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CASE REPORT

# Primary hydatid cyst in the posterior thigh, and its percutaneous treatment

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**Abstract**
Hydatid cyst (echinococcosis) is an endemic parasitic disease, usually encountered in those dealing with agriculture and livestock. The most frequently affected organs are the liver and the lungs. The disease is very rarely encountered in soft tissues. Diagnosing a soft-tissue hydatid cyst may be challenging unless the mass possesses the characteristic features of a hydatid cyst. Soft-tissue hydatid cysts may be treated percutaneously, just like those encountered in the liver. In this case report, we present the radiological findings and modified percutaneous aspiration–injection–reaspiration (PAIR) treatment of a hydatid cyst located in the posterior aspect of the thigh.

**Keywords** Hydatid cyst · Percutaneous treatment · PAIR · Soft tissue · Skeletal

## Introduction

The hydatid cyst is a zoonotic parasitic disease. The most frequently encountered type of the parasite in humans is called *Echinococcus granulosus*. The parasitic eggs are taken in orally and then transferred to the portal system. These eggs are usually kept in the liver, ultimately leading to an infectious process. The second most frequently affected organs in the human body are the lungs, as the parasitic eggs not arrested in the liver migrate through the pulmonary circulation. The liver and the lungs retain these parasitic eggs like a filter and prevent their access to the systemic circulation. Eggs that inadvertently escape these filtering mechanisms gain access to the systemic circulation and may cause infections in various distant organs, such as the spleen, kidneys, bones, heart, and even the brain. The skeletal system and soft-tissue compartments are rarely involved, and infections in these regions are usually due to a secondary extension after its hematogenous spread [1]. In this case report, the radiological findings and percutaneous treatment of a very rare soft-tissue hydatid cyst are presented, together with a thorough literature search.

## Case report

A 32-year-old male shepherd presented with a growing mass in his thigh. He revealed a history of trauma that he had undergone 2 years previously. He reported that a painless mass, which started to grow 6 months following the trauma, had recently become substantially larger with occasional pain. His physical examination revealed no specific findings other than the rather soft mass in his thigh. Ultrasound uncovered an anechoic cystic mass with the dimensions 11 × 9 cm. Magnetic resonance imaging (MRI) examination disclosed a mass, which had been reported to resemble a hematoma. The patient was sent to our Interventional Radiology (IR) Department for further diagnosis and cyst aspiration. The cystic mass was evaluated and reexamined with ultrasound. Then, an intervention was performed to obtain a sample of the cystic fluid material using a 21-G needle. The suspicion of a hydatid cyst arose immediately, upon recognizing that the cystic fluid was highly pressurized and like clear water. Thus, an aspiration of 200 $cm^3$ was performed, with the purpose of

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depressurizing the cystic mass. An ultrasound check done during the aspiration process revealed a detaching germinative membrane, together with echogenic foci, which were thought to resemble hydatid grains (Fig. 1).

The patient was prescribed albendazole (10 mg/kg/day) and was scheduled for percutaneous treatment the following week. After a week of albendazole therapy, the patient came to the IR unit, prepared for the percutaneous intervention. The patient was administered steroids and H-1 receptor blockers to minimize the risk of anaphylaxis, and he was placed under the full control of the anesthesiology team, which administered conscious sedation. Under these circumstances, percutaneous aspiration–injection–reaspiration (PAIR) therapy was administered, with the help of ultrasound and fluoroscopy. A cystogram was obtained, following the half-emptying of the cyst material. No contrast medium leak from the cystic mass was noted (Fig. 2). Next, a 10-F drainage catheter was placed in the cyst, the whole cystic content was aspirated, and a 30% hypertonic NaCl solution was introduced into the cystic cavity. After a 10-min waiting period, the cystic contents were reaspirated, and a portion of the germinative membranes was evacuated through the catheter (Fig. 3). The patient was sent home, with the catheter in place. The next day he returned to the IR Unit, and the cyst was rewashed with hypertonic saline. Then, the catheter was removed. The patient was discharged with a prescription of albendazole (10 mg/kg/day, to be divided into two portions) together with a prophylactic antibiotic.

The patient revisited the hospital with fever and swelling of the thigh, 5 days after his discharge. He stated that he had returned to work immediately following his discharge from the hospital, owing to social and economic factors, and that he neither attended the wound nor complied with the demands of the prescription. The situation was diagnosed as an abscess, because of the clinical findings and swelling of the lesion site. MRI supported the diagnosis because of T2 hyperintense signal in the muscle, skin, and subcutaneous tissues with fluid extending along the deep fascial planes and intermuscular septi (Fig. 4), and the patient underwent an operation. During the operation, the abscess was emptied, and all remaining components of the mass were evacuated (Figs. 5, 6).

## Discussion

Hydatid cyst is a disease that is particularly prevalent in farming and livestock areas, where preemptive healthcare measures are rather low [1, 2]. The infection is spread to the main hosts, namely dogs and other carnivores, when they eat the infected internal organs of the intermediate hosts, for example, sheep, goats, and swine. Infection of the human body happens when the eggs, which are discharged within infected dog feces, are taken in via dirty hands, water, and food. The parasitic larvae (scolices) attach to the human intestines by utilizing

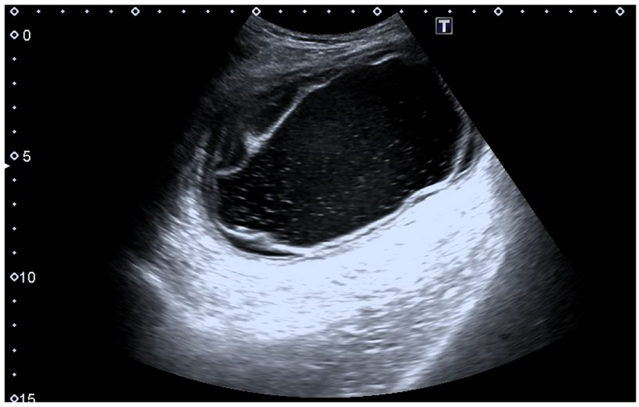


**Fig. 1** Ultrasound reveals detaching germinative membrane, together with echogenic foci, which were thought to resemble hydatid grains

their hooks. Most of the larvae give rise to an infectious process in the liver, whereas those that escape the liver cause an infection in the lungs. Larvae that manage to escape even the lungs, may reach distant organs, such as the spleen, kidneys, skin, muscle tissue, retroperitoneum, bones, heart, and the brain, and cause infections at those sites [1, 3]. The musculoskeletal involvement rates have been reported to be between 1 and 4% [2]. Within the musculoskeletal system, the proximal muscles of the lower extremities are the most frequently encountered sites, and this has been attributed to the high volume and rich vascularization of these muscles [4]. It is well known that the most frequently affected organs are the liver and the lungs [5, 6]. Thus, when the routine ultrasound and computed tomography (CT) examinations of our patient had proved no lesions in these organs, his thigh lesion had not aroused a specific suspicion of a hydatid cyst, and the diagnosis could only be made retrospectively, following the initial aspiration of the cyst. No examples of an association with trauma have been reported in the literature. However, our patient gives a history of trauma 2 years previously, and we think that this

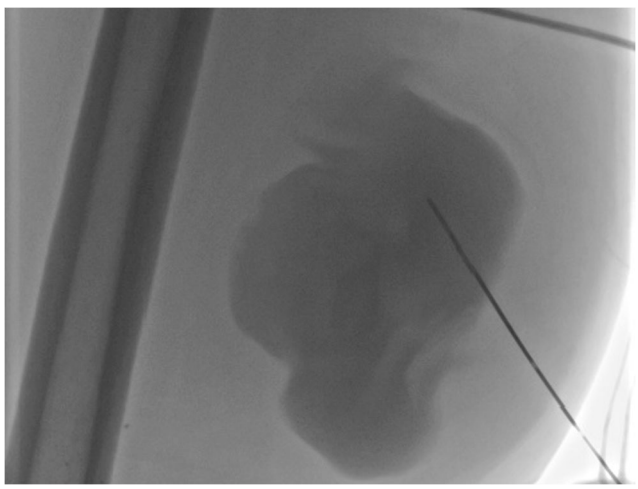

**Fig. 2** Cystogram demonstrates that there was no contrast medium leak from the cystic mass

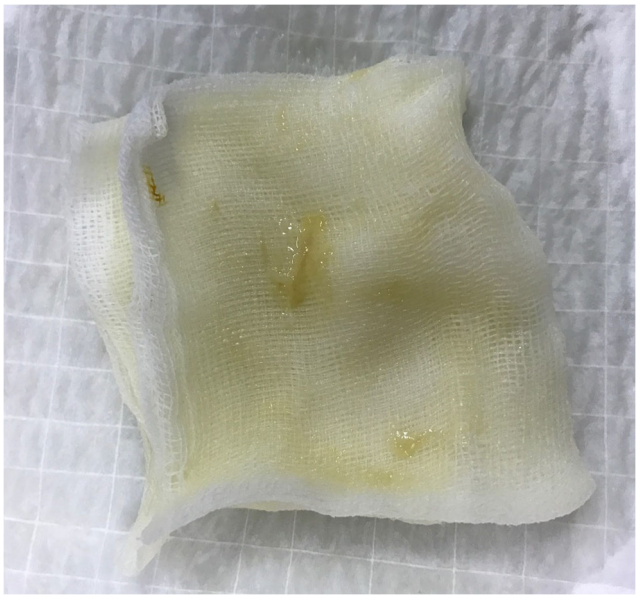

**Fig. 3** A portion of the germinative membranes was evacuated through the catheter

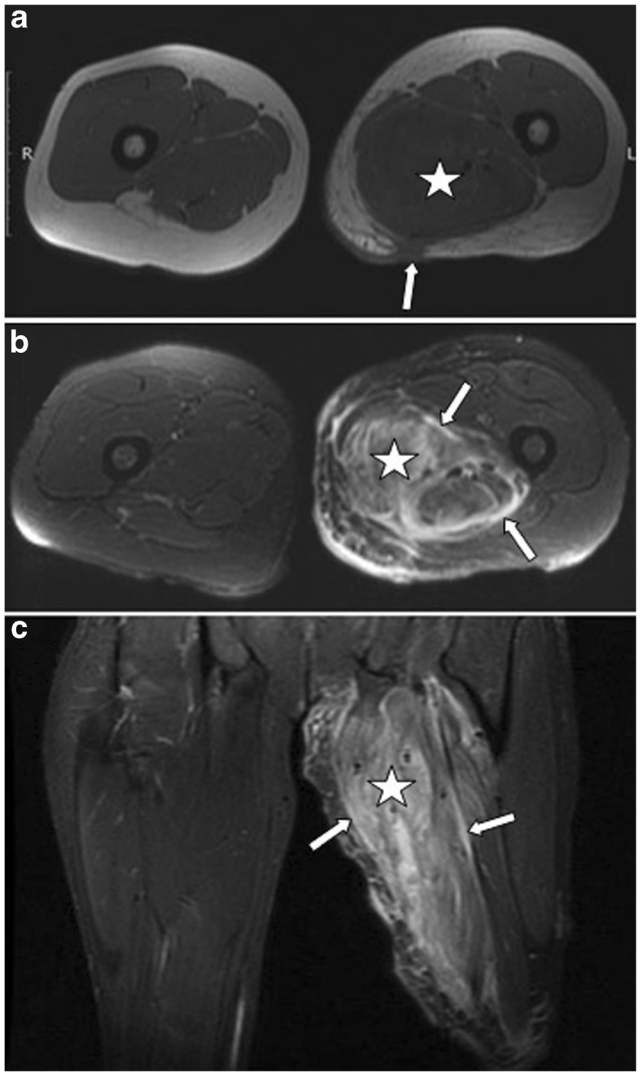


**Fig. 4** **a** Axial T1-weighted MR image shows edema and thickening of the subcutaneous tissues and muscles (*asterisk*) with trace of our previous interventional procedure (*arrow*). **b** Axial T2 fat-saturated and **c** coronal inversion recovery MR images show high signal intensity along the fascia (*arrows*) and show a large area of hyperintensity (*asterisks*) in the muscles

trauma has had some implication as a predisposing factor in his situation.

Serological tests are not always helpful [7]. No serological tests were done in our patient, because the initial diagnosis of the mass was a hematoma, and we were able to make the diagnosis only after the first aspiration of the rock water content of the cyst. Serological tests may remain positive in endemic regions, even years after treatment [8].

The characteristic radiological findings of hydatid cysts are multiple daughter cysts, the two-layer germinative membrane, and the drooping lily sign, which arises because of the collapse of the germinative membrane [9, 10]. It is emphasized that the first modality of choice in the diagnosis of soft-tissue echinococcosis should be MRI [11]. MRI has a very high contrast resolution, and it is capable of demonstrating the regional dissemination of the lesion, together with its relation to the important vascular and nerve elements. Typical MRI findings of hydatid cysts comprise the endo- and ectocysts internally, and the pericyst externally. The internally located endo- and ectocysts cannot be discriminated by MRI, and they appear as a single layer. Conversely, the externally located pericyst is seen as a distinct layer, so these three membranes give the appearance of a two-layer cystic wall, which is a characteristic MRI finding of the hydatid cyst. The internal layer is the germinative membrane itself, and it is seen as a hypointense layer on T2-weighted MRI, although the externally located third layer represents the response of the host and is highly vascularized, thereby appearing hyperintense on T2. The pericyst shows enhancement in contrast series [12]. At retrospective evaluations, the pericyst looked rather hyperintense on T2-weighted images, but still, there was difficulty in diagnosing the lesion, because it lacked the characteristic findings of hydatid cysts, which are mainly the cartwheel appearance, the honeycomb pattern, and the detached membrane sign.

Pre-operative diagnosis is of utmost importance in cases of soft-tissue hydatidosis because these cystic masses are clinically hard to discriminate from other soft-tissue tumors [1, 5]. A biopsy is typically the first-hand procedure in the diagnosis of soft-tissue masses, but in cases of echinococcosis, it may be a source of problems. The issue is that a biopsy performed on a hydatid cyst may cause the vital scolices to enter the blood circulation and cause a fatal anaphylaxis or give rise to the infection of various other tissue compartments [5]. For these reasons, a biopsy is contraindicated in cases of hydatidosis [5].

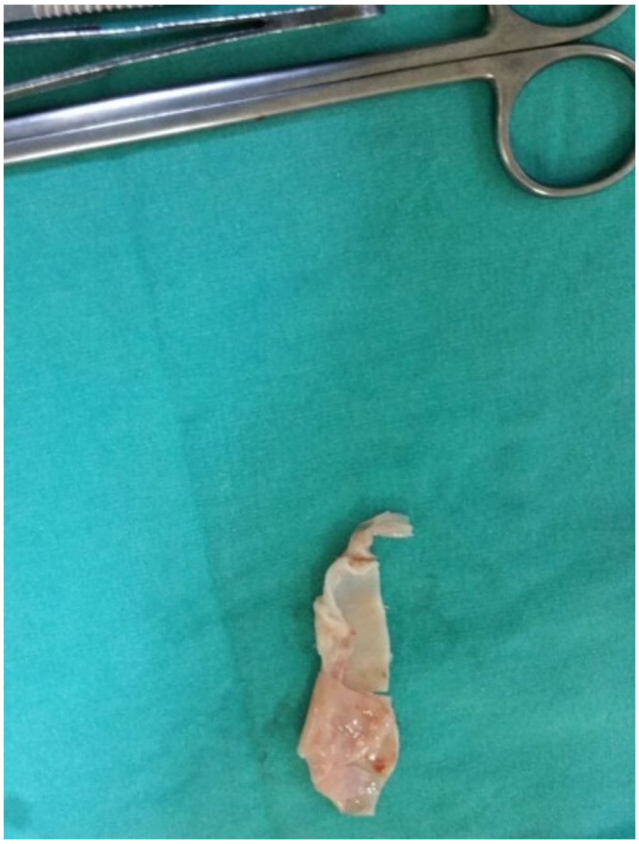

**Fig. 5** Germinative membranes, which were evacuated operatively

True cystic lesions (synovial and ganglion cysts, distended bursae, skin appendage lesions, lymphatic malformation, hydatid cysts), cyst-like lesions (necrotic soft-tissue tumors, myxomas, glomus tumors, peripheral nerve sheath tumors, hemangiomas, and vascular malformations) and Morel–Lavallee lesions (seromas, hematoma, closed lacerations, and other superimposed infections) should be considered in the differential diagnosis of soft-tissue cysts in the thigh region [13, 14]. Synovial cysts are a result of herniation of the synovial membrane. Ganglion cysts contain fluid or mucinous material and have a fibrous lining. Synovial and ganglion cysts demonstrate similar appearance on MRI [13]. Bursae are cystic lesions containing synovial fluid that reduces friction between moving structures such as tendons, ligaments, bone, and skin. Skin appendage lesions are usually diagnosed clinically, but sometimes they are seen incidentally on MRI, having a truly cystic appearance. Lymphatic malformations occur frequently in the pediatric population and seldom in the thigh. Hemangiomas and vascular malformations may manifest as soft tissue with a cystic appearance [13]. Typically, intramuscular myxomas are well-defined ovoid lesions with homogeneous fluid like signal intensity on MRI [13]. The review article by Diviti et al. [14] described Morel–Lavallee lesions as post-traumatic soft-tissue degloving injuries. It was indicated that these lesions could escape from the eye, usually in early-stage evaluations.

Irrespective of whether they are treated surgically or percutaneously, hydatid cyst patients require a medical therapy regimen, consisting of scolicidal drugs and prophylactic antibiotics, both before and after the procedure. The most frequently used scolicidal drugs are mebendazole and albendazole, which are both benzimidazole derivatives. These drugs act by preventing the utilization of glucose by the parasite and thus prevent the formation of adenosine triphosphate [15]. Albendazole therapy must be started preoperatively and must be continued for 3 months postoperatively. Furthermore, a 3-week course of albendazole usage must be followed by a week of interval, and liver function tests must be performed during this time. The proposed dose for albendazole is 10–15 mg/kg/day, and administering this dose in two equal parts is recommended. It has been shown that cyst vitality does not cease with therapies shorter than 3 months, and a minimum of 3 months of albendazole therapy has become a routine prerequisite [16].

Some previous studies have emphasized that the therapy of soft-tissue hydatid cysts is conclusively surgical [5, 17]. Until

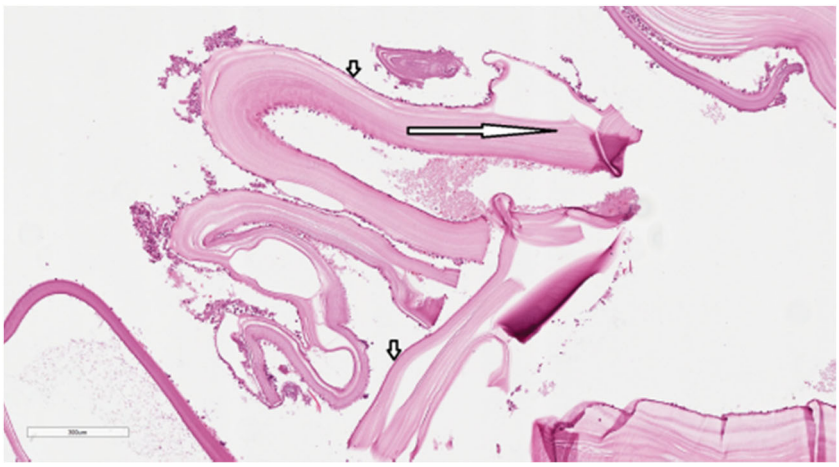

**Fig. 6** The wall of the hydatid cyst consists of a laminated membranous structure (*long arrow*), which is lined by the germinal epithelium (*short arrows*). H&E, ×100

the 1980s, puncture of hydatid cysts was a preferentially avoided procedure, owing to the risk of leakage and anaphylaxis. In 1981, Niron and Özer [18] performed a coincidental and problem-free puncture of a hydatid cyst, which had been previously diagnosed as a simple cyst, leading to the basis and reason for diagnostic and therapeutic puncture procedures of hydatid cysts. Akhan et al. [19], who are among the pioneers of percutaneous treatment procedures of hydatid cysts, have stressed that percutaneous therapy of hydatid cysts is a very reliable and effective means of treatment, and it must be considered a highly viable alternative to surgery. The authors also highlighted that no serious complications were encountered during these percutaneous procedures [19]. The percutaneous approach to therapy, which has been utilized in hepatic hydatid cysts, has proven to be almost equally as effective in many other body sites infected with echinococcosis. In this article, we present a case of hydatidosis with the cystic mass located in the thigh region. The cystic mass was treated percutaneously using the PAIR method.

The acronym PAIR consists of the initial letters of the following words: puncture (entrance to the cystic cavity), aspiration (evacuation of the cystic content), injection (the intracystic administration of a scolicidal, i.e., sclerotic substance), and reaspiration (the complete emptying of the cystic contents) [20]. During the aspiration of the cystic content, some of the cystic fluid remains in the cyst with the purpose of keeping the needle inside the cyst, and then a scolicidal substance is injected into the cyst through this needle. Reaspiration is carried out after waiting for 10–15 min. Because of the risk of dislocating the needle out of the cystic cavity, all cyst aspiration procedures, scolicidal substance injection, and reaspiration must be performed under ultrasound guidance. It may sometimes involve considerable difficulty, both radiological and serological, in differentiating a simple liver cyst from a type 1 hydatid cyst. In such circumstances, the most reliable finding is the demonstration of the detachment of the germinative membrane during cyst aspiration [8]. Similarly, in our patient, the diagnosis could only be made during the aspiration phase. The Gharbi Classification types 1, 2, 3, and (fluid containing) 4, are accepted as active, and they are treated percutaneously, because they may grow over time, or rupture with trauma, or become infected. The PAIR method is preferred for hydatid cysts with diameters less than 5–6 cm and of types 1 and 2, whereas cysts with relatively larger diameters, complicated cysts, and cysts of types 3 and 4 require a modified PAIR procedure. In children and peripherally located type 1 cysts, the upper limit for PAIR may be elevated up to 8 cm in diameter [8]. There are very few articles in the literature concerning the percutaneous treatment of soft-tissue hydatid cysts. Akhan et al. [19] reported 4 cases, in which 6 cysts received percutaneous treatments. The authors mentioned that no major complications were encountered in these cases and no recurrences were detected after a 34.8-month follow-up [19]. Likewise, Yucesoy et al. [21] indicated that although it requires a long catheterization, the percutaneous treatment of giant, soft-tissue hydatid cysts is a very effective means of therapy.

In conclusion, it should be emphasized that hydatid disease must always be on the list of considerations in the evaluation of soft-tissue masses in endemic areas and people dealing with farming and livestock. In such situations, diagnosis must be assured, through imaging modalities before biopsy or surgery. Even though there are currently only a few literature reports on the issue, percutaneous therapy is a safe and reliable alternative to surgery. Especially in large cysts, percutaneous therapy must be performed by catheterization, and the catheter must be kept in place until the daily discharge falls below 10 $cm^3$. During this time, special care should be taken to maintain cleanliness of the procedure area, and antibiotics must be used to prevent the formation of an infection.

## Compliance with ethical standards

**Conflicts of interest** The authors declare that they have no competing interests and sources of funding.

**Ethical statement** All procedures were followed in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the 2008 revision of the 1975 Declaration of Helsinki.

**Informed consent** Informed consent was obtained from patients before they were included in the study.

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