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Case Report

The Effect of Preoperative Albendazole Treatment on Intramuscular Cyst Structure in a Patient with Simultaneous Cystic Echinococcusis of Liver and Vastus Lateralis Muscle

Mahmut Kursat Ozsahin, *Bedri Karaismailoglu, Cumhur Deniz Davulcu, Erdem Sahin, Murat Hız

Orthopaedics and Traumatology Department, Cerrahpasa Medical Faculty, Istanbul University-Cerrahpasa, Istanbul, Turkey

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***Correspondence**
Email:
bedrikio@hotmail.com

Abstract
We report a 44-years-old woman with cystic echinococcosis (CE) who presented with simultaneous involvement of liver and vastus lateralis muscle to Istanbul University-Cerrahpasa, Istanbul, Turkey in 2020. Although she underwent surgery for the liver, the intramuscular mass was ignored. While the patient was under post-operative albendazole treatment, she was investigated for the mass on her right thigh which was excised later. The comparison of magnetic resonance imaging before and after albendazole treatment showed that albendazole eliminated the intramuscular vesicle structures by accelerating the degeneration process of the cyst. The comparison of pathology samples sent from both liver and intramuscular CE also revealed that the albendazole has left the laminar membrane intact, degenerate the germinative membrane, thereby reducing the intra-vesicle pressure and also caused the scolex structures to disappear. To the best of our knowledge, this is the first paper to report the effect of preoperative albendazole treatment on the structure of intramuscular CE.

Introduction

Cystic echinococcosis (CE) is a zoonotic and parasitic disease of humans and animals caused by *Echinococcus granulosus*. It is generally encountered in Mediterranean countries (1). The two most frequently affected organs are the liver and lungs while muscular tissue is rarely involved because of its high lactic acid level and contractility (1). The treatment options include the combinations of chemotherapy, puncture, and

surgical excision but no specific treatment guideline for the timing and duration of chemotherapy has been established so far (2).

We report a case of CE patient who presented with simultaneous involvement of liver and vastus lateralis muscle. She underwent albendazole treatment after the excision of liver cyst and intramuscular cyst was revealed after she completed her albendazole treatment. Thus, available data made it possible to compare the cyst structure before and after albendazole treatment both radiologically and pathologically.

Case Presentation

A 44-years-old female patient was referred to Istanbul University-Cerrahpasa, Istanbul, Turkey in 2020 with a mass in her right thigh which was present in a smaller diameter for ten years. She did not seek any healthcare for her mass until the mass got larger in diameter during the last few months. Four months before, the patient was also diagnosed with liver CE as a result of abdominal ultrasonography (USG) and computed tomography (CT). Multiple cysts between 20-50 mm in size were found in the right lobe of the liver, located in segment.

The patient underwent surgery and after partial cystectomy and germinative membrane dissection, the cavitary area was washed with povidone-iodine and the patient received albendazole treatment in the postoperative period. Before the index surgery, magnetic resonance imaging (MRI) of the thigh was also made to evaluate the mass in her right thigh. The MRI showed a lesion with a size of 180x100x75 mm consisting of cystic components within the right vastus lateralis muscle (Fig. 1). The patient did not show the result of MRI to any physician and she just continued her follow-ups for her excised liver CE. After the patient recovered and completed her albendazole treatment (three 1-month oral doses (10 mg/kg/d) separated by 14-day intervals), she applied to our clinic for her thigh mass. A new MRI was ordered and a mass of 150x70x55 mm was detected along proximal and middle one-third of her right thigh. The complete blood count and biochemical tests were within normal limits. The surgical excision of the mass was planned. The patient had already received albendazole treatment for 3 months preoperatively, after her liver CE surgery.

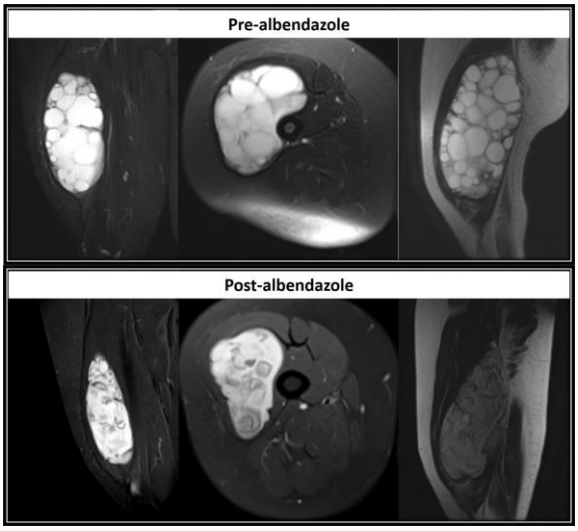

Fig. 1: Preoperative magnetic resonance imaging sections of the intramuscular hydatid cyst before and after albendazole treatment

The mass was reached by a 20 cm longitudinal skin incision on the right thigh anterolaterally. Total excision was performed without damaging the cyst membrane. The surgical area was washed, the incision was closed through a hemovac drain. The mass was sent to pathology and the result was compatible with CE. In the macroscopic examination of the removed cyst, daughter cysts were seen and it was found that the hydatid sand structure was denser and more caseified than normal (Fig. 2). The patient continued to use 10 mg/kg/d albendazole for two months in the postoperative period with the recommendation of the infectious diseases department.

Fig. 2: (a) Opening the mass during the pathological evaluation and (b) the appearance of daughter cysts

The comparison of MRI sections showed that albendazole eliminated the vesicle structures by accelerating the degeneration process of the cyst and created a heterogeneous unilocular encapsulated cyst structure compared to the previous multilocular appearance. According to the WHO-IWGE classification, the grade of CE appearance changed from CE2 to CE3b after albendazole treatment (Fig. 1) (3). Based on the comparison of pathology samples sent from both liver and intramuscular CE; the albendazole has been found to leave the laminar membrane intact, degenerate the germinative membrane, thereby reducing the intra-vesicle pressure. The scolex structures were observed in the material sent from the liver, but not in the specimen sent from muscular tissue after the use of albendazole (Fig. 3).

Discussion

Larvae of *E. granulosus* adhering to various tissues lead to the formation of cystic structures. A CE is composed of an outer laminated, acellular membrane lined by a thin, germinal membrane. Germinal layer generates brood capsules and protoscoleces into a central cavity filled with a clear "hydatid" fluid. It is surrounded first by an acellular laminated layer, then by the host reaction. There is no standard for the medical management of CE and the timing, dose, and duration of treatment varies widely between studies (1).

Although several studies analyzed the effect of preoperative albendazole use on the cysts, the results were different for distinct tissues. Some suggested preoperative albendazole treatment in liver CE, claiming that preoperative 3-month albendazole treatment decreased the viable cyst percentage (1), while others suggested immediate surgery and not to use preoperative albendazole in pulmonary CE, claiming that albendazole decreased the tensile strength of the cuticular membranes which may lead to their perforation (4).

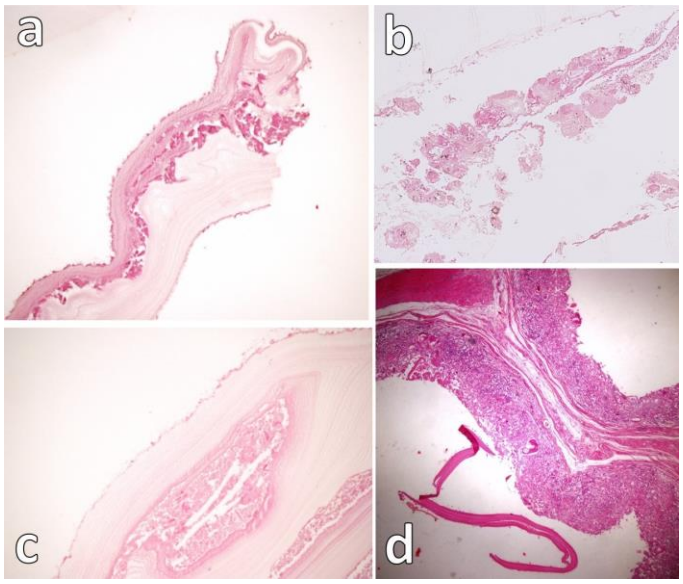

Fig. 3: (a, b, c) Laminar membrane of CE in the liver, adjacent germinal layer and scolex. (d) Laminar membrane of intramuscular cyst hydatid and adjacent CE cuticle. (HEx100) (The photos belonged to the pathological evaluation of the patient in this case report.)

However, no study is available analyzing the effect of preoperative albendazole treatment on intramuscular cyst structure and a wide variety of chemotherapy application methods are available for intramuscular CE in the literature. Some authors preferred to use a short course (a few days) of preoperative albendazole treatment as a prophylaxis for anaphylaxis and dissemination (2). Some authors used longer courses (between 1-3 months) of preoperative albendazole treatment to decrease the viability of the cyst and the risk of recurrence (5). Besides, some authors immediately removed the cyst and applied postoperative chemotherapy to not to weaken the capsule of the cyst with preoperative albendazole treatment (6). Manouras et al evaluated the effect of preoperative 28-day albendazole treatment via MRI (7). They found that cysts showed solidification or calcification with volumetric reduction, and preoperative albendazole facilitated complete resection of the cyst and could reduce the recurrence rates and postoperative complications. The MRI sections and pathological findings of our case also supported their conclusion.

It is quite challenging to surgically remove the cysts without rupture. The cyst rupture or spillage should be avoided to prevent local or distant spread and anaphylaxis (8). Albendazole provides cellular autolysis in the germinal layer by inhibiting tubulin, glucose absorption, and increasing lysosomal activity, although its mechanism of action is not fully understood. Thus, degenerative changes such as volume reduction in the cyst, condensation of the intravesicular fluid, membrane detachment, and calcification can occur (9).

Our case suggested that in case of an intramuscular CE, the use of albendazole in the preoperative period may decrease the risk of rupture during removal of the cyst by decreasing the viability of the cyst and intracystic pressure, so that cyst rupture or spillage can be avoided.

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Conflicting interests

The authors declare that there is no conflict of interest.

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