

Case Report: Primary Localization of a Hydatid Cyst in the Adductor Brevis Muscle

Ali ACAR¹, Osman RODOP², Ercan YENİLMEZ¹, Orhan BAYLAN³, Oral ÖNCÜL¹

GATA Haydarpaşa Eğitim Hastanesi, ¹Enfeksiyon Hastalıkları ve Klinik Mikrobiyoloji Servisi, ²Ortopedi ve Travmatoloji Servisi, ³Mikrobiyoloji ve Klinik Mikrobiyoloji Servisi, İstanbul, Türkiye

SUMMARY: We present a rare case of primary muscular hydatidosis in the left thigh of a 20 year-old man, who presented with painless mass. Ultrasound and magnetic resonance imaging examinations revealed a multilocular intramuscular cyst in the posteromedial compartment of the left thigh mainly occupying the adductor brevis muscle. This site of localization has not been reported previously. The patient was treated successfully by preoperative and postoperative dual treatment of albendazole together with surgery. Hydatid disease should be included in the differential diagnosis of muscular masses, regardless of its location, especially in endemic areas.

Key Words: Echinococcosis, adductor brevis muscle, albendazole

Olgu Sunumu: Adductor Brevis Kasını Tutan Primer Kist Hidatik

ÖZET: Bu yazıda, sol uylukta ağrısız kitle ile başvuran ve primer kas hidatik kisti tanısı alan 20 yaşında bir erkek hastayı sunuyoruz. Ultrason ve MRI görüntüleme yöntemleri ile sol uyluk posteromedial bölgede büyük ölçüde adductor brevis kas içinde yerleşmiş multioküler kistik kitle tespit edildi. Literatürde daha önce bildirilmiş adductor brevis kas tutulumlu kist hidatik olgusuna rastlanmadı. Preoperatif ve postoperatif albendazole tedavisi ve kistin cerrahi yöntemle çıkarılması ile hasta başarılı olarak tedavi edildi. Özellikle endemik bölgelerde, kaslarında kitle tespit edilen hastaların ayırıcı tanısında kist hidatik yer almalıdır.

Anahtar Sözcükler: Ekinokokkozis, adductor brevis kası, albendazole

INTRODUCTION

Hydatid disease is a parasitic infestation by a tapeworm of the genus *Echinococcus*. Four species of *Echinococcus* produce infection in humans; *E. granulosus* and *E. multilocularis* are the most common, causing cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively. The two other species, *E. vogeli* and *E. oligarthrus*, cause polycystic echinococcosis but they only rarely have been associated with human infection (6). These infections are cause of substantial morbidity and mortality in most of the world, including parts of Europe (notably Greece and Turkey), South America, Africa, Australia, Russia, China and the Middle East (3, 6).

Echinococcosis predominately affects the liver followed by lungs; these two organs are account for 90% of cases but cysts may develop in any part of the body. The muscle involvement is extremely rare; its occurrence even in endemic areas is less

than 5% of the total incidence of echinococcosis (8, 10). We report our experience in the management of a patient who developed hydatid cyst in the adductor brevis muscle. We would like to point out that, this zoonotic infection should be included in the differential diagnosis of muscular masses, especially in endemic areas, such as Turkey.

CASE REPORT

A 20 years old male was admitted to our hospital with a painless mass on his left thigh at proximal posteromedial location. The patient was living in a rural area in Turkey where sheep farming is widespread and contact with dogs is common. He had noticed the swelling on his leg seven months ago, without history of trauma, surgery or any additional diseases. His general health condition was good with no fever or weight loss. Our physical examination revealed a well-delineated 15x6 cm round mass on the posteromedial left thigh. The non tender mass was dense with smooth surface and there was no sign of erythema. Other physical findings and laboratory data including total blood cell count, erythrocyte sedimentation rate, serum and urine biochemical parameters, were normal. Ultrasound (USG) examination of the mass revealed a multilocular cyst. Magnetic resonance imaging (MRI) revealed a 12 x 8,5 x 17 cm, well-delineated oval multiloculated cystic lesion containing daughter vesicles,

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Yazışma /Corresponding Author: Ali Acar
Tel: (90) (216) 542 20 20 Fax: -
E-mail: draliacar@yahoo.com

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in the posteromedial compartment of the left thigh mainly occupying the adductor brevis muscle (Figure 1). The mass did not have any connection to neurological and vascular structures of thigh. Serology for *Echinococcus* by ELISA (Ridascreen Echinococcus IgG; R-Biopharm, Darmstadt, Germany) was positive, measuring 2.5 IU (cut-off >1.1). Based on these findings, a diagnosis of hydatid cyst was performed.

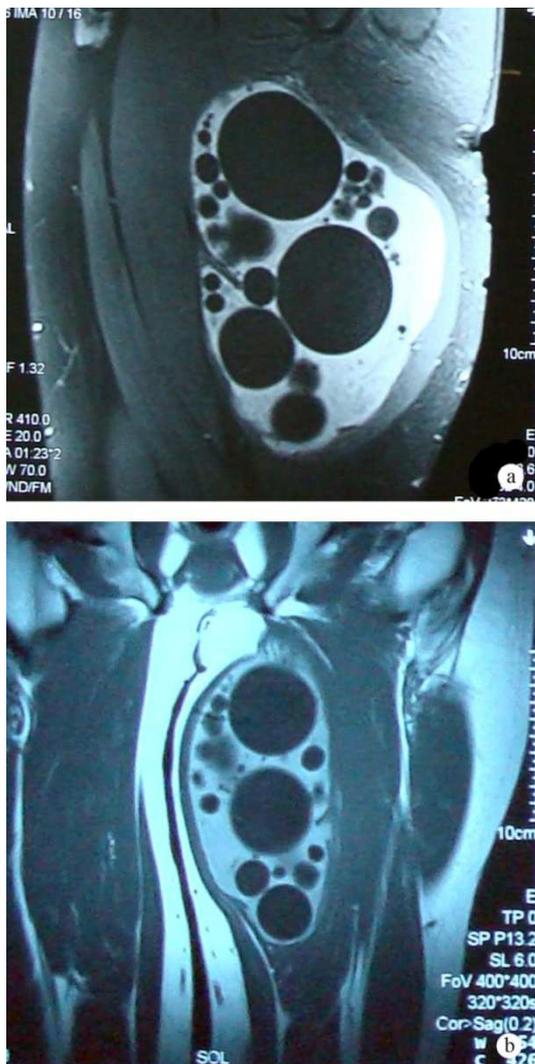


Figure 1. MRI of the left thigh. Sagittal TSE T2 weighted (a) and coronal TIRM weighted (b) images show the multiloculated cystic lesion with multiple daughter cysts in the adductor brevis muscle.

We also performed abdominal USG, chest X-ray and cranial MRI in order to rule out any involvement of the body. All imaging studies detected no other cyst in other parts of the body. So the hydatid cyst in the adductor brevis muscle in our patient assumed to be primer muscle hydatidosis. According to literature, preoperative medication consisted of a 15 mg/kg/d dose of albendazole was applied for one month in order to decrease the count of viable cyst (2, 13). After this medical

treatment, surgery was planned. In operative process before the incision, scolicedal agents (20% hypertonic saline solution) were injected into the cystic cavity. After 15 minutes, an incision was made onto mass at the posteromedial aspect of proximal left thigh. By using sharp and Blount dissection border of the cyst was exposed. Then cyst wall was opened with a 5 cm. incision and its content was aspirated. There were multiple daughter cysts in aspiration material (Figure 2). After the aspiration process, povidone iodine solution was applied into the cavity and irrigation was performed with isotonic saline. Finally, the drain was fixed and tissues were repaired. The diagnosis of a hydatid cyst was confirmed with macroscopic and microscopic histopathological examinations after removal of the mass. Albendazole treatment was continued for two months postoperatively with the same dosage.



Figure 2. Photograph of excised hydatid cyst shows the numerous daughter cysts.

DISCUSSION

Hydatid disease rates are generally higher in sheep-farming areas with poor sanitation particularly in areas with uncontrolled dogs. Dogs in these areas usually acquire infection by eating the entrails of infected butchered sheep or other herded animals. Humans become infected by ingesting eggs via close contact with infected dogs or from contaminated environment. After ingestion, the eggs hatch in the small intestine releasing hooked larvae. These larvae penetrate small intestine wall and circulate through the body. In cystic echinococcosis, the most common involved organ is the liver followed by the lungs (6). Other sites of involvement, in descending order of frequency, are the spleen, skin, muscle, kidney, retroperitoneum, bone, heart, and brain (3, 6).

Primary skeletal muscle hydatid cyst is very uncommon, because implantations at this site require passage through the filters of the liver and lung. In addition, intramuscular growth of cysts is hindered by muscle's contractility and lactic acid content (7). The patient with muscular echinococcosis should be evaluated in order to determine whether there is another focus of dissemination since the disease is generally located in other parts of the body, especially hepatic and pulmonary re-

gions (3, 7). We excluded the other organ involvement by the carefully clinical and radiological examination of the patient, and the localization in the adductor brevis muscle was therefore assumed to be primer muscle hydatidosis.

Diagnosis of human echinococcosis remains highly dependent on imaging techniques to detect the cystic space occupying lesion (3). USG is particularly useful in the diagnosis of hydatid cyst when the daughter cysts and hydatid sand are demonstrated (12). However, in muscular hydatid cyst, the USG findings are sometimes indistinguishable from those of soft tissue abscess leading to misdiagnosis (1, 4). Computed tomography has an advantage over ultrasound for better documentation of site, size and structure of cyst. MRI may show an intense rim which has been proposed as a characteristic sign of hydatid disease (12). Serologic tests are then typically used to confirm the diagnosis. Indirect immunofluorescence antibody test, ELISA, immunoelectrophoresis, and immunoblot test are the commonly used techniques (15). In our case, the diagnosis was established by radiological, serological and histopathological examinations.

The treatment of human muscle hydatidosis is principally surgical; however, the cysts' contents spillage may occur during the surgery resulting in anaphylaxis and/or secondary echinococcosis (9).

For this reason, some authors suggest use of albendazole preoperatively to prevent these complications (2, 13). Our patient was treated by surgical excision of the cyst combined with preoperative and postoperative antihelminthic therapy (albendazole 800mg/d) and there were no complications.

Although primary hydatid disease involving in several muscle groups of thigh including soleus (7), quadriceps (11), adductor magnus (14), sartorius (5) have been reported in the literature, this is the first case of hydatid cysts involving the adductor brevis muscle in our knowledge. Hydatid disease should be included in the differential diagnosis of muscular masses, regardless of its location, especially in endemic areas.

REFERENCES

1. **Bedioui H, Nouira K, Daghfous A, Ammous A, Ayari H, Rebai W, Chebbi F, Ksantini R, Fteriche F, Jouini M, Kacem MJ, Menif E, BenSafta Z**, 2008. Primary hydatid cyst of the psoas muscle: description of 9 cases in Tunisia and review of the literature. *Med Trop*, 68: 261-266.
2. **Bildik N, Cevik A, Altıntaş M, Ekinçi H, Canberk M, Gülmen M**, 2007. Efficacy of preoperative albendazole use according to months in hydatid cyst of the liver. *J Clin Gastroenterol*, 41: 312-316.
3. **Craig PS, McManus DP, Lightowlers MW, Chabalgoity JA, Garcia HH, Gavidia CM, Gilman RH, Gonzalez AE, Lorca M, Naquira C, Nieto A, Schantz PM**, 2007. Prevention and control of cystic echinococcosis. *Lancet Infect Dis*, 7: 385-394.
4. **Durakbasa MO, Kose O, Islam NC, Kilicoglu G**, 2007. A primary hydatid cyst of the gracilis: a case report. *J Orthop Surg*, 15: 118-120.
5. **Duygulu F, Karaoğlu S, Erdoğan N, Yildiz O**, 2006. Primary hydatid cyst of the thigh: a case report of an unusual localization. *Turk J Pediatr*, 48: 256-259.
6. **Eckert J, Deplazes P**, 2004. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev*, 17: 107-135.
7. **Garcia-Alvarez F, Torcal J, Salinas JC, Navarro A, Garcia-Alvarez I, Navarro-Zorraquino M, Sousa R, Tejero E, Lozano R**, 2002. Musculoskeletal hydatid disease: a report of 13 cases. *Acta Orthop Scand*, 73: 227-231.
8. **Kazakos CJ, Galanis VG, Verettas DA, Polychronidis A, Simopoulos C**, 2005. Primary hydatid disease in femoral muscles. *J Int Med Res*, 33: 703-706.
9. **Khuroo MS**, 2002. Hydatid disease: current status and recent advances. *Ann Saudi Med*, 22: 56-64.
10. **Merkle EM, Schulte M, Vogel J, Tomczak R, Rieber A, Kern P, Goerich J, Brambs HJ, Sokiranski R**, 1997. Musculoskeletal involvement in cystic echinococcosis: report of eight cases and review of the literature. *AJR Am J Roentgenol*, 168: 1531-1534.
11. **Ozkoç G, Akpınar S, Hersekli MA, Ozalay M, Tandoğan R**, 2003. Primary hydatid disease of the quadriceps muscle: a rare localization. *Arch Orthop Trauma Surg*, 123: 314-364.
12. **Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS**, 2000. Hydatid disease: radiologic and pathologic features and complications. *Radiographics*, 20: 795-817.
13. **Stankovic N, Ignjatovic M, Nozic D, Hajdukovic Z**, 2005. Liver hydatid disease: morphological changes of protoscoleces after albendazole therapy. *Vojnosanit Pregl*, 62: 175-179.
14. **Vicidomini S, Cancrini G, Gabrielli S, Naspetti R, Bartoloni A**, 2007. Muscular cystic hydatidosis: case report. *BMC Infect Dis*, 7: 23.
15. **Zhang W, Li J, McManus DP**, 2003. Concepts in immunology and diagnosis of hydatid disease. *Clin Microbiol Rev*, 16: 18-36.