young, and majority of patients are less than 40 years of age (3,4). Echinococcal cysts always carry the risk of the rupture with anaphylactic potential, and risk of spillage to healthy viscera. The rupture may occur within the boundaries of the pericystic layer, into the pleural space, or into a neighboring organ, bronchus, or blood vessel. Many factors may be responsible for the rupture, such as trauma or coughing increasing intrathoracic pressure or expansion of the cyst size. No correlation had been found between cyst size and intracystic pressure (5). Pulmonary hydatid cysts usually remain asymptomatic until the time of rupture, and the clinical presentation in these patients is directly related to intact or ruptured cyst status. Rupture may occur during antihelminthic therapy or percutaneous aspiration and trauma can lead to severe complications, such as massive hemoptysis and tension pneumothorax, lung abscess and asphyxia (6). Pulmonary hydatidosis primarily affects children and young adults who may be infected again, and usually they have great lung capacity for expansion. Thus it is important to always use the most conservative surgical methods possible. With complicated cysts, the clinical picture is variable and depends on the nature of the perforation. Often the cyst ruptures into a bronchus. In most cases, solid remnants of the collapsed parasitic membrane are left in the cavity as a source of recurrent infection (7, 8). Surgery remains the treatment of choice for hydatid cysts of the lung. More radical surgical procedures may be needed in complicated cases (6).

In this study we retrospectively assessed a series of patients with pulmonary hydatid cyst that were treated at our center, and we review the clinical presentation, diagnosis, surgical treatment, and postoperative outcome in patients with ruptured cysts. The aim was to review the problems encountered in treating ruptured pulmonary hydatid cysts and delayed surgical therapy in pulmonary hydatid disease.

### Material and Methods

The medical records of 58 patients with complicated pulmonary hydatid cysts who had underwent operation in teaching and private hospital in Rasht, Iran between January 1996 and December 2009, were reviewed. The series included Thirty eight cases were male and 16 females with a mean age 34 years (range, 4 to 69 years). The follow-up times for all 58 patients ranged from 10 months to 3 years. The pulmonary cysts were diagnosed by various combinations of chest roentgenogram, thoracic computed tomography and intraoperative findings. Each patient also underwent abdominal ultrasonography to assess for concomitant hepatic hydatid cysts. Serologic testing was not used routinely for diagnostic workup. Any cyst that had ruptured into the bronchus or pleural cavity, with or without infection, was defined as complicated. A ruptured cyst was considered to be infected when the patient exhibited accompanying problems of purulent sputum, leukocytosis, fever, and with or without lobar and segmental pneumonia.

All of the 58 operations performed with posterolateral thoracotomies. A total of 74 surgeries were performed on the 58 patients. The individuals with right pulmonary and liver-dome cysts underwent right posterolateral thoracotomy with phrenotomy. Twenty-eight individuals required two surgeries; the second operation was performed at least 28 days after the first operation. For the period between the first and second operation, each patient was informed about possible complications and warned that a second surgery may be needed sooner than planned if the cyst ruptured. The individuals with liver-dome cysts underwent thoracophrenotomy. Eight of the 58 patients presented with pneumothorax and empyema. These patients required closed-chest tube drainage initially, and further surgical procedures were performed after their general condition had stabilized. In 46 patients (79.31%) the specific surgical treatment for pulmonary hydatid cysts was cystotomy with evacuation of laminated membrane and closure of Bronchial openings without capitonage. For this procedure the edges of the surgical wound and the lung surface were protected with saline 3% impregnated gauze. Due to risks of leakage into the bronchial system and postoperative tracheobronchial irritation or pulmonary edema, no scolicedal agent was injected into the cyst. The cyst contents were evacuated by needle aspiration, and the cystic membrane was removed with ring forceps. The remaining cavity was irrigated with 3% saline solution. Bronchial openings in the cavity were stitched up with absorbable sutures in a figure of 8. After the bronchial openings were closed, the residual cavity was left open. In 8 patients with complicated cysts that exhibited a thickened or liquified pericyst layer and had damaged the adjacent parenchyma, a pericystectomy was performed. Decortication was performed in 4 patients (5.9%). wedge resection was performed in 6 patients (10.34%) and lobectomy in 2 patients (3.44%). none of the patients required pneumonectomy. Before surgery, all patients were placed on albendazole (800 mg/d in adults, 10 mg/kg/d in children) for at least 8 day. After surgery, all patients were placed on albendazole antihelminthic therapy (800 mg/d in adults, 10 mg/kg/d in children) for at least 2 months. Data related to symptoms, preoperative complications, surgical procedures, postoperative morbidity, hospitalization time, and cyst recurrence were collected from each patient's records.

### Research Finding

of 58 patients who were studied , 40 patients (68.96% ) were male and 18 patients (31.034%) were female . the Average age of patients were 4-63 years, Therefore most of the patients were at the age groups of 20-39 years. Chest X Ray in 100% of patients and thoracic CT scan in 63% of patients was used For diagnosis of disease. Assessment of clinical findings, chest roentgenograms, thoracic computed tomography, led to the correct preoperative diagnosis of pulmonary hydatid disease in 52 patients (89.65%). The other 6 patients were diagnosed intraoperatively. In 6 of these patients, the respective misdiagnoses before surgery were pneumonia and two lung abscess. Most common location of involvement of the cyst in the lungs was right lower lobe in 34 cases (58.62%) and left lower lobe in 12 cases (20.68%) , left upper lobe in 7 case (12.06%) , right upper lobe in 4 cases (6.89%) and right middle lobe in one case (1.72%). Fourteen patients (68.96%) had solitary pulmonary cysts and 18 patients (31.03%) had multiple lesions. Twenty patients (34.48%) had bilateral pulmonary hydatidosis, 5 patients (8.62%) had unilateral multiple pulmonary hydatidosis, and 13 patients (22.41%) had unilateral lung lesions and concomitant liver hydatidosis. Of the 18 patients with bilateral pulmonary hydatid cysts, 6 patients also had cysts in the liver .The most common symptom was chest pain, followed by cough and dyspnea, hemoptysis, sputum, and fever, respectively ([Table 1](#)) The most common preoperative complications were the rupture into tracheobronchial tree and parenchymal complications in 11 patients (18/96%) ([Table 2](#))

Surgical approaches in 38 patients were posterolateral thoracotomy and in 20 patients was anterolateral thoracotomy. In 18 patients with bilateral cyst, one stage operation was performed with anterolateral thoracotomy. In two patients, contralateral side operated on month later. Surgical procedures in 50 patients were evacuation of cyst with cystostomy and Closure of the Bronchial Opening. Capitonage was performed in 16 patients. Wedge resection in 6 patients and Lobectomy in 2 patients was performed. In 20 patients (34.48%) postoperative complications were developed. The most common postoperative complications was prolonged air leak ([Table 3](#)). There was no perioperative or postoperative mortality. In two patients, prolonged air leak controlled with segmentectomy and in three patients with autologous blood pleurodesis. Atelectasis treated with chest physiotherapy and bronchoscopy. Empyema and pleural effusion treated with thoracostomy tube. Hemoptysis stopped spontaneously. The hospitalization times ranged from 7 to 22 days (mean, 11 days). During follow-up recurrence develop in two patients and controlled with antihelminthic therapy.

### Discussion

The hydatid cysts may remain asymptomatic for a long time. As they enlarge, the cysts may rupture and patients complain of cough, expectoration of membranes, hemoptysis, and thoracic pain in cases of pulmonary cysts. The clinical picture with complicated cysts is variable and depends on the nature of the rupture. But in most uncomplicated cases of pulmonary hydatid cysts; lung cysts are either an incidental finding or the patient presents with dry cough, dyspnea, and chest pain. Ruptures into a bronchus are the common complication of the cyst. In most of above cases, remnants of the collapsed parasitic membrane are left in the cavity as a source of recurrent infection (7, 8). Such patients present with expectoration of hydatid fluid and remnants of parasitic membrane, recurrent hemoptysis, purulent sputum, or fever, or a combination of some or all of these symptoms. Expectoration of cystic contents can lead to severe complications, such as asphyxia, acute respiratory failure, massive hemoptysis, and anaphylactic shock (4, 9). Complicated hydatid

cysts, especially those that ruptured into the pleura, are difficult to diagnose radiologically. The rupture into pleura usually misdiagnose as empyema and effusion. In this condition, after chest tube insertion, hydatid material usually observed within the tube. (14) Although the computed tomographic diagnosis of ruptured pulmonary hydatid cyst was the most difficult because of infection, in the planning of the operation it was successful in detecting the localization, contents, and borders of the lesion (10). In contrast to rupture into a bronchus, rupture of a hydatid cyst into the pleural cavity usually causes pneumothorax, pleural effusion, or empyema. One of our patients developed pleural effusion in this way .Cyst rupture into the pleural cavity can also result in tension pneumothorax (11). this complication occurred in one of our patients .The documented rates of simple pneumothorax in patients with pulmonary hydatidosis ranged from 2.4% to 6.2% (4, 12, and 13). Empyema is reported to occur in 7.6% of patients with hydatid disease of the lung (12). In our series, pneumothorax occurred preoperatively in 4 of patients and empyema occurred preoperatively in 6 patients. Some authors contend that the treatment of hydatid disease regimens with oral mebendazole or albendazole are effective against cysts (14,15). Research has shown that 73% to 75% of patients respond to medical management to some degree; however, the reported cure rates are only 25% to 30% (14,16). Reports in the literature of treatment with mebendazole or albendazole have not been documented in the complicated hydatid cysts (10,17). Medical therapy is not effective in complicated pulmonary hydatid cysts because solid remnants of the collapsed parasitic membrane are left in the cavity are a source of recurrent infection and must be remove from the cavity (3,7,8). Operation must be performed early because of the high complication rate in patients who were operated later on. The choice of surgical technique depends on the conditions encountered during the operation. When parenchymal destruction is present, resection must be performed. Only the destroyed parenchyma should be removed. Under other conditions, cystotomy and evacuation and closure of bronchial opening are our surgical methods. In cases of trapped lung caused by infection (empyema), decortication of the pleural peel must be performed. The type of operation was not correlated with morbidity or mortality. Single-lung ventilation is preferred to prevent aspiration of cystic material and respiratory complications, and, in turn, morbidity and mortality. Lung-conserving procedures are optimal for pulmonary hydatidosis. We believe that the most appropriate procedures for pulmonary hydatidosis are open surgery involving removal of the cyst membrane, closure of the bronchial openings.Video-assisted thoracic surgery(VATS) is suggested for selected patients (18). It is possible to remove the cystic membrane thoracoscopically, but uncontrolled spillage of cyst contents may cause anaphylaxis, pleural hydatidosis (if the cyst is intact), or pleural bacterial spread (if the cyst is infected).We did not use VATS in our patients. However, complicated hydatid cysts tend to cause significant pleural thickening and parenchymal destruction; therefore, more radical surgical procedures, such as decortication, segmentectomy, and lobectomy may be required in these patients (4,17,19,20). Reports in the literature note resection rates of 19% to 32% for complicated pulmonary hydatid cysts and 0% to 7% for uncomplicated pulmonary hydatid cysts,(4,19,20) . In a study of 43 patients with pulmonary hydatid cysts and associated pleural complications, Aribas and colleagues (12) Found that decortication was needed in 30 patients (69.8%) and pulmonary resection was needed in 6 patients (14%). In our series, 6 of the patients with complicated cysts required decortication. Complicated pulmonary hydatid cysts are associated with higher postoperative morbidity and mortality than uncomplicated cysts (12, 17, and 20). In complicated cases, infection and inflammation of the adjacent lung parenchyma may affect wound healing and lead to postoperative complications such as prolonged air leakage, empyema, and pneumonia. In addition, many patients with complicated pulmonary hydatid cysts require preoperative antibiotic therapy and supportive treatment. These are the main reasons why complicated cases

tend to have a higher morbidity and mortality rate and require longer hospitalization than uncomplicated cases. There was no mortality in either of our patient groups, and we attribute this to successful management of infectious complications. However, the patients with complicated cysts had higher morbidity and a longer hospital stay. Safioleas and colleagues (13) reported the same hospitalization trend in 42 patients with pulmonary hydatidosis (a 12-days median hospital stay for uncomplicated cases versus a 21-days median hospital stay for complicated cases).

### Conclusion

surgery is the safest curative treatment for pulmonary hydatid cysts. Although surgical treatment is effective in patients with complicated as well as uncomplicated cysts for treatment of pulmonary hydatidosis, surgical intervention before rupture of the cysts is essential. Ruptured cysts are associated with increased morbidity, more extensive surgery, and longer hospital stays. Regardless of whether symptoms are present, all pulmonary hydatid cysts should be surgically treated as soon as they are diagnosed in order to avoid complications.

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Table 1. Clinical Manifestations of Pulmonary Hydatid Disease in 58 Patients

| Clinical Manifestations         | no | %     |
|---------------------------------|----|-------|
| Chest pain                      | 16 | 27.58 |
| dyspnea                         | 14 | 24.13 |
| cough                           | 15 | 25.86 |
| hemoptysis                      | 11 | 18.96 |
| Expectoration of cystic content | 5  | 8.62  |
| Sputum production               | 11 | 18.96 |
| Fever                           | 12 | 20.68 |
| Weight loss                     | 4  | 6.89  |
| bilioptysis                     | 1  | 1.72  |

Table 2. preoperative complications

| Preoperative Complications           | NO | %     |
|--------------------------------------|----|-------|
| recurrent Pneumonia                  | 6  | 10.34 |
| Simple hydropneumothorax             | 2  | 3.44  |
| Tension hydropneumothorax            | 1  | 1.72  |
| Empyema                              | 4  | 6.89  |
| Bronchobiliary fistula and pneumonia | 1  | 1.72  |
| Pleural effusion                     | 1  | 1.72  |
| Infected cyst                        | 3  | 5.17  |
| Infected cyst and pneumonia          | 1  | 1.72  |
| Allergic episode                     | 1  | 1.72  |

Table 3. Postoperative Complications

| Postoperative Complications | NO | %    |
|-----------------------------|----|------|
| Prolonged air leak (>7)     | 5  | 8.62 |
| Empyema                     | 3  | 5.17 |
| Pneumonia                   | 3  | 5.17 |
| Atelectasis                 | 3  | 5.17 |
| Hemoptysis                  | 1  | 1.72 |
| Pleural effusion            | 2  | 3.44 |
| Wound infection             | 3  | 5.17 |