

Imaging Characteristics of Three Primary Muscular Hydatid Cyst Cases with Various Patterns

Primer Kas Kist Hidatidinde Farklı Paternli Üç Olgunun Görüntüleme Karakteristikleri

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ABSTRACT

Primary muscular hydatid cysts comprise less than 0.5% of the cases in endemic populations. These cysts appear as slow-growing masses of the soft tissue, sometimes with inflammatory signs and fistulization. In this report, we present three cases of primary muscular hydatid cyst in which preoperative diagnosis were achieved by characteristic radiological appearances of the cysts. Primary muscular hydatidosis should be kept in mind in the differential diagnosis of a cystic mass of a skeletal muscle in endemic areas.

Key words: echinococcosis; hydatid cyst; imaging; muscle

ÖZET

Primer kas kist hidatidleri, endemik bölgelerde olguların %0,5'inden daha azını oluşturmaktadır. Bu kistler, bazen enflamasyon bulguları ve fistülizasyon gösteren yavaş büyüyen kitleler olarak görülürler. Bu yazıda, kistlerin karakteristik radyolojik görünüşleri ile operasyon öncesi tanısı konulan primer kas kist hidatidli üç olguyu sunuyoruz. Endemik bölgelerde iskelet kasına ait bir kistik lezyonun ayırıcı tanısında primer kas kist hidatidi akılda bulundurulmalıdır.

Anahtar kelimeler: ekinokokkozis; kist hidatid; görüntüleme; kas

Introduction

Hydatidosis is a zoonotic infection caused by *Echinococcus granulosus*. Human Cystic Echinococcosis (CE) is still endemic in some areas of the world including Mediterranean countries and it is still a healthcare problem in Turkey. Muscular localization of the hydatid cyst (HC) is very rare and is usually secondary to hepatic or pulmonary disease, and can cause a variety of diagnostic problems. Primary muscular HC comprises less than 0.5% of

the cases in endemic populations. These cysts appear as slow-growing masses of the soft tissue, sometimes with inflammatory signs and fistulization.¹⁻³

In this report, we present imaging findings [ultrasonography (US), computed tomography (CT), and magnetic resonance (MR) imaging] of three cases with various patterns of primary muscular HC.

Cases

Case 1

A 65-year old man was admitted to our surgery clinic because of a cystic mass within his left thigh musculature. He had realized a progressively growing mass in his left thigh for three years. He was living in a village and had contact with both sheep and dogs since his childhood. There was no history of trauma, fever or weight loss. Laboratory results showed an erythrocyte sedimentation rate of 12 mm/h (Westergren) and a total leukocyte count of 7,000/mm³. A hemagglutination inhibition test revealed that the titer of antibodies to *Echinococcus* was 1:165. Physical examination revealed a diffuse, non-tender cystic swelling occupying nearly the entire anterior aspect of the thigh with the rigidity of the muscles. Chest X-ray was normal. Abdominal US revealed no abnormality. US and CT examinations were performed. US examination showed a 5 x 4 cm mass composed of multiple thin-walled cysts (Fig. 1a). These clusters of cysts were localized between the femur and vastus muscles, with no involvement of the muscle or bone. No calcifications were present. CT scan demonstrated the cystic structure close to vastus muscles (medius, intermedius, lateralis) (Fig. 1b). The lesion was defined as a type-II HC according to the Gharbi classification.

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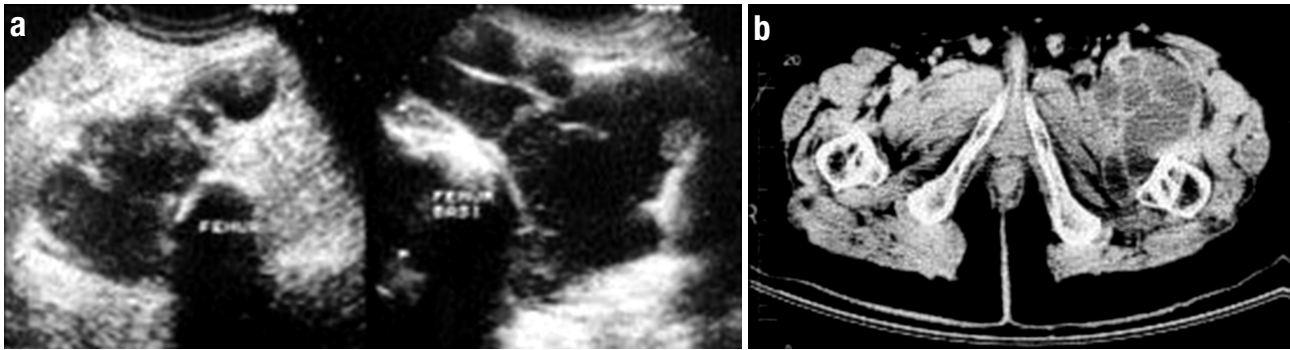


Figure 1. (a) Transverse and sagittal ultrasound images show multiseptated, hypochoic cystic mass, (b) contrast-enhanced axial computerized tomography scan demonstrates well-defined, multiloculated cystic lesion in the left vastus lateralis muscle.

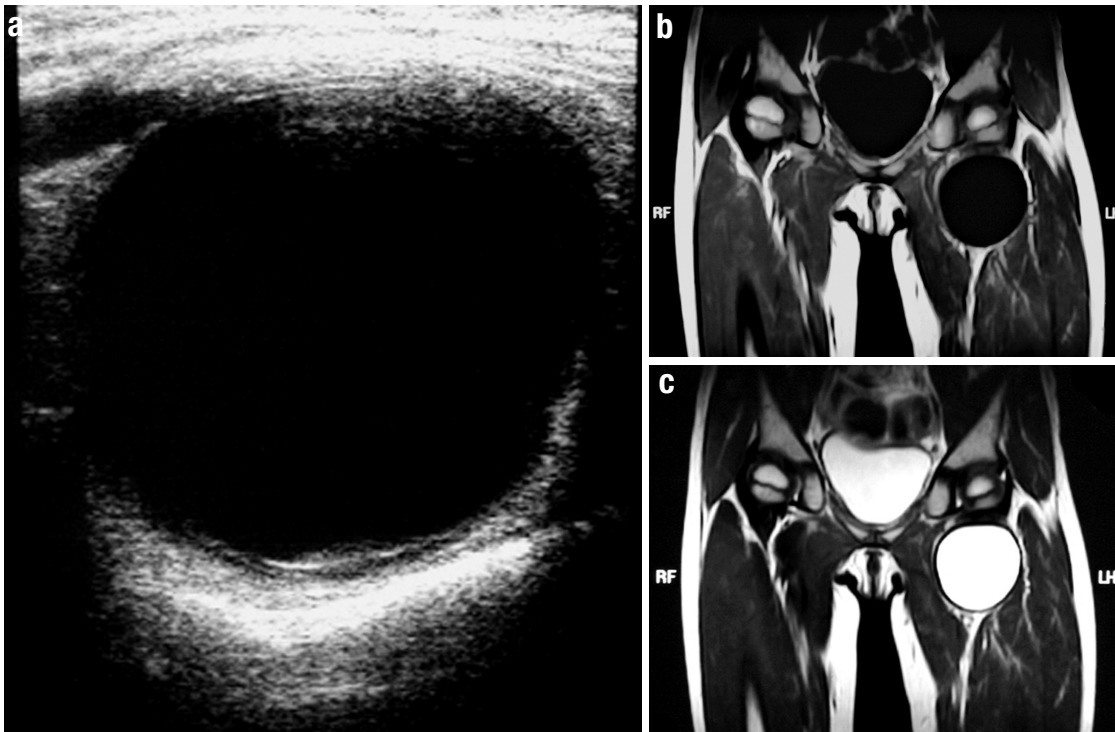


Figure 2. (a) Ultrasound image of the left thigh shows a unilocular cyst with anechoic, clear fluid content and smooth regular wall. Coronal T1-weighted (b) and T2-weighted (c) magnetic resonance images demonstrate a smooth, thin-walled, round cystic mass limited to the intermuscular area in the proximal thigh.

Case 2

A 4-year-old girl presented with painless mass in her left thigh. She was from the rural area. There was no history of trauma, abdominal pain, chest pain, urticaria, fever, or weight loss. Physical examination revealed a diffuse, non-tender, cystic swelling of approximately 5x5 cm, fixed to adductor muscle area with no evidence of local inflammation. Hematologic indices, including the eosinophile count, electrolytes, and total IgE were normal. However, the hemagglutination tests for echinococcosis were positive. Casoni's intradermal test was positive. Radiographs showed the femur was normal and there was a soft tissue mass at the

distal left thigh without calcification. CT showed no lung, abdomen, or brain involvement. US showed a thin-walled, anechoic, unilocular cystic mass in the left thigh (Fig. 2a). On MR imaging, a unilocular cyst with a low-intensity rim in all spin-echo sequences was found in the vastus medialis muscle and was considered a characteristic sign to differentiate it from other non-parasitic epithelial cysts. Cyst wall was thin, regular, not septated, and did not show enhancement. Cyst intensity was homogeneous (hypointense in T1- and hyperintense in T2-weighted images) and we could not see any daughter cysts in the cyst (Fig. 2b, 2c). The lesion did not show peripheral enhancement after administration

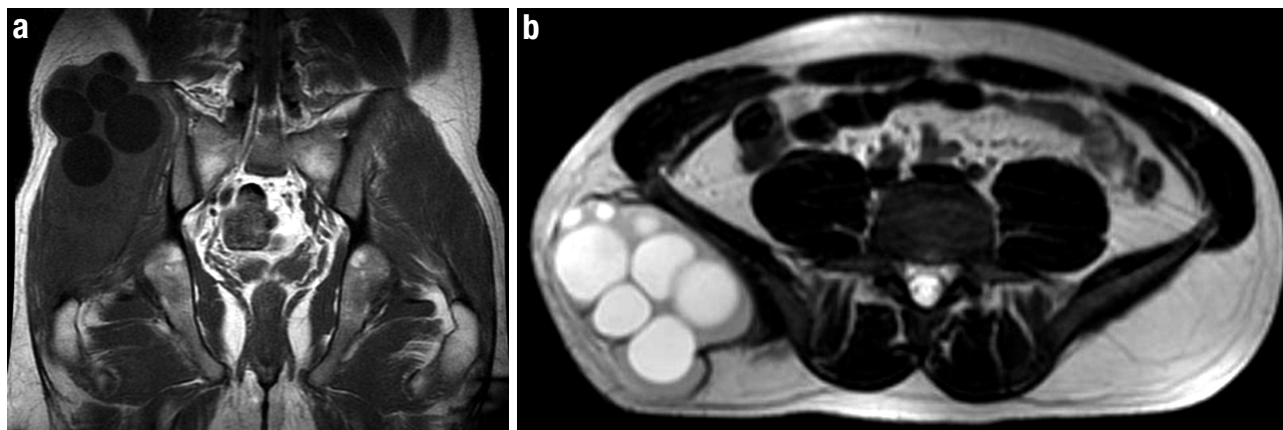


Figure 3. (a) Coronal T1-weighted and (b) axial T2-weighted magnetic resonance images show a well-defined cyst in the right gluteal region containing daughter cysts.

of gadolinium-DTPA. The chest X-ray as well as the ultrasound examination of the abdominal and pelvic cavity was normal. The lesion was defined as a type-I HC according to the Gharbi classification.

Case 3

A 43-year-old male patient was admitted to the hospital with a history of pain in the right gluteal region. There was not a history of fever, weight loss or trauma in the gluteal region. On physical examination, we located a mass in the upper part of the right gluteus muscle, however we did not demonstrate any fluctuation, erythema, ecchymosis, fever or lymphadenopathy. Anti-echinococcal IgG was positive. Radiographic examination of the pelvic region were unremarkable. In the MRI examination we demonstrated a mass of 3×6×6 cm in the right gluteus medius muscle, compatible with a HC (Fig. 3a, 3b). There wasn't any other lesion. The lesion was defined as a type-III HC according to the Gharbi classification.

In all the cases, postoperative histopathological examinations, established the diagnosis of an echinococcus cyst. Patients tolerated the surgery well and postoperative recovery was uneventful. Albendazole was given for 3 months postoperatively.

Discussion

Hydatid cyst is still a health care issue in most of the Mediterranean countries. Isolated primary hydatidosis of skeletal muscle is extremely rare. HC in an unusual location may be defined easily, if there is multiorgan involvement in the same patient.³ However, in the absence of multiple organ involvement, the diagnosis may be rather difficult. The reported soft tissue

hydatid cysts are usually secondary and have a predilection for the lower limbs.^{1,3,5} Primary hydatidosis of skeletal muscle is therefore rare, with reported prevalences of 0.5–4.7%¹⁻⁷, because the cyst uses oxygen for growth and muscles usually contain lactic acids.^{3,5} Soft tissue hydatid disease is unusual, even in endemic areas.⁸ HCs grow slowly, like benign tumors. The contractile nature of the muscles diminishes the growth process of the cysts.^{3,5,7} The tropism to the muscles of the neck, trunk and the root of the limbs can be explained with the increased vascularization and the decreased muscular activity in these areas.^{2,6,9} The main clinical finding of muscular HCs is a palpable mass.⁴

There are several classifications based on the radiological analysis of the morphology and the structure of the HC. They correspond to various developmental stages. In this report, we prefer the five patterns described by Gharbi et al. because they are simple to use with US, CT, and MR imaging (Table 1). Primary HCs of the lower extremity muscles are usually solitary and unilocular or multilocular. In our first and third cases, the lesions were located in the intramuscular region as an expansile multiloculated lesion with multiple daughter cysts. In the second case, it was located outside the

Table 1. Gharbi classification of hydatid cysts

Gharbi	Radiological characteristics
Type 1	Unilocular cyst + wall + internal echogenicities
Type 2	Detached membrane
Type 3	Multivesicular, multiseptated cyst, daughter cysts
Type 4	Heterogeneous cyst, no daughter vesicles
Type 5	Cyst with a wall calcification

muscles as an expansile unilocular cystic mass limited to the intermuscular area. In extensive literature search on primary intermuscular HC we could retrieve only three cases.⁸⁻¹⁰

Muscular HC is diagnosed by using the symptoms and the imaging studies like plain X-rays, US, CT and more recently MR imaging.^{8,9} Symptoms can arise from a mass effect or from the complications of the cyst. Symptoms can include vague pain, cough, low-grade fever, fullness, palpable mass and swelling. Specific diagnosis may be established by examining the fluids aspirated from the cyst, but this is not routinely recommended due to the risks of leakage, spread of the disease and anaphylactic shock.¹¹ US is a useful diagnostic tool showing the size, localization, and the type of the cysts. The sensitivity of US is 95%, and if vesicular fibrils are present, the sensitivity of US increases to 100%. Sonographically they have a thin or thick wall resembling the pericyst with internal echoes. Multiple echogenic foci due to hydatid sand may be evident for the snow storm sign. Simple cysts do not show internal structure. CT scan should be performed in suspicious cases, in order to demonstrate the cysts in relation with the adjacent organs.¹² On CT scan they appear as a well-defined cystic lesion with daughter cysts, that may contain wall calcification, septae or debris without the enhancement on intravenous contrast. MR imaging can adequately demonstrate most features of HC other than calcifications. In addition, MR imaging is superior to US and CT, depicting an exact compromise of adjacent soft tissues. MR imaging typically shows a thin, low intensity rim, probably representing the pericyst which is rich in collagen and is generated by the host. A low-intensity rim (rim sign), more evident on T2-weighted images, has been described as a characteristic sign in muscular HC; that is not a common finding in HCs located elsewhere in the body.²⁻⁶

The differential diagnosis varies with the types of HCs. Differential diagnosis of the muscular HC includes abscesses, chronic hematomas, lymphangiomas, synovial cysts, and necrotic malignant soft tissue tumors.^{7,11}

In conclusion, muscular HC is still an uncommon and important entity more frequently seen in rural areas. HC in soft tissues may present with a variety of patterns. HC should be included in the differential diagnosis where a cystic lesion in an unusual anatomic location is identified in a patient who lives in or has

come from an area in which the disease is endemic. The combination of the patient's history and clinical findings, imaging methods and serological tests may be helpful in the diagnosis of HC.

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