



Tehran University of Medical
Sciences Publication
<http://tums.ac.ir>

Iran J Parasitol

Open access Journal at
<http://ijpa.tums.ac.ir>



Iranian Society of Parasitology
<http://isp.tums.ac.ir>

Short Communication

Concomitant Occurrence of Hepatopulmonary hydatid Cysts in Turkey

Emel Türk Arıbaş¹, *Bayram Metin², Ahmet Dumanlı³, Olgun Kadir Arıbaş⁴

1. Department of Clinical Microbiology Infectious Diseases, Medical Faculty, Ufuk University, Ankara, Turkey
2. Department of Thoracic Surgery, Special Acıbadem Kayseri Hospital, Kayseri, Turkey
3. Department of Thoracic Surgery, Medical Faculty, Afyon Kocatepe University, Afyon, Turkey
4. Department of Thoracic Surgery, Medical Faculty, Gazi University, Ankara, Turkey

Received 09 Jan 2021
Accepted 11 Apr 2021

Keywords:
Hydatid cyst;
Hepatopulmonary local-
ization;
Surgery;
Turkey

***Correspondence**
Email:
drbaymet@hotmail.com

Abstract

Background: We aimed to report the demographic characteristics with diagnosis and treatment methods in patients with concomitant hepatopulmonary hydatid cysts.

Methods: Over a ten-year period (from 2002–2020) in Konya, Turkey, surgery was performed on 52 patients with hepatopulmonary hydatid cyst. Main outcome measure(s) were 52 hydatid cysts patients, which had cysts both in the liver and lungs, were investigated regarding their age, gender, cyst localization, suppuration, symptoms, and treatment methods.

Results: Seventeen of the patients were males. Their mean age was 39.7 ± 18.8 years. The most common occupation was housewifery. The most common symptom was coughing and none of the patients with concomitant hepatopulmonary hydatid cysts were asymptomatic. The pulmonary hydatid cysts were mostly encountered in the right lung and the majority of the hepatic hydatid cysts were observed in the right lobe. The mean hospitalization time of the operated patients was 17.12 ± 6.7 days.

Conclusion: In patients with hydatid cysts localized concomitantly in the right lung and subdiaphragmatic area, right thoracotomy for the pulmonary cyst and a transdiaphragmatic approach for the hepatic cyst is a safe, effective, and comfortable method.



Introduction

H ydatid cysts, caused by the larvae of the *Echinococcus granulosus*, are common in Africa, Mediterranean countries, Middle Asia and the Middle East, and is an important health problem (1). The incidence of hydatid cyst is 2/100,000 in Turkey (2). Hydatid cyst is a zoonotic infection caused by the larvae (metacestodes) of the parasite. The adult forms of this parasite live in the ileum of dogs and other carnivores. The humans are infected with the consumption of food contaminated with the eggs of the parasite, excreted in the gaita of the dogs.

In humans, the cysts are mostly localized in the liver, lungs, brain, heart, bones, muscles, kidneys, spleen, and other organs respectively. The symptoms depend on the involved organ(s), the size of the cyst and the suppuration (1, 2). Our objective was to evaluate the clinical, radiological, and therapeutic characteristics of the patients with concomitant occurrence of hepatopulmonary hydatid cysts.

Material and Methods

Patients, who had concomitant hepatopulmonary hydatid cysts and treated in the Thoracic Surgery Clinics of the Meram Medical School in Konya Necmettin Erbakan University, Gazi University Medical School and Special Kayseri Acibadem Hospital between 2002 and 2020, were investigated in this retrospective study.

All the participants were fully informed about the aims of the study and permission was obtained from patients, where appropriate. University Ethics Committee gave approval for the study to take place at 2020. This study was conducted according to the Declaration of Helsinki as revised in 2000.

Overall, 52 patients, hospitalized in our clinics, were evaluated in respect of their age, gender, cyst localization, suppuration, symptoms, and therapeutic methods. The patients

are first diagnosed with the detection of the hydatid cysts in the postero-anterior x-ray images. Then a thoracic CT examination was performed for a detailed evaluation of the cystic lesions. The abdominal US was used for the abdominal examination and CT examination was only performed if a detailed evaluation was required. In some complex cases, not definitively diagnosed with these methods, an MR examination was added to the process (Fig. 1).



Fig. 1: CT images of a patient with concomitant hepatopulmonary hydatid cysts

Patients with neurological symptoms underwent also MR examination. Preoperative whole blood, biochemical, blood group analyses, hepatitis marker, coagulation parameter, ECG, respiratory function tests, and hydatid cyst ELISA tests were performed for all patients. Preoperative evaluation for anesthesia was also done. Double-lumen selective intubation was performed in all patients in order to

deflate the ipsilateral lung and to prevent the contamination of the contralateral lung with cyst fluid. Besides a patient, who underwent sternotomy due to the bilateral pulmonary localization of the hydatid cysts, all patients underwent thoracotomy. Phrenotomy was performed in addition to the thoracotomy in 13 patients, who had hepatic hydatid cyst concomitant with hydatid cyst in the right lung. During the operation, in both hepatic and pulmonary cysts, intact cysts were surrounded with gauzes impregnated with a hypertonic saline solution to prevent the spread of the parasite to the neighbouring tissues. The cyst content was aspirated with a suitable needle. Afterwards, cystotomy was performed with an incision at the site of the needle entrance. Following the aspiration of the residual fluid in the cyst, the cyst membrane was removed with the help of an over-clamp. Then, the cyst cavity is irrigated with a 1% povidone-iodine solution in order to eliminate the remaining alive parasites. After the irrigation process, the lung was inflated at the cyst area and the site of the bronchial leakage was detected and closed with 2/0 absorbable sutures. The capitonnage of the cyst cavity was done with quilting sutures placed at 1-2cm intervals. After a radial phrenotomy, performed according to the localization of the hepatic cyst, the fluid in the cyst was aspirated and the cyst membrane was removed (Fig. 2).

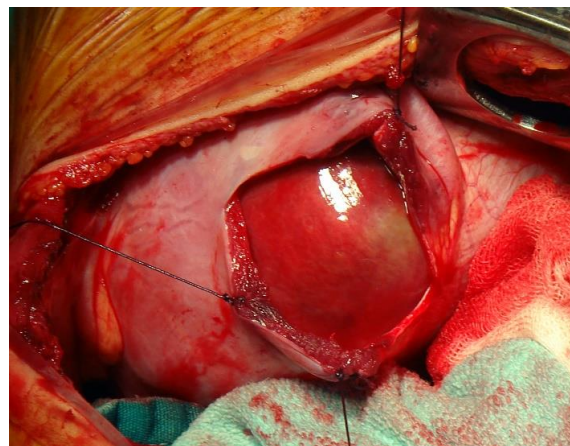


Fig. 2: Operation site of a patient with right thoracotomy and phrenotomy

Then the bile leakage was checked, and the leaking sites were sutured and supported with the omentum in necessary cases. The phrenotomy area was closed with #1 suture after the placement of an abdominal drain for the drainage of the subdiaphragmatic region. Then, the thoracotomy site was closed following the placement of two thoracic drains in the chest cavity. Postoperative complications were encountered only in 8 patients. The mean postoperative hospitalization time was 17.12 ± 6.7 days.

Results

Demographic distribution of the patients according to age, gender and occupational distribution was examined (Table 1).

Table 1: Demographic distribution of the patients

Age	8-72 (39.7±18.8)
Gender, Age by Gender	Males: 17 (32.69), Mean Age: 30.6±17.2 Females: 35 (67.31), 40.7±15.8
Jobs of Patients	Housewives: 61.53% Students: 12.9% Pensioners: 9.92% Shopkeepers: 7.69% Workers: 7.96%

The most common symptoms of the patients were cough (70.0%), flank pain (63.46%), sputum (46.15%) and these symptoms are followed by fever (30.48%), haemoptysis (23.07%), dyspnea (23.07%), nausea (19.23%), salty water expectoration (9.51%), and cyst membrane expectoration (1.90%). Interestingly, all patients with concomitant hepatopulmonary cysts were symptomatic. Regarding the pulmonary localization of the hydatid cyst, 38 patients (73.08%) had unilateral and 14 patients (26.92%) had bilateral localization. Overall, 31 of the unilaterally localized cysts (59.61%) were in the right lung and 7 (13.46%) were in the left lung. In 19 patients (36.53%) the cysts both in the liver and lungs were solitary lesions. 6 patients (11.53%) had a solid pulmonary cyst and multiple hepatic cysts; 11 patients (21.15%) had multiple pulmonary cysts and a solid hepatic cyst; 13 patients (25%) had multiple cysts both in the liver and lungs. Thirty six of the hepatic cysts (69.23%) were localized in the right lobe, 8 (15.38%) in the left lobe and the remaining 8 (15.38%) cysts were localized in both left and right lobes.

The mean diameters of the pulmonary and hepatic cysts were 5.2 ± 2.5 cm and 7.0 ± 3.0 cm respectively. Thus, we could demonstrate that the size of the hepatic cysts was larger than the pulmonary cysts. According to the findings of the surgery and the radiological imaging, 40.38% of the cysts are alive and 59.62% are perforated.

Discussion

In humans, 66% of the hydatid cysts are localized in the liver, 5%-15% in the lungs, and less than 2% in the spleen. Localization in other organs is rather rare. Although the hepatic involvement is more common in adults, pulmonary involvement is more frequently encountered in children. The rate of the concomitant hepatopulmonary hydatid cysts is between 4%-25%. As the hydatid cyst embry-

os can hardly pass through the filtering system of the hepatic and pulmonary capillaries, the rate of hydatid cysts in other organs is very low (1-3). Humans, who are the main host of the parasite, are infected through the gastrointestinal and respiratory tract (4, 5). Furthermore, the parasite might arrive in the lower lobe of the right lung through the transdiaphragmatic path after the traumatic rupture of a hepatic cyst and as a result of a congenital malformation, hepatobiliary surgery and, biliary obstruction (5,6). In our patients, we observed that most of the pulmonary cysts were localized in the lower lobe of the right lung, which supports transthoracic contamination.

In patients with hydatid cyst, the symptoms depend on the size and localization of the cysts and, suppuration. The most common symptoms in the pulmonary cysts are cough, mucopurulent sputum, chest pain, hydroptysis and fever (2). Although the most common symptoms in the isolated hepatic hydatid cysts are pain in the upper quadrant of the right abdomen; loss of appetite, nausea, vomiting, anaphylactic reactions, hepatitis, symptoms of cholangitis due to the obstruction of the bile ducts with daughter vesicles can be observed. Most of the cases is diagnosed incidentally during the examination for other reasons. Segmental or lobar liver atrophy might develop depending on the compression on the portal vein and biliary canal caused by the cyst (7, 8). During the physical examination of the patients with hepatic hydatid cyst, findings of hepatomegaly, palpable mass in the upper quadrant of the right abdomen and abdominal distension may be detected (7, 8).

Regarding the localization of the cysts, although the most common site of the pulmonary cysts is the right hemithorax, there are multiple cysts in 30% of the patients and they exhibit a bilateral distribution in 20% of the patients. Most of the cysts in the right hemithorax are in the lower lobe (2, 9). In our study, 73.08% of the patients had unilateral and 26.92% had bilateral pulmonary cysts.

59.61% of the unilateral cysts are localized in the right lung and 13.46% in the left lung. On the other hand, according to the literature, most of the hepatic cysts are localized in the right lobe (61.8%). The majority of the cysts are in the right lobe because this lobe is relatively larger, and it receives a larger amount of blood through the portal vein (10). In our study, the distribution of the hepatic cysts is consistent with the literature and 69.23% of the cysts were in the right lobe.

The pulmonary hydatid cysts, which have an uncomplicated structure and are relatively small in the early stages, do usually not cause any symptoms and are diagnosed incidentally during a radiological examination due to another reason. The pulmonary x-ray imaging displays intact cysts with regular margins and round homogeneous opacity. If the cyst ruptures into a bronchus or pleura and if air pene-

trates between the pericystic tissues, also called endocyst and exocyst or the cyst membrane collapses in the cavity, findings such as double arch sign, rising sun sign, water lily sign can be observed. If the cyst becomes suppurated, it may be confused with the lung abscess. Thoracic CT and/or MR examinations are done for a more detailed evaluation of the cyst detected with the x-ray examination. The findings of the calcification, observed in the hepatic cysts, are not observed in pulmonary cysts during the CT examination. Intact cysts have regular margins, round and homogeneous structures and hyperdense walls (2, 9). The hepatic cysts are usually evaluated with US, tomography, and magnetic resonance methods. The hepatic cysts are grouped in 5 types according to the Gharbi's classification (Table 2) (7).

Table 2: Gharbi's classification of hydatid cysts

<i>Type</i>	<i>Characteristics of Cyst</i>
Type 1	Unilocular cyst, wall and internal echogenicities
Type 2	Detached membrane (water lily sign)
Type 3	Multivesicular, multiseptated cyst, daughter cysts (honeycomb pattern)
Type 4	Heterogeneous cyst, no daughter vesicles
Type 5	Cyst with a partial or complete wall calcification.

The hydatid cysts are also classified in 3 groups as active, transitional, and inactive. Type 1 and Type 2 are active, Type 3 is transitional, and Type 4 and Type 5 are inactive cysts (7). We did not encounter any calcified cyst in the livers of our patients.

The most common therapeutic modality for both hepatic and pulmonary hydatid cysts is surgery (2, 7). Posterolateral thoracotomy, bilateral thoracotomy and sternotomy are the preferred techniques in pulmonary cysts. Bilateral thoracotomy can be performed simultaneously or as a second session according to the general condition of the patient (2, 7). The video-assisted thoracoscopic surgery (VATS), considered as a minimally invasive approach,

became gradually popular in the treatment of the pulmonary hydatid cysts (2, 11). Methods used for the preservation of the parenchyma (e.g. enucleation, cystotomy-capitonnage, pericystectomy and wedge resection (if needed)) are also preferred in the surgery of the pulmonary cysts. Surgery, percutaneous intervention, pharmacotherapy and follow-up are the therapeutic alternatives for the hepatic cysts. The surgical modalities consist of open surgery and laparoscopic surgery. Radical and conservative surgical methods are two subtypes of the open surgery. During the radical surgery, the pericystic membrane is also removed with the cyst and liver resection is carried out if required. In contrary, during the conservative approach,

the pericyclic membrane is preserved and only the cyst content is removed. The residual cavity is closed with techniques such as omentoplasty, external drainage or capitonnage (7).

Conclusion

Phrenotomy simultaneously with the right thoracotomy in order to render a second surgical intervention, anesthesia and an additional laparotomy incision unnecessary, is an effective intervention in patients, who have cysts in the right lung and concomitantly in the liver.

Acknowledgements

There was no financial support and technical or other assistance received for this manuscript.

Conflict of interest

The authors declare that there is no conflict of interest.

References

1. Dalal U, Dalal AK, Singal R. Concomitant Lung and Liver Hydatid Cyst Managed as One-Stage Surgery. *Maedica (Bucur)*. 2017; 12(1): 19-22.
2. Metin B, Aribas OK, Dumanli A, Aribas ET. Comparison of Pulmonary Hydatid Cysts between Men and Women. *Kuwait Med J*. 2016; 48(4): 312-316.
3. Singal R, Goyal S, Goyal R, Mittal A, Gupta S. Primary splenic hydatid cyst in a young boy an uncommon entity. *West Indian Med J*. 2011; 60: 374-376.
4. Yılmaz M, Işık F. Approach to pulmonary hydatid cyst in childhood. *Hydatid Cyst of the Lung*. 2016; 5: 69-78.
5. Borne J, Shaw JH. Hepatobronchial fistula caused by hydatid disease The Dunedin Experience 1952-79. *Thorax*. 1981; 36: 25-28.
6. Gulamhussein MA, Patrini D, Pararajasingham J, et al. Hepatopulmonary Fistula: a life-threatening complication of hydatid disease. *J Cardiothorac Surg*. 2015; 10: 103.
7. Mihmanli M, Idiz UO, Kaya C, et al. Current status of diagnosis and treatment of hepatic echinococcosis. *World J Hepatol*. 2016; 8(28): 1169-1181.
8. Nunnari G, Pinzone MR, Gruttaduarra S, et al. Hepatic echinococcosis: Clinical and therapeutic aspects. *World J Gastroenterol*. 2012; 18(13): 1448-1458.
9. Garg MK, Sharma M, Gulati A, et al. Imaging in pulmonary hydatid cysts. *World J Radiol*. 2016; 8(6): 581-587.
10. Alan B, Kapan M, Teke M, Hattapoğlu S, and Arıkanoğlu Z. Value of cyst localization to predict cystobiliary communication in patients undergoing conservative surgery with hydatid cyst. *Ther Clin Risk Manag*. 2016; 12: 995–1001.
11. Ocakcioğlu İ. A 3.5 cm single incision VATS Cystotomy and Cappitonnage for a 15 cm hydatid cyst. 2014, August 26. <https://www.ctsnet.org/article/35-cm-single-incision-vats-cystotomy-and-cappitonnage-15-cm-hydatid-cyst>