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A Case of Primary Adductor Muscle Hydatidosis: "Water-lily Sign" on Magnetic Resonance Imaging

Primary hydatidosis of muscle is very rare and can cause a variety of diagnostic problems. Its diagnosis is based on clinical, laboratory and radiological findings. As this infestation resembles a soft tissue tumor on clinical examination, the preoperative radiological diagnosis is very important to avoid unnecessary biopsy. The objective of this article is to present the pathognomonic "Water-lily sign" that has not been previously reported in this muscle group.

To the best of our knowledge, our case is the second reported case of this pathognomonic sign in magnetic resonance imaging of primary intramuscular hydatidosis.

Keywords: echinococcosis, magnetic resonance imaging, muscle

Introduction

Human hydatid disease is an infestation caused by the larval stage of Echinococcus granulosus.[1] It is acquired by ingestion of food or drink contaminated by the larvae. The incidence is highest in Mediterranean countries.[2] Hydatidosis frequently affects the liver and lungs accounting for approximately 90% of cases.[1] It is rarely seen in the spleen, heart, brain, bone and muscle.[2] Musculoskeletal hydatidosis is rare, accounting for only 3% of all cases[3] and primary hydatidosis of the skeletal muscle is extremely rare, even in endemic areas.[2,4-6] Magnetic resonance imaging (MRI) findings of muscle hydatidosis have been described in a number of case reports. Comert, et al., (2003) described the "Water-lily" sign in MRI of primary hydatidosis of the sartorius muscle for the first time.[1] In this case report, we describe the MRI findings, specially the pathognomonic "Water-lily" sign of primary hydatidosis in the adductor muscle of a 50-year-old woman.

Case Presentation

A 50-year-old housewife living in a village in Zanjan, northwestern Iran, presented with a six-month history of a slowly enlarging mass in the anteromedial aspect of her left thigh. Physical examination revealed a 6×5 cm, mobile, painless, firm mass. She was otherwise well before the onset of these symptoms. There was no significant past medical or family history. Ultrasound revealed a cystic mass with echogenic septations. To define the extent and characteristics of the mass, MRI of the left thigh was performed with a 1.0 TMR system (Intera Philips) using a body coil. The MRI study revealed a well-defined cystic mass measured 6×5×3 cm in the adductor muscles of the left thigh. The mass— the mother cyst—was hypointense on T1W spin echo sequence and hyperintense on T2W images. It contained smaller daughter cysts, which were hypointense to the

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surrounding fluid in all pulse sequences (Figs. 1 and 2; black arrows). The collapsed cyst membranes seen in the dependent part of the mother cyst were hypointense on both T1 and T2 pulse sequences, producing the "Water-lily sign" (Figs. 2 and 3; white arrows). The thin, incomplete wall surrounding the mother cyst was low signal intensity on T1W and T2W images (Figs. 1 and 2; white arrowheads). The second incomplete wall, showed as a high signal in T2W image noted in the periphery of the inner wall (Fig 2; black arrowheads). This wall was probably a result of the host reaction. The MRI characteristics of the lesion were regarded as diagnostic for hydatid cyst. The adjacent cortical bone showed no erosion. Chest radiograph, abdominal ultrasound exam and a computed tomography (CT) of the brain did not reveal any other organ involvements. A subsequent serology for hydatidosis was positive.

To avoid a possible anaphylactic reaction and seeding of the daughter cyst, biopsy was not performed. The lesion was completely excised in surgery. Histopathologic examination revealed a cyst wall with an outer chitinous (laminar fibrous) layer, surrounded by inflammatory cells consistent with hydatidosis (Fig. 4).

Discussion

Echinococcus granulosus infection does not usually present until adulthood due to the slow growing nature of the parasite in tissues such as muscle and bone,[7,8] although it can be acquired at any age. Diagnosis is made on clinical grounds with radiological and laboratory tests providing supporting evidence. Of these immunoelectrophoresis is the gold-standard.[5] Primary muscular hydatidosis is rare. Only a few cases have been reported.[2,4-6] Constant contraction of muscles and the production of lactic acid retard the growth of the cyst. Areas of predilection such as the neck, trunk and the root of limbs may be a result of relatively less muscle contraction in these areas and increased vascularization.[9-11]

In soft tissues the germinal layer of the cyst undergoes proliferation producing multiple vesicles in the mother cyst. On T1W images, the actual cysts present as a hypointense signal compared with the intracystic fluid. On the T2W images, the cyst gives low or high signal intensity.[2,12-14] In our case, the daughter cysts were low signal in both T1- and T2-weighted images compared with the intracystic fluid.

The "Rim sign" differentiates hydatidosis from non-parasitic cysts in the liver and lung. This represents the parasitic membranes (germinal or end cystic layer and pertinacious or ectocystic layer) and a membrane rich in collagen as a response of the host to the parasite (pericyst). The rim presents as low intensity especially in the T2W images.

The thickness of the cyst wall depends on the tissue in which the cyst is located—less developed in mus-

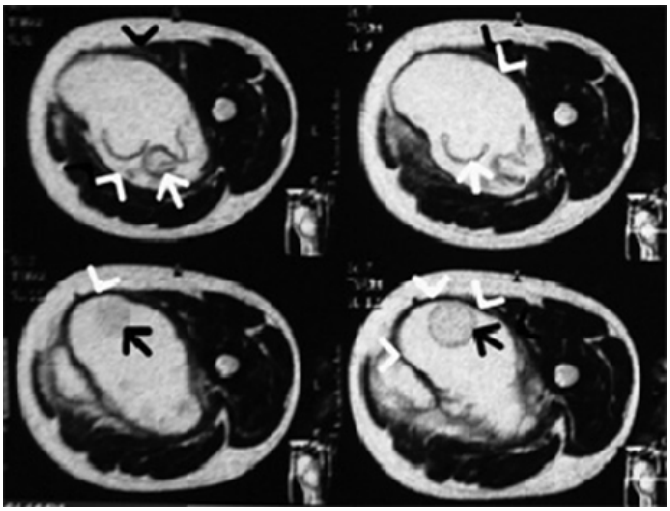

Fig. 1. MRI of hydatid cyst within adductor muscle containing daughter cysts. T1-weighted spin-echo axial images demonstrate multiple daughter cysts showing lower signal intensity than the mother cyst (black arrows), an incomplete hypointense rim noted in the periphery of the mother cyst (white arrow heads).

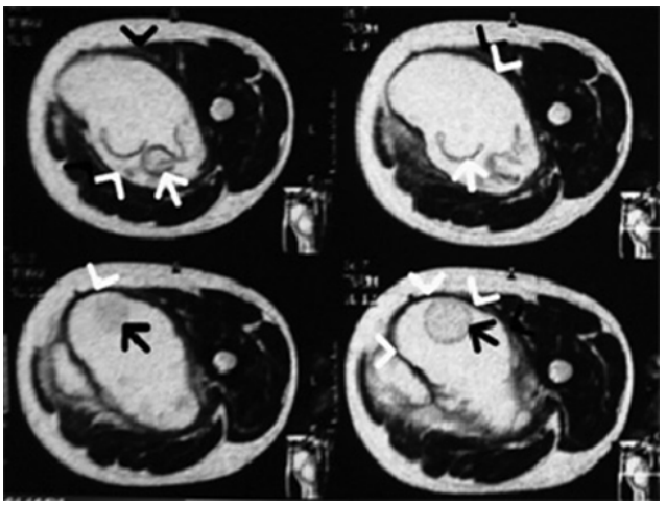

Fig. 2. MRI of the left thigh demonstrating "Water-Lily" sign. Axial T2-weighted images demonstrate collapsed membrane in the dependent portion of the mother cyst (white arrows) producing the "Water-Lily" sign, incomplete double layer of the mother cyst is also noted (white and black arrow heads). The daughter cyst has lower signal intensity than the mother cyst in T2-weighted images.

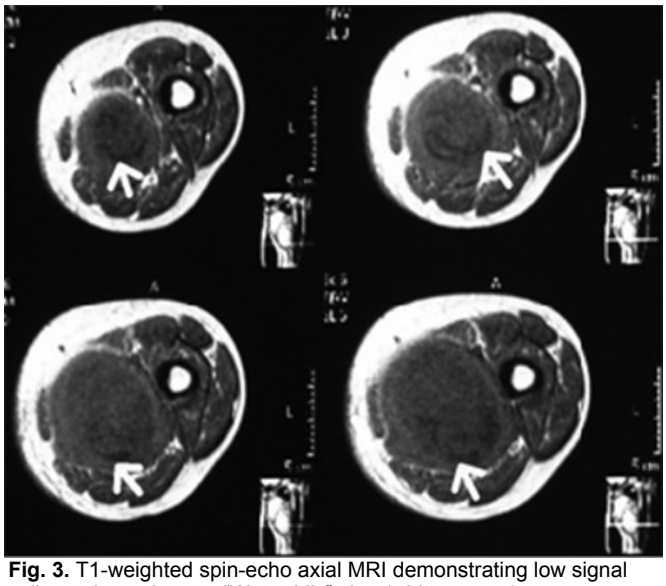

Fig. 3. T1-weighted spin-echo axial MRI demonstrating low signal collapsed membranes "Water-Lily" sign (white arrows).

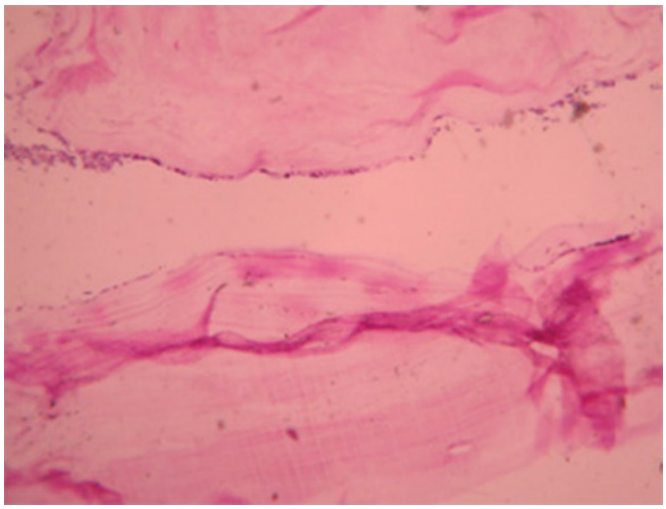

Fig. 4. Histopathologic examination of the left thigh mass revealed a cyst wall with an outer chitinous (laminar fibrous) layer, surrounded by inflammatory cells consistent with the diagnosis of hydatidosis.

cle and absent in bone, and sometimes visible in the brain.[2,12,14,15] We also noted a double layered rim in the superomedial portion of the cyst in this case—similar to the two cases described by Memis, et al,[16] and the case described by Comert, et al.[1] The inner layer was low signal in both T1- and T2-weighted images and the outer layer was isointense compared to the muscle in T1W and was hyperintense in T2W images. Cysts show an enhanced ring with gadolinium which is attributed to the vascularization of the pericyst. In infected cysts, a greater enhancement has been described due to the increased vascularity of this layer. Tarhan, et al,[17] noted detached membrane in their case but Comert, et al, first described the "Water-lily sign" as a pathognomonic sign for intramuscular hydatid cyst.[1] We also noted this sign in our case. Rarer forms of hydatidosis are the unilocular cyst (possibly the early step in development), and complex atypical and solid lesions (representing a hot abscess or soft tissue tumor resulting from the secondary infection or rupture of the cyst).[2]

Controversy exists as to the role of MRI in assessing the viability of the cyst with various findings being reported. Hydatid fluid release ceases and parasitic membranes collapse when the scolaces and hexacant embryos die. This death has been reported as hypointense daughter cysts in contrast to the mother cyst on T2W images.[12,18,19] Others favor proton density-weighted images generated by gradient-echo sequences as a sign of biological activity.[18] Yet other authors have reported the "Snake" or "Serpent" sign as representing a non-viable cyst. In this case, the collapsed paracystic membranes are seen as hypointense on all sequences.[20] However, some authors claim that there are no distinguishable features between sterile and fertile cysts. They argue that the varieties of MRI features are too broad to be of any diagnostic value.[18]

In summary, hydatid disease in soft tissues may present with a variety of patterns. Recognizing these patterns is important in making a differential diagnosis. MRI is an accurate method in characterizing hydatidosis in the typical forms—in particular the "Water-lily sign" that is pathognomonic of intramuscular hydatid disease on MRI. Complications can also be seen on MRI, which can delineate the exact extent of the disease and thus providing good surgical outlines. MRI may possibly also be useful in evaluating the vitality of the cyst.

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