CASE REPORT

Giant Pleural Hydatid Cyst: A Case Report

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ABSTRACT

Introduction: Echinococcosis is a zoonotic disease caused by the larva stage of a Taeniidae parasite of *Echinococcus*. *Echinococcus granulosus* (*E. granulosus*) causes cystic echinococcus (CE) and is the most common species causing disease in humans. A pleural hydatid cyst should be considered initially with a corresponding chest radiograph. It can develop into fatal complications, including secondary infection, severe bleeding, bronchial rupture and pleural or pericardial cavities. This case report shows a rare case of a pleural hydatid cyst.
Case: We reported a 59-year-old woman with shortness of breath and a productive cough for three months. The chest radiography and computed tomography (CT) scan revealed a cystic lesion measuring 14.2 x 18.5 cm with edge calcification thick around 3 mm. The pleural histopathology revealed that the cyst wall was made of fibrocollagen with a red oval echinococcus. The patient received oral albendazole/ABZ (400mg BID) for three months, paracetamol (500mg TID), and n-acetylcysteine (200mg TID). After three months, the patient had no complaints, only shortness of breath with heavy activity.
Conclusion: It is important to be aware of this condition to avoid inappropriate and potentially dangerous underdiagnoses and treatment, which could endanger the patient.

INTRODUCTION

Cysts and cavities in the lung are the most common abnormalities seen on a computed tomography (CT) scan of the thorax and can be a diagnostic challenge. In such circumstances, distinguishing a cyst (a circular air space pathologically lined by a thin epithelium or fibrous tissue, usually 2 mm) from a cavity (wall thickness > 2 mm) influences the diagnostic process. Other radiological characteristics include size, inner wall contour, contents, and location.[1]

The larva form of echinococcus causes a hydatid cyst. The larva stage of a parasite belonging to the Taeniidae family and the *Echinococcus* species causes the zoonotic disease echinococcosis. The most frequently damaged organs are the lungs and liver. Aside from hair, teeth, and nails, hydatidiform cysts can affect practically any organ in the body. The lungs are the second-most frequent site of hematogenous spread in adults and likely the most frequent site in children.[2]

There are four different species of *Echinococcus* known to cause public health problems. *Echinococcus granulosus* (*E. granulosus*) causes cystic echinococcus (CE) and is the most common species causing disease in humans. *Echinococcus multilocularis* (*E. multilocularis*) is rare, the most lethal species, and causes alveolar echinococcus (AE). *Echinococcus vogeli* (*E. vogeli*) and *Echinococcus oligarthrus* (*E. oligarthrus*) cause polycystic echinococcus. CE is the most common presentation in humans, contributing to more than 95% of the estimated 2–3 million global cases. CE is endemic in many parts of the world, especially the Mediterranean countries, Central Asia, including the Tibetan highlands, North and East Africa, Australia, and South America.[3,4]

Hydatid pleural cysts are extremely uncommon. This case was chosen as a lesson in making a diagnosis

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and providing appropriate therapy. It is believed to be helpful to all of us.

CASE

A 59-year-old female patient first arrived at the emergency room Dr. Mohammad Hoesin General Hospital, Palembang, with the main complaint that her shortness of breath had worsened seven days after being admitted. She also complained of a productive cough for three months. The patient has a background in gardening and frequently wears no gloves. The patient acknowledged that she often worked without gloves when farming and gardening.

Vital signs showed blood pressure of 140/90 mmHg, tachycardia, tachypnea, and normal body temperature. Thorax examination revealed a slight increase in right hemithorax dynamic movement, with a decreased right lung vocal fremitus compared to the left lung hemithorax. On percussion, it was found that the right lung was dull at intercostal space II and below. The vesicular sound was decreased from intercostal space II down to the right lung hemithorax compared to the left lung hemithorax. Blood examination showed hemoglobin 12.5 g/dL, leucocyte 11,820/mm^3, thrombocyte 363,000/L, eosinophil count was normal, and erythrocyte sedimentation rate was 20 mm/hour. Amoebae and helminths were not found on stool microscopy.

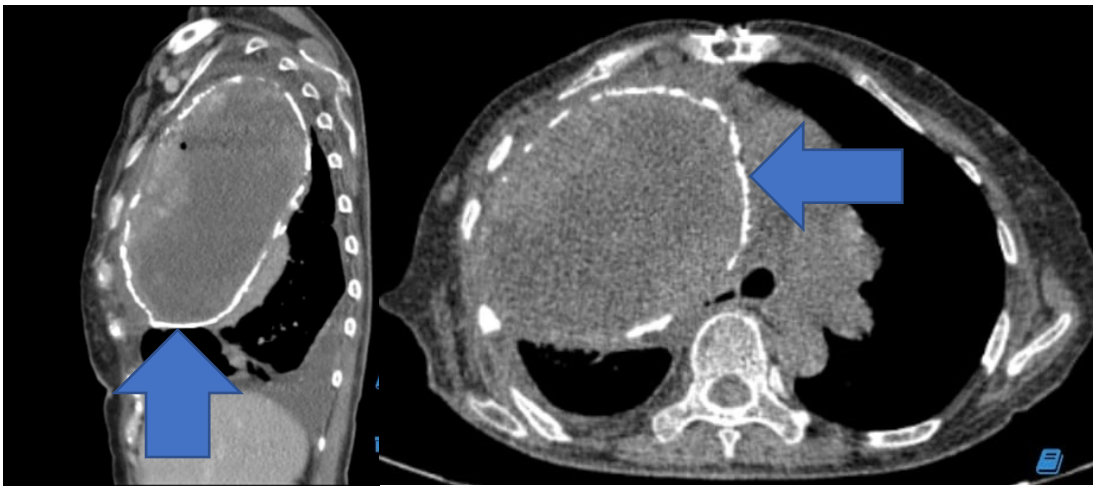
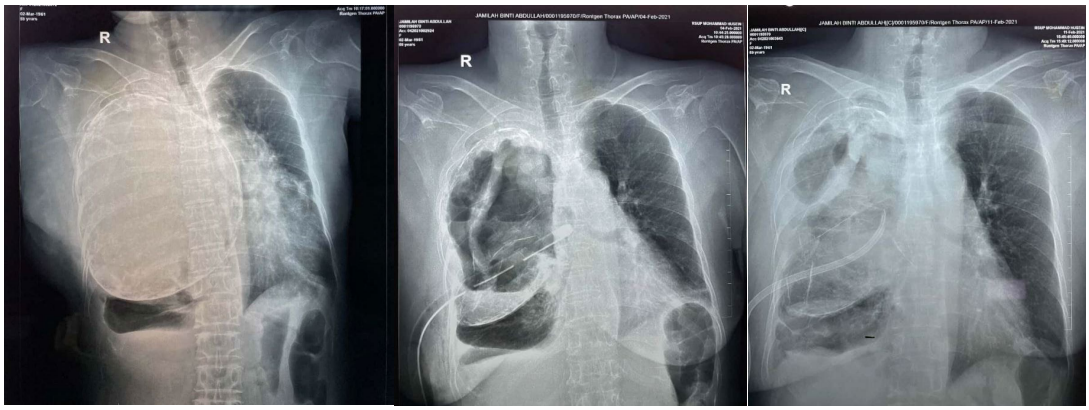


Figure 1. Thoracic CT scan showed a cystic lesion

Thoracic CT scan (Figure 1) revealed a cystic lesion measuring 14.2 x 18.5 cm with edge calcification thick around 3 mm and no contrast enhancement. We planned cyst resection by open thoracotomy based on clinical and CT scan findings. We collected 1,000 ccs of pus in the intraoperative setting for pus culture. The cyst was sticky on the lung parenchyma and pericardium, which led to its identification. Resection was performed, and a tissue sample was collected for histopathology diagnostics.



(A) (B) (C)

Figure 2. Chest X-ray before surgery (A), two days after surgery (B), and two weeks after surgery (C)

The cyst size was reduced dramatically, with little pleural effusion, two days after surgery. Two weeks after surgery, inhomogeneous lesions still existed. The patient's clinical condition improved with a respiratory rate of 18-20 breaths per minute and less frequent coughing. Pus culture was negative. However, pleural histopathology revealed that the cyst wall was made up of fibrocollagen with a red oval hydatid cyst/echinococcus, as seen in Figure 3. The laboratory result after surgery was within normal limits. The patient received oral albendazole/ABZ (400mg BID) for three months, paracetamol (500mg TID), and n-acetylcysteine (200mg TID). After three months, the patient had no complaints, only shortness of breath with heavy activity. On a polyclinic visit, the patient's vital signs were normal. A lung examination revealed a vesicular breath sound in the right lung. The patient can function normally.

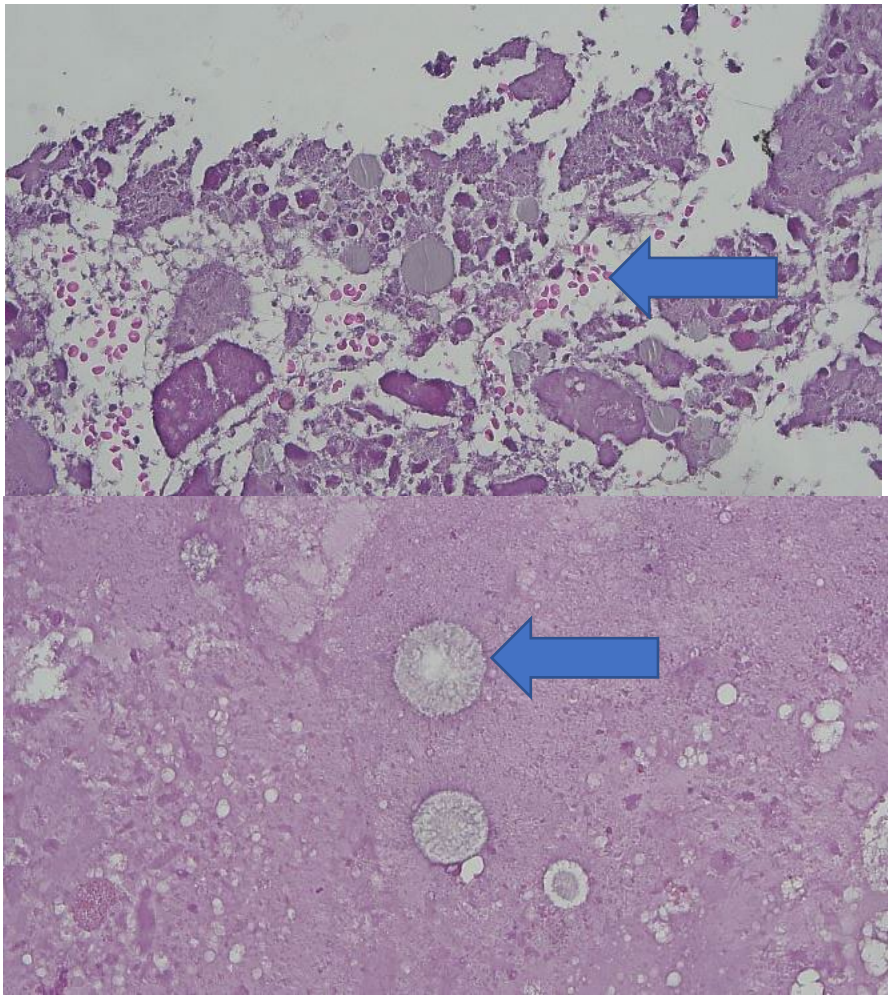


Figure 3. Echinococcus (blue arrow)

DISCUSSION

The most frequent abnormalities detected on CT images are lung cysts and cavities, making diagnosis difficult. The diagnostic procedure in such cases is influenced by the distinction between cysts and cavities (wall thickness >2 mm), focal or multifocal disease, and diffuse involvement. Cysts are spherical air pockets pathologically bordered by thin epithelium or fibrous tissue. Other radiological traits include size, inner wall shape, composition, and location.[2]

Bullae, blebs, pneumatoceles, infectious cysts, congenital cystic lesions, and infections like hydatidiform disease are the most common types of cystic air spaces that manifest as localized or multifocal lesions. Bullae are typically described as 1-mm-thick walls surrounding 1-cm-diameter air sacs within the lung. Most of the time, airway obstruction, high pressure at the lung apex, and congenital abnormalities are linked to blebs, typically seen near the height of the lung. Conversely, the cystic lung cavity known as a pneumatocele grows in size over a few days to several weeks (with trapped air) before eventually healing. Lung cysts can develop as a result of infections like hydatid disease.[1]

Thoracic CT scan with and without contrast in this patient revealed a hypodense (cystic) lesion measuring 14.2 x 18.5 cm with calcification at the periphery of the

right hemithorax with no contrast enhancement, which pushes the trachea and heart to the left, assumed to be a large pulmonary hydatid cyst or thoracic empyema. The CT scan expertise did not explain further the contour of the inner wall of the cyst or the nature of the cyst's contents. The cyst's location, whether in the lung or extra-pulmonary area, is also difficult to determine, especially if the cyst is large.

A zoonotic disease known as a hydatidiform cyst or human echinococcosis is brought on by the larval stage (metacestode) of parasites from the Taeniidae family genus *Echinococcus*. The most frequently damaged organs are the lungs and liver. The liver is the most common site for hydatidiform cysts (60–80% of cases), followed by the lung (10–30%), which is probably the most frequent place in children. Lung cysts are often single and unilateral, common in the right lung, especially in the lower lobe.[5,6]

The small intestine of the main host is where adult worms live. They have three proglottids, each containing many eggs, and two rows of hooklets in their scolex, which serve as their means of attachment to the intestinal mucosa. Dog feces produces eggs that adhere to grass or animal hair. If extensively dispersed, this egg can endure in the environment for at least a year. Adult tapeworms do not cause illness in their hosts. The intermediate host mistakenly swallows eggs from contaminated soil or grass or ingests water or food contaminated with infected dog feces. The eggs hatch in the small intestine and are then discharged. The embryo then passes through the intestinal wall to the visceral capillary layer, where it travels before developing into cystic metacestode, typically found in the liver and lungs. The parasite then develops into a cyst filled with fluid. Each protoscolices inside the cyst can develop into an adult worm once its definitive host has consumed it.

Cysts can be problematic for intermediate hosts because they can contain hundreds or thousands of protozoa (especially in humans). Secondary cysts, daughter cysts or mature worms, can be formed from protoscolices. While adult worms are found in definitive hosts when organs infected with protoscolices are consumed, daughter cysts are detected in intermediate host organs. Development into an adult worm in the intestine of the ideal host takes 4–7 weeks to complete its life cycle. Direct human-to-human transmission of echinococcus does not happen since it requires two different species of animals to end the life cycle. The life cycle is finished when the carnivore consumes a cyst on an intermediary host. The tapeworm larva can grow into an adult tapeworm, which eventually releases new eggs and continues the process. By consuming eggs from contaminated food, drink, or soil, humans can become intermediate hosts by contracting the disease through contact with affected dogs. Inhaling eggs can also result in primary lung illness, which eventually gives rise to fresh eggs and keeps the cycle going.[7]

In this case, the patient's chest X-ray showed the impression of a right lung cyst. By the literature, the right lung is the most common location. Thoracic CT scan with contrast was performed to confirm the diagnosis of a lung cyst. The thoracic CT scan result was a large pleural hydatid cyst, which supports the picture of hydatid cyst disease. Cysts more than 10 cm in size are also known as giant cysts. During surgery, the cyst was located in the pleural cavity, and the cyst wall had adhesions in the superior lobe lung and pericardium.

Lung involvement in hydatidiform illness occurs via several different routes. Ingestion of eggs combined with raw fruit, vegetables, and water can cause disease in humans, as can direct contact with feces or soil or through animal hair that contains worm eggs. After ingestion, it penetrates the stomach and intestines to aid the embryo's discharge. The source then uses its hook to pierce the intestinal wall and adhere to the jejunal and duodenal walls. The portal circulation is then used to transport it to the liver. However, embryos with a diameter of 0.3 mm or less can travel via the hepatic vein, sinusoid, and inferior vena cava into the right heart before eventually settling in the lungs. The majority of embryos are caught in the hepatic sinusoids. In addition, germs can also enter the lungs through other pathways like the thoracic duct, lymphatic channels, internal jugular vein, and right-side heart. The liver and lungs are primarily affected by hydatid larvae through this route. The liver's venous anastomosis is a third potential path. Direct lung exposure through breathing air infected with echinococcus eggs is another potential.[6]

The patient's history of gardening and farming without gloves was gleaned from the anamnesis. It is believed that dirt contaminated with worm eggs was the cause of the sickness. A hydatidiform cyst is a cystic, cavity-filling lesion brought on by the larva form of echinococcus. Three layers of hydatidiform fluid make up hydatidiform cysts. The pericyst, ectocyst, or adventitial layer is the outermost layer created by the host tissue as a defense against parasites. The laminated membrane, also known as the exocyst, is the following layer, and the germinative layer, also known as the endocyst, is the innermost layer.

The cyst's germ layer is the most active. The germinal layer produces an outer laminated membrane, a mother capsule, and inner protoscolices. The mother capsule, a fluid-filled vesicle-like structure containing protoscolices, is created by the germinal layer of the hydatidiform cyst. After ingesting the final host, protoscolices, dormant forms, develop into adults. The hydatidiform cyst's physical integrity is preserved by this

laminate layer of carbohydrates produced from the germ layer. It also serves to shield the germ layer from host immunological action. Hydatidiform cyst fluid is colorless, odorless, and sterile. This carbohydrate-rich laminate layer is formed from the germ layer. It maintains the physical integrity of the hydatidiform cyst and protects the germ layer from host immune attack.[8]

The organ's consistency and the surrounding tissue's suppleness affect how quickly a hydatidiform cyst grows. Because the lungs are softer than the liver, lung cysts grow more rapidly than liver cysts. It is also possible that negative pleural pressure can hasten cyst growth. Children's lung cysts enlarge more quickly than those of adults because of the more elastic nature of their lung tissue. The cyst's diameter increases at a fluctuating rate. According to Bloomfield, *et al*. (1966), it takes roughly 16–20 weeks.[9] Borrie, *et al*. (1962) reported that hydatidiform cysts could reach 1-2 cm in diameter in six months and up to 6 cm in one year.[10] Multiple cysts in the same patient may exhibit different growths. Romig, *et al*. (1986) reported that most (86%) of the cysts had an increase of 40% within one year.[11] Cysts can be static, collapse, or disappear in 34% of cases.

The clinical picture of a hydatidiform cyst depends on location, size, and complications. Small, uncomplicated cysts located peripherally are often asymptomatic and discovered incidentally on chest radiography. Unruptured cysts cause coughing, hemoptysis, or chest pain. Depending on the location and size of the cyst, small cysts may remain asymptomatic. Still, cysts that enlarge to 20 cm in diameter or more may produce symptoms by compressing adjacent structures. A mediastinal cyst that ruptures into adjacent structures can cause bone pain, bleeding, or airflow limitation. The hydatidiform disease causes many complaints in the lungs after cyst rupture. Cysts may rupture spontaneously or as a result of trauma or secondary infection. The patient suddenly developed chest pain, cough, fever, and hemoptysis after the cyst ruptured. The ruptured cyst into the bronchioles and the expelling of the germinative membrane or hooklet of the parasite are called hydatoptysis. Sometimes the patient feels a salty taste in the mouth.[6,7]

The patient's chief complaint was shortness of breath. This symptom has been felt from 2-3 years ago, but it was not so severe that it was considered a common complaint. The patient admitted that the shortness of breath was getting worse three months before admission, and shortness of breath was felt after activity. In addition to shortness of breath, the patient complained of a cough with phlegm, but no bloody cough, chest pain, or coughing that tasted salty in the mouth. Systemic complaints such as fever, symptoms of itching, and no visible bumps on the body indicate that the cyst was not ruptured.

The diagnosis of hydatidiform cysts is suspected in places where the condition is endemic. Radiology and serology are the two main diagnostic techniques utilized to confirm the diagnoses. Blood eosinophilia usually increases in 25% of infected individuals. Additionally noted were leukocytosis and an elevated erythrocyte sedimentation rate. In ruptured cysts, eosinophilia and peripheral blood leukocytosis are more frequent. However, these routine tests are general and have room for improvement.[3]

Naseer, *et al*. reported chest CT characteristics for hydatid cysts from 84 patients.[12] The characteristic is a hypodense cyst with a well-defined perceptible cyst wall with normal surrounding parenchyma (21.4%) or consolidation (34.6%). Radiological signs of hydatid cysts can be categorized as uncomplicated or difficult, depending on whether there are problems such as rupture, complete rupture, or superinfection.

A simple cyst looks like a homogeneous lesion with smooth, variable-thickness margins. Density measurements on CT showed a low Housefield Unit (HU) by the fluid image. Cysts toward the periphery might be oval or polycystic, but cysts in the center are round. A ruptured or infected cyst is called a complicated cyst. Hydatid cyst rupture can happen in up to 47.5% of cases. As a result of the presence of mucus, an infection, or hemorrhagic material, complicated cysts exhibit higher HU values.[3]

Complete or contaminated cyst ruptures fall within the category of ruptured cysts. The separation of the pericyst from the endocyst was cited as a sign of contamination. A confined break brought on by an allergy or infection is unlikely to occur since pericytes encircle the cyst's contents. An air crescent sign (meniscus sign), an inverted crescent sign, and an air bubble sign are imaging indicators of a polluted rupture. The rupture is considered complete when there are indications of a bronchus link. Superinfection is the most frequent complication of a ruptured hydatid cyst. Superinfected hydatid cysts may exhibit high HU values and be challenging to identify from pyogenic abscesses. The radiologic indicators for an infected cyst are the ring enhancement sign, air bubble sign, and air-fluid level.[3]

A CT scan with contrast may reveal a thin, elevated edge if the cyst is intact. The cyst's contents are homogeneous and have a density similar to water. The cystic form of the lung mass can be explained by CT scans, which can give a precise location for organizing the surgical removal of complex cysts. A hydatid cyst's magnetic resonance properties can vary depending on its developmental stage, whether it is uni- or multilocular,

and if it is living, infected, or dead. It also details the severity of the mother and daughter cysts and response alterations in the host tissue and capsule.[7]

Chest X-ray in this case revealed a homogeneous radiopaque image with a rounded shape. It can be concluded that the type of cyst in the patient was an uncomplicated cyst. The IgG enzyme-linked immunosorbent assay (ELISA) is the most precise serological test. It takes almost five years after surgery for a specific IgG ELISA to turn negative. To confirm seroreactivity, immunoglobulin tests utilizing specific echinococcal antigens may be helpful. However, these tests are not routinely accessible.[13]

The echinococcus IgE test must be performed to confirm the diagnosis of a hydatid cyst. Due to the lack of a test facility, this test could not be performed for the patient. The diagnosis was only made based on radiological results and the results of anatomical pathology leading to hydatid cysts, which were then treated with empiric therapy, namely ABZ.

Surgery is the first option to treat hydatid cysts, followed by antiparasitic drug therapy. The most popular surgical treatments for pulmonary hydatid disease are thoracotomy and cyst resection. A median sternotomy, however, is preferred when the illness is bilaterally present.[14] The main goal of surgery is the complete removal of the germinative membrane after inactivation using a hypertonic saline solution to prevent local contamination.[15] If this is not performed carefully, there is a much higher likelihood of a reoccurring illness.[16] However, capitonnage is not required following cystectomy. Only the bronchial apertures should be sutured, and the cavity should be left free because capitonnage produces lung deformity, lengthens the operating time, and increases morbidity.[17,18] The surgical procedure and patient management for primary pleural hydatidosis follow the same guidelines. Chest drainage and ABZ therapy must be administered after the cysts, and pleural fluid has been inactivated with hypertonic saline solution.[19]

Surgery indications are large cysts, infected cysts attached in vital locations, and cysts that have a substantial effect. Surgery is the preferred treatment because the parasite can be removed completely without spilling its contents, and the patient can be cured. During surgery, it is very important to minimize spillage of cyst contents to prevent intraoperative spread and recurrence. For liver cysts, aspiration with helminticide injection and re-aspiration have been recommended. It is not applicable for lung cysts because they cause many complications and are rarely indicated.[6] The patient performed a cystectomy followed by hypertonic saline irrigation.

Medical therapy for pulmonary hydatidiform cysts includes benzimidazole drugs such as ABZ or mebendazole (MBZ). Medical treatment includes smaller cysts, patients with contraindications to surgery (poor surgical risk, refusal of surgery, and multi-organ disease), multiple cysts, recurrent cysts, and patients with hydatid fluid spills during surgery. Therapy should generally be started on day 4. ABZ is preferred due to better bioavailability and fewer adverse effects than MBZ. However, antiparasitic treatment is recommended as anti-infection therapy, while the main course of treatment remains surgical.[20]

ABZ also achieves higher plasma and intracystic drug concentrations. Plasma concentrations are 10–40 times higher than those of MBZ. The usual recommended dose for ABZ is 10–15 mg/kg/day, taken twice daily, and for MBZ, it is 40–50 mg/kg/day, taken three times daily. The optimal duration of medication in pulmonary hydatidosis is unknown, but it should be taken for at least 3-6 months. The drug is given in 14-day intervals over a month to avoid hepatotoxicity. However, continuous therapy is more effective than interval therapy without increasing side effects. Pharmacological contraindications are a large cyst that may rupture, spinal depression, and pregnancy, especially in the first trimester.[6]

It is challenging to assess the efficacy of therapy, which typically calls for ongoing follow-up and imaging. It has also been evaluated whether serological titers should be used to track treatment. In most patients, during the first three months following surgery, serological tests reveal higher titers, which may be related to antigen liberation during cyst management. Three months following surgery, patients who had no recurrence demonstrated a drop in antibody titers according to serological tests. Early or late relapses are characterized by persistently high antibody titers or early decreases and subsequent rises. No serological testing has consistently been reliable for keeping track of individuals receiving treatment for hydatid disease.[7]

ABZ medication should be administered to all surgical patients for three to six months to avoid disease recurrence. Without anti-helminthic therapy, the surgical hydatid cyst recurrence rate is significant (11%). Following surgery, follow-up chest X-rays should be performed every three months for up to a year afterwards. For the first three months, the liver function and chest X-rays should be taken every month.[6]

CONCLUSION

The larva stage of echinococcus causes a giant pleural hydatid cyst. This disease was diagnosed according to a chest CT scan which revealed a smooth and thick hypodense (cystic) lesion measuring 14.2 x 18.5 cm with calcification at the periphery of the right hemithorax with no contrast enhancement. Pleural histopathology revealed that the cyst wall comprised of fibrocollagen with a red oval hydatid echinococcus. The first option for treatment of this disease was surgery followed by medical therapy, such as ABZ 400 mg BID for three months. It is important to recognize this case to manage the patient appropriately. Improper diagnosis and treatment can harm the patient.

Consent

Written informed consent was obtained from the patient.

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Conflict of Interest

The authors declare there is no conflict of interest.

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Author's Contributions

Writing the manuscript and collecting data of the patient: SS, AF and SB. Reviewing and revising: ZA, AR, LA, RP, and TS. All authors contributed and approved the final version of the manuscript.

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