Percutaneous treatment with the Örmeci technique for hydatid disease located in the spleen: Single center experience for twenty six years

Necati Örmeci' 💿, Çağdaş Kalkan' 💿, Fatih Karakaya' 💿, Ayşe Erden² 💿, Kenan Serdal Köse³ 💿, Ali Emrehan Tüzün', Xheni Deda' 💿

¹Department of Gastroenterology, Ankara University School of Medicine, Ankara, Turkey ²Department of Radiology, Ankara University School of Medicine, Ankara, Turkey ³Department of Medical Statistics, Ankara University School of Medicine, Ankara, Turkey

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ABSTRACT

Background/Aims: Hydatid disease remains an important global socioeconomic health problem, particularly in the endemic areas. Although half of the patients show no symptoms, hydatid cysts should be treated because of their fatal complications. The aim of this study is to present the long-term results of percutaneous treatment of hydatid disease using the Örmeci technique.

Materials and Methods: Forty-nine patients with 54 cystic lesions were diagnosed with hydatid disease. Twenty-seven of the 54 hydatid cysts located in the spleen were punctured with a 22-gauge Chiba needle through the parenchyma of the spleen under sonographic guidance as a one-step procedure. For every 1 cm of the long diameter of the cyst lesion, 3 cc of fluid from the cysts was aspirated. For each centimeter of the long diameter, 2 cc of pure alcohol (96%) and 1 cc of polidocanol (1%) were injected into the cysts. Five out of 27 patients did not participate in the follow-up.

Results: The 22 patients who were treated using the percutaneous Örmeci technique were followed up for a mean±SD (median) of 50.32±65.30 (26.00) months (minimum 4 and maximum 298 months). All patients except one were successfully treated. No deaths or major complications were noted. Seven patients experienced minor complications.

Conclusion: Percutaneous treatment with the Örmeci technique is a safe, effective, cheap, and reliable method that does not interfere with splenic functions, and this outpatient procedure should be the method of choice for a surgery alternative.

Keywords: Hydatid cyst, percutaneous treatment, PAIR, Örmeci technique

INTRODUCTION

Hydatid disease remains an important global socioeconomic health problem, particularly in the endemic areas. It is caused by Echinococcus granulosus (E. granulosus). The eggs of mature parasites shed from the bowels of definitive hosts such as dogs or rodents and may infect an intermediate host, that is, humans. After passing through the stomach and the bowel wall, the larvae of the parasite enter the portal vein and liver, where they predominantly remain (70%). Some larvae pass through the liver and enter the lung (Lemman's filters) or can infest any other organ. The spleen is the third most common location (0.9%-8%) of the *E. granulosus* larvae infestation (1,2). The cyst created by the larvae grows 1-2 cm in diameter every year and compresses the vital organs and vessels. Due to its slow growth rate, the cyst is often (35%) asymptomatic. When located in the spleen, it can cause pain (65%) in the left upper quadrant. It can be perforated into the peritoneal cavity, bronchopleural spaces, stomach, and colon (3,4). In addition, it may cause anaphylaxis and sudden death (5).

Here, we present the results of a long-term follow-up of 22 patients with a hydatid cyst located in the spleen after percutaneous treatment using the Örmeci technique and also review the literature.

MATERIALS AND METHODS

Since 1991, 49 patients with 54 cysts who applied to the Ankara University Medical School Hospital were diagnosed with splenic hydatid disease. Of these patients, 22 were not treated, and 12 patients with 13 cysts refused to be treated due to their asymptomaticity. These patients received no follow-up. Of the remaining 10 un-

ORCID IDs of the authors: N.Ö. 0000-0002-8909-2102; Ç.K. 0000-0003-1603-5032; F.K. 0000-0001-5677-5532; A.E. 0000-0002-9518-5428; K.S.K. 0000-0002-6734-3847; X.D. 0000-0003-4391-0322.

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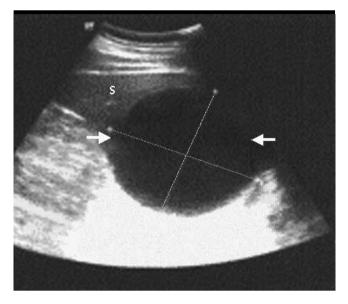


Figure 1. Gharbi type 1 hydatid cyst (arrows) 96x95 mm in size, located in the lower part of the spleen (S), before treatment

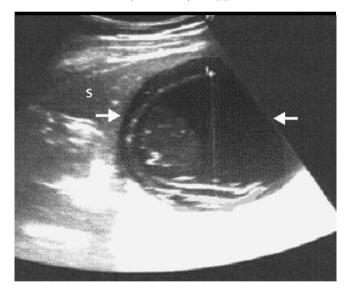


Figure 2. Germinative membrane of the cyst (arrows) is detached and riddled after treatment S: spleen

treated patients with 12 cysts, 10 cysts were <4 cm in diameter. One cyst was Gharbi type 4, and one patient had multiple cysts including one splenic cyst before and after treatment (Figure 1-3). Twenty-seven patients who were diagnosed with hydatid disease underwent percutaneous treatment with the Örmeci technique. Five out of the 27 patients did not participate in the follow-up after treatment (Table 1). We evaluated 22 patients (12 females, 10 males) mean±SD age 40.77±14.20 (young-

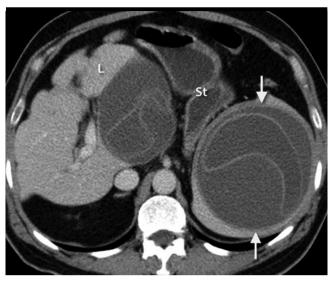


Figure 3. A 74-year-old man with splenic and hepatic hydatid cysts with collapsed membranes. A contrast-enhanced computed tomography shows a hydatid cyst roughly 96 mm in diameter in the spleen (arrows). Note also a hydatid cyst in the liver (L) parenchyma. The cysts were treated with percutaneous injection of absolute alcohol and 1% ethoxysclerol under US guidance. Germinative membranes in both cysts are detached due to treatment and exhibit serpentine appearances in cystic masses St: stomach

est 18, oldest 73) years. All patients signed an additional informed consent before the enrollment. The study conducted according to the principles of the World Medical Association Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects", (amended in October 2013). All patients were examined by ultrasonography, and some of them were also examined by CT or MRI to make a differential diagnosis for non-parasitic splenic cysts. All cysts were classified according to the classification described by Gharbi et al. (6) or the WHO (7). Out of the 27 cysts in the spleen, 11 were located in the upper third, nine in the middle third, and seven in the lower third of the spleen (Table 2). Among the 24 cysts that were followed up, 18 were Gharbi type 1 (81.8%) and six were Gharbi type 3 (18.2%).

All interventions were performed under sonographic guidance in the sonography unit of our department, which was fully equipped for emergencies. All patients were administered sedoanalgesia by anesthesiologists. The patients were laid down in the right lateral position. The cysts were punctured with a 22-gauge Chiba needle through the splenic parenchyma under sonographic guidance as a one-step procedure. For every 1 cm of the long diameter of the cyst lesion, 3 cc of fluid from the

Table 1. Patient flow chart

Total Number		n=49 patients		
		n=54 cysts		
Treated n=27 patients		Not treated n=22 patients		
n=22 patients	n=5 patients	n=10 patients	n=12 patients	
n=24 cysts	n=5 cysts	n=12 cysts	n=13 cysts	
small cysts n=2		small cysts n=10	type 1: 7	
		Gharbi 4, n: 1	type 3: 2	
		multiple cysts n=1	type 4: 3	
			type 5: 1	
			Small cysts: 11(≤4 cm)	
			>4 cm: 2	

Table 2. Characteristics of the patients

Characteristics						
Age: Mean±SD (Youngest, Oldest) year	40.77±14.20 (18, 73) year					
Gender: F/M	12/10					
Localization	Upper: 11	Middle: 9	Lower: 7			
Type of Cysts						
Gharbi Type I: Number (%)	18 (81.8%)					
Gharbi Type III: Number (%)	6 (18.2%)					
Cyst size (cm): (min, max)	Minimum: 37.65 mm (Before Treatment)	Maximum: 89.25 mm (Before Treatment)	Minimum: 21.11 mm (After Treatment)	Maximum: 82.11 mm (After Treatment)		

cysts was aspirated, which is almost the same amount of cc in volume for the Gharbi type 1 and Gharbi type 2 hydatid cysts. Just after the aspiration in Gharbi type 1 and 2, without aspiration Gharbi type 3, for each centimeter of the long diameter of the cysts, 2 cc of pure alcohol (96 %) and 1 cc of polidocanol 1% (ethoxysclerol 1%, Kreussler Pharma, Wiesbaden, Germany) were injected into the cysts. Polidocanol was chosen as a sclerosing agent to destroy the germinative membrane of the cyst, to enhance the sclerosing effect of pure alcohol, and to occlude the relationship between the cyst and vessels, if there was one. The needle, within its sheath, was left inside the cyst for 5 min and then drawn back. The patients were observed for 3 h; if no major adverse events occurred, the patient was discharged. To the best of our knowledge, this is the first report on the percutaneous treatment of splenic hydatid disease using polidocanol (8,9).

All patients were followed up with sonography the day immediately after the procedure, twice in the first 6 months, and then every 6 months. A routine blood chemistry and whole blood count were also performed during the follow-up periods.

Before the treatment, clear fluid-like rock juice and high pressure of the cyst were accepted as evidence of the hydatid cyst viability. During the ultrasonographic examination, a decrease in the cyst diameter, detachment of the germinative membrane for Gharbi type 1 and 2 hydatid cysts, development of pseudosolid pattern, disappearance of daughter cysts in Gharbi type III, disappearance of the whole cystic lesion, and calcification of the cyst wall were accepted as effective treatment.

Statistical analysis

A statistical analysis was performed using Statistical Package for Social Sciences for Windows version 11.5

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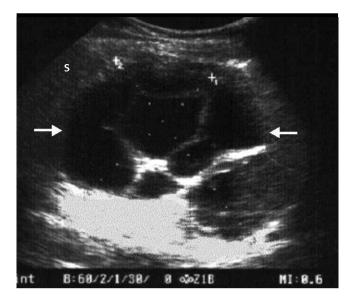


Figure 4. Gharbi type 3 hydatid cyst (arrows), 105x78 mm in size, located in the middle 2/3 of the spleen (S), before treatment

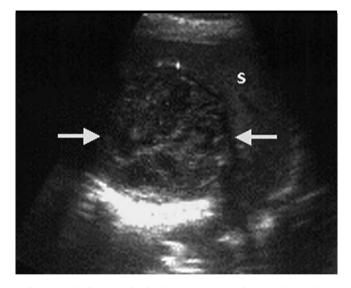


Figure 5. Gharbi type 4 hydatid cyst, 70 mm in diameter, located in the middle 2/3 of the spleen (S). The cyst solidified (arrows) and its diameter decreased after treatment

(SPSS Inc.; Chicago, IL, USA). We used a paired sample t-test to compare the pretreatment and post-treatment cyst diameter. Mean±standard deviation and median were used for continuous variables, and frequencies and percentages were used for categorical variables as descriptive statistics. Statistical boundary was given as 0.05.

RESULTS

Twenty patients had primary splenic cysts (90.9%), but two patients (9.1%) had both splenic and liver cysts (sec-

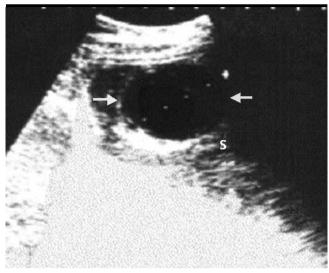


Figure 6. Gharbi type 1 hydatid cyst (arrows) 48 mm in diameter located in the upper third of the spleen (S) before treatment



Figure 7. Gharbi type 5 hydatid cyst 39 mm in diameter, which solidified, calcified, and decreased in diameter after treatment

ondary cysts). Eleven of the 22 patients (50%) had no symptoms. Thirteen patients reported pain in the upper left quadrant. Seven patients had meteorism. No abnormality was detected in the patients' physical examinations. To diagnose the cystic lesions, we mainly used ultrasound examination and sometimes hemaglutination inhibition test (in 14 patients). CT and/or MRI examinations were also used. In addition, during the initial puncture, aspirated material similar to rock juice proved that



Figure 8. A 54-year-old woman with splenic and hepatic hydatid cysts. A contrast-enhanced computed tomography shows a hypoattenuating lesion with peripheral wall calcification (Type V; white arrows) treated 26 years ago. There is also a hydatid cyst (black arrow) in the liver (L) parenchyma with peripheral focal areas of calcification

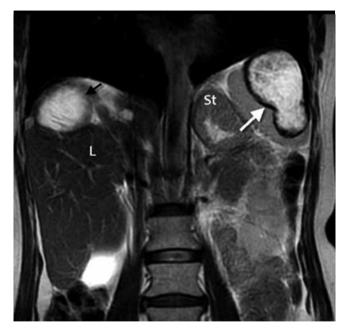


Figure 9. Coronal T2-weighted TSE magnetic resonance image shows a slightly heterogeneous hyperintense splenic mass with a low-signal intensity rim (white arrow), typical of a hydatid cyst. The black arrow indicates a hydatid cyst located at the dome of the right hepatic lobe (L) St: stomach

the lesion was a hydatid cyst. Membrane detachment was observed after the injection of scolocidal agents in all patients with Gharbi type 1 and Gharbi type 2 cysts (Figure 2, 3). Furthermore, during the injection of polido-

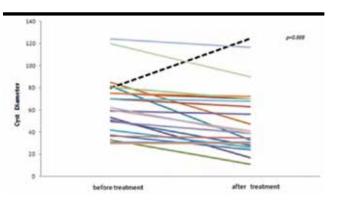


Figure 10. Changing cyst diameter before and after treatment

canol and pure alcohol, the germinative membrane was punctured, and the color became whitish. A sonographic examination showed a gradual decrease in the cyst size in all patients during the follow-up. Three out four Gharbi type 3 cysts became degenerated and solidified (Figure 4, 5). After solidification, some of the cysts might have calcified (Figure 6-9). Twenty-two hydatid cysts in 22 patients, except one cyst, were successfully treated. Treatment failed in one patient with a Gharbi type 3 cyst located at the hilum of the spleen (no degeneration or solidification). The patient underwent splenectomy 3 months after the ineffective percutaneous treatment.

We did not experience any major complications, such anaphylactic reaction, splenic rupture, bleeding, or splenic vein thrombosis, and dissemination during the treatment and follow-up period. One patient had urticaria and temporary low-grade fever. Among the five patients who experienced a temporary ALT increase, four experienced a temporary AST increase; one patient presented with direct bilirubinemia (one-fold), and the levels returned to normal after 3 days. Another successfully treated patient, without consulting our group, underwent a splenectomy himself 8 months after the percutaneous treatment due to persistent upper left quadrant pain.

The mean diameter of the cysts before the treatment was 63.45 ± 25.80 mm. After treatment, it was 51.61 ± 30.50 mm. Only one patient showed an increase in cyst diameter after treatment. The amount of decrease in the cyst diameter was 11.84 ± 18.99 mm per patient (p=0.008) (Figure 10).

DISCUSSION

The spleen is the third most common location for hydatid cysts (0. 5%-8%), which itself is a rare disease. An isolated splenic involvement (primary) is seen in 2.6% cases,

whereas secondary invasion in any other organ such as the liver or lung is found in 20%-50% cases (10,11). In our study, 72% of the cysts were primary cysts and 28% were secondary cysts. The most rational explanation for the location of splenic hydatid disease is systemic circulation. Another explanation could be the retrograde flow through the portal vein, and a third explanation could be direct contagion from a neighboring organ's hydatid cyst (1,12). Most patients with splenic hydatid disease may be asymptomatic because the cysts are slow-growing lesions, enlarging by only 1-2 cms in diameter every year. In our group, 44% of the patients had no symptoms. However, some patients may present with pain and/or meteorism. Some patients reveal palpable mass on the upper left quadrant and/or urticaria (1).

Splenic hydatid disease can be diagnosed by ultrasonography, CT, and/or MRI. Serologic tests such as indirect hemagglutination and immunoblot tests have 95%-100% specificity and 83.3%-87.7% sensitivity (13). Hydatid cysts are classified by ultrasound examination according to the Gharbi or WHO classification. Differential diagnosis should include parasitic and non-parasitic disorders such as congenital, neoplastic, traumatic, and degenerative cystic lesions (14). Hydatid disease should be treated wherever it is located, as it may lead to sudden death due to an anaphylactic reaction, fistulize pleural or peritoneal spaces, lead to cavity infections, portal hypertension due to pressure of the splenic vein, systemic hypertension due to the pressure on the renal artery, or may give rise to mortality and morbidity (1).

Benzimidazole derivatives achieve high concentration in the blood, bile, and liver tissue. There is no correlation between serum and intracystic albendazole concentration. However, cyst fluid concentrations of albendazole and mebendazole are much lower than plasma concentrations. Intracystic drug concentrations are not associated with gender and type of treatment. There is a trend toward higher intracystic concentrations in calcified and smaller-sized cysts (15). Benzimidazol carbamates can be effective in almost one-fifth to one-fourth of patients with small cysts (16). Franchi et al. (17) reported that a long-term (1-14 years) follow-up of 929 hydatid cysts treated with mebendazole or albendazole for 3-6 months recurred in 78.6% cases 2 years after treatment discontinuation. They can be used before and after surgical treatments and PAIR (18,19). They may have adverse effects, such as increased serum ALT and AST levels, leucopenia, abdominal pain, headache, meteorism, and alopecia, in almost 20% of patients.

In the 18th century, when Morgagni documented that splenectomy does not lead to death, total splenectomy became the gold standard in treatment of splenic hydatid disease (20). However, total splenectomy may cause septicemia in 0.2%-0.5% cases, particularly in children. Of those children who experience septicemia, 60% die (20). Additionally, splenectomy has the potential to provoke thromboembolism, pulmonary hypertension, atherosclerosis, and weak immunization (20). The mean hospital stay of patients who undergo splenectomy varies between 2.3 and 9.8 days. The mortality and morbidity rate for this procedure is 3.8% and 14.2%-40%, respectively (11,21-27). Hydatid disease is a benign illness that does not require a radical surgery and lymphadenectomy. Total splenectomy should be preferred in patients with central or hilar location of the cyst, multiple cysts, or giant splenic cysts (22,28).

Since 1960, spleen-preserving surgical techniques, such as partial splenectomy, enucleation, omenthoplasty, rederoofing, cystojejunostomy, and internal or external drainage, have been suggested to protect against the drawbacks of total splenectomy (20,22). Operation time, hospital stay, and recurrence rates are similar for both total splenectomy and spleen-preserving surgery (20). Spleen-preserving surgical techniques should be preferred in patients with small, solitary cysts, and cysts located in the peripheral zone of the spleen. Laparoscopic techniques may have lower morbidity and should therefore be preferred in patients with small and superficially located cysts. Laparoscopic treatment of hydatid cysts is not recommended in patients with infected cysts, complicated cysts, and second or more interventional cysts (27).

Percutaneous treatment of hydatid disease may be an alternative therapy to surgery due to a short hospital stay, less morbidity, and no mortality and recurrence (8,29). The Örmeci technique may have several advantages over the PAIR technique: a) It is an outpatient intervention and does not require hospitalization; b) there is no abscess because a draining catheter is not used; c) morbidity is lower because a thinner 22-gauge Chiba needle is used; d) it is not necessary to use mebendazole or albendazole before or after treatment; e) the amount of sclerosing agent required is less than that in the PAIR technique; and f) the Gharbi type 3 cysts can be treated successfully with this technique.

Akhan et al. (30) treated 12 hydatid cysts with the PAIR technique and draining catheters were used in four. The

abscess occurred in four patients and two of them underwent splenectomy. The hospital stay ranged between 1 and 18 days (30). In another study, 12 patients with 20 splenic hydatid cysts were treated with the PAIR technique and seven of them used a draining catheter. The mean hospital stay was 2.3 days. The abscess was found in 16.6% of the patients (29).

In our study, there was no mortality. There were no major complications such as bleeding, anaphylaxis, abscesses, or perforation. One patient underwent splenectomy after 3 months because of ineffective treatment.

One potential limitation of this study is that as the patients were required to pay for the follow-up, some of them opted out.

In conclusion, percutaneous treatment of splenic hydatid disease is a safe, effective, and reliable method that does not interfere with splenic functions. It is an outpatient procedure and does not require the use of a draining catheter for bigger cysts and albendazole to prevent recurrence. There were no deaths or major complications. Percutaneous treatment specifically with the Örmeci technique should be the method of choice as a surgery alternative.

Ethics Committee Approval: The authors declared that the research was conducted according to the principles of the World Medical Association Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects", (amended in October 2013).

Informed Consent: Written informed consent was obtained from the patients who participated in this study.

Peer-review: Externally peer-reviewed.

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