

Pulmonary Hydatid Cysts Causing Massive Hemoptysis and Hydatoptysis Treated Surgically: A Case Report and Review of Literature



Ikram Chaudhry*, Ahsan Cheema, Hadi Mutairi, Miral Mashhour, Alaa AL Sharif, Shoukat Bojal and MS AlQahtani

Department of Thoracic Surgery, Pathology and Hepatobiliary Surgery, King Fahad Specialist Hospital Dammam, Saudi Arabia

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***Corresponding author:** Ikram ul haq Chaudhry, Department of Thoracic Surgery, Pathology and Hepatobiliary Surgery, King Fahad Specialist Hospital Dammam, Saudi Arabia, Email: drihc007@gmail.com

Abstract

A 52 years old male nonsmoker, presented with a history of shortness of breath, cough, hemoptysis and upper right upper quadrant abdominal pain for the last one month. No history of loss of weight or appetite. Patient was diagnosed six months ago as hydatid disease of liver and lung and he was commenced on oral scolicial agent Albendazole by referring hospital. Computed tomographic scan of thorax and abdomen Demonstrated large bilateral pulmonary and hepatic cysts with typical radiological findings of hydatid cyst. Pulmonary cysts were excised by staged procedure and later on hepatic cyst were drained by interventional radiologist.

Keywords: Echinococcus granulosus liver; Lung; Hemoptysis; Hypertonic saline; Surgery

Background

Parasitic infestation by Echinococcus Granulosus was described by Rudolph in 1808 as a hydatid cyst also this is known as echinococcosis and hydatosis [1]. There are four other species of Echinococcus which can affect human but among all

Echinococcus Granulosus is the most common cause of zoonotic parasitic infection (Table 1). Epidemiologically, this disease prevails almost all over the world but there are endemic areas as Australia, New Zealand, South and Central America, Middle East, Sub Saharan Africa, Russia, China, Turkey [2,3].

Table 1: Epidemiological features of different Echinococcus species.

Species	Geographical	Human Infection	Definite Host	Intermediate Host
E Granulosus	All over the world	Common in human	Dog and other Canines	Sheep, goats cattle, camels, horses
E Multiloculariari	Russia, western China, Central Europe (Northern Japan, America, Africa)	Less common infection in human but can be severe, limited to animal hosts mostly	Primarily dogs, wolves, coyotes, cats	Deer, Bison, Rodents ,moose
E Vogeli	Central and South America	Intermediate	Wild dogs and Canines	Rodents. peacocks
E Oligarthrus	Central and South America	Rare cases reported	Wild animals Pumas, Jaguars	Rodents, rabbits

Case

A 52 years old male smoker presented to our tertiary care hospital with history of cough and hemoptysis. He was diagnosed six months ago in the referral hospital as case of

hydatid disease of lung and was commenced on Albendazole 800 mg daily. He had scattered hemoptysis for 3-4 times in a month but prior to admission he had massive hemoptysis. On clinical examination he had dyspnea with the respiratory rate of 24/min, no cyanosis or jaundice, breath sounds were decreased

over right hemi thorax. Laboratory tests revealed Hemoglobin (8gm/dl), liver enzymes Alanine Transaminase (ALT) 28U/lit Aspartate Transaminase (AST) 16U/lit, Alkaline phosphatase 300 U/lit. Chest radiograph and CT scan of thorax and upper abdomen showed large bilateral pulmonary and hepatic cysts. Immunoglobulin G for echinococcus was positive. As the patient had massive hemoptysis and was symptomatic urgent surgery was planned. Surgical approach was right thoracotomy and we found a very large cyst densely attached and compressing the lung parenchyma. A purse string suture was placed and in the center of cyst and surrounding lung tissues were covered with

gauze soaked in hypertonic 14% saline. A 14 F catheter was inserted in the cyst and tightly snugged to avoid any spillage then 200 ml of 14% hypertonic saline was installed in the cyst cavity and left for five minutes after that fluid was sucked, catheter was removed and purse suture snugged. With meticulous dissection cyst was removed in total sparing the lung parenchymal tissues (Figures 1 & 2). The left side cysts were removed as staged procedure after four weeks in similar fashion. Histopathology report showed dead daughter cysts and no live parasite (Figure 3).

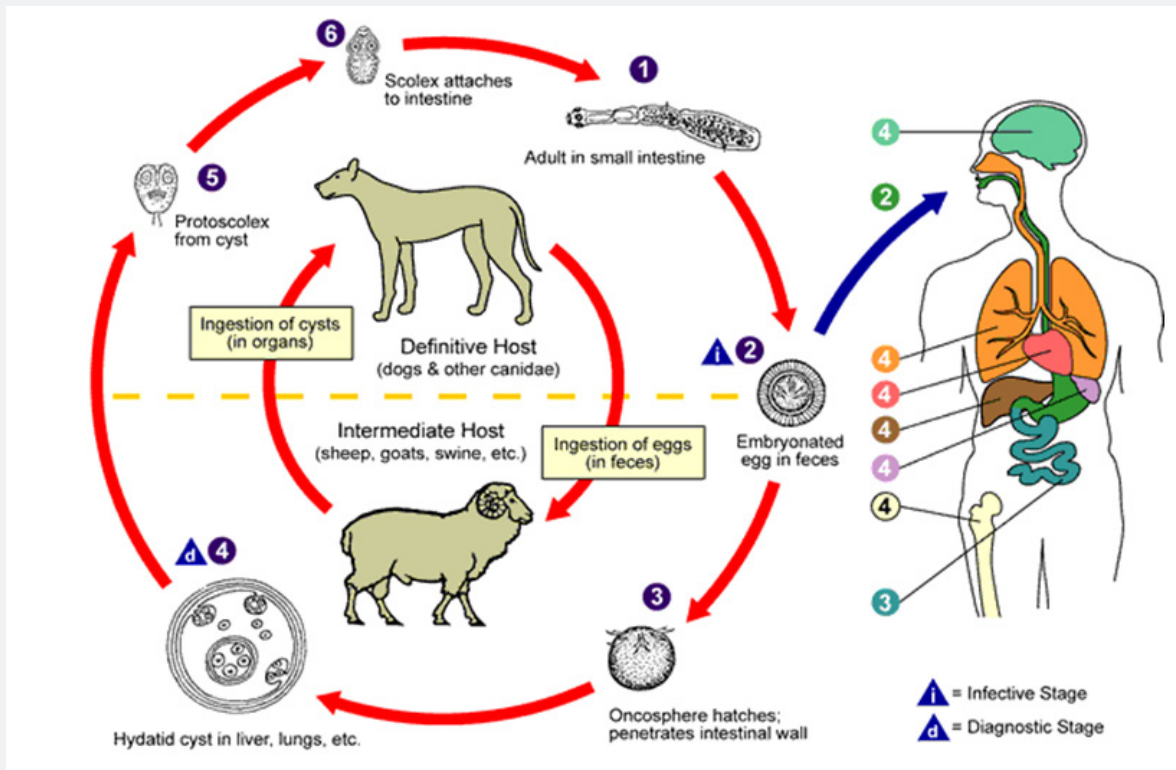


Figure 1: Lifecycle of Echinococcus Granulosus (Tapeworm) and effected human organs. (CDC Center for Disease Control and Prevention).

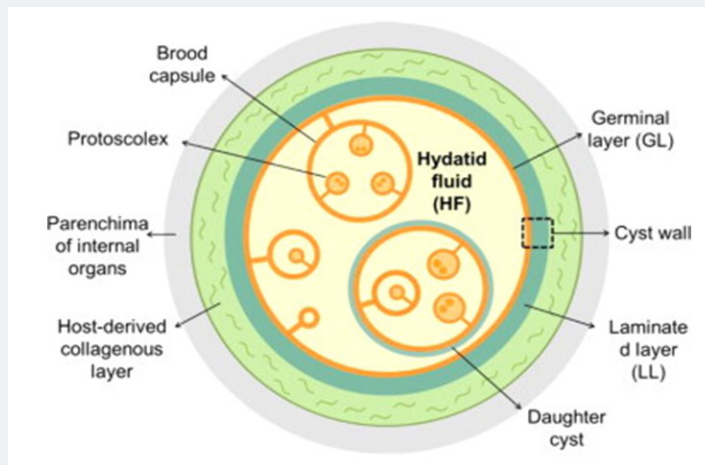


Figure 2: Trilayer structure of the Hydatid Cyst.

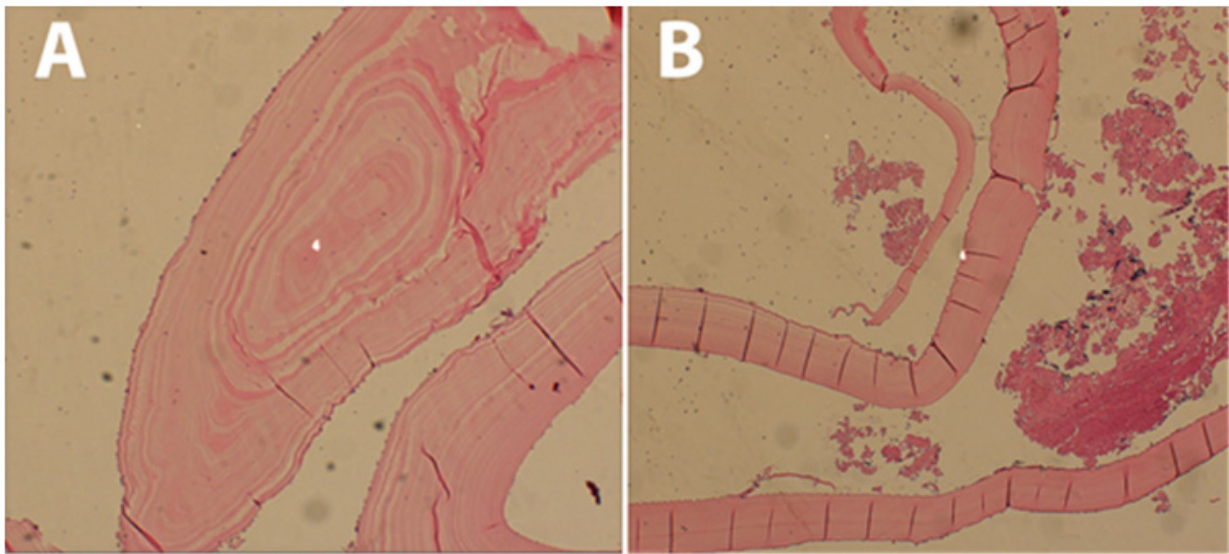


Figure 3: (A) Cyst cavity showing a germinal layer (B) H&E 10 X showing Cyst wall.

Discussion

Echinococcosis also known as hydatidosis is the most common parasitic disease caused by larval cestodes. This parasite harbors in two hosts: primary and intermediate to complete its life cycle. Livestock animals like sheep, goat, horse, pigs, camels as well as humans are intermediate host while the Carnivorous animals like wolves, dogs, are definite host (Figure 4A-4F). Humans are infected accidentally by ingesting the vegetables, fruits and water contaminated by soil with dog stool containing eggs. Echinococcus eggs can survive for a year outside in the atmosphere and are the main source of contamination

[4-6]. On ingestion eggs hatch to oncosphere larva in the small intestine thereafter, enter in to the blood stream and is carried to the liver by portal circulation or can by pass to pulmonary system and then develop to metacestodes. These fluid filled cysts are composed of hundreds of protoscolices, which are the source leading to formation of daughter cysts or mature worm (2-3 mm long). This life cycle is usually completed in 2-7 weeks. Human to human spread is not possible as carnivores' (definite host) is required to complete the life cycle. Hydatid cysts are tri layers, (pericyst, laminated and germinal) filled with nutritious fluid that promote larval growth [7,8] (Figure 5).

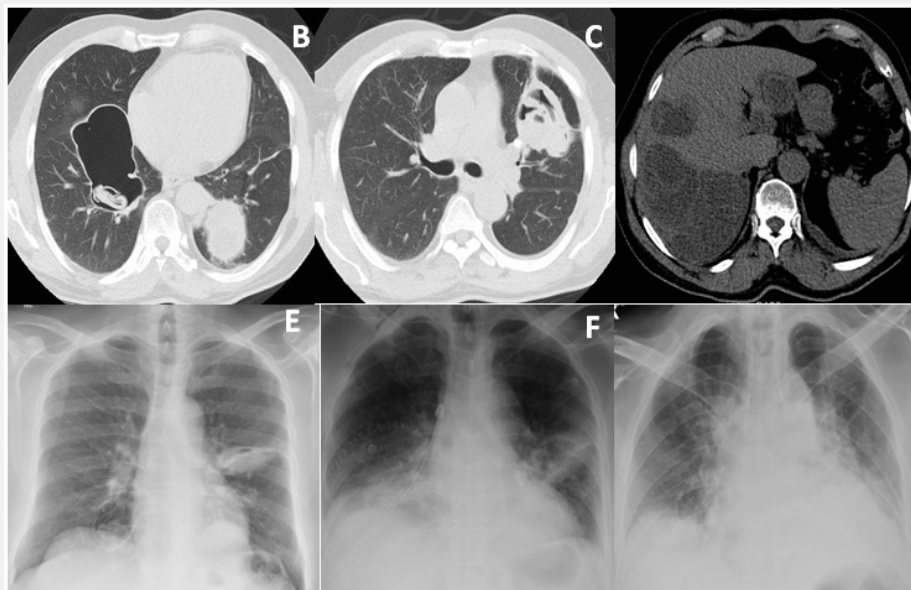


Figure 4: (A) CT Scan of Thorax showing a large cystic lesion in the Right lung. (B) CT Scan of Thorax showing a large cystic lesion in the Left lung. (C) CT Scan of Liver shows multiple cystic lesions. (D) Pre-operative chest x-ray. (E) Chest X-ray after right cyst excision. (F) Chest X-ray after bilateral staged thoracotomy.

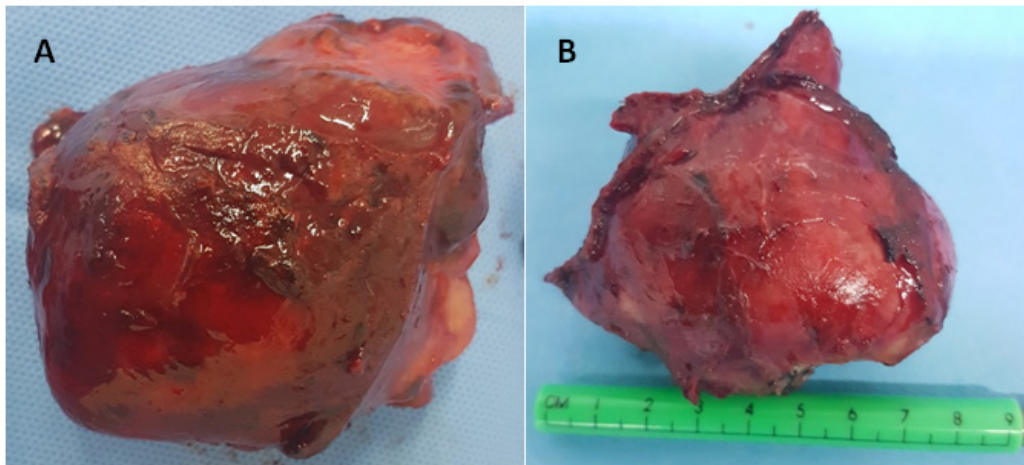


Figure 5: (A) Macroscopic view of the Right Hydatid cyst after excision. (B) Left Hydatid cyst after excision.

The best diagnostic modality is imaging chest x-ray and CT scan of thorax and abdomen [9,10]. Serological methods, Weinberg complement fixation and Casoni tests were used in the past and their yield is very poor. ELISA and IHA are most widely used serological tests [11]. Patients with pulmonary hydatid disease are initially asymptomatic. Most of the symptoms are either due to mass effect of cyst on neighboring structures or rupture of cyst leading to anaphylactic reaction, hemoptysis, hydatoptysis, infection, pneumothorax and empyema (Table 2). Clinically patient can present with chest pain, fever, tachycardia, hemoptysis, and hydatoptysis. Pulmonary hydatid cysts increase in size faster than in other parts of body probably due to elastic nature of lungs and negative intrathoracic pressure [12-14].

Treatment is chemotherapy and surgery. The medical treatment is very beneficial in patients with multisystem (disseminated) disease and can be given as neoadjuvant therapy or adjuvant therapy in patients at risk of spillage during the surgery. The commonly used chemotherapeutic drugs are Mebendazole, Albendazole, Benzimidazole and Praziquantel. Albendazole is preferred because of its better bioavailability and the dose is 15mg/kg body weight per day in two daily doses approximately 800mg daily dose for minimum 3-6 months. It is not recommended in pregnancy particularly in first trimester due to its teratogenicity [15-18]. Horton et al. [19] treated 500 patients of Echinococcosis giving those 800 mg of Albendazole daily two and half cycles with 14 days interval. After evaluation of 250 patients only 47 required surgery and resected specimen demonstrated that only five patients have viable parasite. Recurrence rate in patients who received pre-operative Albendazole as compared to those who did not receive Albendazole was 18.75% & 4.16 % respectively. In medical literature Little et al. [20] reported recurrence rate of 22%, Mottaghlani and Sadi observed 11.3%. Medical treatment alone is not enough to eradicate the disease once there are cystic lesions in lung and liver [19,20].

Radical Surgical resection of host tissues and entire cyst is mandatory if patient is symptomatic or any signs of infection or invasion in to surrounding structures. Postoperative chemotherapy for 1-2 years is recommended. Surgical approach is designed according to the location of the cysts. Most common surgical approaches are thoracotomy, median sternotomy and video assisted thoracoscopic resection. The basic surgical principal is that spillage of cyst contents should be avoided and adjacent tissues should be packed with gauze soaked in hypertonic saline solution (15%,20%) to avoid contamination. In case of cyst rupture during surgery or if cyst is infected, after the removal of germinal layer its recommended to wash the cyst cavity with hypertonic saline solution [21]. Dekak et al. [22] reported 202 patients who underwent surgery out of 422 cases. Enucleation, capitonage, segmentectomy, and lobectomy were the procedures performed [22]. Biswas & Burhan et al. [23] reported 26 and 24 cases respectively which were treated surgically. Ashok et al. [24] reported 33 cases out of 72 who required surgery [24].

Liver is the most common site of hydatid cyst formation, right lobe and left lobes are affected 60-75% and 20% respectively. Hepatic cysts generally remain asymptomatic for long time. The most common complication is intrabiliary rupture 3-17% or rarely can rupture in to pericardial, pleural or peritoneal cavity, and neighboring organs in 20-50% of cases. Sometimes cysts develop secondary bacterial or fungal infection. The best diagnostic modality is Ultrasound and Spiral CT scan. ERCP can be helpful for diagnosis of biliary rupture or communication. Liver hydatid cyst rupture can be categorized as contained, communicating or direct. Reported incidence of communicating rupture in to biliary system is 44-64% [25]. The patient with intrabiliary rupture present with right upper quadrant pain, obstructive jaundice, fever, cholangitis and sepsis (Table 2). There are multiple treatment modalities for the management of liver hydatid disease and surgery is reserved for complicated

cysts. Rupture of the cyst into adjacent organs, complicated with biliary fistula, compression of adjacent vital structures and cysts with infection or hemorrhage require surgical intervention. Surgery is also recommended for cysts with many daughter vesicles that are not amenable to percutaneous treatment (WHO stage CE2 and CE3b) [26] (Table 3). If the cyst diameter is more than 10cm and or percutaneous facilities are not available then surgery is advisable. The other management

options recommended by WHO are drug therapy (Albendazole) for stage CE1 and CE3 a provided the cysts are less than 3cm. If because of any reason medical management is not feasible then percutaneous treatment with puncture, aspiration, injection and respiration (PAIR) is the alternative. Combination treatment with PAIR and Albendazole is recommended for stage CE1 and CE3 that are more than 5cm in size [27-29].

Table 2.

Hepatobiliary		Pulmonary	
Symptoms	Complications	Symptoms	Complications
Right Upper Quadrant Abdominal Pain With Rebound Tenderness	Rupture: Intrabiliary Pericardial Peritoneal Pleural Adjacent Organs	Chest Pain	Mass Effect Of The Cyst
Hepatomegaly	Infection: Bacterial, Fungal	Fever	Rupture Of The Cyst: Lung Trachea Pericardium
Fever With Chills	Hemorrhage	Tachycardia	Anaphylactic Reaction
Cholangitis	Biliary Fistula	Hemoptysis	Hemoptysis
Sepsis		Hydatoptysis	Hydatoptysis
Anorexia			Lung Infection
Weight Loss			Pneumothorax
Nausea, Vomiting			Empyema
Obstructive Jaundice (Late)			Bronchopleural Fistula

Table 3.

WHO-IWGE 2001	Gharbi 1981	Description	Stage
CE 1	Type I	Unilocular, anechoic cystic lesion with double line sign	Active
CE 2	Type III	Multiseptated, "rosette-like" "honeycomb" cyst	Active
CE 3 A	Type II	Cyst with detached membranes (water-lily sign)	Transitional
CE 3 B	Type III	Cyst with daughter cysts in solid matrix	Transitional
CE 4	Type IV	Cyst with heterogeneous hypoechoic/hyperechoic contents. No daughter cysts.	Inactive
CE 5	Type V	Solid cyst with calcified wall	Inactive

Conclusion

In conclusion we treated a patient who had simultaneous hepatic and bilateral pulmonary hydatid cysts who presented with hemoptysis and right abdominal upper quadrant pain. The pulmonary hydatid cysts were resected as staged procedure and liver cysts were treated with PAIR and Albendazole therapy for six

months. Our improvised technique of installation of hypertonic saline in the cyst prior to surgical dissection is effective and safe.

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