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Preoperatively Manifestations as Univesicular Mass-like Lesion and Postoperatively as a Multivesicular Lesion with Multiple Daughter Cysts: A Very Rare Case Report

Manouchehr Aghajanzadeh [a*], Ali Alavi Foumani [b], Azita Tangestaninejad [b], Seyed Aliaskhar Fakher Mosavi [b], Omid Mosaffaee Rad [a], Zahra Sadin [c] and Mohaya Farzin [c,d]

[a] Department of Thoracic Surgery, Guilan University of Medical Sciences, Rasht, Iran.
[b] Department of Pulmonology, Guilan University of Medical Sciences, Rasht, Iran.
[c] Inflammatory Lung Diseases Research Center, Department of Internal Medicine, Razi Hospital, School of Medicine, Guilan University of Medical Sciences, Rasht, Iran.
[d] Department of Physiology, Razi Clinical Research Development Center, Guilan University of Medical Sciences, Rasht, Iran.

Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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**Corresponding author: E-mail: Maghajanzadeh2003@yahoo.com;*

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ABSTRACT

Pulmonary vesicular hydatid cysts and uncomplicated cysts with echinococcosis *granulose* and daughter cysts are very rare. A 58-year-old man presented to our center with cough, hemoptysis, and right-side chest wall pain. Chest x-ray and computerized tomography scan revealed a cystic-like appearance mass in the hill of the right lung. FOB and BAL were normal. With right side PL- --thoracotomy, the cystic mass of the lung was removed. Hundreds of 1to3 cm size daughter cysts and laminated membrane was discovered after opened the cystic mass. Pathologic examinations revealed the hydatid cysts with daughter cysts on the lung cystic mass. To the best of our knowledge, this is probably a rare presentation of a lung hydatid cyst as a mass-like and univesicular cyst, which is full of a daughter cyst.

Keywords: Daughter cyst; lungs; univesicular hydatid cyst; cystic mass.

1. INTRODUCTION

“An infection with the cestode Echinococcus granulosus causes hydatid disease. Cystic echinococcosis is seen in some countries in words. It is a public health problem in endemic areas, such as South and Central America, the Middle East, Africa, Russia, China, Australia, and New Zealand. and is endemic in Iran” [1,2,3]. “Adult worms mature in the dog's intestine (definitive host) and the eggs are released in the stool” [4,5,6]. Adult worms mature in the dog's intestine (definitive host), releasing eggs in the stool [3,4,5]. “Animals like sheep get this disease via ingestion of contaminated vegetables. Humans are accidental hosts; oncospheres hatch in the duodenum, penetrate the intestines, and are carried via the bloodstream to various organs” [3,4,5]. Most often, it affects the liver and lung tissue [1,3,4,5]. “Hydatid disease mainly affects the liver (75%) and the lung tissue (15%) and occurs only 10% in other organs” [2-6,7,8]. “Pulmonary hydatid disease may accompany complications, including cyst rupture into the pleural space, lung mass, hemoptysis, and suppuration” [9,10,11,12,13,14]. “Patients may develop sudden onset of chest pain, cough, fever, and hemoptysis after cyst ruptures, urticaria wheezing, and anaphylaxis secondary bacterial infection. Infection resulting in difficulty in differentiation disease” [2-4,7,8,9,13].

“Hydatid disease should be in the differential diagnosis when a cystic lesion detected in an endemic area as bronchogenic cyst, lung carcinoma, sarcoma of the lung, metastasis, hematoma, mesothelioma, granulomatous lesions, and pulmonary abscess” [2-4,7,8,9,13,15,16]. Daughter cysts and calcifications are rarely seen in pulmonary hydatid cysts [15,16]. “Chest imaging is the principal investigational modality for pulmonary hydatid cysts. Computed tomography (CT) and magnetic resonance imaging (MRI) of the lungs are the various valuable modalities in the diagnosis of thoracic hydatid cyst” [7,8,9,10,11,12,13]. The treatment is surgery [1,6,10,12,13]. “The goal of surgical intervention includes the removal of the entire cyst while preserving the lung parenchyma as much as possible and without allowing intraoperative spillage” [1,9,10,11,12]. The aim of this case report is for three interesting causes, first is the unusual presentation of a univesicular cyst in imaging preoperative and with multiple daughter cysts postoperative; second is the location of the cyst in the hill of the lung, and third is present as a cystic mass lesion.

2. CASE REPORT

A 58-year-old man was admitted to our referral hospital in pulmonology department with complaint of dry cough, milled hemoptysis, right side chest pain, night sweeting and Lowe grade fever for three months. The symptoms in recent two weeks was sudden expectoration mild sputum with mixed with blood. In examination the respiratory sounds were normal but crackles were present in right hemi thoraces. Chest roentgenogram and computed tomography (CT scan) demonstrated cystic mass like lesion in the right hill of lung (14 × 12 × 11 cm), (Figs. 1,2,3,4). FOB performed show small compression on the right upper lobe, others were normal. BAL was normal in pathology evaluation. Patient refereed to thoracic surgery ward for excision of that lesion. After general anesthesia with one lung ventilation, five intercostal space was opened in right up position with muscle saving procedure. After pneumolysis the cystic mass in the hill of lung was walling of with wet sponge in normal saline for prevention of spillage cyst content of cystic mass when rupture occurs,

because in endemic area as Iran all cystic masses should be in diffracted from hydatid disease. The cystic mass was resected with safe margin as wedge resection. After chest -tube insertion and closed of chest wall the cystic mass was operated. After opened the mass there was ruptured laminated membrane with more than hundreds of 1 to3cm daughter cysts in cavity (Figs. 5,6,7). Ultrasonography of abdomen was normal for cystic or mass lesions. Albendazol 800 mg prescribe for three cores of 28 days with 14 day interwall. The patient discharged on the fifth day postoperatively without any complications. As a result, the pathologic examinations of ruptured laminated membranes and daughter cysts confirmed a hydatid cyst. On 6 and 12 month follow up, patient condition is well.

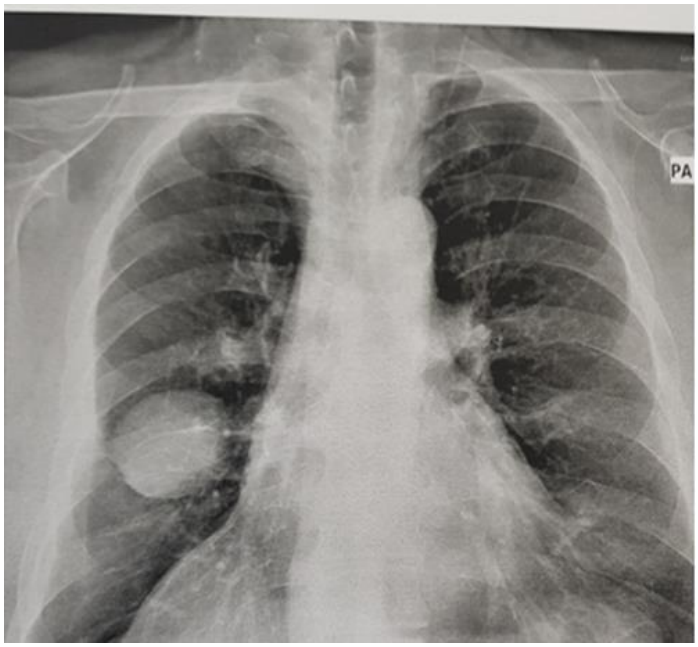

Fig. 1. CXR show a round cystic mass like lesion in RL lobe

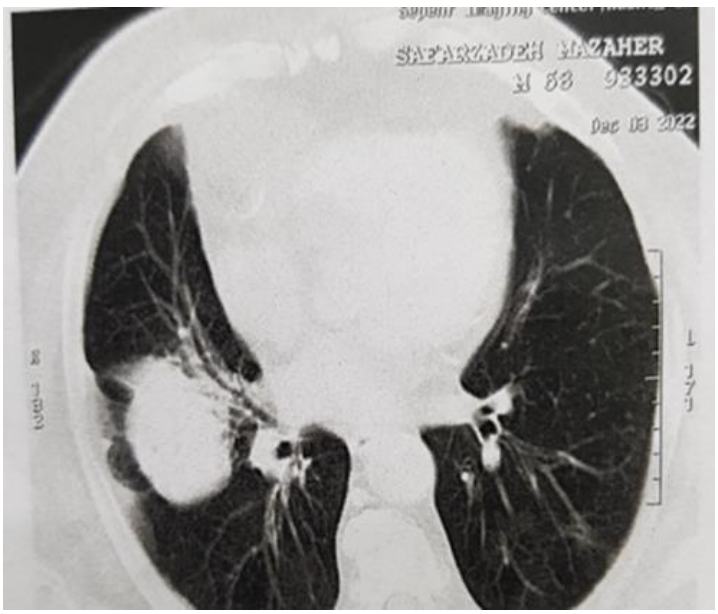

Fig. 2. CT-scan shows mass like lesion in the RL lobe

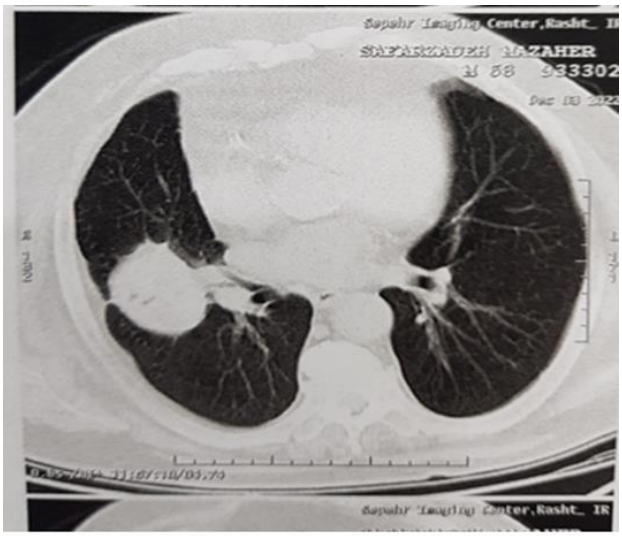

Fig. 3. CT-scan shows mass like lesion in the RL lobe

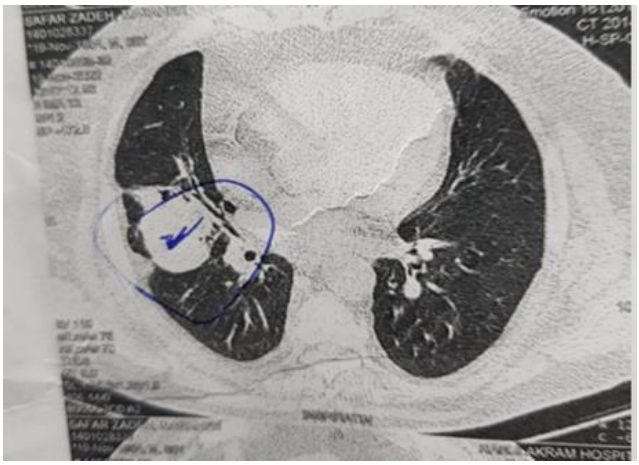

Fig. 4. CT-scan show mass like lesion in the hill of lung with pleural thickening

Fig. 5. Hydatid cyst content

Fig. 6. Hydatid cyst content with daughter cyst

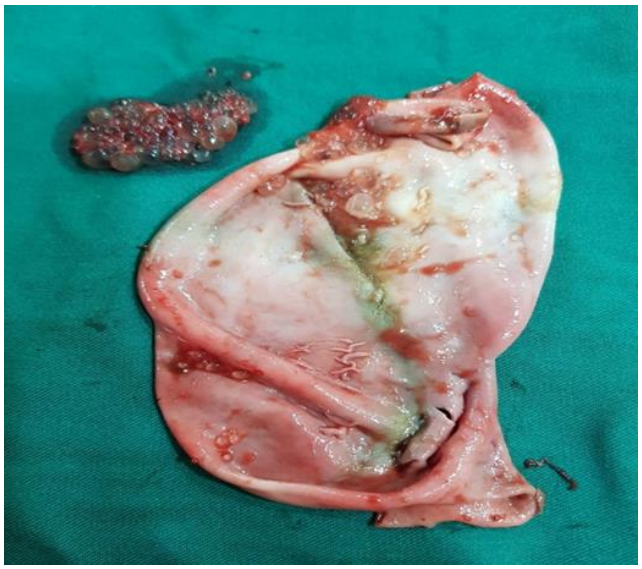

Fig. 7. Lamminated membrane and daughter cysts

3. DISCUSSION

Hydatid cyst disease is still a significant health problem in certain countries such as South and Central America, the Middle East, Africa, Russia, China, Australia, and New Zealand. and Central Europe occurs in immigrants from endemic areas, and in these areas, hydatid cysts are endemic [1-3,4,5,9]. In Iran, hydatid cysts are endemic [1,15,16].

“Hydatid disease mostly affects the liver (75%) and the lung tissue (15%) and occurs only 10% in other organs” [2,4,5,6,7,8]. “Pulmonary hydatid cysts are usually asymptomatic, while symptoms may appear with increasing size over a period of time or in cases where cysts are in the central portion of the lung or other complications” [3,9,10,11,12] as our case. Hydatid disease is caused by an infection with the Echinococcus granulosus in endemic areas such as Iran [1,3,7,11,12]. “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” [3,7,11,12]. “Adult worms mature in the intestine of the definitive host as a dog, and the eggs are defecated with stool” [3,4,5,6]. Animals like sheep get this disease by ingesting contaminated vegetables and water. Humans are accidentally infected. Oncospheres hatch in the duodenum mucosa of sheep, penetrate the intestines, and are carried via the bloodstream to various organs

such as the liver, lungs, and all organs [3,4,5,6,7]. Daughter cysts can also sporadically or rarely be seen in pulmonary hydatid cysts [3,4,7,9,11,12]. “The germinative layer secretes hydatid fluid and generates a brood capsule. If the inner layer and the brood capsules split up, daughter cysts can be produced” [13]. “Due to the early presentation of symptoms and treatment, in PHC, there is enough time for forming the daughter cysts; hence, the occurrences of daughter cysts in the pulmonary hydatid cysts are uncommon” [15]. We present one case with a cystic mass lesion and univesicular on CT-scan in the hill of the right lung before surgery with hundreds of daughter cysts with laminated membrane postoperative, like such cases rarely reported in the literature [15].

Univesicular cysts may contain no daughter cysts [15,16] and are called unilocular; cysts with daughter cysts are called multivesicular. In the literature [16], Chest X-ray and CY-scan can show cyst mas lesion as a round opacity in a hemi thorax [15,16]. A CT scan can detect pulmonary hydatidosis with or without daughter cysts [15,16], but a CT scan cannot show small sizes of daughter cysts as in our case before surgery. “Repeated mechanical trauma such as heartbeats, respiratory motions, separation of some parts of the laminated membrane from the pericyst layer into the cyst cavity, and the formation of daughter cysts. Daughter cysts may develop directly from the end cyst, resulting in multisystem or multivesicular pulmonary hydatid cysts” [13,15,16,17]. In this case, daughter cysts were present in the mass-like cystic lesion. Daughter cysts rarely can be seen in pulmonary hydatid cysts. The germinative layer secretes the hydatid fluid and brood capsule. Daughter cysts can be produced by spitting up germinative and brood capsules [18]. “Because of the early presentation of symptoms and early treatment of pulmonary hydatid cysts, there is not enough time to form the daughter cysts. Therefore, the daughter cysts in the pulmonary hydatid cysts are uncommon” [18]. We present one case of intact lung hydatid cysts with a CT scan without any daughter cysts, but postoperative, there were more than a hundred daughter cysts rarely reported in the literature. Cysts without daughter cysts are called univesicular, and those with daughter cysts are called multivesicular. A CT scan can show pulmonary hydatid cysts with or without daughter cysts [18] but in the majority of cysts, due to the very small sizes of daughter cysts, CT -scan is not detected before surgery [18] as in our case. There are two theoris for the formation of daughter cysts [18]: Repeated mechanical trauma, such as pulsation of heartbeats and respiratory motions and opening of bronchioles into the cyst cavity [18]. In our case, daughter cysts were not present in the hydatid cyst, but it present in the mass cystic lesion postoperative with cut the mass.

It is required for Differential diagnosis of lung masses, which most commonly present with malaise, weight loss, and cough, especially in endemic areas such as Iran [15].

“Hydatid cysts of the lung should always be kept in the differential diagnosis when a cystic lesion is detected in a patient from an endemic area. Differential diagnosis for pulmonary HCs can be listed as bronchogenic cyst, lung carcinoma, sarcoma of the lung, metastasis (when cysts are multiple), hematoma, mesothelioma, granuloma, and lung abscess” [19,20]. “CT scans and MRIs can help cysts from masses in some patients, but a definitive diagnosis is not possible” [4,5,7,15]. Transthoracic needle biopsy is dangerous or contraindicated in endemic areas, such as our case, because it can rupture cyst and produce anaphylaxis and disseminate cyst content [3,4,7,11].

Surgical treatment. For patients who are cardiovascularly stable and able to undergo a major operation, surgery is the treatment of choice because the parasites can be excised completely, and the patient will be cured [1,2,3,10,11,15]. Our surgical options for lung cysts include wedge resection, evacuation, bronchial opening closure with precystectomy, rarely lobectomy, and capitonnage for obliteration of remnant cavity [1,3,7,11,12]. During surgery, it is important to prevent intraoperative spillage of cyst contents for dissemination, allergic reactions, and recurrence [1,3,7,11,12,15]. In our patient we don’t use any sporicidal agents in pulmonary hydatid cysts. Medical therapy with Albendazole is Used in patients who cannot tolerate undergrowing surgery or in disseminated disease and when there is intraoperative spillage of hydatid fluid [17,20,21,22]; after surgery, we prescribed Albendazole 800 mg in three cores of 28 days with 14 days interval.

4. CONCLUSION

In the endemic areas, all cystic mass lesions of the lung should be differentiated from hydatid

cysts. MRI and CT scans help with diagnosis, but FOB can help with diagnosis of lung mass or sometimes can see the priciest. The only way for a definitive diagnosis is surgery such as VATS or thoracotomy.

CONSENT

Informed consent was obtained from all individual participants included in the study.

ETHICAL APPROVAL

This study was approved by the ethics comity of Arya hospital and the Inflammatory Lung Diseases Research Center of Razi Hospital, Guilan University of Medical Sciences, Rasht, Iran.

DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of this manuscript.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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