A novel modified PAIR technique using a trocar catheter for percutaneous treatment of liver hydatid cysts: a six-year experience

Alaaddin Nayman
Ibrahim Guler
Suat Keskin
Tuba Berra Erdem
Hale Borazan
Ahmet Kucukpan
Huseyin Ozbiner
Abdussamed Batur
Ersern Ertekin
Bahadir Feyzioglu
Osman Koc
Hasan Emin Kaya
Osman Temizoz
Adil Kartal
Orhan Ozbek

Hydatid disease is a parasitic infection caused by the larval stage of the tapeworm *Echinococcus E. granulosus* is the most common cause of hydatid disease in humans and is found throughout the world. It is endemic in large sheep raising areas like the Mediterranean region, the Middle East, Southeast and Central Russia, Northern China, South America, Australia, and New Zealand (1). Hydatid disease usually affects the liver (50%–70%) and less frequently lung, peritoneum, kidney, brain, mediastinum, heart, bone, soft tissues, spinal cord, spleen, pleura, adrenal glands, bladder, ovary, scrotum, and thyroid gland (2). Treatment approaches include medical, surgical, and minimally invasive procedures. Medical treatment with albendazole or mebendazole alone has a low rate of success and high rate of relapse, making this treatment option controversial (3). The surgical approach has been the gold standard therapy for the hydatid disease for a long time (4). However, in recent years, percutaneous treatment of the hydatid cyst emerged as a potential alternative to surgery, because of its efficiency, reliability, and low morbidity and mortality rates. The puncture, aspiration, injection, and reaspiration (PAIR) technique, which involves puncture of the cyst wall, aspiration of cyst contents, instillation, and reaspiration of the scolicidal agent, has gained international recognition. PAIR technique can be achieved by using a coaxial catheter system to aspirate the cyst content and infuse scolicidal agent at the same time (5). Another technique can be performed by catheterization (3, 6). In our study, we performed the PAIR procedure by directly entering into the hydatid cyst cavity through a single puncture using a trocar catheter instead of placing a catheter through stiff wire after puncturing with a Seldinger needle. The primary goal of the current study was to determine the success and reliability of this technique in patients with hydatid disease.
with albendazole intolerance, were given 10 mg/kg per day albendazole orally for three weeks before the procedure to prevent dissemination during the procedure. Each patient was examined by an anesthesiologist during preoperative evaluation and underwent US examination to determine cyst type, location, number, and size. Complete blood count and coagulation parameters were checked before the intervention, an INR value less than 1.5 and platelet count higher than 100,000/mL were considered appropriate.

Heart rate, arterial oxygen saturation, and noninvasive blood pressure were monitored. After catheterization of a peripheral vein, lactated Ringer solution 5 mL/kg was administered, and oxygen 4 L/min was applied via face mask. Diphenhydramine HCl 20 mg and methyl prednisolone 1 mg/kg were applied intravenously to all patients for prevention of allergic reactions and decreasing the anaphylaxis risk. Because of an anaphylaxis risk, epinephrine was also prepared. Midazolam 0.03 mg/kg and fentanyl 50 μg were used for initial sedation and additional midazolam 0.01 mg/kg and fentanyl 25 μg were applied when needed.

Procedure
All procedures were performed by two radiologists who have at least three years of experience in nonvascular interventional radiology. Patients were positioned in supine or lateral decubitus position on a fluoroscopy table with C-arm equipment. Povidone iodine and appropriate draping were used to attain a sterile condition after shaving the abdominal wall covering the hepatic region. Local anesthesia (prilocaine hydrochloride) was administered before the puncture of the cyst. We used number 11 scalpel blade for skin incision after local anesthesia. A 6F trocar type all-purpose drainage catheter (Skater™, Angiotech Pharmaceuticals) was then placed into the cyst through US guidance. A stopcock was attached to the catheter to prevent air escape into the cyst during aspiration and injection. All cyst content was aspirated and the cyst was filled with a mixture of nonionic contrast medium and saline solution (1/2 contrast medium + 1/2 saline solution 0.09% NaCl) equal to the amount of aspirate. A cystogram was then obtained in two planes (anteroposterior and lateral) to check any communication between the cyst cavity and the biliary tract. If cystography showed communication between the biliary tract and the hepatic hydatid cyst cavity, alcohol was not used because of the risk of inducing secondary sclerosing cholangitis. The content of the cyst cavity was aspirated again after the cystograms, followed by injection of absolute alcohol (98% ethyl alcohol) in a volume equaling two-thirds of the aspirate. After 20-30 minutes, the procedure was completed by aspiration of the alcohol from the cyst cavity and fixation of the catheter to the skin.

Results
Our study included 374 patients (161 males and 213 females; mean age, 48 years; age range, 7–81 years) with a total of 493 cysts. Of the cysts, 317 were Gharbi type I (WHO CE 1) and 176 were Gharbi type II (WHO CE 3A). All patients were treated with albendazole for prevention of peritoneal dissemination of the cyst content, except six patients who were intolerant to the drug. Trocar catheter was applied successfully in all cysts. Mean diameter of the cysts was 76 mm, ranging from 33 mm to 221 mm. In cystograms obtained from 493 cysts, cys-
Cystobiliary fistulization was observed in 13 cases (Fig. 1). These cysts were considered inappropriate for alcohol injection and referred to surgery. In the remaining 480 cysts of 361 patients, 469 cysts (97.7%) were treated successfully (Figs. 2, 3), while 11 cysts (2.2%) recurred. A distended appearance or lack of reduction in cyst size was considered as a sign of recurrence. All recurrences were detected in the first month. A second percutaneous intervention was performed for all of the 11 recurrent cysts and nine of them were treated successfully. Cystobiliary fistulas were detected during the second intervention in the remaining two recurrent cysts and these cases were referred to surgery (Fig. 4). Cysts with reduced size and loss of distended appearance after the first month showed no increase in size at six-month, one-year, or two-year follow-up exams. Mean follow-up time was 21.6±1.8 months and there were no further follow-ups after two years. During the follow-up the patients were evaluated by US (additionally by MRI for multiple cysts), and serologic tests were not utilized.

Complications were usually minor and transient. Minor complications were fever (n=14, 3.7%), urticaria-like reactions (pruritus, mild skin rash, transient allergic reaction) (n=23, 6.1%), and biliary fistula (n=7, 1.9%). The only major complication was anaphylaxis observed in two cases (0.55%), which quickly resolved by immediate intervention of the anesthesiology team. These two patients had been given albendazole prophylactically. No deaths occurred related to percutaneous treatment per se or its complications. No alcoholemia was seen in any of the cases during or after the procedure.

All patients were hospitalized overnight because of catheterization; 46 patients were hospitalized 2–17 days (mean, 1.6 days) due to complications. Prolonged hospitalization of the patient who stayed in the hospital for 17 days was due to patient’s multiple cysts which required four sessions of the procedure. This patient had urticaria as well.

Discussion

Our results suggest that our novel modified PAIR technique may have easy application, high success rate, lower severe complication rate, and lower cost. In addition, to the best of our knowledge, this is the largest study on percutaneous treatment of hydatid cysts in the literature.

The gold standard treatment of the hydatid cyst remains controversial. In WHO informal working group on echinococcosis 2010 guidelines, it is stated that possible methods for the treatment of the hydatid cyst have not been compared and there is no “best treatment option” (7). Traditional treatment is surgical; however, mortality (0%–6.3%), complication (12.5%–80%), and recurrence (2.2%–22%) rates of this modality are high (8–10). Medical treatment alone is not efficient, but it can be used as an adjunct before or after the percutaneous treatment as a prophylaxis for abdominal dissemination (9, 11). Percutaneous treatment of the hydatid cyst is a reliable, efficient, and comfortable option, which has been used widely in the last two decades. In our study, treatment of the hydatid cyst was performed effectively by single puncture catheterization as a modification of the PAIR procedure.

In a meta-analysis, the success rate of hydatid cyst treatment by the PAIR procedure with albendazole or mebendazole prophylaxis was found to be higher than that of surgery. The success rates of the PAIR and surgery were reported as 95.8% (737/769) and 89.8% (855/952), respectively, and the difference was statistically significant. The success rate was 97.7% (469/480) in our study. In addition, lower recurrence rates were reported in percutaneous treatment compared with surgery (12). In our study, the recurrence rate was 2.2%.

Previous studies in which the patients were given albendazole or mebendazole preoperatively and postoperatively varied in terms of dose and duration of the medication. In studies reporting the length of medical treatment period precisely, patients were given albendazole or mebendazole for a median of 7 days (range, 4 hours to 15 days) before the drainage and for a median of 28 days (range, 4 hours to 6 months) after the procedure. Albendazole 10–20 mg/kg per day or mebendazole 10–50 mg/kg per day have been used in differ-
the catheter is placed into the cyst cavity. Manipulation of the dilator and catheter over the wire during the procedure, for example, may be superior to catheterization by Seldinger technique (via Seldinger needle, or guiding needle) owing to its easier steady within the cavity of the cysts that are located in regions hard to reach such as the dome of the liver, or clogging of the needle tip by membranes during the PAIR steps. Catheterization by Seldinger technique is a more controlled and comfortable method than the PAIR procedure (15). Our method may be superior to catheterization by Seldinger technique (via Seldinger needle, stiff wire, dilator, catheter) owing to its easier application and lower cost.

The potential complications of pre- and postprocedure include fever, hypotensive reaction, vasovagal reaction, nausea, vomiting, biliary fistula/rupture, cavity infection/abscess, peritoneal leakage, subcapsular hematoma, active arterial hemorrhage, intracystic bleeding or gallbladder hemorrhage, pleural effusion/pneumothorax, transient hypernatremia, and other unclassified reversible complications. Lethal anaphylactic shock is the most feared complication of the percutaneous treatment of the hydatid disease. The rates of reversible and irreversible anaphylactic shock were reported as 1.67% (99/5943) and 0.03% (2/5943), respectively in a meta-analysis by Neumayr et al. (16). No fatal anaphylactic shock occurred in our study, but reversible anaphylactic shock was encountered in two cases (0.55%, 2/370).

Surgical treatment of the hydatid cyst is associated with prolonged hospitalization. The length of stay in hospital was reported to be 10 days on average (range, 4.6-15 days) (17, 18). Laparoscopic surgery of the hydatid cyst, which requires a shorter hospitalization than laparotomy, became popular in recent years and the length of hospitalization for this method was reported as 4.66 days (range, 2-8 days) in a meta-analysis (19). The mean length of hospital stay in the literature for percutaneous treatment of the hydatid cyst is 2.4 days (range, 1.6–4.2 days); however, outpatient percutaneous treatment has also been reported (12, 15). In our study, the mean length of hospitalization was 1.6 days (range, 1–17 days), consistent with the previous literature.

The larger series in the literature mostly discuss surgical treatment options, whereas percutaneous treatment series include fewer patients. To the best of our knowledge, our study is the largest series reporting percutaneous treatment in the literature (20).

Our study has some limitations. The main limitation of this study is its retrospective and nonrandomized nature. The fact that we included consecutive subjects with Gharbi type 1 and 2 cysts, but not subjects with Gharbi type 3 or 4 is another limitation. Exclusion of patients with cystobiliary fistulas from PAIR procedure can be considered as another limitation as well.

In conclusion, this study confirms that it is possible to treat hydatid disease in a minimally invasive manner with a new modified PAIR technique. Percutaneous treatment of hydatid cyst with a trocar catheter may be at least as effective and applicable as the standard catheterization technique and it may be an effective alternative to conventional surgical treatment of the hydatid cyst.
PAIR procedure or standard catheterization method, considering its cost-effectiveness and low complication rates.

Conflict of interest disclosure
The authors declared no conflicts of interest.

References