



# Pulmonary hydatid cyst mimicking pneumonia, lung mass, pneumothorax, pleural effusion, pulmonary abscess, and pyopneumothorax

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## Manuscript details:

Received: 04.09.2022

Accepted: 05.11.2022

Published: 31.12.2022

## Cite this article as:

Ehsan Hajipour Jafroudi, Manouchehr Aghajanzadeh, Ali AlaviFoumani, Azita Tangestaninejad, Yousha Pourahmadi, Mahsa Mousazadeh MD (2022) Pulmonary hydatid cyst mimicking pneumonia, lung mass, pneumothorax, pleural effusion, pulmonary abscess, and pyopneumothorax., *Int. J. of Life Sciences*, 10 (4): 308-316.

Available online on <http://www.ijlsci.in>

ISSN: 2320-964X (Online)

ISSN: 2320-7817 (Print)



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## ABSTRACT

The lung is the second most common site of involvement with echinococcosis granulosis in adults after the liver (10-30% of cases), and the most common site in children. Rupture of a pulmonary hydatid cyst (PHC) may cause some unique presentations in these patients. The clinical presentation and complications of PHC depend on whether the cyst is intact or complicated. Presentations of the complicated PHC group are unusual such as: mass, pneumonia, pleural effusion, pulmonary abscess, and pneumothorax. The aim of this study was to review the problems of the complicated PHC ones. Between 2002 and 2021, 59 patients with a total of 82 ruptured PHC were gone under the operation. The medical records for 59 patients of pulmonary hydatidosis were retrospectively investigated. Data related to symptoms, preoperative diagnosis, surgical procedures, postoperative morbidity, hospital stay were gathered. Among these patients, 36 patients (61%) were male. The median age of the patients was 36 years (range, 12 to 60 years). The clinical manifestations were as follow: Productive cough in 15 patients (25.42%), Dyspnea in 14 patients (23.72%), chest pain in 22 patients (37.28%), fever in 12 patients (20.33%), and hemoptysis in 11 patients (18.64%). Preoperative tools for diagnosis were: chest X-ray, Thoracic Computed Tomography (CT-scan), and Magnetic Resonance Imaging (MRI) which led to the correct preoperative diagnosis of complicated pulmonary hydatid disease in 22 patients (37.28%). Right lower lobe was the most common location of these cyst in the lungs in 37 patients (63%). In 39 cases (66.10%) rupture of cyst occurred into tracheobronchial tree, while in 20 patients (33.89%) rupture of cyst occurred into pleural space. Furthermore, recurrent pneumonia occurred in 11 patients (18.64%). Also lung abscess were observed in 11 patients and lung mass in 17 patients. Postoperative complications were occurred in 18 patients (30.50%). Reoperation performed for bronchopleural fistula in 5 patients. Median hospital stay for the patients was 10 days. Surgery is the primary mode of treatment for patients with complicated cases of pulmonary hydatid cyst. Immediate treatment should be performed in any patient who is diagnosed with pulmonary hydatid cyst because any delay in the treatment will increase morbidity, mortality, and hospital stay.

**Keywords:** *Echinococcosis granulosis, hydatid cyst, pulmonary hydatid cyst*

## INTRODUCTION

Hydatid disease is a serious health problem in some areas in the world such as: Mediterranean region, the Middle East, New Zealand, and South Africa. In some countries like Iran and South Africa, it is endemic (Morar *et al.*, 2003). Although it can be found in any part of the body, it is more common in the liver (65% to 75%) and lung (15%). Concomitant pulmonary and liver hydatid cysts occur in 4% to 25% of the patients with hydatid cyst (Morar *et al.*, 2003). It is imperative to investigate the possibility of new cysts in other parts of the body in patients diagnosed with either a pulmonary or hepatic cyst (Morar *et al.*, 2003).

Intrapleural rupture occurs in 5% of cases and may present with an acute clinical picture as an intense chest pain, persistent cough, severe dyspnea, cyanosis, shock, and suffocation (Yekeler *et al.*, 2012). The diagnosis of complicated pulmonary hydatid disease is difficult because it can mimic many other pulmonary and pleural diseases, including pneumonia, bronchiectasis, tuberculosis, abscess, lung tumors, pleurisy, and empyema (Köksal *et al.*, 2004; Lal *et al.*, 2013; Ekinci *et al.*, 2015). Liver hydatid cysts can also rupture into the peritoneum causing peritonitis, or trans-diaphragmatically into the pleural space or bronchial tree which can cause pulmonary hydatidosis or a bronchial fistula (Köksal *et al.*, 2004; Lal *et al.*, 2013; Cobanoğlu *et al.*, 2011; Işık *et al.*, 2018). The most frequent complication of pulmonary hydatid disease is the rupture of the cyst into tracheo-bronchial tree (Puri *et al.*, 2011; Ekinci *et al.*, 2015; Işık *et al.*, 2018; Asgary *et al.*, 2016). If a cyst ruptures into the pleura or pulmonary parenchyma, it can be life-threatening as tension pneumothorax or pleural effusion (Puri *et al.*, 2011; Kürkçüoğlu *et al.*, 2002). The diagnosis can be made based on the clinical presentation, Chest X-ray, and CT-scan of the chest (Ekinci *et al.*, 2015; Aghajanzadeh *et al.*, 2018). Although bronchoscopy is unnecessary in patients with a typical clinical and radiological picture, it can be performed either there is suspicion of a tumor or an atypical presentation (Alavi *et al.*, 2010; Cakir *et al.*, 2010). Serological tests are less reliable in such conditions and they are not used due to the high possibility of false positive and false negative responses. Surgery is highly recommended for these cases. The aim of surgical intervention in pulmonary hydatid cysts is mainly the removal of the laminated membrane without causing more intraoperative

contamination and prevention of an intrapulmonary residual cystic cavity (Yilmaz *et al.*, 2009; Aarons *et al.*, 1999; Kuzucu *et al.*, 2004). Here, we evaluated patients with pulmonary hydatid cysts which presented with rare and unusual presentation of pneumonia, lung mass, lung abscess and tension hydro/pneumothorax during nineteen years follow up.

## MATERIAL AND METHOD

The medical records of 59 patients with unusual presentation of pulmonary hydatid cyst who had undergone operation in both teaching and private hospital in Rasht, Iran between January 2002 and December 2021, were reviewed. The series included 36 male cases and 23 females' cases. The median age of patients was 36 years (12 to 60 years). The pulmonary cysts were diagnosed by chest roentgenogram, thoracic computed tomography, and intraoperative findings. All patients also underwent abdominal ultrasonography to assess for concomitant hepatic hydatid cysts. 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 patients underwent posterolateral thoracotomies with one lung intubation. Patients with right pulmonary and liver-dome cysts underwent right posterolateral thoracotomy with phrenotomy. Three patients presented with simple pneumothorax (Fig. 1,2), six patients with simple pleural effusion (Fig. 3,4,5,6), and another six patients with pyopneumothorax (empyema) (Fig. 7,8,9).

These patients required chest tube drainage initially, and further surgical procedures were performed after their general condition had stabilized. Lung abscess present in eleven patients (Fig. 10,11,12), and pulmonary mass present in seventeen patients (Fig. 13,14,15).

Twenty eight of patients underwent fiberoptic bronchoscopy for lung mass, recurrent pneumonia, and lung collapse (Fig. 16,17,18). In one patient with recurrent pneumonia fiberoptic bronchoscopy was curative treatment (Fig. 19, 20, 21).

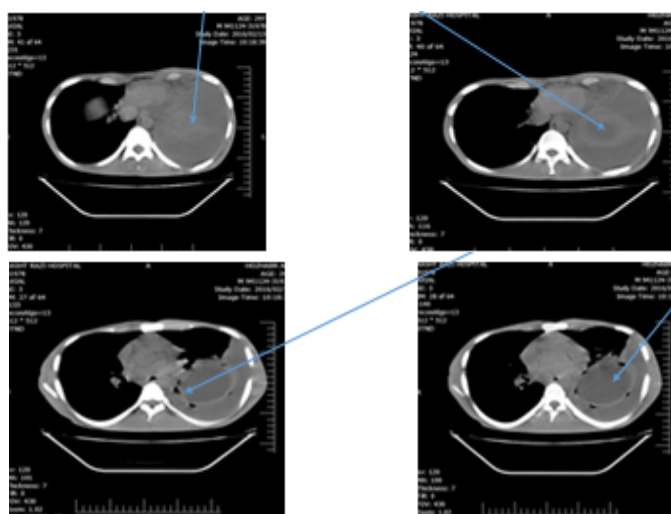
Diagnostic thoracotomy performed in 19 patients because of lung mass, recurrent pneumonia, and non-

expansion of lung and severe air leakage after chest – tube insertion. Bronchial openings in the cavity were stitched up with absorbable sutures in a figure of eight. After the bronchial openings were closed, the residual cavity was left open. Decortication was performed in 18 patients (30.50%), wedge resection was performed in 22 patients (37.28%), segmental resection in 8 patients (13.55%), and lobectomy in 4 patients

(6.77%). After surgery, all patients were treated with antihelminthic therapy such as albendazole (800 mg/d in adults, 10 mg/kg/d in children) in three course of 28 day with 14 day interval. Data related to symptoms, preoperative diagnosis, complications, Intra-operative diagnosis, surgical procedures, postoperative morbidity and mortality, and hospitalization stay, were collected from each patient's records.



**Fig. 1,2: CXR show simple pneumothorax**



**Fig. 3,4,5,6: show simple pleural effusion**



**Fig. 7,8,9: CXR show hydro- pneumothorax**

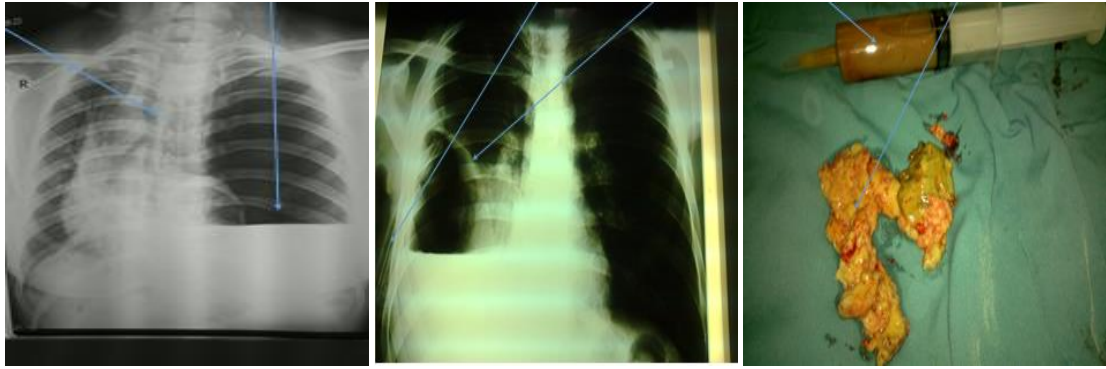


Fig. 10,11,12 CXR show empyema with content of empyema (infected laminated layer of cyst)



Fig. 13,14,15: show CT-scan of chest with lung abscess and its contents due to hydatid cyst

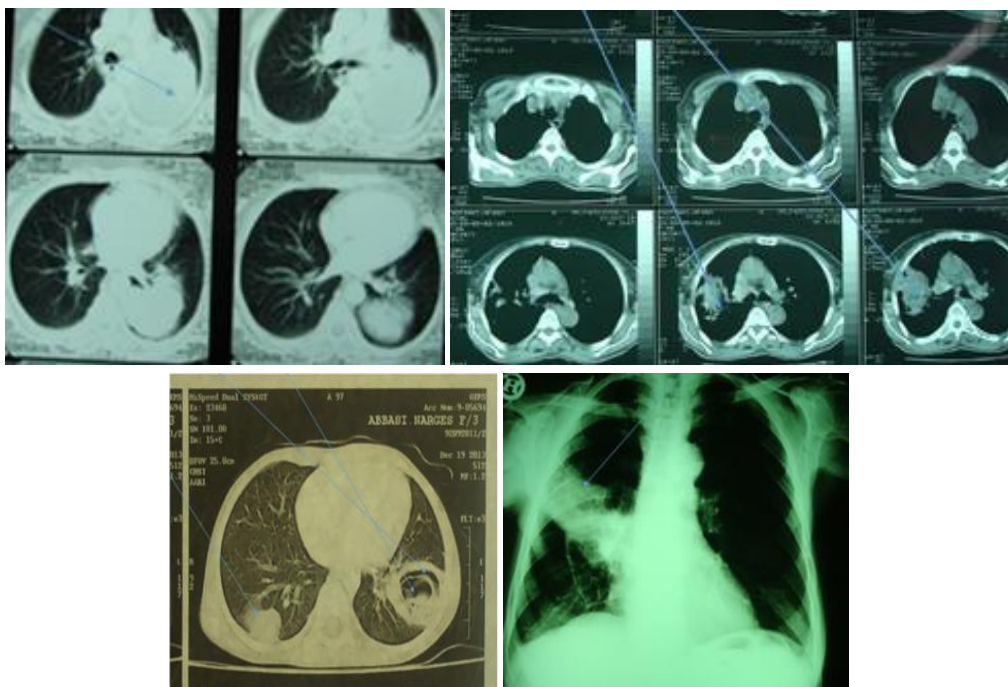
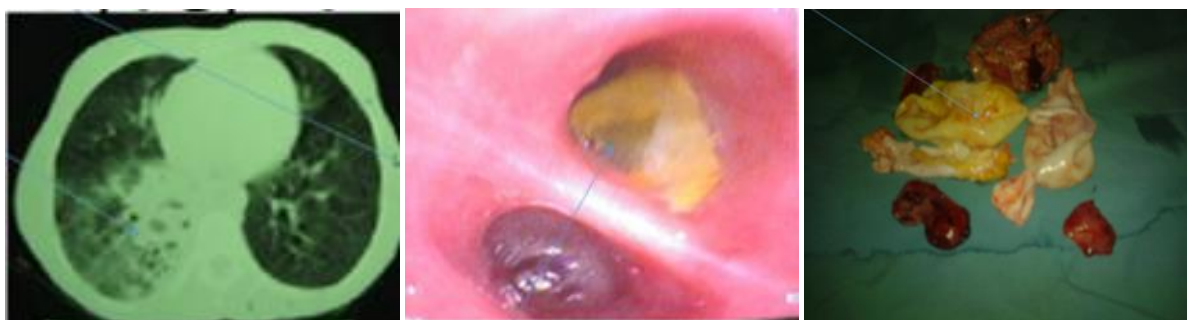


Fig. 16,17,18: CT -scan and CXR of chest show lung collapse and mass due to hydatid cyst



**Fig. 21,22,23: Show recurrent pneumonia in 35-year-old, Bronchoscopy performed, membrane was removed**

**RESULT**

Thirty six of patients (61%) were male and 23 patients (39%) were female. The median age of the patients was 36 years (12 to 60 years). Chest X Ray and chest CT scan were used in 100% of patients as diagnostic tool. Assessment of clinical findings, chest roentgenograms, and thoracic computed tomography, led to the correct preoperative diagnosis of pulmonary hydatid disease in forty patients (67.79%). The other nineteen patients (32.21%) were diagnosed intraoperatively. In these patients, the respective

misdiagnoses before the surgery were pneumonia, lung mass, pneumothorax, pleural effusion, empyema, and lung abscess. The most common location of the cyst in the lungs was right lower lobe in thirty-six cases (61%) and left lower lobe in eighteen cases (30.50%). The other were left upper lobe, right upper lobe and right middle lobe. The most common symptom was chest pain, followed by cough and dyspnea, hemoptysis, malodor sputum, and fever (Table 1). The most common preoperative complications were the rupture into tracheobronchial parenchymal tree and the pleural space (Table 2).

**Table 1. Clinical Manifestations of Pulmonary Hydatid Disease in 59 Patients**

Clinical Manifestations	No	%
Chest pain	22	37.28 %
Dyspnea	14	23.72 %
Cough	15	25.42 %
Hemoptysis	11	18.64 %
Expectoration of cystic content	5	8.47 %
Sputum production	11	18.64 %
Fever	12	20.33 %
Weight loss	4	6.77 %

\*\*\*One patient may present more than one Clinical Manifestations

**Table 2. Preoperative complications**

Complications	NO	%
Lung mass	17	28%
Recurrent Pneumonia	11	18%
Simple pneumothorax	3	5%
Tension hydropneumothorax	5	8%
Empyema (pyopneumothorax)	6	10%
Simple Pleural effusion	6	10%
Lung abscess (Infected cyst)	11	18%
Allergic episode	3	5%

\*\*\*One patient may present more than one Preoperative complications.

**Table 3. Postoperative Complications Postoperative**

Complications	NO	%
Empyema	8	36.36%
Pneumonia	4	18.18%
Atelectasis	7	31.81%
Hemoptysis	5	22.72%
Pleural effusion	4	16.16%
Massive and prolong Air leakage	6	27.27%
Wound infection	9	40.90%
Prolonged Moderate air leakage	8	36.36%

\*\*\*One patient may present more than one Postoperative Complications Postoperative.

Surgical approaches in 34 patients was posterolateral thoracotomy, while in eight of these patients unfermented phrenotomy and cyst evacuation had been done because of the simultaneous liver hydatid cyst. Furthermore, surgical approach was anterolateral thoracotomy in 20 patients because of simultaneous bilateral cyst lung. One stage operation with anterolateral thoracotomy was performed in five patients. General anesthesia with one lung ventilation performed in 40 patients. Surgical procedures in 32 patients was evacuation of all elements of cyst with cystostomy and Closure of the Bronchial Opening. Capitonage of the remnant cavity was performed in eight patients. In other patients cavity remained open. Wedge and segmental resection in 12 patients and Lobectomy in 2 patients was performed because of destruction of the lobe. Postoperative complications were developed in 22 of patients. Prolonged air leak was among the most common postoperative complications (Table 3).

There was no intraoperative or postoperative mortality. In 2 patients, prolonged air leak controlled with segmentectomy and in 3 patients with autologous blood pleurodesis. Atelectasis was treated with chest physiotherapy and bronchoscopy. In addition, empyema and pleural effusion were treated with thoracostomy tube. Hemoptysis stopped spontaneously. The hospitalization stay ranged from 7 to 14 days (mean, 8 days). During follow-up no recurrence was detected.

## DISCUSSION

The hydatid cysts of lung and liver may remain asymptomatic for a long time. When cyst become enlarge, they may rupture spontaneously, with trauma

or any diagnostic procedures( Chacko *et al.*, 2009; Yekeler *et al.*, 2012; Aghajanzadeh *et al.*, 2014). Pulmonary cyst may rupture into tracheobronchial system, pulmonary parenchyma, or in the pleural space ( Chacko *et al.*, 2009; Aghajanzadeh *et al.*, 2014). These patients complain of cough, expectoration of membranes, hemoptysis, dyspnea, thoracic pain, and anaphylaxis (Ekinici *et al.*, 2015 ; Aghajanzadeh, *et al.*, 2018; Asgary *et al.*, 2016 ). In addition, this would be an either an incidental finding or presents as a dry cough, dyspnea, and chest pain in most of the uncomplicated cases (Ekinici *et al.*, 2015; Işık *et al.*, 2018 ).

The remnants of parasitic membrane in the collapse cavity could be considered as a source of recurrent infection for the patients( Ekinici, *et al.*, 2015 ; Işık, *et al.*, 2018). Expectoration of hydatid fluid and remnants of parasitic element will present with recurrent hemoptysis, purulent sputum, or fever in case of pulmonary abscess in these patients (Ekinici, *et al.*, 2015; Aghajanzadeh, *et al.*, 2018; Asgary, *et al.*, 2016). Moreover, expectoration of cystic element can lead to some fetal complications, such as asphyxia due to membrane or cyst fluid, acute respiratory failure, massive hemoptysis, and anaphylactic shock (Aghajanzadeh, *et al.*, 2018; Asgary, *et al.*, 2016; Kuzucu, *et al.*, 2004).

When a cyst rupture into the pleura space, the diagnosis become difficult via radiologic tools). These complications can be misdiagnosed as empyema or hydropneumo thorax (Köksal, *et al.*, 2004 ; Chacko, *et al.*, 2009 ; Yekeler, *et al.*, 2012 ; Kürkcüoğlu, *et al.*, 2002). In these complications, hydatid material would be detected following chest tube insertion ( Yekeler, *et al.*, 2012 ; Kürkcüoğlu, *et al.*, 2002), while CT scan may

not help the diagnosis ( Kürkçüoğlu, *et al.*, 2002 ; Ekinçi, *et al.*, 2015 ; Asgary, *et al.*, 2016 ; Ramos, *et al.*, 2001). In addition, FOB can be considered as both diagnostic tool and intraoperative treatment in some cases( Alavi, *et al.*, 2010 ; Cakir, *et al.*, 2010 ; Taha, *et al.*, 2005 ) due to their ability in detecting the cause, content, localization, and borders of the lesion( Asgary, *et al.*, 2016 ; Ramos, *et al.*, 2001).

Generally, complications such as pneumothorax, pleural effusion, and hydropneumothorax may occur following rupturing a cyst into a pleural space (Yekeler, *et al.*, 2012; Kürkçüoğlu, *et al.*, 2002; Aghajanzadeh, *et al.*, 2020). In this regard, 6 of our patients developed pleural effusion, six hydropneumothorax, and 6 empyema. Cyst rupture into the pleural space can also result in tension pneumothorax ( Yekeler, *et al.*, 2012 ; Aghajanzadeh, *et al.*, 2020 ; Cobanoğlu, *et al.*, 2011 ; Kuzucu, *et al.*, 2004 ). This complication occurred in 18 patients (30.50%) of our patients .The rates of simple pneumothorax in patients with pulmonary hydatidosis ranged from 2.4% to 6.2% ( Köksal, *et al.*, 2004 ; Ekinçi, *et al.*, 2015 ; Aghajanzadeh, *et al.*, 2018 ).

Empyema has been reported to occur in 7.6% of patients with hydatid disease of the lung (Asgary, *et al.*, 2016; Yilmaz, *et al.*, 2009; Aarons, *et al.*, 1999). In our series, simple pneumothorax presents preoperatively in six of our patients and empyema present preoperatively in six patients and lung abscess (Infected cyst) in eleven patients.

Complicated pulmonary hydatid cysts can mimic several pleural and pulmonary disease such as non-resolving pneumonia due to the remnant membrane of the cyst after the parenchymal or bronchial rupture. This membrane would be considered as a foreign body and recurrent pneumonia would present subsequently (Kürkçüoğlu, *et al.*, 2002; Ekinçi, *et al.*, 2015 ; Cobanoğlu, *et al.*, 2011 ; Alavi, *et al.*, 2010 ; Aghajanzadeh, *et al.*, 2014). In our study 11 patients present the recurrent pneumonia. In some of them rupture cysts present as a lung abscess with fever malodor sputum with air fluid level in their X-ray (Asgary, *et al.*, 2016 ; Yilmaz, *et al.*, 2009 ; Aghajanzadeh *et al.*, 2014 ). Generally, in our study 11 patients present with lung abscess. In addition, mass like lesions are another rare presentation of PHC this complication, which may occur after incomplete evacuation of content of the cavity (Ekinçi, *et al.*, 2015;

Işık, *et al.*, 2018; Asgary, *et al.*, 2016 ). Seventeen cases in our study present with lung mass (Fig 13,14,15,16). According to the previous studies, it is possible that their manifestation would be similar to tuberculosis or bronchiectasis (Ekinçi, *et al.*, 2015; Asgary, *et al.*, 2016 ; Aghajanzadeh, *et al.*, 2014 ); however, in our study we didn't detect such complications.

There is no specific clinical finding in complicated PHC cases (Aghajanzadeh, *et al.*, 2014; Chacko, *et al.*, 2009 ; Asgary, *et al.*, 2016 ). Laboratory findings or serologic tests are not diagnostic too( Aghajanzadeh, *et al.*, 2014 ; Asgary, *et al.*, 2016 ; Yilmaz, *et al.*, 2009 ). Bronchoscopy is not indicated in patients with a typical clinical and radiological picture. In such case is this could be dangerous because of the risk of cyst rupture (Alavi, *et al.*, 2010; Cakir, *et al.*, 2010; Taha, *et al.*, 2005). It can be performed when there is a suspicion of tumor or the possibility of a recurrent pneumonia, mass, lung abscess, and lung collapse( Yilmaz, *et al.*, 2009 ; Cakir, *et al.*, 2010 ; Taha, *et al.*, 2005 ).

In uncomplicated cases, Some researchers recommended medical treatment of hydatid cysts with oral mebendazole or albendazole (Asgary, *et al.*, 2016; Gil-Grande, *et al.*, 1993). Some reports showed that 73% to 75% of patients respond to medical management to some degree; but cure rates are only 25% to 30%( Alavi, *et al.*, 2010; Yilmaz, *et al.*, 2009; Cakir, *et al.*, 2010; Kuzucu, *et al.*, 2004; Gil-Grande, *et al.*, 1993). In the complicated hydatid cysts the treatment with mebendazole or albendazole have not been recommended( Asgary, *et al.*, 2016; Gil-Grande, *et al.*, 1993; Gulsun, *et al.*, 2010). Medical therapy is not effective in complicated cases PHC because the parasitic membrane which left in the cavity are a source of recurrent infection and must be removed from the cavity ( Aghajanzadeh, *et al.*, 2014; Lal, *et al.*, 2013; Cobanoğlu, *et al.*, 2011; Asgary, *et al.*, 2016 ; Gulsun, *et al.*, 2010 ). Operation must be performed early after diagnosis because delay in the operation may develop high complication rate( Aghajanzadeh, *et al.*, 2014; Asgary, *et al.*, 2016; Ramos, *et al.*, 2001; Gulsun, *et al.*, 2010; Mahmood, *et al.*, 2011 ).

Choosing the appropriate surgical procedures depends on the findings during the operation( Aghajanzadeh, *et al.*, 2014; Cobanoğlu, *et al.*, 2011; Asgary, *et al.*, 2016). All patients underwent posterolateral and anterolateral thoracotomy for surgical treatment of

PHC. When parenchymal destruction is present, resection of involvement portion as wedge, segmental resection, and lobectomy is indicated (Aghajanzadeh, *et al.*, 2014; Cobanoglu, *et al.*, 2011; Asgary, *et al.*, 2016). Because of benignity of this disease, only the destroyed parenchyma should be removed (Aghajanzadeh, *et al.*, 2014; Cobanoglu, *et al.*, 2011; Asgary, *et al.*, 2016; Yilmaz, *et al.*, 2009). In other patients, our choice of surgical methods are cystotomy and evacuation of all content of cavity and closure of bronchial opening in the cavity with resection of fibrotic tissues or pricystectomy subsequently. In patients with trapped lung caused by infection, decortication of the pleural peel must be performed (Aghajanzadeh, *et al.*, 2014; Asgary, *et al.*, 2016; Mahmood, *et al.*, 2011). Our choice for general anesthesia is single-lung ventilation for preventing aspiration of cystic material and respiratory complications. Lung-conserving procedures are optimal for pulmonary hydatidosis (Aghajanzadeh, *et al.*, 2014; Yekeler, *et al.*, 2012; Lal, *et al.*, 2013). Some researchers recommended that the most appropriate procedures for PHC are the open surgery and removal all portions of involved lung as we did in our study (Puri, *et al.*, 2011; Chacko, *et al.*, 2009; Yilmaz, *et al.*, 2009). Video-assisted thoracic surgery (VATS) is suggested for selected patients (Asgary, *et al.*, 2016 ; Ramos, *et al.*, 2001). We did not use VATS surgery in PHC.

Complicated hydatid cysts cause significant pleural thickening and parenchymal destruction; therefore, more difficult surgical procedures, such as decortication, segmentectomy, and lobectomy may be required in these patients (Köksal, *et al.*, 2004; Chacko, *et al.*, 2009; Yilmaz, *et al.*, 2009; Aarons, *et al.*, 1999; Kuzucu, *et al.*, 2004). We used above procedure in our patients. Resection rates of for complicated pulmonary hydatid cysts are 19% to 32% and 0% to 7% for uncomplicated pulmonary hydatid cysts (Cobanoglu, *et al.*, 2011; Yilmaz, *et al.*, 2009) .We performed decortication, wedge and segmental resection, and lobectomy while pneumonectomy in our cases wasn't performed.

In a study of patients with PHC and pleural complications, they found that decortication was needed in 30 patients (69.8%) and pulmonary resection was needed in 6 patients (14%) (Safioleas, *et al.*, 1999). In our series, 9 of the patients with complicated cysts required decortication, while twelve

patients needed wedge and segmental resection and two patients needed lobectomy. Morbidity and mortality in Complicated PHC are higher than uncomplicated cysts postoperative (Aghajanzadeh, *et al.*, 2014; Ekinci, *et al.*, 2015; Cobanoglu, *et al.*, 2011; Asgary, *et al.*, 2016 ; Aarons, *et al.*, 1999 ; Kuzucu, *et al.*, 2004). In complicated cases, infection of the adjacent lung parenchyma may lead to postoperative complications such as prolonged air leakage, empyema, atelectasis, and pneumonia. In addition, many patients with complicated pulmonary hydatid cysts require preoperative antibiotic therapy and supportive treatment. Complicated cases have a higher morbidity and mortality rate and need longer hospitalization than uncomplicated cases (Aghajanzadeh, *et al.*, 2014; Asgary, *et al.*, 2016; Safioleas, *et al.*, 1999). There was no mortality in our patients. Safioleas and colleagues (Safioleas, *et al.*, 1999 ) reported hospitalization time with pulmonary hydatidosis as a 12-days median hospital stay for uncomplicated cases versus a 21-days median hospital stay for complicated cases. In a report by Asgary and colleagues (Asgary, *et al.*, 2016) the hospital stay was from 7 to 22 days (mean, 11 days). The hospitalization stay of our patients ranged from 7 to 14 days (mean, 8 days).

## CONCLUSION

Surgery is a choice for curative treatment for pulmonary hydatid cysts. Although surgical treatment is effective in complicated cysts. Ruptured hydatid cysts are associated with increased morbidity. Also more difficult surgery and hospital stays are longer because of the surgical treatment before rupture of the cysts is essential. All pulmonary hydatid cysts should be surgically treated as soon as they are diagnosed for prevention of serious complications.

**Conflict of Interest:** None of the authors have any conflicts of interest to disclose. All the authors approved the final version of the manuscript.

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