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Treatment of Hepatic Hydatid Cyst in a 7-Year-Old Boy Using a New Type of Radiofrequency Ablation Electrode

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Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Conflict of interest: None declared

Patient: Male, 7
Final Diagnosis: Echinococcosis
Symptoms: Cough • fever
Medication: —
Clinical Procedure: Medical treatment and RFA of liver cyst
Specialty: Pediatrics and Neonatology

Objective: Unusual setting of medical care

Background: Radiofrequency ablation has been established as a treatment method for malignancies and some particular cystic lesions, especially in adults. Experience with radiofrequency ablation in the treatment of hydatid cysts, especially in children, is limited.

Case Report: Although echinococcosis is rare, especially in children, we describe a 7-year-old boy with lung and liver cystic echinococcosis. Diagnosis was established by clinical history and imaging findings and confirmed by positive antiechinococcal antibodies. After 6 months of chemotherapy with albendazole, the liver lesion remained and a radiofrequency ablation under computed tomography guidance was obtained. The procedure was performed with a new type of ablation electrode in order to minimize handling and procedure duration and to achieve the best clinical result in only 1 session. This type of electrode provides the ability to simultaneously drain and ablate the cyst and the ability to monitor the desired ablation temperature in real time.

Conclusions: Our patient is the first pediatric case with hepatic hydatid cyst treated successfully with the use of a new type of radiofrequency ablation electrode under computed tomography guidance.

MeSH Keywords: Child • Echinococcosis, Hepatic • Pulsed Radiofrequency Treatment

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/904432>



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Background

Human echinococcosis is a parasitic disease caused by tapeworms of the genus *Echinococcus*. The 2 most important forms, are cystic echinococcosis (CE) caused by infection with *Echinococcus granulosus*; and alveolar echinococcosis (AE) caused by infection with *E. multilocularis* [1]. Humans are infected through ingestion of parasite eggs in contaminated water, food, or soil or through direct contact with animals. A number of animals act as intermediate hosts of *Echinococcus*. They become infected by ingesting the parasite eggs in contaminated material, and the parasite then develops into larval stages in the viscera. Carnivores are infected through the consumption of viscera of intermediate hosts and act as definitive hosts for the parasite.

Cystic echinococcosis is a rare disease, especially in pediatric patients [2]. According to guidelines [3,4], chemotherapy combined with minimal invasive techniques seems to be the treatment of choice for most cases. Based on evidence provided mainly by an experimental study in animal models [5], we decided to perform percutaneous radiofrequency ablation (RFA) instead of the puncture, aspiration, injection, reaspiration procedure (PAIR) to treat hepatic hydatid cyst. The procedure was performed with a new type of electrode that assures drainage and ablation at the same time.

Case Report

A 7-year-old boy presented to the Emergency Department due to a fever that began 4 days before, severe coughing, and mild

abdominal pain. His parents reported taking the child on a trip to a rural area of Albania a few months ago. Past medical history was otherwise unremarkable. Physical examination revealed crackling in the right upper lung lobe and mild splenomegaly. Laboratory investigation showed mild leukocytosis with eosinophilia (17 000 white blood cells/ μ l, absolute eosinophil count 1585 cells/ μ l). Chest x-ray revealed consolidation involving the right lung upper lobe. Abdominal ultrasound showed mild hepatomegaly and splenomegaly. At part VIII of the liver, a 4-cm cyst was found. He immediately started antimicrobial therapy and underwent a lung computed tomography (CT) scan that revealed a consolidation with air bronchogram in the right lung upper lobe. Lack of response to the empirical antimicrobial medication, the reported travel to an endemic echinococcosis area, and the imaging findings were indicative for cystic echinococcosis, so albendazole (15 mgr/kg) was added to his medication. The diagnosis of echinococcosis was confirmed with enzyme-linked immunosorbent assay (ELISA) detection of IgG antibodies against *E. granulosus* antigens (titer 1/4000).

Reevaluation of our patient 2 months after chemotherapy with albendazole revealed complete recession of the lung hydatid cyst (Figure 1) but no response of the hepatic hydatid cyst. Albendazole treatment was continued. Our patient completed 6 months of chemotherapy medication with albendazole and was then reevaluated. Classification of the hepatic hydatid cyst was made according to World Health Organization (WHO) classification with the use of ultrasound. The cyst presented as uniformly anechoic with fine echoes settled in it, representing hydatid sand, stage CE 1. According to treatment guidelines, our patient had all indications to undergo the puncture,

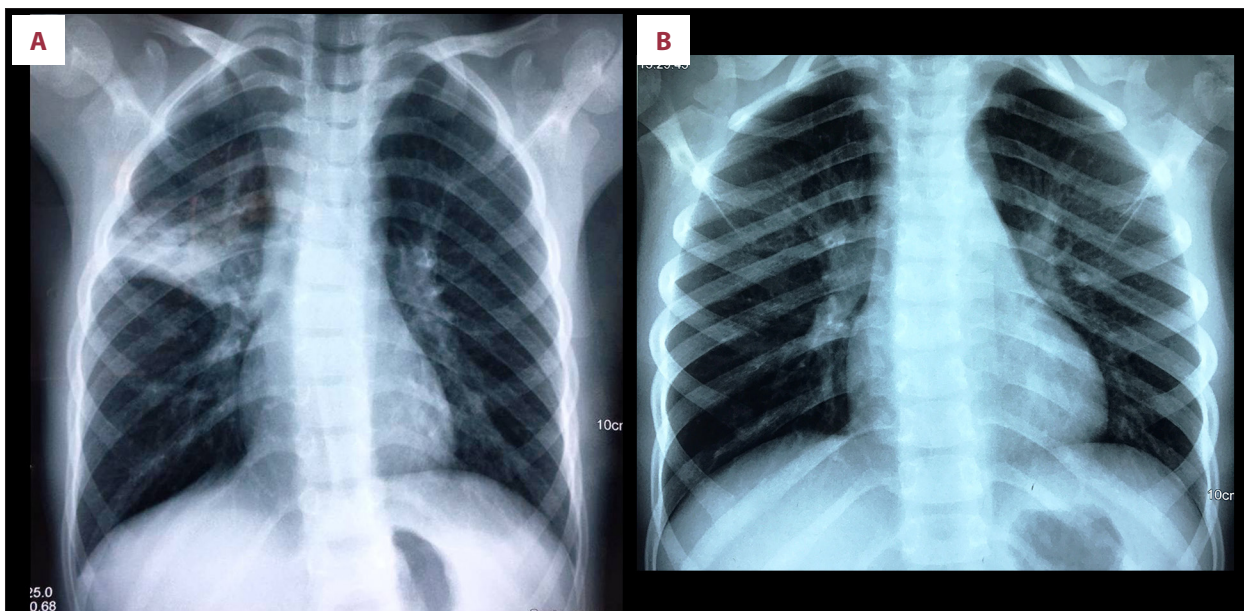


Figure 1. (A) Chest x-ray at the time of diagnosis revealed consolidation involving the right lung upper lobe. (B) Normal chest x-ray 2 months after chemotherapy with albendazole.

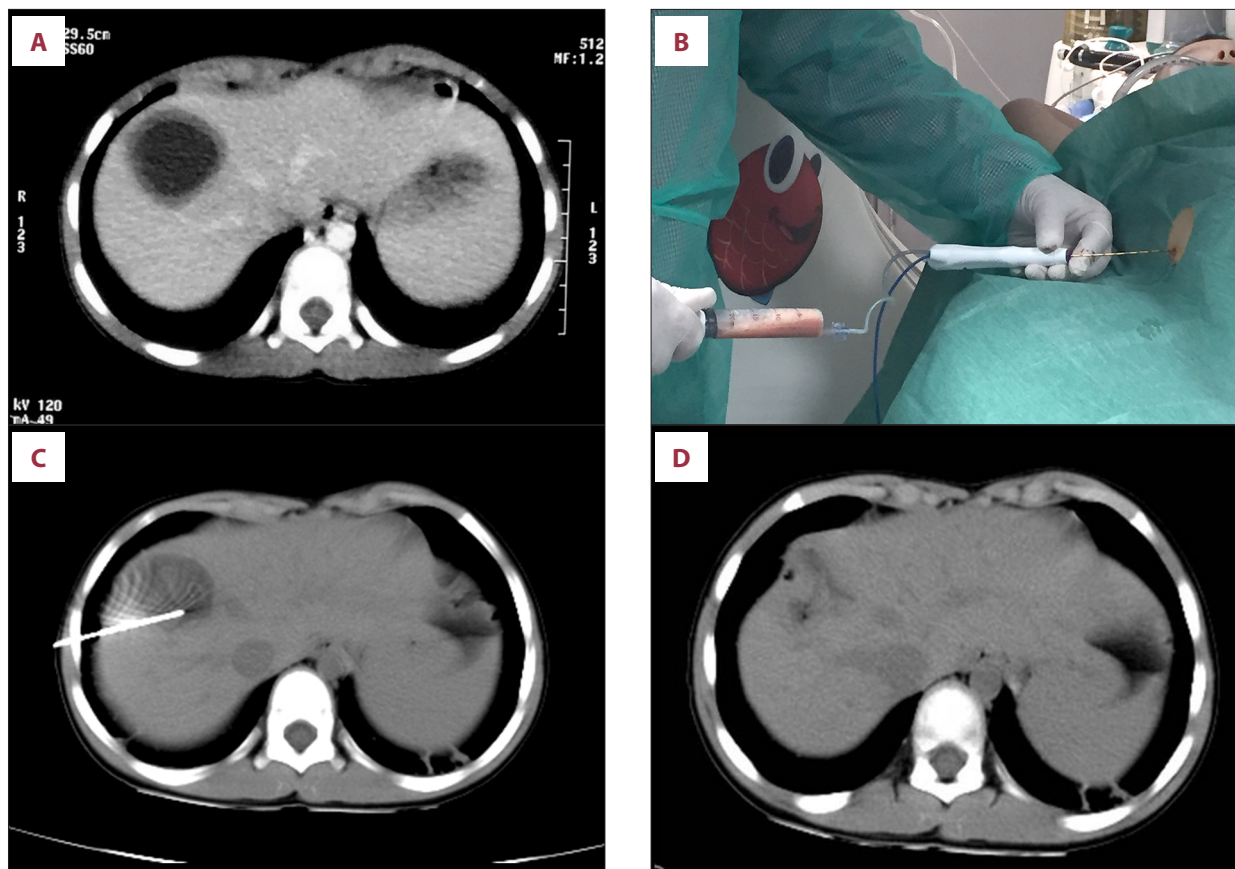


Figure 2. (A) CT image before the ablation reveals a 4-cm hydatid cyst at part VIII of the liver. (B) Patient in supine position with the CystTip electrode inside the lesion, ablating the cyst and simultaneously aspirating cyst contents, (C) CT image displaying the radiofrequency ablation electrode inside the hydatid cyst after 3 min of ablation – minor artifacts (D) CT image immediately after the RFA, indicating decrease in cyst size.

aspiration, injection, reaspiration procedure (PAIR). Instead of PAIR, we decided to perform radiofrequency ablation (RFA) of the hepatic cyst under computed tomography guidance, which is a promising new method. Our patient underwent pre-procedural blood tests, including measurements of hemoglobin concentration, international normalized ratio, partial thromboplastin time, and platelet count. His parents were informed and signed a written consent form before the procedure. The procedure was performed under general anesthesia due to the possibility of anaphylaxis, although he was treated with albendazole for 6 months [6].

The radiofrequency ablation procedure was performed by an interventional radiologist (LT) with 20 years of experience in CT-guided percutaneous interventions.

The imaging modality used for percutaneous electrode guidance was spiral CT (Brightspeed Elite 16 RT, GE healthcare, USA) with a neonatal low-dose scan protocol (80 kV, 33 mAs, slice width 5 mm, feed/rotation 16, kernel B40s, CT dose index 0.9) and the minimum number of scans (n=12). The patient

was placed in supine position and the ablation route was chosen by the interventional radiologist. Ablation was performed with a CystTip™ electrode diameter (1.5 mm, length 15 cm) (RF Medical Co. Ltd, Seoul, Korea) connected to a radiofrequency generator M-3004 (200W Multifunctional Generator®, RF Medical Co. Ltd., Seoul, Korea). This new type of electrode gives the ability to aspirate and ablate a cyst simultaneously (Figure 2). At the same time, we can measure internal temperature and cyst wall temperature in real time by moving temperature sensor. A grounding pad was placed on the patient's skin in an area of good electrical conductivity (typically the thigh) to complete the electrical circuit and avoid burn injuries. Pulse radiofrequency ablation energy was applied for 10 min at 90–110 W. The maximum tissue temperature ranged from 90°C to 110°C. Initially, ablation was performed for 5 min in a full-content hydatid cyst, then aspiration of the contents of the cyst occurred simultaneously with the ablation procedure, which was completed in 10 min.

The ablation was designed to induce coagulation necrosis of the germinal layer of the cyst. Technically successful ablation

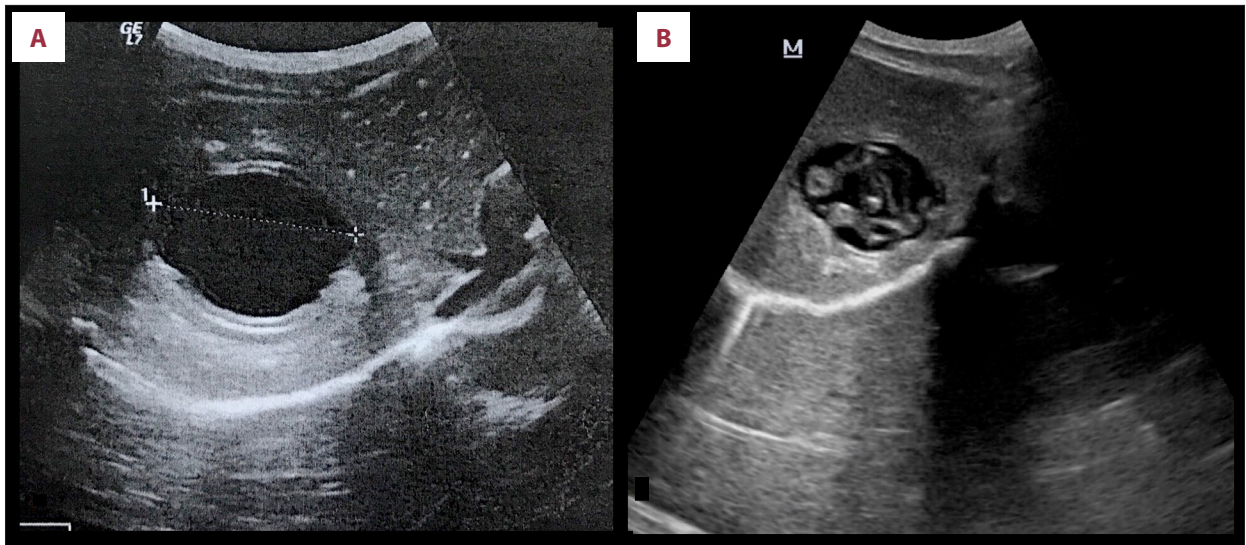


Figure 3. (A) Ultrasound imaging of the hydatid cyst at the time of diagnosis. Uniformly anechoic cyst with fine echoes settled in it, representing hydatid sand, stage CE 1. (B) Follow-up 3 months after RFA reveals an inactive cyst in stage CE4.

occurs when a decrease in cyst size is noted. To evaluate the immediate response of the lesion to ablation, and to identify any complications, a spiral CT was performed immediately after the procedure. A post-procedural CT revealed decreased cyst size <2 cm due to overlapping of the cyst wall, and no complications were noted. Our patient was hospitalized for 24-h monitoring and was discharged the next day, as no complications occurred.

Follow-up was performed with clinical examination and ultrasonography at 1 month and 3 months after the ablation when the cyst was classified as CE4 stage, with diameter of 2.8 cm. (Figure 3). One month after the ablation, the absolute eosinophil count was normal (450 cells/ μ l) and echinococcal antibodies had decreased (titer 1/780). No recurrence of the ablated cyst occurred during 3-year follow-up.

Discussion

Cystic echinococcosis (CE) is a zoonosis caused by *Echinococcus granulosus*. This tapeworm can occasionally cause illness in intermediate hosts, such as humans. Cysts contains thousands of protoscoleces and many daughter cysts. Cysts act as a space-occupying lesions on adjacent tissues. Cyst fluid contains parasite antigen that can sensitize the host [1]. Urgent anaphylactic reaction takes place if a cyst ruptures spontaneously or during surgical procedures [6]. Mean annual incidence rates vary from less than 0.01 cases per 100 000 inhabitants to higher levels in endemic areas, such as Mediterranean countries. In Greece, the mean annual incidence rate is estimated at 0.16/100 000 inhabitants, whereas in Albania it is estimated at 0.6/100 000 inhabitants. The organs most frequently affected

are the liver and lungs [7,8]. Treatment is determined according to the standardized ultrasound classification as proposed by the WHO [3,7,9]. CE1 and CE2 are active cysts containing viable protoscoleces. CE3 has been subdivided into CE3a (detached endocyst) and CE3b (predominantly solid with vesicle cysts). CE4 and CE5 are inactive. CE4 has mixed hypo- and hyperechoic content with absence of daughter cysts, giving the appearance of a ball of wool (wool sign), and CE5 has arch-like, thick, partially or completely calcified walls. In CE1 and CE3a <5 cm, the treatment of choice is albendazole [10], whereas in cysts with diameter >5 cm, albendazole is combined with conventional percutaneous procedures such as PAIR [11]. PAIR with ethanol or hypertonic saline is established as a therapeutic option for hepatic lesions, while experience in lung echinococcosis is limited [12–15]. Surgery [16], and in some cases PAIR, may cause post-procedure complications such as cyst infection or abscess [10], biliary fistulae [16], and chemical cholangitis due to injection of hypertonic saline or ethanol during the procedure.

The use of RFA as a treatment for hydatid cysts instead of PAIR was first reported by Brunetti [17] and then by Bastid [18]. CT-guided RFA in children is a well-known minimally invasive method to treat osteoid osteomas and lesser malignancies [19]. RFA causes protein denaturation via high temperatures exceeding 60°C. Thus, destruction of the germinal layer is achieved without injection of any scolicidal agent, minimizing the possibility of complications. RFA is easy to perform; it consists of just 1 step with 1 puncture, and aspiration, injection, and reaspiration are not needed.

Brunetti [17] reported the treatment of 5 patients with 6 CE3b hepatic cysts. The procedure was effective in all patients.

Efficacy assessment was based on the presence of scolices in post-procedure cyst fluid microscopic examination. Relapse was noted 12 months after the procedure in cases where full ablation was not achieved due to the cyst structure. Complications, specifically abscess, occurred in 1 patient. The poor results achieved in the study may be due to application of the method in an inappropriate group of patients. In our case, unlike Brunetti et al., we evaluated RFA efficacy according to decreased cyst size, ultrasonographic staging impairment, and decrease in eosinophils and echinococcal antibodies. Bastid et al. [18] treated 1 patient with a CE2 hepatic cyst; in 6-month follow-up, no mid-term complications or relapse occurred. Limited data present RFA as an effective and safe procedure, with a high success rate in treatment of cystic hepatic lesions [20–23]. No preliminary animal studies were performed before the application of this technique in human CE. Lamonaca et al. [5] published an *ex-vivo* experimental study to evaluate RFA for treatment of cystic echinococcosis in animal models. Nine liver and 8 lung cysts were treated with RFA. The success rate was 100% in both hepatic and lung cysts. The study revealed the efficacy of RFA in treating explanted hydatid cysts. Cyst volume reduction after PAIR or pericystectomy is estimated to be 65% or more at 6–19 months after treatment [24]. Lamonaca observed the same level of volume reduction immediately after RFA treatment. Evaluation of RFA effectiveness in our patient confirms the results of the study performed in animal models [5]. We noted immediate decrease in cyst volume and satisfactory immediate necrosis

of the cyst wall. If we did not have the ability to use the ablation electrode called CystTip, an alternative would have been use of a conventional RFA electrode with a second aspiration needle in place, to aspirate it first and then ablate (with the RFA electrode still in place), but the technique can cause inappropriate ablation and an unnecessary second puncture.

Complications, such as biliary fistula, after liver hydatid cyst surgery or PAIR [25] are decreased with RFA because coagulative necrosis is accomplished without detaching the endocyst from the pericystium.

Conclusions

We described a pediatric patient with hepatic hydatid cyst treated with a new radiofrequency electrode called CystTip. This type of electrode ensures simultaneous drainage and ablation of the cyst, thus minimizing interventions needed and procedure duration, thereby reducing complications. Hydatid cyst treatment is achieved in a single session, since RFA causes thermal necrosis of the entire cyst wall. Furthermore, RFA avoids the use of hypertonic saline or ethanol, thus minimizing the risk of complications such as chemical cholangitis.

Conflict of interest

None.

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