

## Evaluation of Patients with Cystic Echinococcosis

### Kistik Ekinokokkozis Olgularının Değerlendirilmesi

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#### ABSTRACT

**Objective:** Cystic echinococcosis (CE) is a globally prevalent zoonotic disease.

**Methods:** Demographic, clinical, laboratory, and follow-up data of patients between May 2009 and 2015 were retrospectively analyzed by screening data from a hospital automation system.

**Results:** A total of 238 (females, n=139 and males, n=99) patients with a mean age of 40.6±20.58 years were included. Less than half (40.8%) of the patients were living in the countryside. Hepatic involvement of CE was most frequently (72.2%) seen. A majority (75.6%) of the patients were symptomatic, but abdominal pain was the most frequently seen symptom. For diagnosis, in all patients, imaging modalities were used, while in 66% of the patients, serological methods were also employed. The patients received both medical and surgical treatments (78.5%, n=187), only surgical treatment (10.5%, n=25), or only medical treatment (8.8%, n=21). Surgical treatment was performed for patients with hepatic (n=139/176, 80.6%), pulmonary (n=78/94, 82.9%), splenic (n=7/9; 77.7%), and mesenteric (n=6/7, 85.1%) cysts, and patients cases with brain, bone, muscle, omentum, bladder, and adrenal cysts had undergone surgical intervention.

**Conclusion:** Publication of regional data is important in terms of epidemiological considerations and may aid in the formulation of standard treatment approaches.

**Keywords:** Cystic echinococcosis, hydatid cyst, zoonotic diseases

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#### ÖZ

**Amaç:** Kistik ekinokokkozis (KE) dünya genelinde yaygın, önemli bir zoonotik hastalıktır.

**Yöntemler:** Mayıs 2009-2015 tarihleri arasında, hastanemizde KE tanısı ile takip edilen hastaların demografik, klinik, laboratuvar bulguları, hastane otomasyon sistemi verileri taranarak, retrospektif olarak incelendi.

**Bulgular:** Çalışmaya 139'u kadın 99'u erkek olmak üzere 238 hasta dahil edildi. Yaş ortalaması 40,6±20,58 idi. Hastaların % 40,8'inde kırsal kesimde yaşam öyküsü vardı. En sık tutulan organ karaciğer (%72,2) idi. Hastaların %75,6'sı semptomatik olup en sık görülen semptom karın ağrısı idi. Tanıda tüm hastalarda görüntüleme yöntemine başvurulurken, %66' sında serolojik yöntem de kullanılmıştı. Hastaların % 78,5'i (n=187) hem medikal hem cerrahi tedavi, %10,5'i (n=25) yalnız cerrahi tedavi, %8,8'i (n=21) ise yalnız medikal tedavi almıştı. Karaciğer tutulumu saptanan 173 hastanın %80,6'sına (n=139), akciğer tutulumu saptanan 94 hastanın %82,9'una (n=78), dalakta tutulum saptanan 9 hastanın %77,7' sine (n=7), mesenterde kist saptanan 7 hastanın %85,1'ine (n=6); beyin, kemik, kas, omentum, mesane ve sürrenalde kist saptananların ise tümüne cerrahi uygulandığı tespit edildi.

**Sonuç:** Bölgesel verilerin yayınlanması, epidemiyolojik açıdan önem taşıdığı gibi standart tedavi algoritmalarının oluşturulmasına yardımcı olabilir.

**Anahtar Kelimeler:** Kistik ekinokokkozis, hidatik kist, zoonotik hastalıklar

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## INTRODUCTION

Cystic echinococcosis (CE) is a zoonotic disease that creates problems in many countries including Turkey (1, 2). In developing countries in particular, the disease results in health problems and economic losses in both animals and human beings (3). CE is more widely seen in Australia, New Zealand, South Africa, Central and South America, and in almost all Asian countries where the number of stray dogs is very high and where people are mostly raise livestock (4). The worldwide prevalence and incidence of the disease are 1-7% and 0-32/100,000, respectively (5). Its prevalence and incidence in Turkey are 50-400/100,000 and 3.4/100,000, respectively (4).

*Echinococcus granulosus*, which is an etiological agent of CE, lives in the bowels of hosts such as dogs and wolves. When ova excreted in fecal material are ingested by intermediate hosts such as sheep, cattle, and goats or incidentally ingested by human beings the disease occurs. Its parasites settle in almost every organ, including mainly the liver, followed by the lungs, kidney, spleen, brain, skeleton, and heart, and lead to the onset of the disease (6).

In this study, we aimed to analyze the epidemiological and clinical features of patients with CE and evaluate diagnostic methods and applied treatment modalities.

## METHODS

The study had a retrospective design. Patients with CE followed up at the clinics of the departments of General Surgery, Chest Surgery, Pediatric Surgery, and Infectious Diseases of Afyon Kocatepe University Hospital were screened using data from the hospital automation system. Data of all accessible patients were reviewed. Medical and epidemiological characteristics, clinical findings of

the patients, number and location of cysts, diagnostic serological and imaging modalities performed, medical and surgical therapies applied, and notification of the disease were investigated.

## Statistical analysis

All patient data were recorded in Statistical Package for Social Sciences 20.0 for Windows (IBM Corp.; Armonk, NY, USA) for analysis. Continuous variables were expressed as medians and categorical variables as frequencies and percentages. Ethics committee approval and informed consent of the patients were obtained.

## RESULTS

The medical files of 686 patients with a diagnosis of CE whose records were in the hospital automation system were investigated. Patients (n=448) with partially accessible data and those with the initial diagnosis of CE whose diagnosis could not be confirmed in subsequent analyses were excluded, and 238 patients (females, n=139, 58.4%; males, n=99, 41.6%) with a mean age of 40.6±20.58 years were included.

Ninety-seven (40.8%) patients were living in the countryside.

Hundred and eighty patients (75.6%) were symptomatic at admission, and 24.4% of them were diagnosed during randomly performed radiological examinations. Symptomatic patients had complaints of abdominal pain (65.6%, n=118), coughing (23.3%, n=42), chest pain (19.4%, n=35), nausea-vomiting (14.4%, n=26), dyspepsia (11.1%, n=20), fever (7.8%, n=14), dyspnea (7.8%, n=14), hemoptysis (6.7%, n=12), jaundice (3.9%, n=7), hydatidosis (3.9%, n=7), bone discharge (0.8%, n=2), back and loin pain (0.4%, n=1), dysuria (0.4%, n=1), and fainting (0.4%, n=1).

Single and multi-organ involvement were detected in 174 (73.1%) and 64 (26.9%) patients, respectively. The number of organs involved is shown in Table 1.

**Table 1.** Number of organs involved

Single organ involvement (n=174)		Multi-organ involvement (n=64)	
% (n)	Organs	% (n)	Organs
64.3 (112)	Liver	54.6 (35)	Liver and lungs
29.3 (51)	Lung	7.8 (5)	Liver and kidney
1.7 (3)	Omentum	4.6 (3)	Liver and bone
1.1 (2)	Bone	4.6 (3)	Liver and spleen
1.1 (2)	Kidney	4.6 (3)	Liver, lung, and spleen
0.5 (1)	Mesentery	3.1 (2)	Liver and mesentery
0.5 (1)	Spleen	3.1 (2)	Liver, mesentery, and spleen
0.5 (1)	Brain	3.1 (2)	Lung and muscle
		1.5 (1)	Liver and adrenal
		1.5 (1)	Liver and omentum
		1.5 (1)	Liver and bladder
		1.5 (1)	Liver, lung, and brain
		1.5 (1)	Liver, spleen, and omentum
		1.5 (1)	Liver, pancreas, and mesentery
		1.5 (1)	Lung, spleen, and omentum
		1.5 (1)	Liver, lung, spleen, and kidney

**Table 2.** Surgical procedures used on liver cysts in the patients studied

% (n)	Type of Surgical procedures
7.6 (18)	Partial cystectomy and drainage
7.6 (18)	Partial cystectomy and omentopexy
3.3 (8)	Partial cystectomy and capitonnage
7.6 (18)	Cystectomy and drainage
0.4 (1)	Cystectomy and capitonnage
0.8 (2)	Total cystectomy
5 (12)	Segmentectomy
0.8 (2)	Lobectomy
15.1 (36)	Unknown
80.6 (139)	Number of patients operated
100 (173)	Number of patients

**Table 3.** Surgical procedures used on lung cysts in the patients studied

% (n)	Type of Surgical procedure
4.6 (11)	Cystectomy and drainage
5.9 (14)	Cystectomy and capitonnage
6.3 (15)	Cystectomy, capitonnage, and decortication
2.9 (7)	Cystostomy and capitonnage
1.7 (4)	Drainage
1.7 (4)	Drainage, capitonnage, and decortication
1.3 (3)	Enucleation
1.3 (3)	Lobectomy
6.7 (16)	Unknown
82.9 (78)	Number of patients operated
100 (94)	Number of patients

**Table 4.** Surgical procedures used on cysts in other organs in the patients studied

Organs	% (n)	Surgical procedures (n)
Spleen (12)	77.7 (7)	Splenectomy (6)
		Cystostomy and drainage (1)
Bone (5)	100 (5)	Debridement (3)
		Partial cystectomy and drainage (1)
Mesentery (7)	85.1 (6)	Small bowel resection (1)
		Cystectomy (5)
Omentum (6)	100 (6)	Cystectomy (6)
Muscle (3)	100 (3)	Cystectomy (3)
Brain (2)	100 (2)	Cystectomy (2)
Adrenal (1)	100 (1)	Cystectomy (1)
Bladder (1)	100 (1)	Cystectomy (1)
Pancreas (1)	100 (1)	Cystectomy (1)

The number of cysts in a patient ranged between 1 and 12 (mean  $n=2\pm 1.79$  cysts). The diameter of the cysts was at least 0.4 mm and at most 25 cm (median diameter, 6.1 cm).

Diagnostic serological tests were performed in 66% (n=157) of the patients. These patients had undergone an indirect fluorescent antibody test (IFAT) combined with an indirect hemagglutination (IHA) test (90.4%, n=142), only IFAT (57%, n=9), or only the IHA test (3.8%, n=6).

Diagnostic imaging tests had been performed in all patients. Chest radiograms (posteroanterior view) (90.8%, n=216), upright plain abdominal radiographs (14.7%, n=35), abdominal ultrasonograms (US) (60.5%, n=144), thoracic US (6.7%, n=16), abdominal computed tomography (CT) (66%, n=157), thoracic CT (48.7%, n=116), abdominal magnetic resonance imaging (MRI) (12.6%, n=30), cerebral CT and MRI (0.8%, n=2) had been obtained from the patients.

Patients had undergone surgical (87.4%, n=208) or medical (89.1%, n=212) treatment. Patients had also undergone both medical and surgical treatment (78.5%, n=187), only surgical treatment (10.5%, n=25), or only medical treatment (88%, n=187).

Some patients with hydatid cysts (139/173, 80.6%), pulmonary involvement (alveolar echinococcosis) (78/94, 82.9%), splenic cysts (7/9, 77.7%), mesenteric cysts (6/7, 85.1%) and all patients with brain, bone, muscle, omentum, bladder, and adrenal cysts had undergone surgical treatment. Surgical treatments applied are shown in Table 2-4.

Thirty-nine (18.8%) surgical interventions had been performed with the indication of recurrent CE. In 19 (9.1%) patients with CE, postoperative complications had developed, which consisted of abscess (4.6%, n=11), incisional hernia (0.8%, n=2), biliary fistula (0.4%, n=1), surgical field infection (0.4%, n=1), pneumothorax (0.4%, n=1), and pneumonia (0.4%, n=1). Mortality secondary to surgical intervention was not detected.

As medical treatment, all patients had received albendazole.

We got contact with Provincial Directorate of Public Health in Afyon. The number of EC patients was 238 who were registered in the online system of notification of infectious diseases between May 2009 and May 2015. We found notifications of only 13 (5.5%) patients.

## DISCUSSION

Cystic echinococcosis is a frequently encountered parasitic disease in Turkey. Globally, it is more frequently seen in women, and its incidence increases with age (5). According to the database of the Turkish Republic Ministry of Health, the disease is most frequently seen between the age range of 45 and 64 years in Turkey. In our study, in compliance with the literature (the results are similar), our study population consisted of 58.4% of female and 41.6% of male patients and the disease was mostly detected among patients aged between 40 and 61 years.

Risk factors for the disease include being engaged in agriculture, ecological chance, poor hygiene, lower socioeconomic status, illegal and improper animal slaughter, uncontrolled increase in the dog population, inadequate information about the disease,

and lack of disease control programs (1, 6, 7). In our study, 40.8% of the patients had a history of rural life. However, because our study had a prospective design, we could not obtain information about the schooling level of the patients, hygiene conditions, and conditions for animal slaughter.

Cystic echinococcosis can lead an asymptomatic course or present with various symptoms and signs. Symptoms arise as a result of the mass effect of the enlarging cyst. However, in asymptomatic patients, a diagnosis can be incidentally made by detecting the cyst during radiological examinations (6). In our study, in compliance with the literature, most patients were symptomatic and the most frequently reported complaints were abdominal pain, coughing, nausea-vomiting, and dyspeptic problems. Asymptomatic patients were diagnosed during radiological examinations.

The disease involves the liver and lungs in 50-70% and 20-30% of the patients, respectively; however, rarely, it can involve almost every organ. Frequently, only a single organ is affected; alternately, cysts can be detected in more than one organ at the same time (1, 6). In compliance with the literature, we detected single and multi-organ involvement at the same time in 73.1% and 26.9% of the patients, respectively. The cysts were localized in the liver (72.7%) and lungs (39.5%) and less frequently in the omentum, mesentery, bone, kidney, muscle, adrenal, brain, and pancreas.

Splenic echinococcosis is seen at an incidence ranging between 0.5% and 4%. Splenic cysts usually develop secondary to the intraperitoneal or systemic spread of the contents of ruptured hepatic cysts. Rarely, isolated splenic involvement is seen (8). In our study, splenic echinococcal cysts were detected in 12 patients, and our incidence of splenic echinococcosis (5%) was close to the upper limit of the incidence rates cited in the literature. Only one of our patients had isolated splenic involvement, and one patient developed an echinococcal cyst. The cysts developed as a result of spread from the liver (n=10) or omentum (n=1), which might stem from the delayed diagnosis.

Rarely, bone involvement is seen (0.5-4%) (6, 9). The long bones, vertebral column, pelvis, and ribs are the most frequently involved parts of the skeleton (10, 11). Fifty percent of vertebral involvement is seen in the thoracic region (9, 12). In our study, bone involvement was seen only in five patients. Two of them had isolated primary bone involvement, while the remaining three had bone involvement that appeared secondary to direct spread from the liver. Cysts were localized in the pelvis (n=3), thoracic vertebra (n=1), and ribs (n=1). One of the patients was asymptomatic, while discharge from the left hip (n=2), back pain (n=1), chest pain, and dyspnea (n=1) were seen in other two patients. Though very rarely seen, in regions where the disease is endemic, bone involvement should be kept in mind. In patients with unexplained back, chest, and hip pain, echinococcal cysts should be investigated.

In our study, in 8 (3.4%) patients with renal involvement including two with isolated renal involvement, renal echinococcosis was detected. The renal involvement of CE has been seen in 4% of the cases cited in the literature. In isolated renal involvement in particular, preoperative diagnosis is extremely challenging even

in endemic regions. The disease can be confused with tumors. Therefore, during surgery, complications such as unexpected cyst ruptures and anaphylactic reactions can develop (13, 14). As confirmed by our study, the consideration of CE in the etiology of patients with renal cysts is important for the prevention of potential intraoperative complications.

In patients with CE, central nervous system (CNS) involvement is rare and is seen in nearly 1.6-5.2% of the patients (15, 16). In our case series, CNS involvement was seen in only 2 (0.8%) patients. CNS involvement is rare in adults, and 75% cases of CNS involvement are pediatric cases (17). In our study, our patients with CNS involvement included a 12-year-old child and a 56-year-old adult. Cerebral CE is radiologically seen as single, round, and unilocular lesions (18, 19). In endemic regions in particular, if similar radiologic images are seen, CE should be kept in mind.

In our case series, in a male patient, a cyst that was 5 cm in diameter in the retrovesical recess, which occupied the region between the sigmoid colon and bladder and adhered to the anterolateral wall of the bladder, was detected. Retrovesical CE is extremely rare. As seen in our patient, as it leads to nonspecific symptoms such as abdominal pain and dysuria, which can be seen in many urinary system disorders, it is very hard to discriminate CE from other causes. In male patients in particular, it is easily confused with cystic degenerations encountered in prostatic hyperplasia (20, 21). Therefore, particularly in regions where CE is endemic, these atypical locations should not be overlooked.

Adrenal involvement is very rare, and it was seen at an incidence of 0.06-0.18% in an autopsy series (22). Adrenal cysts are mostly asymptomatic. The presence of a cyst has been demonstrated during autopsy or radiological procedures (23, 24). Adrenal cysts and hydatid cysts are encountered in 6-7% of patients with adrenal cysts (22, 25). We also detected adrenal involvement in our 51-year-old female patient (0.4%). Adrenal gland and concurrent hepatic involvement were seen. The patient was asymptomatic, and the diagnosis was incidentally made during a radiological examination.

The diagnosis of CE is based on serological and radiological assessments. IFAT and IHA tests are sensitive in 80-100% of the patients, while their sensitivities drop to 50-56% in alveolar CE. However, their rates of specificity range between 88% and 98% in hydatid cysts and between 25% and 56% in alveolar CE. Therefore, negative serological test results are not enough to eliminate the diagnosis of CE. For making a diagnosis, serological and radiological methods should be used in combination (6). In our study 157 patients underwent serological examinations and negative results were detected in 46 patients. In all 46 patients with negative serological test results, a diagnosis of CE was made based on the radiological test results. The diagnosis of 37 of these 46 patients was histopathologically confirmed.

A standard treatment approach does not exist for CE. The Echinococcus Study Group of the World Health Organization has emphasized that treatment should be determined in consideration of the characteristics of the cyst, accessibility to necessary equipment or experience of the surgeon who may perform the treatment, and patient's compliance to long-term follow-up and

has recommended four different treatment approaches: total surgical excision, percutaneous drainage, medical treatment with benzimidazole derivatives such as albenazole, and "watchful waiting" (26). Total surgical excision has been recommended for complicated symptomatic cysts, cysts compressing vital organs, and percutaneously undrained cysts; however, there is no consensus about the surgical technique to be applied (26, 27). As an alternative to total surgical excision, percutaneous drainage is successful particularly in pediatric patients with unilocular cysts and cysts larger than 5 cm in diameter. In this procedure, percutaneous access into cysts is achieved under the guidance of a radiological modality, and the cyst is aspirated. Then, a scolicidal agent such as 20% saline and 95% ethanol or cetrimide is injected into the cyst and left in situ for 15-20 min. Later, this fluid is aspirated. To prevent relapse, 4 h before and 1 month after the procedure, albendazole is administered (6, 26). In our study, 91.2% of the patients had undergone surgical procedures and in only 4 patients with alveolar CE, percutaneous drainage yielded successful results. No standardization was observed among the surgical methods reported in the literature, and different treatment approaches were used based on clinical manifestations and the surgeon's experience. In 9.1% of the patients, postoperative complications developed. In a comprehensive meta-analysis published in 2015, surgical treatments for CE were evaluated, and a complication rate of 28.3% was indicated, which was apparently higher than our complication rate (28).

Monotherapy with benzimidazole derivatives is recommended in patients with inoperable hepatic and pulmonary CE, cysts in multiple organs, or cysts smaller than 5 cm in diameter. Another indication is peritoneal cysts (26). To this end, most frequently, albendazole is preferred. It is recommended that the daily dose of 15 mg/kg is given in two divided doses. Another alternative is mebendazole, which is given at a daily dose of 40-50 mg/kg in three divided doses (6, 26). It has been reported that the treatment response rate of 30% was achieved when patients were treated for 3-6 months, while the prolongation of treatment to up to 18-30 months accomplished a 32.7% decrease in the number of cysts and 49% cystic degeneration, which amounted to an overall response rate of 81% (26, 29). The most important reason why albendazole treatment does not have the desired level of success is its poor absorptive capacity. Therefore plasma and intracystic concentrations are inadequate. (26). However, in recent years, many authors reported that an encapsulated lipid formulation of albendazole may achieve higher intracystic and intraplasmic concentrations that will increase treatment response rates (30). In our study, only medical therapy was successful in 21 (8%) patients.

Another treatment approach in the treatment of CE is "watchful waiting." According to the classification of echinococcal cysts proposed by the Study Group on Echinococcosis of the World Health Organization, echinococcal cysts are classified based on ultrasonographic characteristics in five stages. CE1 and CE2 denote activated cysts, while CE4 and CE5 denote inactivated cysts. CE3 was further classified as CE3a and CE3b. CE3b cysts do not require surgical treatment unless they give rise to complications. CE3b cysts poorly respond to medical therapy. In a study, "watchful waiting" was employed for CE3b cysts, and these cysts remained stable with time, while albendazole treatment

activated cysts only temporarily. During the follow-up period, the incidence of unwanted events did not significantly differ between both treatment methods. Therefore, "watchful waiting" was found to be suitable for long-term follow-up (31).

Cystic echinococcosis is among the reportable diseases in category C in Turkey from 2005 onwards. However, serious deficiencies exist in the reporting process. Every year, nearly 500 cases are reported to the Ministry of Health and Social Welfare. When the official data of the ministry are analyzed, between 2008 and 2012, a total of 3006 cases were reported. However, according to the official data of Social Security Institution that reimburses the hospital expenses of patients, a total of 12,556 surgical interventions were performed with the diagnosis of CE between these years (32). Similarly, in our study, the number of reportable CE cases was very few. The number of cases of CE reported from all hospitals in the province of Afyonkarahisar, where our hospital is situated, was only 5.5% of the patients followed up in our hospital. The under-reporting of patients with CE may yield erroneous results on the incidence rates of this disease in Turkey, which eventually leads to the underestimation of this disease. Therefore, more effective training programs are needed on reporting this disease.

## CONCLUSION

In conclusion, CE is an important zoonotic disease that is more frequently encountered in Turkey than presumed, and because of its atypical locations, it can be misdiagnosed. In particular, in female patients with unexplained abdominal pain who were referred from rural areas, CE should be considered. It should not be forgotten that serological tests are not sufficient for making a diagnosis, and the diagnosis should be supported with imaging modalities. As was the case in our study, the publication of regional data may aid in the formulation of standard treatment algorithms.

**Ethics Committee Approval:** Ethics committee approval was received for this study from the ethics committee of Afyonkarahisar Clinical Research Ethics Committee (Decision Date: 30.07.2015, Decision Number: 2015/10).

**Informed Consent:** Informed consent is not necessary due to the retrospective nature of this study.

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**Hasta Onamı:** Çalışmanın retrospektif doğası gereği hasta onamına gerek yoktur.

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