



## Treatment of Liver Hydatidosis with Puncture, Aspiration, Injection, and Reaspiration Technique: Two Case Report Hydatidosis Treatment with PAIR.

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### ABSTRACT

Liver hydatidosis is a considerable health problem in the developing countries, such as Iran. There are different therapeutic approaches for the treatment of this disease including; laparoscopy, open surgery, as well as puncture, aspiration, injection, and reaspiration (PAIR) technique. Herein, we presented the case of two pediatric patients with multiple hydatid cysts in the liver, who were treated through the PAIR technique with no complication or relapse in the follow-up.

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### 1. Introduction

Hydatid disease is an infection that most commonly affects the lung and liver [1]. This disease arises from a parasite that affects both human and herbivores in its larval stage (*Echinococcus granulosus*) and can finally develop cystic lesions in any abdominal organs. In the pediatric patients, this disease mostly affects the lungs [2], and there are rare cases of multiple liver lesions in this population [3].

The symptoms and signs of hydatid disease depend on the location and size of the cysts [4]. The patients inflicted with liver hydatidosis are usually asymptomatic, and many of the cysts are incidentally detected on ultrasonography [5]. Ultrasound imaging is the main and best diagnostic method for this condition; in addition, the serologic tests are mostly applied for assurance [4]. The hydatid cyst consists of three layers. The outer layer or pericyst represents the host cell response to parasite. The other two layers are middle laminated membrane and inner germinative layers, which belong to the parasite itself [6].

The cyst fluid is crystal clear and contains daughter vesicles and antigenic proteins [7]. There are various therapeutic methods for the treatment of the patients with hydatidosis. These methods include surgical, laparoscopic, percutaneous, and pharmaceutical therapy [8]. The puncture, aspiration, injection, and reaspiration (PAIR) method is a therapeutic approach, which is performed by the guidance of ultrasound [9].

The application of this method in the therapeutic process results in excellent outcomes and minimal complications [10]. Surgery is the most commonly used and known method of treatment for hydatid disease; nonetheless, it leads to significant morbidity, hospital stay duration, and cost [11].

Although PAIR has been demonstrated to have excellent treatment outcomes, surgery is still the treatment of choice for the patients with hydatid disease.

In this regard, PAIR procedure is mostly applied for the surgically inaccessible sites [4] or for the patients who cannot undergo surgery. Additionally, this technique is contraindicated for inaccessible cysts or inactive and calcified cystic lesions [12]. In this case report, we presented two children with multiple hydatid cysts in the liver who showed excellent response to PAIR therapy without any complication or recurrence in the one-year follow-up.

### 2. Case presentation

#### Case 1

A 6-year-old female patient presented with failure to thrive, abdominal bulge, and hepatomegaly that were initiated six months before admission. The liver hydatidosis diagnosis was confirmed by ultrasonography (Fig 1A), according to which the diameter of the biggest liver cyst was 96 mm. Some liver cysts were complicated and solid, and most of them were univesicular. No hydatid cyst was found in other thoracoabdominal organs. After two weeks of outpatient albendazole prescription, the PAIR procedure was performed for the seven cysts under general anesthesia with ultrasound guidance, which led to the successful aspiration of all cysts. The patient showed no anaphylactic reaction or other complications.

#### Case 2

A 10-year-old female patient presented with sudden loss of consciousness. The brain computed tomography (CT) scan revealed single brain hydatid cyst. Her physical examination showed hepatomegaly. The patient was diagnosed with liver hydatidosis through ultrasonographic examination, revealing

two complicated right renal hydatid cysts. The diameter of the biggest liver cyst was 50 mm, and the renal and some of the liver cysts were complicated and solid. The majority of the liver cysts were univesicular. Furthermore, eosinophil count was estimated as 20. In the first step, the PAIR procedure was implemented for 11 univesicular cysts under general anesthesia with ultrasound guidance.

After one month, the follow-up sonography revealed one univesicular active hydatid cyst with a diameter of 46 mm in the right liver (Fig 1B). At this time, the eosinophil count dropped to 9. Subsequently, the PAIR procedure was performed for the mentioned cyst. The patient showed no anaphylactic reaction or other complications in both steps.

In the present study, two cases of hepatic hydatidosis were diagnosed on the basis of the clinical and typical ultrasonographic findings revealing either univesicular or multivesicular cysts in the liver. Prophylactic albendazole administration was initiated before the implementation of the PAIR procedures for two weeks. The patient received a close monitoring for the emergence of such complications as anaphylaxis, bronchospasm, and laryngeal edema. The PAIR technique was performed only for the univesicular cysts.

Percutaneous drainage was implemented under general anesthesia and continuous ultrasonographic guidance (Esoate Mylab 50, Italy). The PAIR procedure was initiated with the puncture of the cysts by 18-22 gauge needle (Gray Angiocath and Shiba needle), which was inserted into the cyst under ultrasound guidance (Fig 2). The suction of the cyst was performed through the instillation of 15% hypertonic saline for 15 min and cyst reaspiration.

Only hypertonic saline was used to wash out the cyst contents and kill scolices. The real-time sonography showed a gradual decrease in the cyst size and the separation of the endocyst (germinal layer) from the pericyst. All of the available univesicular cysts were treated in one session, except for one residual univesicular cyst that was managed in the second session. On the third postoperative day, the patients were discharged in good condition.

After the drainage of the univesicular cysts, the patients were prescribed to use oral albendazole for one year since some of the multivesicular cysts were intact. The follow-up ultrasounds were performed 1 and 3 days, 1 week, and 1, 3, 6, and 12 months after performing the PAIR procedure. The signs of the inactive cyst included decreased dimension of the cysts, septation of the contents, irregularity in the walls of the cysts, and separation of endocyst from pericyst (Fig 1 C). None of the treated cysts disappeared completely; however, all of them became inactive. After one year, both patients showed an improvement in their clinical conditions. Additionally, the first patient had a significant weight gain.

### 3. Discussion

Hydatid disease is a parasitic infection that has two definite hosts, namely dogs and carnivores. Human beings are the accidental hosts, and hydatid cysts can affect almost any organ in body [4]. This infection is still a public health problem in many parts of the world, especially in the underdeveloped and developing countries [1]. The symptoms and signs of this disease depend on the involved organ, complications, and growth stage. Hydatidosis can vary from being asymptomatic to having non-specific symptoms, leading to mortality or severe morbidity [7].

The radiologic features of this condition have a wide spectrum varying from simple cystic lesion to a solid one, and the lesions can be single or multiple.

Ultrasonography, CT scan, or magnetic resonance imaging can be used for the detection and evaluation of the cysts [7]. Imaging (ultrasonography as the modality for the abdominal cysts) is the main and best diagnostic method, which is used to perform the PAIR procedure [4].

Despite the presence of various therapeutic approaches for the treatment of hydatidosis (e.g., surgical, laparoscopic, percutaneous, and pharmaceutical therapy) [8], surgery is still the first method used for the management of this disease. However, this procedure can lead to mortality and morbidity, especially in the patients with multiple and disseminated lesions like our cases.

In a study, Daali et al. investigated the efficiency of surgical technique in the treatment of multiple hydatid cysts of the liver. They reported that this technique resulted in a high rate of morbidity, few number of recurrence, and also one mortality due to the nearly total obstruction of the liver parenchyma, which caused hepatic insufficiency [13]. The surgical removal of multiple hydatid cysts in the pediatrics is rarely reported, and it is recommended to make personalized treatment decision in this regard [14].

The percutaneous drainage of the liver hydatid cyst was first reported in 1985 [15]. Since then, multiple studies have shown the equality or even superiority of this method to the surgical approach [16]. This procedure has been reported to have no significant complications. Nevertheless, the major risk of the percutaneous approach is the spillage of the cyst fluid during the intervention, which could be minimized by using ultrasound or CT scan for monitoring the position of the needle and interaperitoneal approach [15]. Moreover, the patients treated with PAIR show excellent outcomes in the long-term follow-up [17].

The surgical complications vary from 8-16% in the uninfected cysts to 89% in the infected cysts. The most frequent complication is sepsis. Moreover, the mortality rate in the patients who undergo surgery is estimated to be about 0.5-3% [15]. Furthermore, disease recurrence is lower in the percutaneous treatment as compared to the open surgery (3.5% versus 16%) and is almost equal to that of laparoscopic surgery [18].

Laparoscopic surgery seems to be effective and safe for the accessible sites. However, it has its own disadvantages, such as the limited area of manipulation and high risk of spillage during puncture [18]. In our patients, regarding the high risk of surgical treatment due to the extensive dissemination of the cysts and abnormal liver function tests (in one of the patients), it was reasonable to choose the PAIR procedure instead of surgical interventions.

Since the pharmaceutical therapy is rarely effective and has high side effects and relapse rates, the application of percutaneous approach seems to be more effective with lower morbidity, mortality, also cost than the surgical treatment [19]. In the present study, we achieved a good outcome by the employment of the PAIR procedure along with pharmaceutical treatment. These two cases show that PAIR can act as an excellent substitute surgical treatment of numerous hydatid cysts in liver who seem to undergo higher morbidity by surgical treatments. They have excellent response to PAIR therapy with no complication or recurrence in one year follow up.

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**Foot notes****Implication for health policy makers/practice/research /medical education:**

In order to better recognition of best method for hydatid cyst treatment, especially in cases with multiple cysts. It would be interesting for radiologists, surgeons and patients.

**Authors' Contributions**

Seyed Ali Alamdaran: collecting the data and writing the article

Jahanbakhsh Hashemi: writing and editing of the article

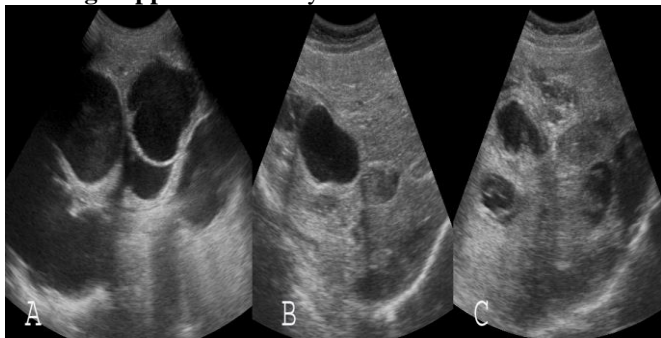
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**Fig 1. Ultrasound images of liver hydatidosis before and after treatment: A) liver hydatidosis with variable size before treatment. B) One month follow up sonography showed one unilocular active hydatid cyst with multiple residual inactive hydatidosis. C) After one year sonography showed multiple residual inactive hydatidosis.**



**Fig 2. photographic view of patient during treatment; three gray Angiocath and one Shiba needle inserted in liver hydatid cysts. Fluid oozing from Shiba needle during aspiration step is obvious. Closed Angiocaths is due to waiting time after injection step.**

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