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Retrospective Analysis of Our Pulmonary Hydatid Cyst Cases

Pulmoner Kist Hidatik Vakalarımızın Retrospektif Analizi

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ÖZ

Amaç: Kliniğimizde akciğer hidatik kisti (AHK) tanısı ile takip ve tedavisi yapılan hastaların verilerini retrospektif olarak değerlendirmek.
Gereçler ve Yöntem: Kasım 2010- Kasım 2020 tarihleri arasında kliniğimizde AHK tanısı ile tedavi edilmiş olan 83 hasta; yaş, cinsiyet, kist lokalizasyonu, boyutu, kistin başvuru esnasında rüptüre olup olmadığı,başvuru sırasındaki semptomları, karaciğer kisti birlikteliği, postoperatif yatış süreleri açılarından değerlendirildi.
Bulgular: Hastalarımızın 33'ü erkek , 50 tanesi kadındı. Hastalarımızın yaş ortalaması 34,71 idi. Hastalarımızdan toplamda 109 kist çıkarıldı, bunlardan 9 tanesi dev kist olarak değerlendirildi. Sağ üst lob da 14, sağ orta lob da11, sağ alt lob da 37, sol üst lob da 16, sol alt lob da 31 kist tespit edildi. 9 hastada bilateral kist mevcuttu. Kistlerin 52 tanesi perfore, 57 tanesi intact idi. En yaygın semptom öksürüktü. 16 hasta ise asemptomatikti.
Sonuç: AHK'i vücudumuzda herhangi bir organı tutabilen zoonotik bir hastalıktır. Karaciğer kist hidatiğinden sonra ikinci sıklıkta görülmektedir. Akciğerin yapısından dolayı kolay büyüme eğilimindedir. Tanı da radyoloji çoğunlukla yeterli olmakta ve AHK'lerin tedavisinde cerrahi ön plandadır.

Anahtar Kelimeler: Echinococcus, Hidatik kist, Akciğer

ABSTRACT

Aim: To retrospectively evaluate the data of patients diagnosed with pulmonary hydatid cyst (PHC) who were followed up and treated in our clinic.
Materials and Method: 83 patients treated with the diagnosis of PCO in our clinic between November 2010 and November 2020; Factors such as age, gender, location and size of the patient, whether the cyst was ruptured at the time of admission, symptoms at the time of admission, relationship with liver cysts, and postoperative hospital stay were evaluated.
Results: 33 of our patients were male, 50 of them were female. The mean age of our patients was 34.71. A total of 109 cysts were removed from our patients, 9 of them were evaluated as giant cysts. There were 14 in the right upper lobe, 11 in the right middle lobe, 37 in the right lower lobe, 16 in the left upper lobe, and 31 in the left lower lobe. Nine patients had bilateral cysts. 52 of the cysts were perforated and 57 were intact. The most common symptom was cough. 16 patients were asymptomatic.
Conclusion: PHC is a zoonotic disease that can affect any organ in our body. It is the second most common after liver hydatid cyst. It tends to grow easily due to the structure of the lung. In diagnosis, radiology is mostly sufficient and surgery is at the forefront in the treatment of AHC

Key words: Echinococcus, Hydatid cyst, Lung

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INTRODUCTION

Reportedly originating from the dogs of Icelandic fishers, hydatid cyst (HC) infection was first described by Thebesius in the 17th century(1). Common in the Mediterranean, the Middle East, and South America where animal breeding is intensive, this parasitic disease is also common in Turkey (2). Larva forms of Echinococcus granulosus taeniae cause a parasitic infection in humans. Echinococcus Multilocularis, Echinococcus Oligarthrus and Echinococcus Vogeli are other Echinococcus with potential to infect humans (3).

Most of the embryos cause parasitic infection most commonly in liver as they are hooked in liver sinusoids. However, embryos smaller than 0.3 mm in diameter can pass through hepatic sinusoids, make their way to the right heart through the hepatic vein and inferior vena cava (IVC), and from here they can settle in lungs (the second most common) where they cause parasitic infection (4). Pulmonary hydatid cyst (PHC), which is separated from liver HCs because of the high elasticity of the lung tissue and the negative pressure effect on the thorax, is a parasitic disease that is still relevant in Turkey(5).

MATERIALS AND METHOD

Based on the permission granted by the Ethics Committee of Necmettin Erbakan University's Meram Faculty of Medicine, we analyzed the data of the patients who sought medical aid in our clinic between November 2010 and November 2020 and were diagnosed with PHC based on an archival review. The age, gender, cyst location and size of 83 patients treated upon the diagnosis of PHC, and whether the cyst was ruptured at the time of admission, their symptoms at the time of admission, association of liver cysts, and postoperative length of stay were evaluated.

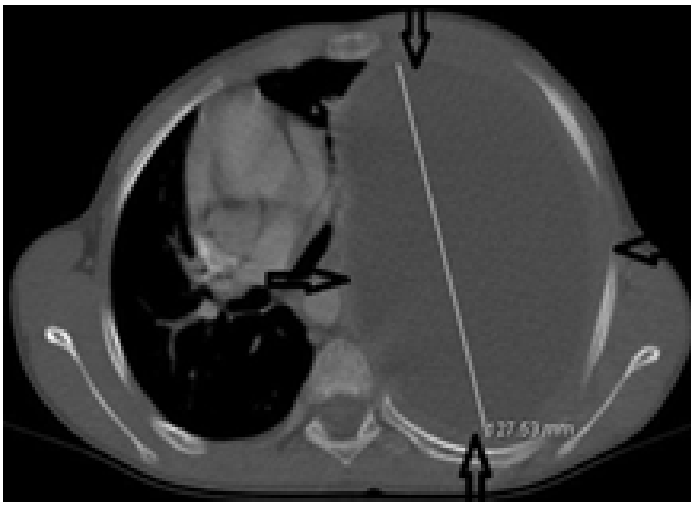


Figure 1. Intact giant cyst

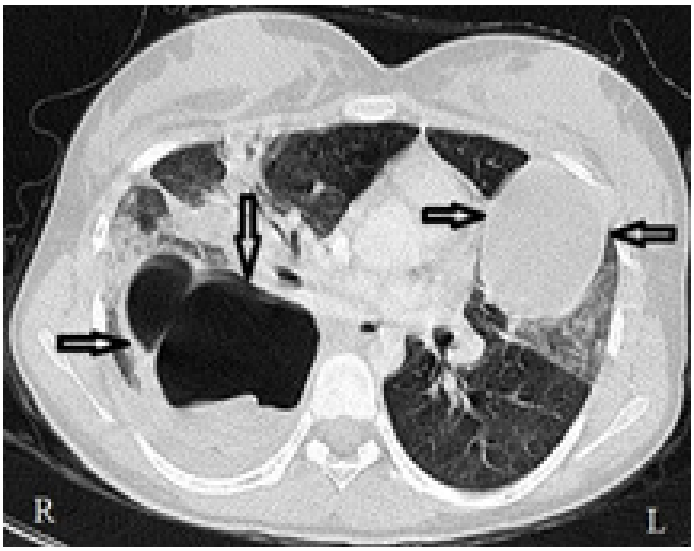


Figure 2. Bilateral cysts (ruptured cyst in right hemithorax, intact cyst in left hemithorax)

RESULTS

33 of our patients were male (39%), 50 of them were female (41%). The youngest was 5 years old, the oldest was 83 years old and the mean age was 34.71 ± 18.35. A total of 109 cysts were removed from our patients, 9 of them (08%) were accepted as giant cysts (Figure 1). Cyst sizes ranged from 1.5 to 14 cm, and the mean AHC diameter was 5.14 ± 8.02 cm. There were 14 cysts (13%) in the right upper lobe, 11 cysts (9%) in the right middle lobe, 37 cysts (34%) in the right lower lobe,

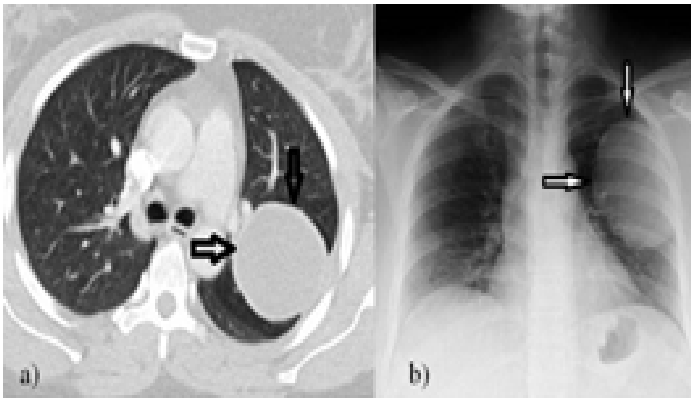


Figure 3. Unruptured intact PHC is seen as a smooth-edged, round or oval homogeneous opacity on its radiography. a) Thorax computed tomography of PHC, b) Postaeroanterior chest radiography of PHC

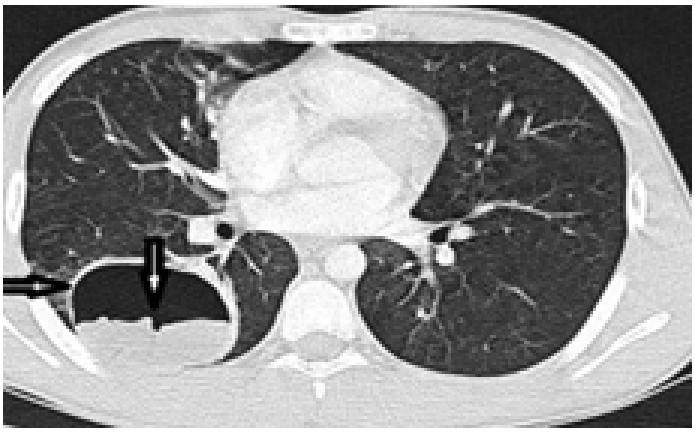


Figure 4. The appearance called water lily sign or lotus flower sign indicates that the AHK is ruptured. It is caused by the appearance of air-fluid level and the floating of the collapsed cyst membrane on the liquid.

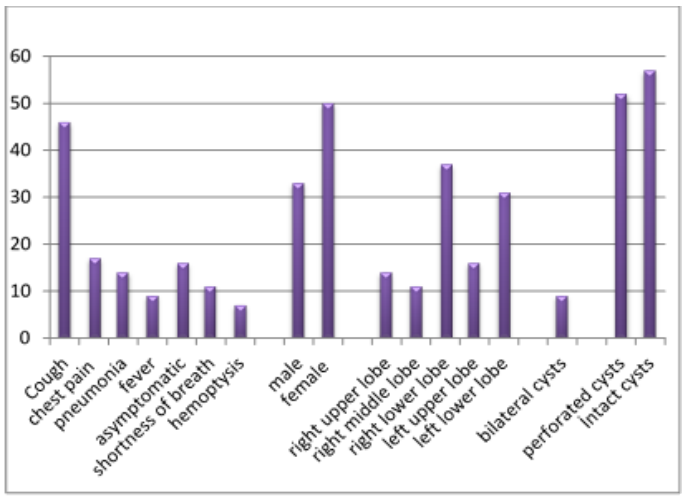
16 cysts (15%) in the left upper lobe and 31 cysts (29%) in the left lower lobe. Nine patients (10%) had bilateral cysts (Figure 2). 52 (47%) of the cysts were perforated and 57 (53%) were intact. Cough in 46 patients (46%), chest pain in 17 patients (20%), pneumonia in 14 patients (16%), fever in 9 patients (10%), shortness of breath in 11 patients (13%), hemoptysis in 7 patients (08%). 16 patients were asymptomatic. (Table 1)

Statistical analysis was performed using PASW for Windows version 17.0 software (SPSS Inc., Chicago, IL, USA). Descriptive data were presented in mean ± standard deviation, median (min-max) or number and frequency.

DISCUSSION

HC, also known as Echinococcosis / Hydatidosisis, is a parasitic disease that can be seen in all countries. It could be endemic in Mediterranean countries including Turkey, and the Middle East and South America, and countries that live on animal breeding(2).

Table 1. Pulmonary hydatid cyst (PHC) datas



Echinococcus is a parasite of the cestod class. There are 4 forms that can cause a disease in humans: Echinococcus Granulosus, Echinococcus Multilocularis, Echinococcus Oligarthrus and Echinococcus Vogeli (6). The most common is Echinococcus granulosus. Although the parasite has different genotypes, the main structure of HC is the same in all genotypes(6,7).

It has a cystic wall made of three layers.

1) Pericystic membrane: It is also known as ectocyst. This membrane actually emerges as a result of an inflammatory response of the host to the cyst. It has a structure that completely pertains to the host.

2) Laminar membrane: It is located between the pericystic layer and the germinative layer.

3) Germinative membrane: It is the live part where protoscolex (hydatid sand) and daughter vesicles are located.

Echinococcus genus need 2 different hosts to complete their life cycle. While the adult form of the cyst settles in the intestines of animals such as wolves, dogs, and foxes, which are billed as main hosts, the larval form settles in animals such as sheep, humans and cattle, causing HC disease (7,8). HC disease is more common in places living on farming and animal breeding where there is poor attention to cleanliness and preventive medicine measures are insufficient. There are publications reporting that the cyst, which is transmitted to humans by oral ingestion of cyst eggs through food and beverages contaminated with the feces of infected animals, can be transmitted to the fetus by respiration and transplacental route(9). Larvae that make their way to the intestinal wall through digestion adhere to the intestinal wall with their suction cups. Being part of the bloodstream, they can reach the liver where they cause the disease most commonly (75%). Lungs are the second most common organs that HC settles in (15%) (5).

The cyst makes it to the lungs by a few routes. The hematogenous route is one of them. Most of the HC embryos that reach the liver through the portal circulation are placed in liver sinusoids. However, embryos smaller than 0.3 mm settle in the right heart and then finally in the lungs through the hepatic vein and vena cava inferior without being stuck in the hepatic sinusoids (4). Through the lymphatics of the intestine, which is the second route, the larvae bypass the liver and enter the jugular vein and the right side of the heart and from there into the lungs (10). In addition, the lungs can be infected through the transdiaphragmatic route and rarely through inhalation (5). PHC is more common in children than liver HC (5,9,11). Hydatid cyst disease can be seen in other organs of the body apart from lungs and liver (9,12).

PHCs differ from liver HC in terms of some characteristics. Because of the elastic structure of the lung, cysts that settle here grow faster than cysts in the liver. Their average annual growth rate is nearly 1 to 50 mm. a year (5). HC can become larger in lungs. Although PHCs larger than 10 cm. in diameter are defined as giant HCs, there are publications in the literature that refer to HCs that cover more than 50% of one lobe of the lung as giant HCs instead of a standard size that would define giant HCs. (2,13). In our study, we defined PHCs with a diameter larger than 10 cm. as giant cysts. Of all 109 cysts removed, 9 (8%) of them were giant cysts. As long as PHC is not ruptured and it is located peripherally, it can be asymptomatic. Clinical symptoms are usually caused by compression of the surrounding tissues in non-ruptured cysts. Our 16 patients (19%) were incidentally identified and they were asymptomatic. The cyst was ruptured in none of them. In literature, it is reported that PHCs are seen more commonly in the lower lobes and can be bilateral at a rate of 20% (9). Similar to the literature, 51% of PHCs were detected in the lower lobes in our study. The rate of bilateral PHC in our study was 10%. Cysts larger than 5 cm. in diameter can often cause symptoms related to bronchial compression (4). In rare cases, it is reported that HC causes mediastinal shift and related symptoms as a result of pressure on the mediastinum (5). Cough is the most common symptom due to cyst compression on the bronchus or irritation of the cyst contents due to cyst rupture (5,13). In our study, the most common symptom was cough (46%).

Cyst perforation is the most common complication for PHCs An increase in intracystic pressure and spontaneous rupture may develop as a result of continuous production of PHC fluid (14). As a result of both pericyst and endocyst rupture, the cyst content is poured into the pleural cavities. PHC can be ruptured into the pleural space or into the intrabronchial area. The perforation into the intrabronchial area may result in pneumonia. 14 of our patients (16%) had pneumonia and all of them had perforated PHC. In literature, it is reported that perforation rates range from 24.7% to 61% (5). The perforation rate was 47% in our study. Peripherally located PHCs are more prone to rupture due to the weaker pericyst tissue and poorer lung tissue support. (4) A PHC rupture may result in cough, hemoptysis, sudden onset chest pain, fever, pneumothorax, hydropneumothorax, empyema, and secondary infection (4,5). Another feared and important characteristic of rupture is the development of hypersensitive reactions ranging from urticaria to anaphylaxis (4,15). No case of anaphylaxis was found in our study . In 7 cases, an allergic reaction in the form of urticaria was encountered and the symptoms were resolved in a short period of time. Hemoptysis can be caused by pressure erosion in the bronchus induced by PHC, and secondary bronchial infection or rupture of the cyst. In PHC, hemoptysis is more common in adults than in pediatric population (4,5). The rate of hemoptysis was 8% in our study (4,5).

Anamnesis is the first step of any diagnosis. One should suspect HC if people who live on farming and have a history of dog feeding reside in an HC-endemic area. Radiological and serological tests are the methods adopted to diagnose. Peripheral blood eosinophilia, leukocytosis and an increased sedimentation rate can be seen when the cyst is ruptured. However, all these findings are non-specific (16). Although Casoni and Weinberg tests are used for diagnosis, their diagnostic value is nearly 52% (17). It is reported that the Casoni and Weinberg tests can remain positive for 2 to 5 years after an PHC operation (17). The rate of seropositivity is higher in liver HCs compared to PHC. The precision of serological test ranges from 85% to 98% in hepatic cysts and from 50 to 60% in pulmonary cysts (5). Posteroanterior lung (PA Ac) radiography and thorax computed tomography (CT) are the most common methods for diagnosis (4,6). In literature, there are studies reporting that it can be diagnosed by PA AC radiography in over 90% of the cases (17). It can also be diagnosed for nearly 100% of the cases with CT (13). In our clinic, we primarily used PA X-ray and Thorax CT. Unlike hepatic HC, the role of ultrasound for AC HC is limited except in cases where lesions are close to the chest wall (4).

The radiological image varies by whether the cysts are ruptured or not. Intact and non-ruptured PHC looks like as a smooth-edged, round or oval homogeneous opacity in radiography (Figure 3). Unlike extrapulmonary cysts, pulmonary hydatid cysts do not become calcific and the formation of daughter vesicles is rare (18). When air enters between the pericyst and the membrane, a crescent-like image appears on the cyst. This image, which is called the crescent or Moon sign, is considered to be a sign that the cyst will rupture (9). The image, which is also called the water lily sign or the lotus flower sign, indicates that the PHC is ruptured. This is caused by the image of air-fluid level and the floating of the collapsed cyst membrane on the liquid (19) (Figure 4). Surgery is the main treatment of PHC. Oral treatment administered with albendazole or mebendazole has not been entirely successful (4,6,20).Medical treatment is recommended for patients who cannot tolerate surgery, whose heart is unstable, have a multi-organ disease, have too many cysts to be removed, and have contraindications for surgery. Patients with a intraoperative transmission risk caused by the rupture of the cyst during the operation are also included in the group with chemotherapy indication (20). In case of ruptured PHC, the operation should be immediately performed because of the risk of complications such as anaphylactic shock and secondary infection as mentioned above.

As a surgical procedure, approaches that spare lung parenchyma should be preferred. Cystotomy and capitonage are the most common methods as parenchymal sparing surgery techniques (21,22). Resections such as lobectomy and segmentectomy are not preferred because they will cause irreversible loss of lung tissue. The reinfection rate is high in countries such as Turkey where the disease is common. Patients may need to undergo multiple operations. Recurrent loss of pulmonary tissues in each operation can lead to serious complications (5).Segmentectomy or lobectomy in PHCs is considered indicated only if there is severe damage to the lung parenchyma and destruction of more than 60% of the lobe, and if the lung parenchyma is not recoverable as a result of suppurating cyst cavities (11,23). Another lobectomy indication is the presence of multiple cysts that cannot be removed from the same lobe (21). For patients with bilateral PHC, bilateral simultaneous thoracotomy, sequential thoracotomy over an intermittent period of time, median sternotomy and Clamshell incision are the surgical options to choose from. Except for patients with normal cardiopulmonary reserve, and elderly and pediatric patients who can tolerate the operation, there are publications that recommend bilateral simultaneous thoracotomy in a single session (1). There is a risk of life-threatening complications that may develop in both lungs following bilateral thoracotomy in the same session. In addition, some studies report that one-stage sequential thoracotomy should be preferred over bilateral thoracotomy. Because liver and lungs have different tissue characteristics, percutaneous treatment for the liver is not suitable for lungs. While the liver has a solid structure, the lungs are soft and full of air. Percutaneous treatment for the lungs can lead to infection with communication from a residual bronchial cyst ¬(25). We primarily prefer surgical treatment for our patients with PHC. Cystotomy and capitonage are the most common methods we adopt as a parenchymal sparing surgery technique. We preferred to perform consecutive thoracotomy at 4-week intervals in our cases with bilateral PHC since we thought the complication rate was lower. We attached priority to the side that was not ruptured, and was large in size, and more in number. We operated the other side in a second session 4 weeks later. We recommend Albendazole treatment for patients who have ruptured PHC before or during the operation in order to minimize the risk of recurrence, or if we cannot operate for various reasons, we recommend 2 cycles of 10-15 mg / kg a day in two doses at 10-day intervals.

CONCLUSION

PHC is a zoonotic disease that can involve any organ in an individual's body. As its tissues serve a filter, it is the second most common disease behind liver hydatid cyst. The elastic structure of the lungs causes PHCs to easily grow. Cough and chest pain are the most common symptoms in patients with PHC who may be clinically asymptomatic. Radiological imaging (PA-AC radiography, chest CT) and serological tests are used for diagnosis. Radiological imaging suffices in most cases. Surgery is the leading mode of treatment for PHCs. Medical treatment is preferred for patients who cannot be operated and refuse surgical treatment.

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