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Hydatid cysts in muscle: a modified percutaneous treatment approach

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Summary
Introduction: Any organ in the human body may be affected by hydatid disease, but the liver and the lungs are most commonly affected. A rare localization of hydatid disease is within muscle tissue. Herein we present three patients with muscular hydatid disease who were successfully treated with a modified percutaneous approach.
Methods: Patients with Gharbi type III cysts were treated on an outpatient basis. All procedures were performed under ultrasound guidance in the ultrasonography unit of our department. After local anesthesia, percutaneous puncture was performed in a one-step procedure. After free drainage stopped, absolute ethanol and polidocanol were injected into the cyst cavity. After the procedure, the patient was observed for at least six hours for any adverse reactions and sent home. Patients were followed-up with ultrasonography. A positive treatment effect was characterized by a reduction of the cyst's pseudo-tumor pattern and size, and by detachment of the germinal membrane.
Results: The three patients in this report had a total of five hydatid cysts in muscle tissue and were all successfully treated with a modified percutaneous approach without recurrence.
Conclusion: Percutaneous drainage without re-aspiration is simple, easy to apply, low cost, repeatable, and does not require hospitalization. There have been no reported deaths associated with the procedure and morbidity is very low. When the technique is applied properly, relapses do not occur. With its low complication rate and its suitability for outpatient treatment, this method can be an alternative to surgery or puncture, aspiration, injection, and re-aspiration (PAIR) in selected patients.
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Introduction

Ecchinococcosis or hydatid disease, a parasitic infection caused by *Echinococcus granulosus* or *Echinococcus multilocularis*, is prevalent in most sheep- and cattle-raising countries in the Mediterranean region and the Middle East, as well as Asia, Australia, New Zealand, and South America. The disease is still an important cause of morbidity in these parts of the world.[1] Most cases of hydatid disease are generally asymptomatic, and when apparent the symptoms are due to compression of adjacent structures by enlarging cysts.

Largely occurring in the liver and lungs, musculoskeletal involvement is a rare manifestation of hydatid disease due to the high lactic acid concentration in skeletal muscle and to mechanical factors such as contractile activity,[2] and usually affects a single muscle. Ultrasonography, computerized tomography (CT), and magnetic resonance imaging (MRI) are useful in the diagnosis and classification of the cysts.[3–8] Current methods of treatment include surgery, percutaneous methods like puncture, aspiration, injection, and re-aspiration (PAIR), percutaneous drainage without re-aspiration, and medical management with benzimidazole derivatives.[9–11] These methods are used principally for liver cysts.

To our knowledge, there have been no published studies of percutaneous aspiration for the management of muscle hydatidosis. Herein we present three patients with hydatid cysts in muscle tissue and their successful treatment with a modified percutaneous technique.

Methods

Intervention

Patients with muscle hydatidosis were classified according to the Gharbi classification (Table 1).[12] Those with Gharbi type III cysts were treated on an outpatient basis. All procedures were performed under ultrasound guidance in the ultrasonography unit of our department, which is fully equipped for emergencies. An intravenous line was established and 50 mg of meperidine was given 20 minutes before the procedure. After local anesthesia, the percutaneous puncture was performed with a 22-gauge Chiba needle in a one-step procedure.

Unlike the PAIR technique, which includes re-aspiration of the sclerosing agent used, this method involves puncturing the cyst and allowing it to drain freely under its own pressure.[11] After free drainage has stopped, 2 mL of absolute (96%) ethanol and 1 mL of polidocanol (aethoxysclerol 1%) for each centimeter of the cyst's longest diameter are injected into the cyst cavity under ultrasound guidance. An advantage of this technique is that type III cysts can also be treated, but without free drainage. The needle is then held in place for five minutes within its sheath and is then withdrawn from the patient. The alcohol mixture remains in the cyst. After the procedure, the patient is observed for at least six hours for any adverse reactions, and is then permitted to go home.

Table 1 The Gharbi classification for hydatid cysts

Type I	Purely cystic lesion
Type II	Germinative membrane is detached within parts of the cyst
Type III	Multicystic lesion separated by septae
Type IV	The cyst is degenerated with a pseudo-solid appearance
Type V	The pseudo-solid cyst and ectocyst are calcified

Follow-up

Patients were followed-up with ultrasonography on the day after the procedure (within 24 hours), twice in the first six months and then every six months thereafter. A positive treatment effect was characterized by a reduction of the cyst's pseudo-tumor pattern and size, and by detachment of the germinal membrane. For each patient, CT examinations were performed before treatment and at least six months after.

Case reports

Case 1

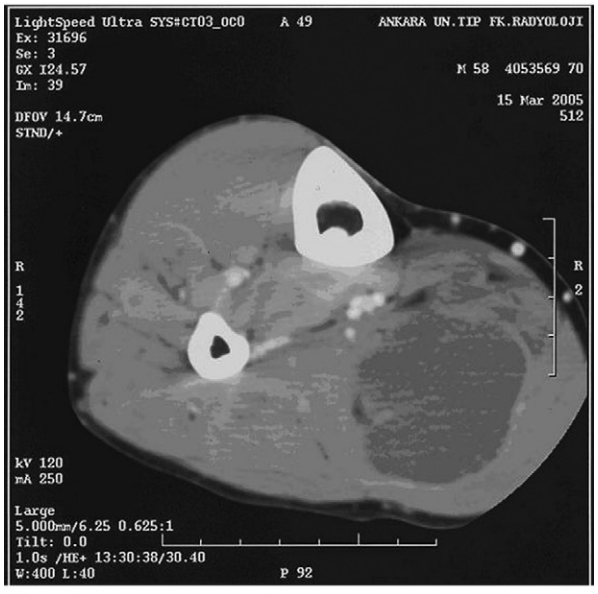
A 51-year-old man presented to the Gastroenterology Unit in June 1997 with pain and a palpable mass in the right gastrocnemius muscle. His medical history was not significant and his physical examination was unremarkable except for the leg mass. All routine laboratory tests were normal. On CT, two adjacent Gharbi type III cysts (multicystic fluid collections with septae) with diameters of 10 cm each were visible in the right gastrocnemius muscle (Figure 1A). Indirect hemagglutination testing was positive for *E. granulosus* at a titer of 1:800. After percutaneous treatment, the hydatid cysts gradually became solid (Gharbi type IV) (Figure 1B) and the pain in the muscle resolved. The patient was followed up for almost eight years with ultrasonography and CT.

Case 2

A 31-year-old man with chronic renal failure was hospitalized in May 2000 with complaints of abdominal discomfort, nausea, and muscle pain in the left upper leg. The patient had undergone nine surgical operations, starting in 1994, for recurrences of hydatid disease in this leg. On physical examination a painful and palpable mass was detected. Routine biochemical tests revealed anemia, and elevated blood urea nitrogen and creatinine levels. Ultrasonography and CT revealed two Gharbi type III cysts in the left gluteus medius, with maximal diameters of 5.6 and 4.8 cm, respectively (Figure 2). In the gluteus maximus and iliopsoas muscles, additional cysts smaller than 5 cm (1.5×1.5 cm, 4×3.5 cm, 2×2 cm, and 4×5 cm) were detected behind the two larger lesions. The indirect hemagglutination test was positive ($>$1:200) for *E. granulosus*. The two large cysts were successfully treated with a modified percutaneous method, and the pain subsided and the palpable mass became smaller. Five years after the procedure, ultrasonography and CT confirmed that the two cysts treated percutaneously had become solid



(A)



(B)

Figure 1 (A) Prior to percutaneous sclerosing therapy, two cysts are seen in the right gastrocnemius muscle with daughter cysts (Gharbi type III). (B) Computerized tomography scan eight years after therapy; the daughter cysts have disappeared in the cyst located posteriorly while the cyst located anteriorly has shrunk.

(Gharbi type IV). There was no recurrence. The other cysts had increased in diameter and further therapy was deferred pending the development of symptoms and/or the cyst sizes becoming large enough to puncture.

Case 3

A 45-year-old man presented to the hospital in February 2003 with the complaint of pain in the left gluteal area. He had previously undergone surgery in September 2002 for a 6×3 cm hydatid cyst in the gluteus maximus muscle. An MRI scan performed at that time revealed a Gharbi type III muscle cyst (Figure 3), which was resected and found to be hydatid on histopathologic examination. After one year, the patient began having muscle pain. Ultrasound revealed a Gharbi type I hydatid cyst measuring 1.8×3 cm. On CT, the cyst was visible between the gluteus maximus and gluteus medius muscles, and serum levels of muscle enzymes were normal. In October 2003, the cyst was treated with a modified percutaneous method; immediately after the intervention the germinative membrane was found to be detached from the pericyst. The patient's pain subsided, and during 24 months of follow-up the cyst gradually solidified. There has been no relapse.



(A)



(B)

Figure 2 (A) Prior to percutaneous sclerosing therapy, two hydatid lesions with daughter cysts were visualized by computed tomography (CT) in the right gastrocnemius muscle. (B) Eight years after treatment, the daughter cysts have disappeared or diminished in size.



Figure 3 Multilocular cystic lesion in the gluteus maximus muscle seen on CT scan.

Discussion

The liver and lungs act as natural mechanical filters for oncospheres following penetration of intestinal mucosa, thus making it quite difficult for them to reach musculoskeletal tissue. The proportion of hydatid disease localized in muscle has been reported at 3—5%, occurring in various anatomic regions including the thigh,[2] quadriceps femoris,[13] biceps femoris,[14] psoas,[15] diaphragm,[16] triceps,[16] myocardium,[17] right ventricular outlet,[18] and extra-ocular muscles.[19] It is believed that local production of lactic acid and mechanical factors such as contractile activity tend to inhibit the embryo from settling in muscle.[2,20] The three patients reported here fulfilled the criteria for muscular hydatidosis with impairment of muscle function and exclusion of liver and lung involvement.[21,22]

Medical treatment with benzimidazole compounds (mebendazole and albendazole) has been used in the management of liver and lung hydatid disease since the early 1970s. Their effects include prevention of the entrance of tubulin into mitochondria, inhibition of glucose uptake with suppression of glycogen deposits in the cyst, and disruption of cyst homeostasis via reduced production of adenosine triphosphate.[23] Intracystic concentrations of mebendazole may be only about 10% of the serum level. For this reason, the drug's effect is limited and approximately 3—30% of cases recur after treatment is stopped.[24] Consequently, benzimidazole compounds cannot be used effectively in muscle or kidney hydatid disease due to their low cystic fluid concentrations.[3,24,25] Also, according to an evidence-based meta-analysis, combination treatment with albendazole and praziquantel was found to be superior to albendazole monotherapy.[26] Thursky and Torresi successfully treated a patient who was at high risk for dissemination during the initial exploratory procedure with muscle resection and combination therapy with albendazole plus praziquantel.[2]

Surgery is the preferred therapeutic modality for muscular hydatid disease. However, disease recurrences are frequent following surgical management. Repetitive surgical operations and accompanying disorders increase postoperative mortality from 0.9% to 3.5%.[1,3,27]

In 1985, the era of non-operative percutaneous treatment of hepatic hydatid cysts began when Mueller and colleagues demonstrated that administration of nitrate compounds and hypertonic serum into the cyst cavity after aspiration of fluid was not dangerous.[28] In a study by Khuroo et al., percutaneous treatment of liver cysts combined with albendazole was superior to surgical cystectomy in terms of hospital stay (4.2 vs. 12.7 days) and complication rate (32% vs. 84%).[29] Similarly, a meta-analysis of 21 trials comparing PAIR plus chemotherapy to surgery concluded that overall cure rate, recurrence, incidence of complications, fever, allergic reactions, and hospital stay were significantly less in the former group.[30]

Percutaneous drainage without re-aspiration is simple, easy to apply, low cost, repeatable, and does not require hospitalization. There have been no reported deaths associated with the procedure and morbidity is very low. When the technique is applied properly, relapses do not occur. The method has been used for hydatid cysts located in the spleen and kidneys.[31,32] The three patients in this report had a total of five hydatid cysts in muscle tissue and were all successfully treated with a modified percutaneous approach without recurrence. Patient 2 had previously undergone nine unsatisfactory surgical procedures for his muscle cysts.

In conclusion, percutaneous drainage without re-aspiration of injected scolicide is a simple, effective, safe, repeatable, and cheap modality for the management of muscle hydatid disease. With its low complication rate and its suitability for outpatient treatment, this method can be an alternative to surgery or PAIR in selected patients.

Conflict of interest: No conflict of interest to declare.

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