



Clinical and surgical evaluation of cyst hydatid cases in a training and research hospital in Şanlıurfa province

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Abstract

Echinococcus, which is a zoonotic agent, is endemic in our country and causes hydatid disease by localizing in various organs in the human intermediate host. This study aims to present the demographic and clinical characteristics of patients treated for hydatid cysts in our hospital, determine the methods used for treatment, and review the complications in detail. Cases reported to have a hydatid cyst in the surgical material pathology result in Şanlıurfa Training and Research Hospital were evaluated between March 2017 and December 2019. All patients were living in Turkey, which is endemic for hydatid cysts. The records of the patients who were operated on with the pre-diagnosis of cyst hydatid after the examinations and whose pathology results were reported as hydatid cysts were reviewed retrospectively. During the study dates, a total of 84 patients, 55 (65.4%) female and 29 (34.5%) male, were operated on for cyst hydatid in our hospital. The mean age of the patients was 28.6 (min: 2-max: 67). Seventeen (20%) of the patients were in the pediatric age group. When the patients were examined in terms of the number of cysts, 65 (77.3%) patients had a single cyst, while 19 (22.6%) patients had more than one cyst. When evaluated in terms of cyst location, liver localization was observed in 35 (41.6%) patients, liver and lung co-location in 4 (4.7%) patients, lung location in 19 (22.6%) patients, spleen location in 2 (2.3%) patients, and brain location in 2 (2.3%) patients. localization, adnexal localization in 6 (7.1%) patients, renal localization in 2 (2.3%) patients, and soft tissue localization in 14 (16.6%) patients. In the pediatric age group, cyst localization was 4 in the liver, 10 in the lung, 2 in the brain, and 1 in soft tissue. Preoperative serology test results of 42 patients were reported as positive (+) (1/320-1/2560). The average duration of hospitalization was 1-27 days. When the postoperative complications of the patients were examined, the biliary fistula was found in 4 (4.7%) patients, prolonged air leak in 2 (2.3%) patients, pneumothorax in 1 (1.1%) patient, empyema in 1 (1.1%) patient, who was operated for liver cyst hydatid and postoperative seroma was observed in 5 (5.9%) patients. The mean specimen diameter in the pathology results was 2-20 cm. Cyst hydatid disease caused by echinococcus is endemic in our country and often requires surgical intervention. Imaging methods and serology are used in the diagnosis, but a negative serology does not rule out this disease. Patients should be followed up for possible complications after surgery.

Keywords: echinococcus granulosus, hydatid cysts, treatment, complication

1. Introduction

Hydatid cyst is a parasitic disease most commonly found in the liver and lung, caused by *Echinococcus granulosus*, which is more common, especially in livestock populations (1). The disease is more common in younger people. It has a global distribution, and Mediterranean, Middle East, Eastern Europe, Australia, South America, and African countries are considered endemic in terms of echinococcal infection (2). Although its prevalence in Turkey is lower than in previous years, it varies between 5-40 per 10,000 people and is higher in regions where animal husbandry is common such as rural areas of Eastern and Southeastern Anatolia (3).

Echinococcus granulosus is transmitted to the intermediate host, humans, by infected food. After the larvae are taken, they are absorbed from the duodenum, come to the liver via the venous route, attach to the sinusoids, and form the liver cyst

hydatid. Larvae exceeding sinusoids cause disease in peripheral organs with systemic circulation (4). The liver and lungs act as a filter for the parasite. Hydatid cysts occur at a ratio of 50-70% in the liver and 20-30% in the lungs. Rarely, parasites that enter the systemic circulation cause disease in other organs and soft tissues (5). The most common involvement is in the spleen after the liver and lung, while cyst localization in the heart and brain are less common (6). Soft tissue and intramuscular hydatid cysts are rare (0.7-0.9%) even in endemic countries. The growth rate of the cyst may vary depending on the organ. Cases of pulmonary hydatid cysts allow the cysts to reach larger sizes than other organs due to the spongy structure of the lung and may manifest symptoms earlier (7). Slow-growing hydatid cysts remain asymptomatic for a long time. While the disease becomes symptomatic with compression on surrounding tissues and organs, rupture of the

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cyst may cause severe allergic reactions (8).

Ultrasonography is considered the most common imaging method in diagnosing liver cyst hydatids, with its easy application and evaluation of cyst sizes, localization, number, and viability (9). Cross-sectional imaging is preferred in pulmonary hydatid cysts. Immunological diagnostic methods for hydatid cyst are considered helpful in the primary evaluation, complementary to imaging methods, and useful in the follow-up after surgery and pharmacological treatment (10).

Hydatid cyst treatment is managed according to the patient, the stage of the disease, and the location. There are different options for treatment, including medical treatment, percutaneous and surgical interventions (11). Life-threatening complications such as cyst rupture and anaphylaxis may occur in the interventional period. Also, an intensive care unit may be required for complications occurring in the postoperative period.

This study aims to present the demographic and clinical characteristics of patients treated for hydatid cysts in our hospital, determine the methods used for treatment, and review the complications in detail.

2. Material and Methods

Cases that were reported to have a hydatid cyst in the surgical material pathology result in Şanlıurfa Training and Research Hospital were evaluated between March 2017 and December 2019. All patients were living in Turkey, which is endemic for hydatid cysts. The records of the patients who were operated on with the pre-diagnosis of cyst hydatid after the examinations and whose pathology results were reported as hydatid cysts were reviewed retrospectively. The study was conducted under the Helsinki Declaration. The study was approved by the Harran University Medical Faculty Hospital Local Ethics Committee. Since percutaneous treatment was not performed in our hospital, patients who required PAIR were transferred to another center and were not included in the study. Age, gender, nationality, serology results, preoperative abdominal and/or superficial ultrasonography (USG), tomography (CT) and magnetic resonance imaging (MRI) findings, number of cysts, pathology reports, surgery performed, duration of operation, anesthesia method applied, postoperative complications, duration of stay in intensive care unit and duration of hospital stay of the patients were recorded from the file records.

3. Results

Between March 2017 and December 2019, 84 patients were operated on for hydatid cyst in our hospital. 65 (77.3%) of the patients were Turkish, and 19 (22.6%) were Syrian. 55 (65.4%) of the patients were female, and 29 (34.5%) were male. The mean age of the patients was 28.6 (min: 2-max: 67). 17 (20%) of the patients were in the second decade, and 24 (28.5%) were in the third decade. Seventeen (20%) of the patients were in the pediatric age group. When the patients were examined in terms

of the number of cysts, 65 (77.3%) patients had a single cyst, while 19 (22.6%) patients had more than one cyst. When evaluated in terms of cyst location, liver localization was observed in 35 (41.6%) patients, liver and lung co-location in 4 (4.7%) patients, lung location in 19 (22.6%) patients, spleen location in 2 (2.3%) patients, and brain location in 2 (2.3%) patients. localization, adnexal localization in 6 (7.1%) patients, renal localization in 2 (2.3%) patients, and soft tissue localization in 14 (16.6%) patients. In the pediatric age group, cyst localization was 4 in the liver, 10 in the lung, 2 in the brain, and 1 in soft tissue. Preoperative serology test results of 42 patients were reported as positive (+) (1/320-1/2560). While the most common complaint in liver localized cysts was abdominal pain, the most common complaint in lung localized hydatid cysts was cough and shortness of breath and a palpable mass in soft tissue hydatid cysts.

General anesthesia was applied to 74 (88%) patients during the operation, spinal anesthesia was applied to 4 (4.7%) patients, and local anesthesia was applied to 6 (7.1%) patients. Emergency surgery was performed in 12 (14.2%) patients due to spontaneous cyst rupture (7 liver hydatid cysts, five lung hydatid cysts). During the operation, laparoscopic surgery was performed in 14 (16.6%) patients, laparotomy in 33 (39.2%) patients, thoracotomy in 21 (25%) patients, craniotomy in 2 (2.3%) patients, and local excision in 14 (16.6%) patients. Simultaneous cholecystectomy was performed in 5 (5.9%) patients who were operated on for liver hydatid cysts. Splenectomy was performed in 2 patients, partial nephrectomy in 2 patients, and endoscopic surgery in 6 patients (one salpingectomy, four laparoscopic cystectomies, one transabdominal hysterectomy + unilateral salpingo-oophorectomy (TAH+USO)), and lobectomy in 2 patients. Operation times ranged from 20 to 200 minutes. 6 (7.1%) patients were followed up in the postoperative intensive care unit. The average duration of hospitalization was 1-27 days. When the postoperative complications of the patients were examined, the biliary fistula was found in 4 (4.7%) patients, prolonged air leak in 2 (2.3%) patients, pneumothorax in 1 (1.1%) patient, empyema in 1 (1.1%) patient, who was operated for liver cyst hydatid and postoperative seroma was observed in 5 (5.9%) patients. The mean specimen diameter in the pathology results was 2-20 cm.

4. Discussion

Hydatid cyst, which is the larval stage of *Echinococcus granulosus*, has been recognized since ancient times. Hippocrates (460-377 BC) reported the presence of hydatid cysts in cattle and pigs and defined the hydatid cyst she detected in the human liver as "a sac filled with water (Jecur aqua re-pletum)".

The initial stage of primary EG infection is mostly asymptomatic. Many infections develop in childhood but do not show clinical signs until adulthood. Since approximately 50% of the cases can continue asymptotically, they cannot

be diagnosed or detected incidentally during autopsy (12).

Symptoms and findings in patients depending on the location and size of the cyst. Small and/or calcified cysts may remain asymptomatic. However, complications such as symptoms due to mass compression in organs, obstruction of blood or lymphatic flow, rupture, and secondary bacterial infections may develop.

When making a diagnosis, it is important to know the occupation, hobbies, living conditions, education, socioeconomic level, and other exposures of the patient. Although nonspecific leukopenia or thrombocytopenia, mild eosinophilia, and abnormalities in nonspecific liver function tests are observed, it has no diagnostic value. Eosinophilia is observed in 15% or less of cases and is usually detected when antigenic cyst fluid is exposed to the environment (13).

Infections caused by echinococcus, a zoonotic agent, have been on the list of diseases which has to be reported in our country since 2005. In the report published by the General Directorate of Public Health in Turkey in 2020, it was reported that the number of cases is increasing (14). In the report, which examines different pieces of literature from Turkey and the world, the female/male ratio of the disease varies between studies (15). In the European Centre for Disease Prevention and Control 2016 echinococcus annual epidemiologic report, the highest number of cases was reported in the age group of 25–44 years; the highest notification rate was reported in the age groups 25 years of age above. In our study, the female/male ratio was 1.8, and the age range was between 2 and 67 years. The mean age was found to be 28.6.

Hydatid cyst disease, which is endemic in our country, is transmitted to humans by consuming food contaminated with parasite eggs and sometimes by direct contact. Respiratory transmission is not observed. After the hatched embryos enter from the intestinal system, they come to the liver with portal circulation and locate here frequently (70-75%). The second organ in which the hydatid cyst is most commonly located is the lung (20-30%). Lung localization is more common in the pediatric age group (16). In our study, 46.4% liver localization and 27.3% lung localization were seen in all patient groups. As per the literature, the most common location was lung (58.8%) in the pediatric age group. Compared to the literature, the lack of liver hydatid cyst and the proportional excess of lung hydatid cyst in our study can be attributed to the exclusion of patients who will be administered PAIR.

In order of frequency, spleen, soft tissue, intra-abdominal, kidney, brain, bone, pancreatic breast, pelvis, joint, bladder, heart, ovary, thyroid, retroperitoneum, incision scar, and common bile duct are among the localizations outside the liver and lungs (17). Intramuscular localization is rare due to muscle contraction and lactic acid. In our study, soft tissue localization was the most common location other than hepatopulmonary location.

Radiological imaging methods and serological tests are used in the diagnosis of hydatid cysts. The sensitivity and specificity of laboratory and serological tests in the diagnosis of hydatid cyst are limited. There are two major EG antigens. Antigen 5 is the major parasite antigen found on the inner surface of the germinal membrane, daughter vesicles, and protoscolices. Antigen B is a highly immunogenic polymeric lipoprotein and shows greater specificity than detection of antigen 5. When these antigens are examined by ELISA, the sensitivity is around 60-90%, and the specificity is approximately 90% (18,19). In another study conducted in our country, while the sensitivity of the UAV, ELISA, and WB tests were 96.7%, 87.1%, and 100%, respectively, the specificities of these tests were 82.2%, 89.2%, and 85.7% (20).

Serological and immunological tests may be negative in the early period; therefore, radiological imaging methods are more reliable (21). Serological tests are used in the diagnosis as well as in the post-treatment follow-up. Our study observed that a preoperative serology test was performed in 50% of the patients, and it was positive.

The most common clinical findings in patients diagnosed with hepatic hydatid cyst are abdominal pain and abdominal mass/hepatomegaly, and these rates are reported to be 56-84% and 31-86%, respectively, in the literature (22). Cysts located in the lung grow faster than cysts in the liver and give symptoms after compression or rupture of the surrounding tissues. The most common symptoms are shortness of breath and chest pain after coughing (23). The most common complaints in our patients were abdominal pain for liver cyst hydatid and cough for lung cyst hydatid. Medical treatment, open and laparoscopic surgical excision, and PAIR for type I-II hydatid cysts with appropriate localization are used in the treatment of hydatid cysts.

The most common complication of hepatic hydatid cyst is 5-25% intrabiliary rupture (23). After surgery in lung hydatid cysts, the most common complications are prolonged air leak, empyema, and pneumonia (23,24). In the study conducted by Tercan et al. (25), atelectasis (3.3%), biliary fistula (3.0%), and abscess (2.5%) were reported as the most common complications. In our study, the biliary fistula rate among hepatic hydatid cysts was 10.2%, and prolonged air leakage between pulmonary hydatid cysts was 4.3%.

Cyst hydatid disease caused by echinococcus is endemic in our country. Patients admitted with various symptoms depending on the location of the cyst. Some patients are diagnosed incidentally without any symptoms due to the elasticity of the tissue. Treatment of hydatid cyst is surgical. Imaging methods and serology are used in the diagnosis, but a negative serology does not rule out this disease. Patients should be followed up for possible complications after surgery.

Ethics Committee Approval

The study was approved by the Harran University Medical

Faculty Hospital Local Ethics Committee (approval no: HRU/21.15.14).

Conflict of Interest

No conflict of interest was declared by the authors.

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References

- Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev.* 2004 Jan;17(1):107-35. doi: 10.1128/CMR.17.1.107-135.2004. PMID: 14726458; PMCID: PMC321468.
- Poyrazoglu OB, Donder Y, Dulger AC, Arikan TB. Celiac disease prevalence in patients with hydatid cyst. *Journal of Turgut Ozal Medical Center.* 2017; 24(4).
- Altay N, Yüce HH, Küçük A, Aydoğan H, Yalçın S, Yildirim ZB. Anesthetic management in hydatid disease: a review of 435 cases. *Clin Ter.* 2014;165(2):e90-3. doi: 10.7471/CT.2014.1689. PMID: 24770834.
- Altay N, Yüce HH, Küçük A, Aydoğan H, Yalçın S, Yildirim ZB. Anesthetic management in hydatid disease: a review of 435 cases. *Clin Ter.* 2014;165(2):e90-3. doi: 10.7471/CT.2014.1689. PMID: 24770834.
- Kayaalp C, Dirican A, Aydın C. Primary subcutaneous hydatid cysts: a review of 22 cases. *Int J Surg.* 2011;9(2):117-21. doi: 10.1016/j.ijso.2010.10.009. Epub 2010 Oct 25. PMID: 21029797.
- Köksal AS, Arhan M, Oğuz D. Kist Hidatik. *Güncel Gastroenteroloji* 2004; 1: 61-67.
- Kammerer WS, Schantz PM. Echinococcal disease. *Infect Dis Clin North Am.* 1993 Sep;7(3):605-18. PMID: 8254162.
- Sarkar M, Pathania R, Jhobta A, Thakur BR, Chopra R. Cystic pulmonary hydatidosis. *Lung India.* 2016 Mar-Apr;33(2):179-91. doi: 10.4103/0970-2113.177449. PMID: 27051107; PMCID: PMC4797438.
- Bhutani N, Kajal P. Hepatic echinococcosis: A review. *Ann Med Surg (Lond).* 2018 Nov 2;36:99-105. doi: 10.1016/j.amsu.2018.10.032. PMID: 30450204; PMCID: PMC6226561.
- Pawlowski Z, Eckert J, Vuitton DA, Ammann RW, Kern P, Craig P, Set al. Echinococcosis in humans: clinical aspects, diagnosis, and treatment. In: Eckert J, Gemmell MA, Meslin FX, Pawlowski Z, eds. *WHO/OIE Manual on Echinococcosis in humans and animals.* Paris: Office International des Épipizooties; 2001: 20-71.
- McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *Lancet.* 2003 Oct 18;362(9392):1295-304. doi: 10.1016/S0140-6736(03)14573-4. PMID: 14575976.
- Brunetti E, Kern P, Vuitton DA; Writing Panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop.* 2010 Apr;114(1):1-16. doi: 10.1016/j.actatropica.2009.11.001. Epub 2009 Nov 30. PMID: 19931502.
- Sayek İ. Kist Hidatik Hastalığı: Klinik Yönleri. Altıntaş N, Tınar R, Çoker R (eds). *Hidatoloji Derneği Yayınları, İzmir* 2004:141-147.
- Kokturk O, Guruz Y, Akay H, Akhan O, Biber Ç, Çağırıcı U, ve ark. Toraks Derneği Paraziter Akciğer Hastalıkları Tanı ve Tedavi Rehberi. *Toraks Dergisi* 2002; 3: 1-16.
- Irmak H, Özkan AT. Türkiye’de Kistik Ekinokokkoz Mevcut Durum Raporu, *Türk Hijyen ve Deneysel Biyoloji Dergisi.* 2020;77:3
- Annual Epidemiological Report for 2016 Echinococcosis European Centre for Disease Prevention and Control, 2018.
- Garg MK, Sharma M, Gulati A, Gorsu U, Aggarwal AN, Agarwal R, Khandelwal N. Imaging in pulmonary hydatid cysts. *World J Radiol.* 2016 Jun 28;8(6):581-7. doi: 10.4329/wjr.v8.i6.581. PMID: 27358685; PMCID: PMC4919757.
- Hakverdi S, Sayar H, Yaldiz M, Erdoğan S, Akansu B, Canda MS. Cukurova Yöresinde Seyrek Yerleşimli Ekinokokkozis (134 olgu) [Unusual localization of echinococcosis in Cukurova (134 cases)]. *Türkiye Parazitoloj Derg.* 2009;33(1):77-81. Turkish. PMID: 19367552.
- Köktürk O. Akciğer hidatik kist hastalığı. In: Ekin N, Uçan ES eds. *Solunum Sistemi Enfeksiyonları.* Ankara, Turgut yayıncılık, 2001:557-604.
- Mono PL. Clinical manifestations and diagnosis of echinococcus. www.uptodate.com 2013.)
- Akisu C, Bayram Delibaş S, Yuncu G, Aksoy U, Ozkoç S, Biçmen C, Sevinç S, Yaldiz S. Akciğer hidatidozunun tanısında IHA, ELISA ve Western Blot testlerinin değerlendirilmesi [Evaluation of IHA, ELISA and Western Blot tests in diagnosis of pulmonary cystic hidatidosis]. *Tuberk Toraks.* 2005;53(2):156-60. Turkish. PMID: 16100652.
- Yenigül AE, Cetin DA. Two rare cases; primary soft-tissue hydatid cyst mimicking soft tissue tumor. *Annals of Medical Research* 2019; 26(2), 293-295.
- Akova A, Özkan S. Karaciğer hidatik kisti. *Göztepe Tıp Dergisi.*2004; 19: 213-215.
- Durceylan E, İliklerden DM. Akciğer Hidatik Kisti Cerrahi Tedavisindeki Klinik Deneyimlerimiz. *Van Tıp Derg.* 2020;27(2): 144-149.
- Aldahmashi M, Alassal M, Kasb I, Elrakhawy H. Conservative Surgical Management for Pulmonary Hydatid Cyst: Analysis and Outcome of 148 Cases. *Can Respir J.* 2016;2016:8473070. doi: 10.1155/2016/8473070. Epub 2016 Aug 24. PMID: 27642249; PMCID: PMC5013219.