



## Primary Cystic Echinococcosis in Psoas Muscle

### CASE REPORT

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### ABSTRACT

Cystic echinococcosis (CE) caused by *Echinococcus granulosus* has been endemic in many parts of the world and involvement of the liver is the most frequently seen form. However, primary musculoskeletal involvement is rare. The radical treatment is surgical resection followed by anthelmintic medical therapy. In this study, a case of a 38-year-old woman with rare localization of CE in the psoas muscle at the proximal thigh is presented.

Keywords: Psoas muscle, cystic echinococcosis, *echinococcus granulosus*

### INTRODUCTION

Cystic echinococcosis (CE) caused by *Echinococcus granulosus* has been endemically seen in many parts of the world, such as the Mediterranean, the Middle East, and Africa. In our country, it is most commonly observed in Eastern and Southeastern Anatolia (1). The localization is mostly in the liver and the rest, by going into microcirculation, is in the lungs and kidneys, and rarely, in the muscles and brain. The ratio of CEs with primary muscle localization ranges from 1% to 5% (2).

### CASE REPORT

A 38-year-old woman presented with pain in the right lower extremity for 6 months and was admitted to our hospital because of a swelling in the front face of the thigh. During the physical examination, palpation revealed a fixed, mild painful mass (Figure 1) in the mid-proximal of the femur anterior about the size of 10 × 10 cm. The neurological examination revealed hypoesthesia in the femoral nerve innervation. A 10 × 12 × 10 cm-sized cystic mass with septation, which could be distinguished prominently as being localized to surrounding tissue, was found on ultrasonographic (USG) examination. No symptoms other than soft tissue expansion and sclerotic areas in places were determined on direct radiography (Figure 2). In magnetic resonance (MR), although slight contrasting in the form of a rim was observed after contrast in the coronal and axial planes around daughter vesicles, along the right psoas muscle, going through the inguinal canal and showing extension until femoral 1/3 intermediary object, a 14 × 10 × 10 cm-sized formation was observed that was compatible with a hydatid cyst, with multiple hypointense cystic appearances compatible with daughter vesicles and distinguished prominently as being localized to surrounding tissue (Figure 3a-c). MR angiography, on the other hand, revealed that it had no relationship with the vascular structure (Figure 4). Serological tests were not performed on the patient. Other than an increase in sedimentation rate in complete blood count and a moderate increase in eosinophils, no pathology was observed. The patient's chest x-ray and abdominal and chest computed tomography (CT) showed no pathology.

Preoperative infectious disease consultation was requested from the patient, with a preliminary diagnosis of primary muscle localized CE. After obtaining informed consent from the patient, the cyst was scraped from the surrounding tissue and was completely excised (Figure 5) under general anesthesia. Because the cysts were ruptured during surgery, the surgical site was washed with hypertonic solution (3% sodium chloride) that was available. Surgical layers were closed in accordance with the procedure. Albendazole (10 mg/kg/day) was given for postoperative 3 months on the basis of the recommendation of the infectious disease specialist. After surgery, CE diagnosis was confirmed histopathologically. On the third postoperative week, preoperative hypoesthesia completely disappeared. At the 18-month postoperative follow-up, no recurrence was detected.

### DISCUSSION

*E. granulosus* is the causative agent for CE. The definitive hosts are Canidae, such as dogs, wolves, and foxes, and the eggs excreted in the feces of these animals infect intermediate hosts via food, beverages, and contaminated hands. Although intermediate hosts are dominantly sheep, other butchery animals (goats, cattle, pigs, camels, etc.)

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Submitted  
24.06.2013

Accepted  
25.10.2013

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This study was presented in  
the 23<sup>th</sup> National Congress of  
Orthopedics and Traumatology,  
2013, 29 October Antalya,  
Turkey.

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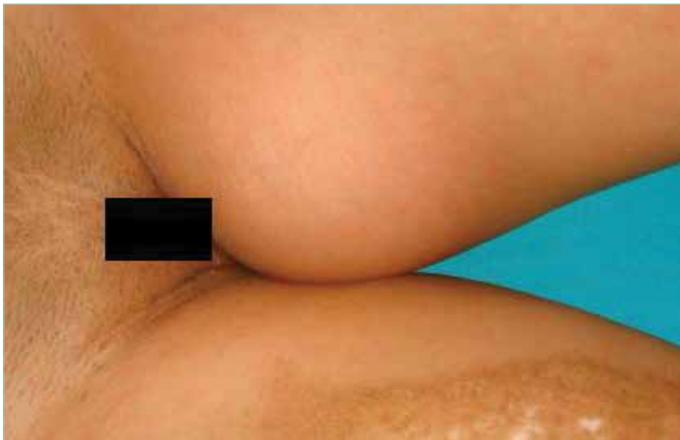
and rarely humans can be the intermediate hosts as well. Humans are an accidental intermediate host for this parasite and almost a dead-end. Because the possibility of a canine eating human flesh is rare, a canine's obtaining hydatid cysts that can be found in various organs of the human is unlikely. The ratio of the most common CE organ involvement is 73% in livers, 14% in lungs, 1-5% in the muscles (3). Muscle localization might be primary or secondary. It is difficult to enter the arterial circulation because the oncosphere leaves the protoscolex in the digestive system and enters the portal circulation and passes through the lung filters; thus, muscle involvement with other organs is rare (4). In our case, because preoperative screening revealed no cysts in other organs, the diagnosis made was primary muscle CE.

Muscle localized CE develops slowly because of high levels of lactic acid in the muscles and contractions and can grow for a long time without showing any symptoms (5). It should be noted that in endemic regions, such atypical localizations can be found in many different anatomical regions apart from the liver.

In the diagnosis of the disease, history taking, radiological imaging techniques (ultrasound, CT, and MRI), and serological methods are used. The cyst wall thickness, presence of daughter vesicles,

and germinal membrane's separation from the pericyst are the characteristic findings on MRI, which is one of the most useful diagnosis methods (6). In our case, daughter vesicles were typically seen in MRI.

In the literature, it is stated that the sensitivity of serological tests is 60-90%. Arazi et al. (7) determined 27% positive indirect hemagglutination test results in a musculature CE case series. Negative



**Figure 1.** The patient's clinical presentation



**Figure 2.** The radiological appearance of the lesion



**Figure 3.** (a) A 14 × 10 × 10 cm-sized mass along the psoas muscle passing through the inguinal canal and causing expansion in the surrounding tissue. (b, c) After the application of contrast, contrast enhancement around the daughter vesicles can be typically seen in the form of a peripheral rim

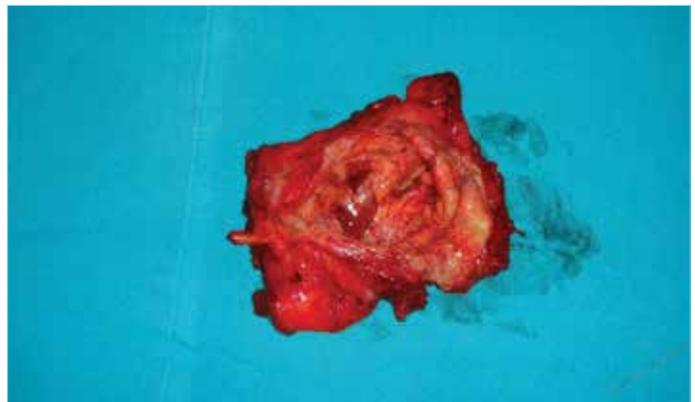


**Figure 4.** The mass was adjacent to major vascular structures on MR angiography

serological test results do not indicate the absence of disease (7). In our case, serological tests were not performed. In addition to avoiding diagnostic biopsies and being prepared during surgery, anaphylactic shock after intraoperative cyst rupture and risk of iatrogenic spread of the disease should be kept in mind (8). Also, in our case, biopsy was not performed.

Although there are studies that recommend anthelmintic treatment administration to patients prior to surgery, a consensus has not been reached yet (7). Our case did not receive any treatment prior to surgery.

Surgical excision and medical treatment are recommended in isolated muscle localized CE cases. If total excision cannot be performed during surgery, cyst drainage, removal of the membrane, and irrigation with hypertonic solution of the pouch are required (9). It has been reported in the literature that applying a protoscolicidal agent such as hypertonic saline into the cyst is useful to prevent recurrence and contamination due to a rupture that may occur during surgery (10). We also completely excised the cyst surgically by scraping it from the surrounding tissue. The surgical site was washed with a hypertonic solution because the cyst ruptured during surgery. As a precaution for probable recurrence, albendazole (10 mg/kg/day) was given prophylactically for postoperative 3 months. Apart from malignancies, abscess, and hematoma in extremity localized masses, CE should also be kept in mind in endemic regions.



**Figure 5.** Intraoperative view and macroscopic view after the removal of the daughter vesicles

## CONCLUSION

In order for successful treatment of CE, careful surgery in conformity with the principles, washing of the surgical site with hypertonic solutions in terms of metastasis to neighboring tissues, and administration of anthelmintic therapy are recommended.

**Informed Consent:** Written informed consent was obtained from patients who participated in this study.

**Peer-review:** Externally peer-reviewed.

**Authors' contributions:** Conceived and designed the experiments or case: İK. Performed the experiments or case: İK, İHK, MK. Analyzed the data: İK, MH. Wrote the paper: İK, İHK. All authors read and approved the final manuscript.

**Conflict of Interest:** No conflict of interest was declared by the authors.

**Financial Disclosure:** The authors declared that this study has received no financial support.

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