

# Muscle hydatid diseases: percutaneous treatment with Örmeci technique

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## ABSTRACT

**Aim:** To present results of percutaneous treatment with Örmeci technique for muscle hydatid diseases.

**Material and Method:** Twelve patients (9 male, 3 female) with 16 hydatid cysts (10 CE Type 1, 6 CE Type3B) were treated by percutaneous treatment with Örmeci technique. The percutaneous puncture was performed under sonographic guidance using a 22-gauge Chiba needle as a one-step procedure in CE type 1 and 3A. However, two to six Chiba needles according to size of the cysts were used in different locations at the same time in the cyst of CE type 2, and 3B). For every 1 cm of the long diameter of the cyst lesion, 3cc of fluid from the cysts was aspirated, which was almost the same amount of cc in volume for the CE type 1 and CE type 3A hydatid cysts. A 2cc of pure alcohol (96 %) and 1cc of povidocanol 1% (ethoxysclerol 1%, Kreussler Pharma, Wiesbaden, Germany) were injected into the cysts right after the aspiration of CE type 1 and type 3A, without the aspiration of CE type 2 and 3B, for each centimeter of the long diameter of the cysts. The total amount of pure alcohol and povidocanol were injected equally among the CE type 2 and type 3B cyst's needles. It was waited for five minutes for all scolexes to be killed and the needle/or needles were taken back. The patients with hydatid disease were followed up mean 34.75 ±14.39 (maximum 65-minimum 15) months.

**Findings:** Fifteen out of 16 hydatid cysts (93.75%) cured. We had two complications of treatment. One patient had an abscess in the cyst after the percutaneous treatment. After the percutaneous drainage, patient was cured well, and he had no symptoms during the follow up. Another patient had torpidity in his leg after the treatment. After three months, he had no symptoms.

**In Conclusion,** Percutaneous treatment with Örmeci technique is outpatient based, successful, safe, repeatable, cheap and it can be used as an alternative treatment in selected patients.

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

## INTRODUCTION

Hydatid disease (HD) is still an important health and economic problem especially in endemic areas such as Turkey, Eastern Europe, Eastern Africa, South America, Australia, and New Zealand; in which livestock grows up. The larvae of *Echinococcus granulosus* causes the HD in Human as an intermediate host. Normally, adult parasite resides in the intestine of carnivores. Several thousand eggs of parasite are casted by feces in every day. Human being is accidentally infected by those eggs. When the egg is ingested by the host, larvae of *Echinococcus granulosus*

hatches and penetrates intestinal wall and comes to the liver by the portal vein. If it pass through the filters of liver and lungs, it may reaches any organ like muscles in the body. When it resides in any organ, grows 1-3 cm in diameter in a year; the bigger the more complications occur. It compresses the vital organs, fistulize into the spaces like peritoneum, pleura, bile ducts. Besides, the disease gives big economic burden on the endemic countries. It was found that the total cost of a single hospitalization, including hospital stay, surgical intervention, personnel,

drugs, and administrative costs ranged from €5,874 to 23,077 (median €11,033) per patient in Pavia, Italy (1).

Hydatid disease mainly locates in the liver (70%) and the lungs (20 %). Musculo-skeletal system is one of the rare locations of this disease; and it varies 0.5-5 % in the literature (2).

Aim of this study is to present the results of percutaneous treatment with Örmeci technique for 12 patients with 16 muscle HD.

### MATERIAL AND METHOD

The study was carried out with the permission of Ankara University Faculty of Medicine Ethics Committee (Date: 26.10.2015, Decision No: 16-689-15). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

Nine hundred forty-two patients with 1367 cysts were diagnosed as hydatid disease among the ones who applied to Ankara University Medical School, Department of Gastroenterology. Sixteen patients with 24 cysts out of 1367 (1.36 %) were diagnosed muscle hydatid cyst (Figure 1). Three patients were not treated because two of them received Albendazole and one patient had type 4 hydatid cyst which is not necessary for the treatment. Four out of twenty-one hydatid cysts located both in the muscles and liver. One patient was lost to follow up. Twelve patients with 16 hydatid cysts included in the study. Nine out of 12 (75%) were males, three of them were females (25%), mean age was 38.83±12.27 year. Ten cysts (62.5%) were CE Type 1, six out of 16 (37.5%) was CE Type 3. Eight cysts (50%) located at upper part of left leg, while five cysts (31.25%) placed upper part of the right leg. One cyst on the right side, another cyst on the left side located at left lower part, lower extremity. One cyst located at right thoracal paraspinal area. Mean diameter of the cysts was 60.15±37.35 mm. Seventeen out of 21 hydatid cysts were in the muscle (80.95%, primary) and four out of 21 cysts (19.05%, secondary) were in the liver in thirteen patients. Baseline characteristics of the patients with muscle hydatid diseases is seen in Table 1.

The patients were diagnosed hydatid disease by ultrasonographic findings which were described by Gharbi HA and WHO-Infomal Working Group on Echinococcosis improved Gharbi's classification for the diagnosis of echinococcosis in 2003 (3,4). Besides that On the MRI examination; occurrence of the hydatid matrix, daughter cysts, floating membranes in the cyst cavity, the wall of hydatid cyst which was shown as characteristically low signal intensity, as "rim" on T2 weighted MR images was accepted as hydatid disease.(5). Similarly, On the CT examination; occurrence of water attenuation cyst with well-defined, all daughter cysts which appear as round,

peripherally located cystic lesions in the mother cyst, the high density fluid surrounding the daughter cysts which appears like "spoked wheel pattern", small, round daughter cysts among the solid matrix in the mother cyst, detached germinative membrane from the ectocyst which appear floating, thin and hypodense membrane which is called "water lily pattern" was accepted as hydatid disease (5).

**Table 1. Basic characteristics**

Parameter	Mean±Std. Dev (max-min)	Number (%)
Age	38.83±12.27 (70-21)	
Follow-up	34.75±14.39 (65-15 months)	
Baseline Diameter	60.15±37.35 (160-25)	
Hydatid Cyst Type 1		10 (62.5%)
Hydatid Cyst Type 3		6 (37.5%)
Gender (Male-Female)		9 (75%) 3 (25%)
Primer-Secondar Cysts		13 (81.3%) 3 (16.8%)
Hydatid Cyst Localization	Upper part	Right 5 (31.25%) Left 8 (50%)
	Lower part	Right 1 (6.25%) Left 1 (6.25%)
	Extermity	
	Medulla Spinalis	1 (6.25%)

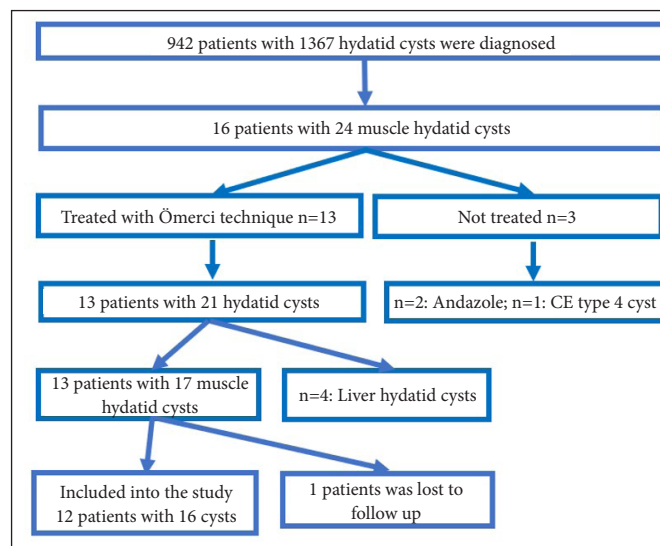


Figure 1. Flowchart of the patients for the study

All alive hydatid cysts were treated by percutaneous way with Örmeci technique. The patients with dead hydatid cyst were excluded from the study. 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 procedures were performed under sonographic guidance in the ultrasonography unit of a gastroenterology department that was fully equipped

against an emergency condition. An intravenous line was established. The patients were positioned according to the location of the cysts as face-down or supine position. All patients were given 5mg of meperidine and 40mg methyl prednisolone as sedo-analgesia just before the procedure. The percutaneous puncture was performed under sonographic guidance using a 22-gauge Chiba needle as a one-step procedure in CE type 1 and 3A. However, two to six Chiba needles according to size of the cysts were used in different locations at the same time in the cyst of CE type 2, and 3B. For every 1 cm of the long diameter of the cyst lesion, 3cc of fluid from the cysts was aspirated, which was almost the same amount of cc in volume for the CE type 1 and CE type 3A hydatid cysts. A 2 cc of pure alcohol (96 %) and 1cc of polidocanol 1% (ethoxysclerol 1%, Kreussler Pharma, Wiesbaden, Germany) were injected into the cysts right after the aspiration of CE type 1 and type 3A, without the aspiration of CE type 2 and 3B, for each centimeter of the long diameter of the cysts. The polidocanol (1%), had been used by our group for the first time to close the connection among the cysts and blood vessels, lymphatic vessels and/or biliary ducts since 1991. The total amount of pure alcohol and polidocanol were injected equally among the CE type 2 and type 3B cyst's needles. We waited for five minutes for all scolexes to be killed and the needle/ or needles were taken back. This technique was reported before by our group(6). Although CE Type 2 and Type 3B are difficult to treat compared to CE Type 1, we added Albendazole 10 mg/kg/day for 6 months right after the percutaneous treatment. All patients were followed up for the function of vital organs for two to three hours and the patients were sent to their home. The patients were followed up one day, three months and six months after and each year by USG in terms of treatment criteria, blood checking and sometimes CT or MRI.

Every patient was evaluated in terms of treatment criteria as follows:

- Diameter of the cysts before and after the treatment
- Detachment of germinative membrane especially in CE Type 1 hydatid cysts
- Degeneration and pseudo-solidification of the cysts
- Whether is there any daughter cyst in mother cyst or not?

A statistical analysis was performed using Statistical Package for Social Sciences for Windows version 11.5.(SPSS Inc.; Chicago, IL, USA). Descriptive statistics were summarized as counts and percentages for categorical variables, mean and standard deviations and median (minimum and maximum) for others. The difference between the two dependent groups was evaluated with a Wilcoxon test. A p value less than 0.05 was considered significant.

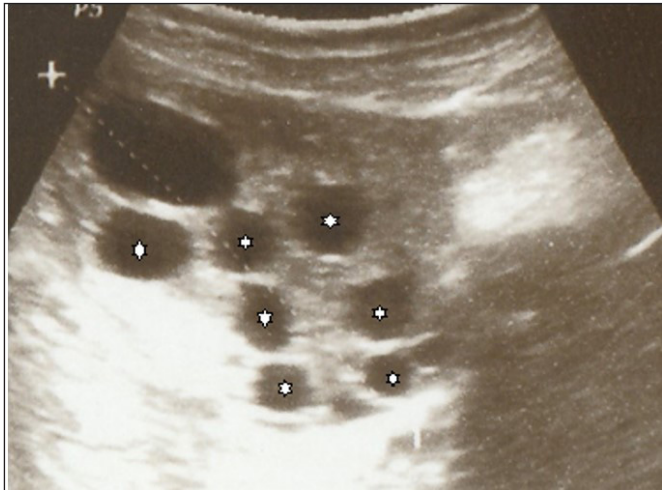
## RESULTS

The patients with hydatid disease were followed up mean  $34.75 \pm 14.39$  (maximum 65-minimum 15) months. Diameter of the cysts were decreased after the treatment compared before in 11 out of 16 cysts ( $P=0.036$ ). Right after injection pure alcohol (96%) and polidocanol 1 % germinative membrane whitened, riddled and detached from ectocyst in 6 (60%) out of 10 Type 1 hydatid cysts. Right after the treatment of type 3 hydatid cyst, daughter cysts distorted, whitened and they showed degenerative changes on the ultrasonographic examinations. (Figure 2). Most of the patients showed pseudo-solidification during the follow up. On the MR examinations, it was shown that daughter cysts degenerated, solidified, and decreased the diameter of the mother cysts (Figure 3A, 3B, 3C, 3D, 3E, 3F, 3G). Fifteen out of 16 hydatid cysts (93.75%) cured. One out of 16 cysts (6.25%) unchanged (Table 2).

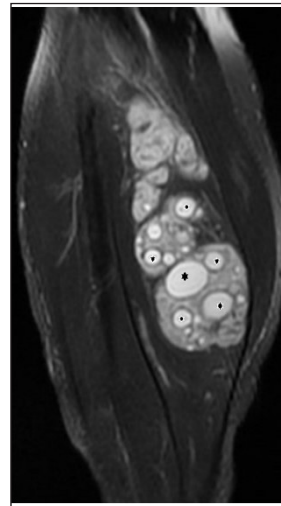


**Figure 2.** Left leg is swollen because of CE type 3 hydatid cyst in musculus gastrocnemius. Two needles were placed into the cyst at the same session

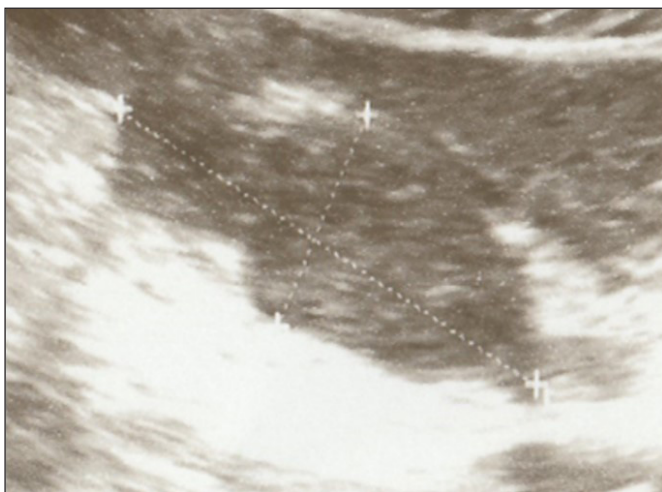
	Mean±Std. Dev	Count (Percent)
Previous diameter	60.15±37.35	
Diameter after the treatment	39.75±36	p=0.036
Unchanged		1(6.25%)
Pseudo-solidification 1/3		2(12.5%)
Pseudo-solidification 2/3		1(6.3%)
Pseudo-solidification 3/3		12(75%)
Detachment of cyst germinative membrane		6(37.5%)
Decreasing of the cyst diameter		11(68.8%)



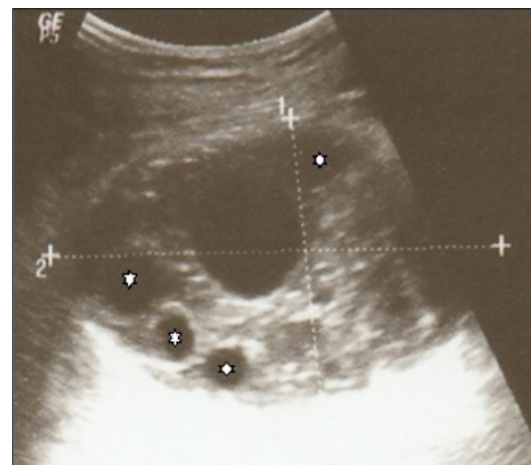
**Figure 3A:** 44-year-old man with cystic echinococcosis (CE). A well-circumscribed multiple daughter cyst (asterisks) with solid content (CE 3b according to WHO) is observed in the muscle. The cysts appear anechoic with smooth contour and do not contain calcification. The solid matrix appears isoechoic with neighboring muscle structures



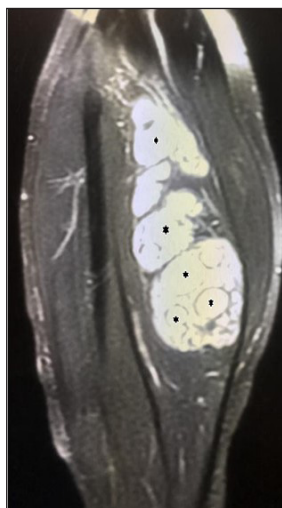
**Figure 4B and 4C.** Two follow-up ultrasonography's performed one- year- periods after the treatment show that the cystic lesion has solidified



**Figure 3B:** One-year follow-up ultrasonography after the treatment shows that the cystic lesions are hardly noticeable, and the lesion is almost completely solidified



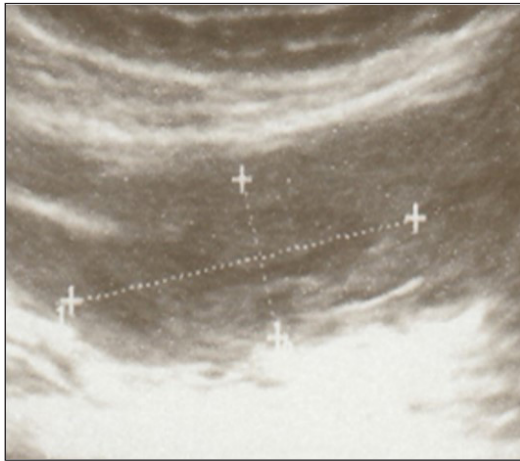
**Figure 5A.** 44-year-old man with cystic echinococcosis (CE 3b according to WHO). (Same case as Fig.3). A Coronal T2-weighted SPAIR (Spectral Attenuated Inversion Recovery) MR image. A well-circumscribed multiloculated semisolid lesion is observed in the crura region. Multiple, uniformly demarcated, and homogeneous internal structured daughter cysts (asterisks) are present in the lesion. The cysts appear hyperintense on T2 weighted image. The walls of the cysts are seen as hypointense on T2 weighted image (rim sign). Solid matrix of the lesion is seen slightly hyperintense relative to neighboring muscle structures on T2 weighted image. Calcification has not been detected yet



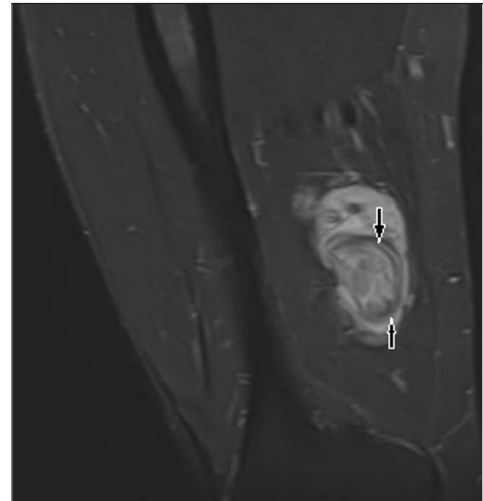
**Figure 4A:** 37-year-old man with cystic echinococcosis (CE). A large, well-circumscribed semisolid cystic lesion (CE 3b according to WHO) containing a big cyst and multiple daughter cyst (asterisks) with different sizes is observed in the muscle



**Figure 5B.** Coronal T2-weighted SPAIR MR image, after 1 year, overall size of the lesion is constant, but there is minimal reduction in the size of the cysts



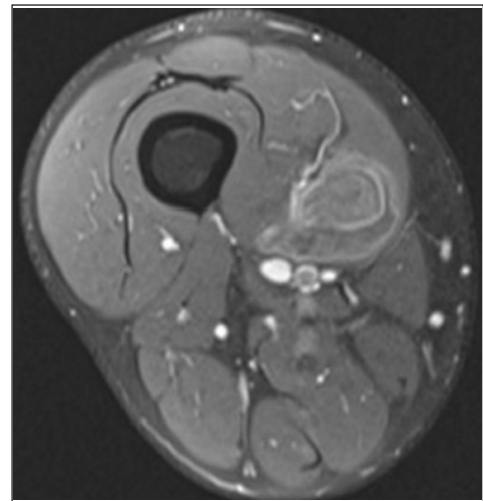
**Figure 5C.** Coronal T2-weighted SPAIR MR image, there is a decrease both number and the size of cysts in the 2nd year follow-up after treatment. In addition, a decrease is also observed in the size of the mass itself



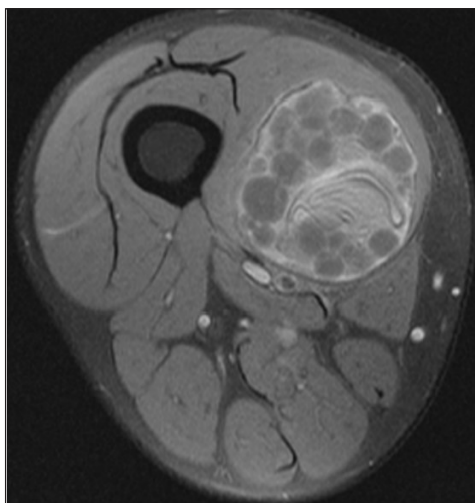
**Figure 6C.** Coronal T2-weighted SPAIR and Fig 6D: Axial post-contrast fat sat T1-weighted turbo spin-echo MR images, one year after the treatment, a decrease in lesion size and the detached endocyst (black arrows) are seen



**Figure 6A.** 37-year-old man with cystic echinococcosis (CE 3b according to WHO). (Same case as Fig.4). A Sagittal T2 weighted turbo spin-echo MR image. The lesion containing cysts of various sizes (daughter cysts, asterisks) and floating membranes (arrows) is observed within the Vastus muscles. The peri cyst (long arrows) of the lesion which appears hypointense on T2-weighted image (rim sign). The daughter cysts appear homogeneous hyperintense on T2 weighted image



**Figure 6D.** axial post-contrast fat sat T1-weighted turbo spin-echo MR images, one year after the treatment, a decrease in lesion size and the detached endocyst (black arrows) are seen



**Figure 6B.** Axial post-contrast fat sat T1-weighted turbo spin-echo MR image, the peripheral type of enhancement is seen on the cyst itself and the cystic components

## DISCUSSION

Muscle hydatid disease is rarely seen in almost 0.5-5 % of all hydatid cysts 5. We found the incidence of muscle HD in 1.36 % among 1367 hydatid cysts in our series. The liver (70%) and the lungs (20%) are the most common locations of the disease because the liver and the lungs are natural mechanical filters of the oncosphere of hydatid cyst. If the oncosphere can pass through the liver and lungs, it can place in any organ like spleen, kidneys, muscles, brain, and bones by the systemic circulation. In primary muscle HD, the cyst located in only muscles. However, in the secondary HD, cysts place not only in muscles, but also in other organs such as the liver, the lungs or peritoneum. In this case series, it was found that the incidence of primary and secondary muscle hydatid cysts was 81.3% and 16.8%, respectively. It is difficult for oncosphere to place in the muscles because of lactic acidosis which occurs in working muscles. The need of

oxygen also increases when the hydatid cyst growing up and oxygenation of the cyst become difficult due to lactic acidosis. Another point is permanent contraction of muscles which gives rise to restrict of it to place in the muscles (2). The most common localizations of the hydatid cyst at the muscles are proximal part of the thigh, pelvis, paravertebral region, shoulder and humerus due to large volume of muscles and rich vascularization (2, 7) Most of the cysts (81.25%) located in proximal parts of the lower extremities in this series.

Usually, the patients with muscle hydatid cyst have no symptoms but one third of the patients may present palpable mass with pain on the thighs or hips. Serologic examinations for antigens of hydatid juice such as ELISA, LATEX agglutination test, immunoblotting test in muscle hydatid diseases is found negative on most of the cases (8). Ultrasonography is the method of choice for diagnosis of the disease. Gharbi (3) described the diagnostic criteria of hydatid disease on ultrasonographic examination in 1981. WHO- Informal Working Group on Echinococcosis improved Gharbi's classification for the diagnosis of echinococcosis in 2003. Ultrasonography has several advantages compared to CT or MR such as easy to find everywhere, no radiation, cheap, high diagnostic sensitivity and specificity (9). It can be decided easily stage of cyst, number of the cyst and distribution of the cyst (9).

Anechoic cyst with posterior acoustic enhancement and double wall sign, hydatid sand (it is dispersed in the cyst and appear as falling snowflakes "snowstorm sign" when the patient turn on right or left side), detachment of germinative membrane from the ectocyst "water lily sign", multivesicular cyst (multiple daughter cyst which are seen as anechoic, round cysts in the mother cyst are some of the characteristics of hydatid disease on the ultrasound examinations (3-5)). (8) Daughter cysts locate at the peripheral areas in the mother cyst. After becoming mature, they fully filled inside of the mother cyst which is named "spoked wheel pattern" (10).

On the CT examination, CE Type 1 cyst appears as water attenuation cyst with well-defined wall. CE type 2; daughter cysts appear as round, peripherally located cystic lesions in the mother cyst. After becoming mature, the high-density fluid surrounding the daughter cysts appears like "spoked wheel pattern". Small, round daughter cysts among the solid matrix in the mother cyst is described as CE type 3b. Detached germinative membrane from the ectocyst appear floating, thin and hypodense membrane which is called "water lily pattern" (CE type 3a). Calcification, infection in the cyst, peritoneal and bones invading of the cysts are shown best on CT examination. (5)

On the MRI examination, hydatid cyst appears hypointense on T1 weighted images and hyperintense on T2 weighted images. The hydatid matrix, daughter cysts, floating membranes in the cyst cavity are clearly seen compare to CT (10). The wall of hydatid cyst is shown as characteristically low signal intensity, as "rim" on T2 weighted MR images (5).

Differential diagnosis should be performed among hydatid disease and single or multiple hemangiomas, fungal, pyogenic or amoebic abscess, neoplasia with hemorrhage and/or necrosis, metastasis, post-surgical sequela and textiloma, lipoma, myositis, Tbc, aneurism, hernia synovial cyst etc. (9,11)

Stojkovic et al. (12) reported that 711 patients with 1308 liver and peritoneal hydatid cyst were treated by benzimidazole derivatives. They estimated that 40 % of the cysts are active or become active after two years. Similarly, Franchi C et al. (13) treated 448 patients with 929 hydatid cysts by mebendazole or albendazole for 3-6 months. After the long term (1-14 years) follow up, 74.1% of patients were shown degenerative changes, however almost 25 % of those cyst relapsed again. Benzimidazole derivatives are not recommended for the treatment of hydatid disease alone. They can be combined with before and after surgical treatment and Puncture, Aspiration, Injection, Re-aspiration (PAIR) Technique.

The combination of surgical and albendazole treatment is the method of choice for the treatment of hydatid diseases (1,11,14-18). Togral et al. (19) treated 5 patients with muscle and bone hydatid disease by the combination of surgical and albendazole treatment. One patient had relapse after 1-9 years follow up. Arazi et al. (11) treated 15 patients (n: 7 muscle, n:8 bone) with hydatid diseases by the combination of Albendazole and Surgical treatment. Four patients with bone hydatid disease had relapsed during the follow up for 30 months. However most of the case treated by surgery related the muscle HD reported cure without relapse (20).

Surgical treatment has some disadvantages such as large incision, longer hospital stays and expensive. Battelli G et al. (21) reported that while a patient who was operated on hydatid disease cost 14 000 USD, compared to the one who was not operated on hydatid disease cost 2500 USD.

PAIR technique had been using since 1985, as an alternative method compared to surgery for the treatment of hepatic hydatid diseases because of short hospital staying, less mortality and morbidity and cheaper than surgery. However, PAIR method and catheterization techniques for the treatment of hydatid disease with extrahepatic localization may have more complication and longer hospital stay. Arslan et al. (22) treated with 27 patients, PAIR (n:20) and Catheter technique (7 patients)

with extrahepatic localizations. Hospital staying was mean 2.3 days (1-14 days), occurrence of abscess was 16.6%.

Percutaneous treatment with Örmeci technique had been using since 1991 (2,23-26). This technique has some advantages such as, outpatient bases, no fistula occurrence, repeatable, less morbidity, cheaper, easy to use compared to PAIR method (27). We have published preliminary report related hydatid cysts in muscle in 2007(2).

In our best knowledge, this is the largest series about percutaneous treatment of muscle hydatid disease in the literature. Sixteen hydatid cysts in 12 patients were treated by percutaneous way. Fifteen out of 16 hydatid cysts (93.75%) cured. Two patients who had CE type 3 hydatid cysts had second puncture due to incomplete treatment of the first attempt. One patient who had CE type 3B hydatid cyst, 135×120×65 mm in diameter could not be cured. Ten CE type 1 and 3 CE type 3 were cured well after the treatment.

We had two complications of treatment. One patient had an abscess in the cyst after the percutaneous treatment. After the percutaneous drainage, patient was cured-well and he had no symptoms during the follow up. Another patient had torpidity in his leg after the treatment. After three months, he had no symptoms. The weak point of this study is to be retrospective study.

## CONCLUSION

Percutaneous treatment with Örmeci technique, is useful in patients with muscle hydatid disease. It is successful, safe, repeatable, easy to perform and cheap. It can be an alternative treatment in patients who had contraindications to surgical treatment or who do not accept the surgical treatment.

## ETHICAL DECLARATIONS

**Ethics Committee Approval:** The study was carried out with the permission of Ankara University Faculty of Medicine Ethics Committee (Date: 26.10.2015, Decision No: 16-689-15).

**Informed Consent:** Because the study was designed retrospectively, no written informed consent form was obtained from patients.

**Referee Evaluation Process:** Externally peer-reviewed.

**Conflict of Interest Statement:** The authors have no conflicts of interest to declare.

**Financial Disclosure:** The authors declared that this study has received no financial support.

**Author Contributions:** All of the authors declare that they have all participated in the design, execution, and analysis of the paper and that they have approved the final version.

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