

**CASE REPORT**

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**Bronchoscope saline injection method (ME Hejazi Method) in ruptured peripheral and central pulmonary hydatid cysts: a case series****Mohammad Esmail Hejazi\*, Sepideh Tahsini Tekantapeh\*, and Saeede Maddahi\*\*****ABSTRACT****BACKGROUND**

This paper mainly focuses on patients with ruptured pulmonary *Echinococcus granulosus* infections (alveolar hydatid disease), who suffered from ruptured alveolar hydatid cyst. In this study we aimed to remove these ruptured central and peripheral pulmonary hydatid cysts by the bronchoscopic saline injection method (ME Hejazi method).

**CASE DESCRIPTION**

In this retrospective study, we evaluated eight patients from an endemic area who were non-surgically treated for ruptured pulmonary hydatid cysts at Imam Reza hospital between 2016-2017. By the bronchoscopic saline injection method (ME Hejazi method), we extracted the entire hydatid cysts of all patients by fiber optic bronchoscopy for the detachment of the underlying membrane from the cavity wall. There were three female and five male patients, with a mean age of  $40 \pm 23$  years (range 17–63 years). Ruptured cysts were located in the peripheral (2) and central (6) parts of the lungs. All of our experiences have been successful without any complications and residual cyst membrane. During the follow-ups, clinical and radiological recovery were seen in these patients.

**CONCLUSIONS**

In the peripheral ruptured hydatid cysts, accurate recognition and location of the cyst is essential and the blind approach is not recommended because it needs several bronchoscopic interventions. Therefore the Hejazi method will be a beneficial and suitable alternative method for surgery in the treatment of patients with ruptured pulmonary hydatid cyst with cyst membrane adhesions.

**Keywords:** Bronchoscope saline injection method, ME Hejazi Method, Pulmonary hydatid disease, Ruptured hydatid cysts

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## Introduction

Human cystic and alveolar echinococcosis (hydatid disease) is still the cause of morbidity and mortality in some parts of the world including Eastern Asia, Middle East, North Europe, and South America. Somehow it is a public health problem in the Mediterranean and Middle East area.<sup>(1,2)</sup> Hydatid disease is a chronic parasitic disease with an animal-human cycle. It is caused by the larval (metacestode) stages of the dog tapeworm belonging to the genus *Echinococcus* (Taeniidae). There are three recognized forms of echinococcosis: cystic, caused by *E. granulosus*, alveolar, caused by *E. multilocularis*, and polycystic, caused by *E. vogeli*.<sup>(2,3)</sup> Among the three forms of hydatid disease, alveolar echinococcosis is associated with poor response and cyst rupture is an expected probability which complicates patient management and treatment.<sup>(4,5)</sup>

Despite current control methods, elimination of this pathogen is difficult. Hydatid control campaigns have made impressive technological improvements in the prevention, diagnosis and treatment of human and animal cystic echinococcosis. Although using these new methods increased the efficiency of hydatid control programs, hydatid disease is still common, the treatment is difficult enough and it will be more complex when we face a ruptured hydatid cyst.<sup>(2)</sup> Human cystic echinococcosis is the most common presentation which is estimated to be more than 95% of whole global cases (2-3 million) compared to alveolar echinococcosis which is less than 0.5 million cases (all reported in the northern hemisphere). The lungs are involved in about 30% of cystic echinococcosis.<sup>(3)</sup>

The main way to diagnose human echinococcosis is by imaging techniques (e.g. computed tomography, magnetic resonance imaging, ultrasound and radiography) which can accurately detect space occupying lesions. Laboratory-based techniques is the other useful way for confirmation of clinical infection by the

aid of specific serum antibodies (IgG antibodies and recombinant antigen B subunits).<sup>(1,2)</sup> In cystic echinococcosis, fluid-filled cysts develop and grow in potential body cavities, mainly in the liver and lungs or other locations. Cystic larvae grow slowly, thus they are well-tolerated by the host. In contrast, alveolar echinococcosis invades the surrounding parenchyma and does not have well-defined external margins.<sup>(4,5)</sup> The treatment of cystic lesions is difficult because most develop rapidly in the lungs. Therefore surgical removal is the principal therapeutic approach to treat ruptured and chronic pulmonary hydatid cysts, but on the other side medico-surgical approaches are expanding, along with percutaneous drainage for hepatic cystic echinococcosis.<sup>(3-5)</sup>

Generally the outlook for these patients is unclear and rarely predictable, so long-term imaging follow-ups seems to be essential to manage their condition and achieve favorable treatment outcomes.<sup>(6)</sup> Pulmonary hydatid cysts are often asymptomatic and can be diagnosed incidentally on plain chest radiographs, but computed tomography (CT) is the preferred imaging modality.<sup>(3,7,8)</sup> The aim of this retrospective study was to share our experiences of removing these ruptured central and peripheral pulmonary hydatid cysts by the bronchoscopic saline injection method (ME Hejazi method).

## CASE SERIES

The eight patients described in this report were seen and evaluated in the Respiratory and Critical Care Medicine ward of Imam Reza Hospital between 2016 to 2017 for complaints of exacerbation of respiratory manifestations (Table 1).

All eight patients were successfully managed nonsurgically by the ME Hejazi method (Figure 1) and all were asymptomatic at one-year follow-up (Figures 2-5). This research was approved by the Tuberculosis and Lung Disease Research Center Ethics Board.

Table 1. Pertinent details of ruptured pulmonary hydatid cyst subjects

	Cases							
	1	2	3	4	5	6	7	8
<b>Gender</b>	M	M	F	F	M	M	M	F
<b>Age (years)</b>	51	17	63	34	23	29	31	38
<b>Cyst type</b>	Peripheral	Peripheral	Central	Central	Central	Central	Central	Central
<b>History</b>	Chronic cough with acute exacerbation	Recurrent purulent cough	Chest wall pain and productive cough	Fever, chills and pneumonia	Abrupt purulent cough and dyspnea	Chronic cough with recent exacerbation	Increased sputum and dyspnea	Pneumonia and purulent cough

Note: M: male; F: female

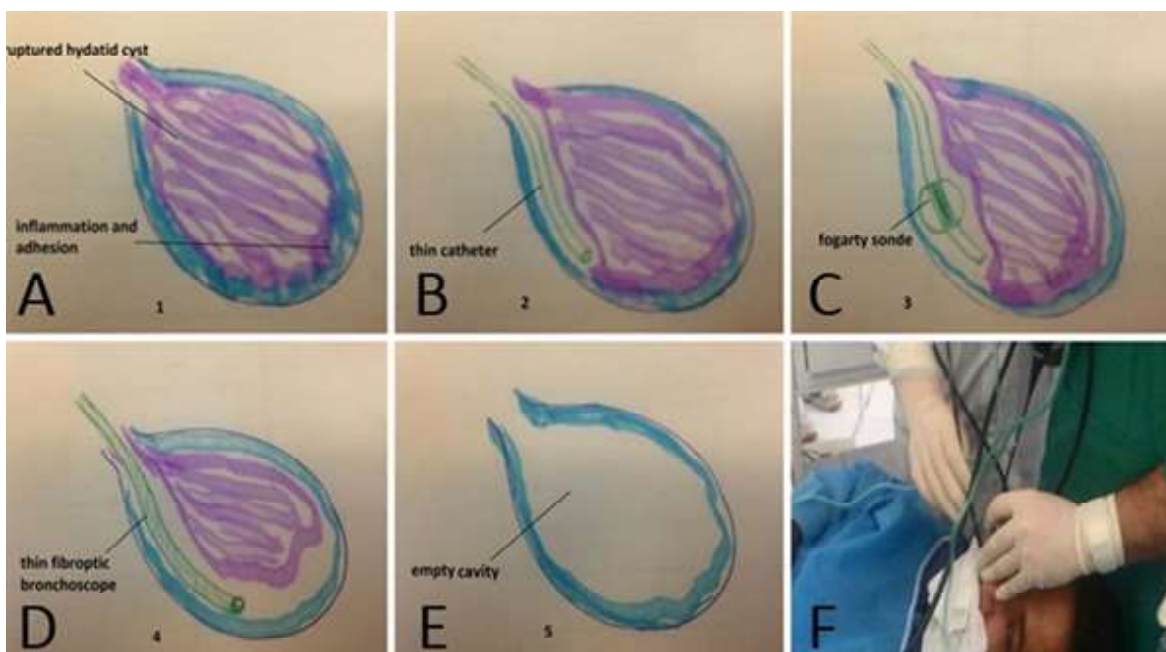


Figure 1\*. Stepwise bronchoscopic saline injection method to extract the ruptured hydatid cyst.

(A) Ruptured hydatid cyst was seen with adhesion and inflammation in cyst base that could not be removed only with bronchoscopic suction. (B) A thin catheter was entered to the space between cyst and lung tissue. (C) Fogarty balloon was inflated to release the adhesion of hydatid cyst. (D) A thin bronchoscope was entered to release the space between cyst and lung tissue. (E) The whole cyst contents was extracted by bronchoscope and the empty cyst was seen. (F) The procedure was performed with two bronchoscopes simultaneously

\* Figure 1; Reproduced from The Clinical Respiratory Journal 2016; 1752-6981, with permission of The Clinical Respiratory Journal Ltd and courtesy of ME Hejazi et al. A novel bronchoscope method (saline injection method) for complete extraction of ruptured pulmonary hydatid cyst (Case report article)



Figure 2. Hydatid cyst white membrane as seen in orifice and in formalin

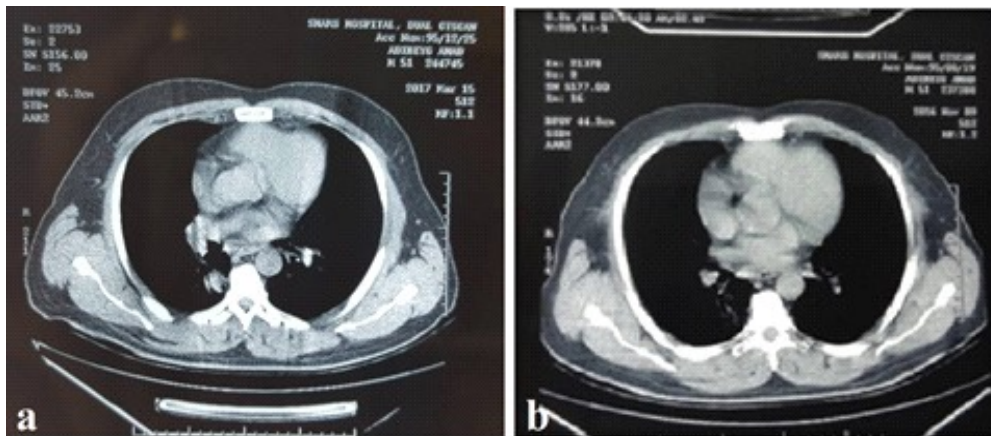


Figure 3. Peripheral hydatid cyst in a 51-year old man before (a) and after (b) ME Hejazi Method

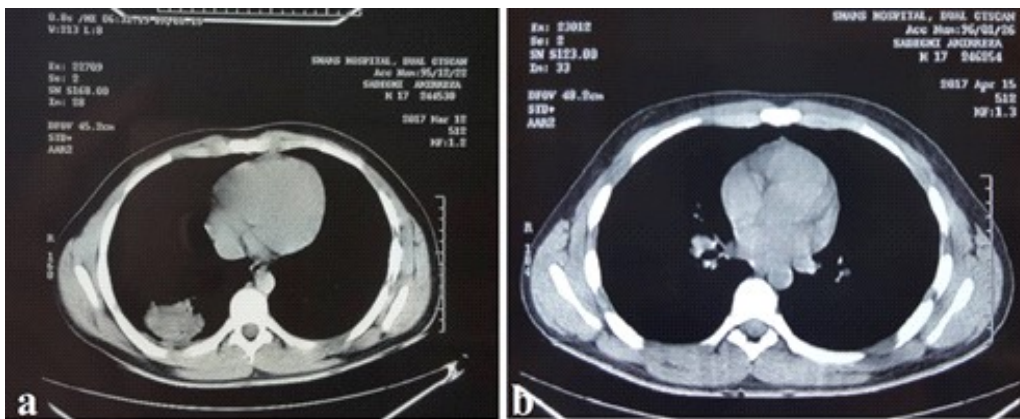


Figure 4. (a) Peripheral hydatid cyst in a 17-year old young man who was referred with recurrent purulent cough. (b) Complete removal of ruptured peripheral hydatid cyst 1 month after ME Hejazi method

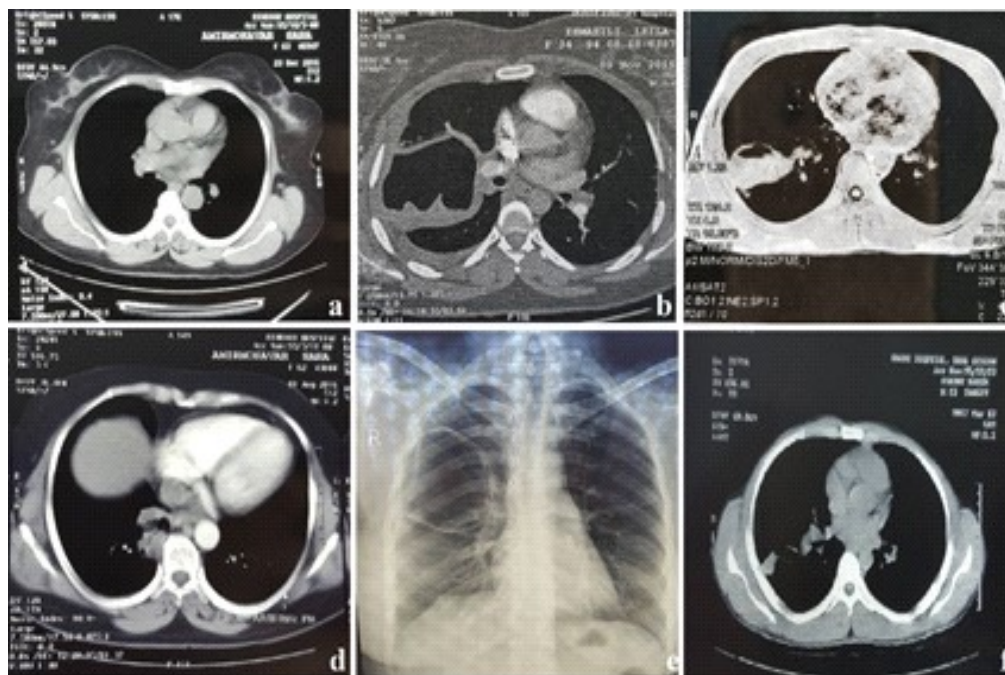


Figure 5. Ruptured central hydatid cysts of 6 patients which were resolved by interventional bronchoscopy method (a-f)

## DISCUSSION

Hydatid cyst infestation commonly involves the liver (55% to 70%), the lungs (18% to 35%), or both the liver and the lungs (5% to 13%).<sup>(9)</sup> A pulmonary hydatid cyst can spontaneously rupture into the pleura or bronchus due to superinfection, chest trauma, or erosion of cyst wall into a bronchus.<sup>(10)</sup> The diagnosis of complicated pulmonary hydatid cysts may not be easy because hydatid cyst disease mimics tuberculosis, lung cancer, empyema, or abscess.<sup>(11)</sup>

Pulmonary hydatid cysts may rupture into the bronchial tree or peritoneal cavity and produce cough, chest pain or hemoptysis.<sup>(12)</sup> Surgery has traditionally been the principal definitive method of treatment. As mentioned above there are some choices for treatment of hydatid cyst but the main treatment of ruptured chronic lung hydatid cyst is surgery, which based on previous review studies, is associated with some complications such as pneumonia, lung atelectasis, deep vein thrombosis, hospitalization, and morbidity. In case that the patient was not a proper candidate for surgical operation or did not agree with surgery because of complications, the routine treatment is a combination of medications which has been proved not so effective.<sup>(3)</sup> In recent years several case reports were presented of incidental hydatid cyst removal by bronchoscope, but there was not any systematic and specific plan for proper clinical action.<sup>(5,13)</sup> Since it seemed an efficient way to use, we started to think about making a well-defined method with a classified plan (as explained above) to remove ruptured hydatid cyst by bronchoscope. Based on Mahomed and Jayaguru's<sup>(14)</sup> method, we designed our saline injection method by bronchoscopy (ME Hejazi method) for removing the ruptured pulmonary hydatid cysts with adhesions which cannot be removed only by bronchoscopic suction. Finally, for the first time, we tried our method on two patients and the results were satisfying. There was not any sign of complications, recurrence

and remaining cyst membrane on the bronchial tree wall. So, we decided to introduce and describe this novel treatment method in a case report published in 2016 which had not been reported until then.<sup>(1)</sup> Due to the satisfying results, we expanded our study and tried it on more patients with ruptured hydatid cyst (Table 1).

During the last three years (2015-2017), we did this method on 8 other patients with the same condition (Figures 3-5). Fortunately it was effective in all cases without any complications and residual cyst membrane. During the follow-ups, clinical and radiological recovery were seen in these patients. Based on cyst membrane position against the main bronchus, hydatid cysts are divided into the two groups of central and peripheral cysts. Observation of the cyst membrane is an important principle for intervention, especially in the peripheral lesions. In these patients use of the Fogarty balloon is not possible. With the aid of a catheter we are able to reach the margins of the cyst and inject the saline, which is more difficult and needs several bronchoscopic interventions. In two of the patients this method was used without fail. The important point about peripheral lesions which should be considered is that if we were not able to see the hydatid cyst membrane, normal saline injection will not be useful. Therefore we recommend that: i) this method should be used before surgical operation in central ruptured hydatid cysts; ii) in the peripheral ruptured hydatid cysts, accurate recognition and location of the cyst is essential and the blind approach is not recommended because it needs several bronchoscopic interventions; and iii) in all of the cases medical treatment is necessary.

## CONCLUSIONS

This novel bronchoscopic saline injection method is curative, cost-effective, minimally invasive and needs no general anesthesia and hospitalization, with less complications in comparison with surgery. Therefore it will be a beneficial and suitable alternative method for



surgery in treatment of patients with ruptured pulmonary hydatid cysts. However, to extract further information, more studies are recommended to be done on this method.

### CONFLICTS OF INTEREST

The authors declared no competing and conflict of interests.

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### CONTRIBUTORS

All authors contributed equally to this work. All authors read and approved the final manuscript.



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