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Evaluation of radiologic diagnostic criteria and treatment options in skeletal muscle hydatid cysts

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Abstract

Aim: To investigate the diagnosis and treatment methods of soft tissue involvement of hydatid cysts (HCs). **Materials and Methods:** Eleven patients who were diagnosed as having HC with muscular tissue (soft tissue) involvement between 2010 and 2016 were evaluated retrospectively. Seven patients had typical HC magnetic resonance imaging (MRI) and four patients had cysts with an unusual appearance. We evaluated how to diagnose the cysts using imaging methods, their characteristic radiologic images, and treatment alternatives against the disease. The patients were treated with antihelminthic chemotherapy preoperatively and postoperatively. **Results:** Of the 11 patients who underwent treatment, 7 were diagnosed using MRI and 4 were diagnosed with histopathologic examinations. The mean follow-up period was 16 (range, 6–24) months, and the mean age was 39.4 (range, 24–56) years. In seven patients, multivesicular appearance with specific MRI findings, T2-hypointense rim appearance, double-rim sign, membrane dissociation, and appearance of daughter cysts were identified as diagnostic criteria. Two patients underwent ultrasound assisted percutaneous aspiration–injection–reaspiration (PAIR) treatment. Seven patients underwent total pericystectomy, and two patients underwent subtotal pericystectomy with serum saline injection into the cyst. Two patients showed signs of mild anaphylaxis, one during the diagnosis and one during treatment. **Conclusion:** There may be difficulties in the diagnosis and treatment of HCs of the musculoskeletal system. It should be known that there are alternative methods in the treatment (cyst excision and PAIR treatment). Clinical, serologic, and radiologic findings should be used in the diagnosis. To avoid complications during the histopathologic diagnosis, MRI should be examined in detail. It is thought that atypical cysts can be diagnosed (double-layer appearance and peripheral rim sign) in addition to typical cysts (detached membrane and multivesicular appearance), and diagnosis and treatment can be planned without anaphylactic complications.

Keywords

diagnosis, hydatid cysts, magnetic resonance imaging, pericystectomy, soft tissue

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Introduction

Hydatid cyst (HC) is an endemic parasitic disease, which is the most common cause of echinococcus granulosus cestode in humans. It is more common in those who are engaged in agriculture and animal husbandry, and those in contact with dogs. It can be consumed in food contaminated by larvae.

Although it is more commonly seen in the liver (50–70%) than in the lung and other internal organs, it is rarely seen in soft tissues (1–4%). One of the reasons why muscles are a rare cyst location is that the cysts require oxygen to grow, but there is no suitable environment due to the accumulation of lactic acid in the muscles, and the growth of cysts slows down as the muscles contract.[1,2]

Clues in imaging modalities may help in the diagnosis of the disease in patients who are difficult to clinically diagnose.[3,4] Preoperative diagnosis of skeletal muscle HC is essential. It should be taken into account that spreading and anaphylactic shock may occur due to spillage during incisional biopsy. Magnetic resonance imaging (MRI) can adequately demonstrate many features of HC other than calcifications. Although serologic markers are generally used in the diagnosis, eosinophilia is an important finding in laboratory findings, but it may not always be seen.[5] There are some serologic tests that can help in the diagnosis (e.g. Western blot and hemagglutination antibody test), preferred treatment in musculoskeletal system HCs; however, cases of anaphylaxis and/or secondary echinococcosis may occur during surgery with the pouring of the contents of the cysts.[6] In preoperative radiologic evaluations, it is especially important with MRI to avoid biopsies or mistaken operative management of the cyst.

Given that HC is a rare disease, there is a lack of sufficient series in the literature, there is a possibility of anaphylaxis, and difficulties can be faced during the pathologic diagnosis, our aim was to describe the detailed imaging features of HC and to present the treatment options.

Materials and methods

Eleven patients who were diagnosed as having HC with muscular tissue (soft tissue) involvement between 2010 and 2016 were evaluated retrospectively. The patients comprised nine men and two women. The mean age of the patients was 39.4 (range, 24–56) years. Findings that might be specific in MRI were recorded. Cysts were seen in seven patients in medial adductor muscles, in two patients in gluteal muscles, in one patient in elbow muscles, and in one patient in thoracolumbar region muscles. Albendazole was given at dosages of 10 and 15 mg/kg/day to patients weighing <60 kg and >60 kg, respectively, for 2 weeks prior to surgery and continued for 3 months postoperatively. During surgery, the operation area was completely washed with 20% hypertonic sodium chloride. T1 and T2 multiplanar fast spin echo images were evaluated in at least two planes in MRI in each patient. Post-contrast images were obtained by giving intravenous gadolinium to all patients. Seven patients were diagnosed using MRI and excision was performed. Cystic contents were diagnosed by puncture in one of four patients, and a histopathologic examination was performed after total excision in two patients. One patient underwent total excision after histopathologic diagnosis with biopsy.

Results

Two patients underwent ultrasound-assisted percutaneous aspiration–injection–reaspiration (PAIR), and seven patients underwent total pericystectomy. Two patients with large diameter (>25 cm) HC and adjacent vascular nerves were treated with 20% serum saline injection and underwent intracystic washing and aspiration followed by subtotal pericystectomy. An attempt was made to define features that might be specific in MRI. Classifications were made according to detailed imaging characteristics. Erythrocyte sedimentation rate and C-reactive protein values were elevated in three of our patients, and no lesions were found in the liver or any other organ. The indirect hemagglutination test was positive in two patients. No recurrence was seen in any patients. A superficial infection was observed in one patient, which improved with antibiotic treatment. One patient who was treated with PAIR also received antibiotics and underwent surgery because of a deep infection and abscess. Mild anaphylaxis-like symptoms were seen in one patient during the diagnosis and one patient during the treatment.

In MRI, the typical multivesicular appearance was seen in 7 of the 11 (63%) patients with intramuscular HC (Figure 1(a)). In 4 of the 11 (36%) patients, unilocular cysts were seen (Figure 2) and detached membranes were seen in 1 patient (Figure 1(b)). A hypointense peripheral rim was seen in T2-weighted images in eight (72%) patients (Figure 1(c)). MRI revealed a typical double-layered appearance in 4 of the 11 patients (36%) (Figures 1(c) and 2). In seven (63%) patients, the mother cysts appeared hypo- and hyperintense in T1- and T2-weighted images, respectively (Figure 1(a) to (c)). In four patients, the mother cyst appeared mildly hypointense with T1 and mildly hyperintense with T2-weighted images. In all seven cases that had a multivesicular appearance, the daughter cyst was hypointense in T1-weighted images. In contrast, the daughter cysts appeared hyperintense in five cases and two were hypointense in T2-weighted images. In both groups with low and high signal intensity, viable scolices and sterile cysts were detected, and there was no association between cyst viability and daughter vesicle T2 signaling.

The MRI characteristics and surgical interventions are summarized in Table 1.

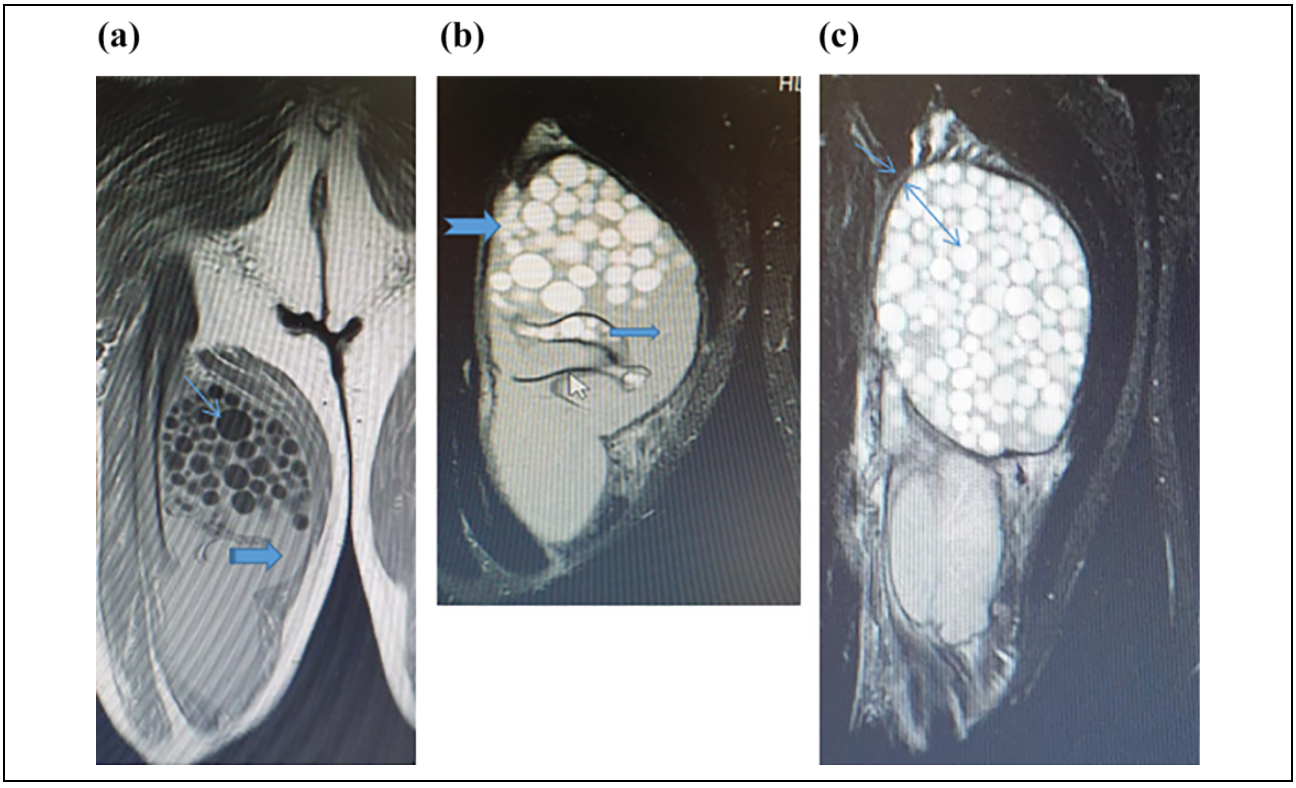

Figure 1. (a) Intramuscular hydatid cyst with typical features. A coronal T1-weighted images reveals a multivesicular cystic lesion within the right adductor muscle. The mother cyst (thick arrow) shows intermediate signal intensity while the daughter cyst (thin arrow) appears hypointense. (b) Coronal T2-weighted fat-saturated MR image of the same patient. The daughter cysts appear hyperintense (thick arrow) compared with the intermediate signal mother cyst (thin arrow). Also note the detached hypointense membranes (white arrow). (c) Same patient as in (a) and (b). The typical double-layered appearance with a hypointense inner rim and hyperintense outer layer is depicted (arrows). MR: magnetic resonance.

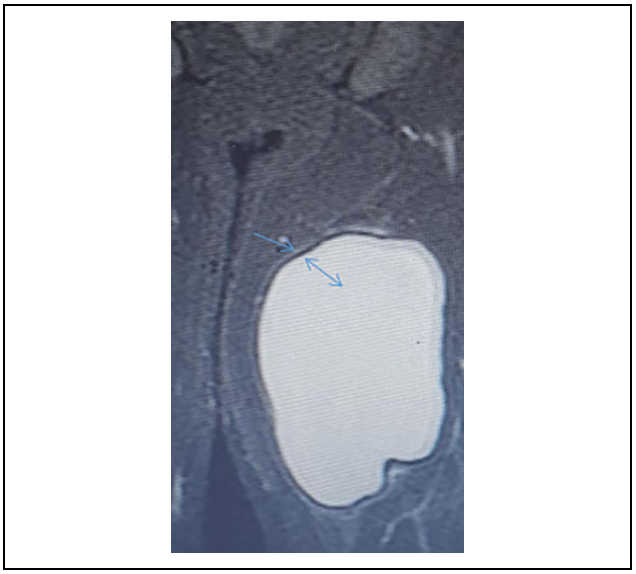

Figure 2. A 30-year-old man presented with a mass in the right medial thigh. Coronal T2-weighted fat-saturated MR image reveals a unilocular cyst that was finally proven to represent intramuscular hydatid cyst in histopathologic analysis. The double-layered appearance (arrows) typical for hydatid cyst was only seen at retrospective evaluation. MR: magnetic resonance.

Discussion

HC is a parasitic disease caused by parasitic echinococcus granulosus and can be seen at any age. The diagnosis of hydatid disease of the musculoskeletal system is difficult clinically and radiographically. Cysts remain clinically silent for a long time and radiographic findings may be nonspecific.[7,8] MRI is capable of adequately demonstrating many features of muscle hydatid disease. Although soft tissue and muscle involvement is not very common, there are some studies in the literature and case series in which HC was seen in the body of the thighs, and adductor, gluteal, and pelvic muscle groups.[9–12] Intramuscular HC in chest wall muscles and pectoralis major, sartorius, quadriceps muscles have been reported.[2,13]

Treatment consists of surgical excision of the diseased sections along with antihelminthic chemotherapy.[14–16] Antihelminthic chemotherapy has been reported to decrease the number of live cysts and the risk of recurrence.[14,17–19] To prevent the spread of infection to healthy tissue during surgical intervention, cysts should be removed without rupture. There are studies reporting the use of alcohol, formalin (10%), silver nitrate (0.5%), povidone iodine (10%), and hypertonic saline (20%) to kill

Table 1. The magnetic resonance imaging characteristics and surgical interventions.

Patient	Age	Location	Multivesicular	Rim	Double-rim	Detached membrane	Surgery method
MAÜ	31	Thigh	+	+	+	+	S. pericystectomy
SB	32	Thigh	▬	+	+	▬	PAIR
SS	42	Lumbar	+	+	▬	▬	S. pericystectomy
AB	32	Thigh	+	▬	▬	▬	PAIR
NI	38	Thigh	▬	+	▬	▬	T. pericystectomy
AA	44	Elbow	+	+	▬	▬	T. pericystectomy
NS	46	Thigh	+	+	+	▬	T. pericystectomy
HK	38	Thigh	▬	▬	▬	▬	T. pericystectomy
MS	56	Gluteal	+	▬	▬	▬	T. pericystectomy
SR	26	Gluteal	▬	+	+	▬	T. pericystectomy
YK	51	Thigh	+	+	▬	▬	T. pericystectomy

S. pericystectomy: subtotal pericystectomy; T. pericystectomy: total pericystectomy; PAIR: percutaneous aspiration–injection–reaspiration.

cysts.[8,16] In our study, we performed a subtotal excision in which we cleaned the cyst with hypertonic saline and washed into the cyst using hypertonic saline and we did not encounter anaphylaxis. The patients who had mild anaphylaxis-like symptoms included one patient in whom cyst excision was performed and one patient who underwent diagnostic cyst puncture. According to some authors, PAIR treatment is a common technique in liver HCs but is not usually applied in muscle or bone HCs.[20] In contrast, we found successful results in patients who underwent PAIR treatment. Marginal resection of muscle HCs has been reported, and high recovery and low recurrence rates have been reported.[18,21] Similarly, we observed no recurrence in any of our patients who underwent marginal resection.

In a series of seven cases reported by Madhar et al., four patients had a typical multivesicular cyst in MRI, and three patients had univesicular cysts.[22] García-Díez et al. detected unilocular cysts in two of seven patients with MRI.[9] In the study conducted by Arazi et al., seven of eight patients with muscular HC had unilocular cysts and one patient had multivesicular appearance.[18] In 17-patient series by Arkun et al., in 5 patients who had only muscle and soft tissue involvement on MRI, 3 patients and 2 patients had multivesicular and univesicular appearance, respectively.[23] In our study, seven patients had multivesicular appearance, which was typical and diagnostic for HC and four patients had univesicular appearance. Univesicular HCs have greater potential for mixing with other pathologies in the differential diagnosis and should accordingly be evaluated with more care.

Although synovial cysts, abscesses, hematoma, and necrotic tumors form unilocular cysts with a similar appearance to HCs, a T2-hypointense rim and double-wall appearance are very characteristic for HCs.[24] The T2-hypointense rim that surrounds cysts has been defined as a characteristic marker for liver and lung hydatidosis.[9] The rim consists of parasitic membranes (endocyst and ectocyst) and a vascularized membrane (pericyst) formed in response to the host. A hypointense peripheral ring representing a non-vascularized ectocyst in T2-weighted images can be seen in soft tissue HCs. Recently, it was shown that the rim contains two layers; the hypointense inner layer of the rim on T2-weighted images represents an acellular laminating membrane ectocyst and the outer hyperintense layer represents the vascularized pericyst.[24,25] In a study investigating the characteristics of soft tissue HCs, the T2-hypointense rim marker was reported in only 28% of patients.[9] In contrast to this study, the presence of a hypointense rim in T2-weighted series was 72% (8/11) in our study. In addition, four patients had a double-layer appearance in the cyst wall. We concluded that the diagnosis of HC could be made in the preoperative period in unilocular cysts without using biopsy in the presence of a hypointense rim and a double-wall appearance; it was thought that anaphylaxis-like symptoms during the diagnosis could be prevented in these patients.

When the correlation between the viability of cysts and MR images was examined, some authors could not identify the diagnostic characteristics of viable cysts and sterile cysts, whereas for others T2-weighted images correlated with high signal intensity and viability, and these were confirmed in microscopy examinations.[9,26] In our study, viability scolices were observed in three of five cases with hyperintensity with T2, and hydatid scolex viability was not detected in two cases. In the microscopic examination, a scolex was found in one of the two cases in which the daughter vesicles were more hypointense than the main cyst in the T2-weighted images; no scolices were detected in the other patient. We could not decisively conclude whether there was a relationship between T2 signaling and viability because we did not have a sufficient number of patients. In our study, there was no association between cyst viability and daughter vesicle T2 signaling.

In some studies, it has been stated that collapsed parasitic membrane appearance (dissociated membrane) secondary to damage or degeneration of HC can be seen in cysts located in the musculoskeletal system, appearing hypointense in all sequences.[27,28] In our 11-patient series,

1 patient had a dissociated membrane appearance in hypointense appearance in all sequences.

There were some limitations of the study. The choice of treatment was made according to the surgeon's preference. Also, the number of patients participating in the study was limited because muscle HCs are rare; there are no large patient series in musculosceletal system, even in endemic areas

Conclusion

In conclusion, in this study, successful results could be obtained through the addition of PAIR treatment and/or perysistectomy to antihelminthic chemotherapy. In addition, we concluded that the detection of a peripheral rim and double-wall appearance in unilocular HCs increased diagnostic accuracy.

Authors' contribution

Writing the article, revision, and accomplishment of the surgeries by HUO and RK; entire intellectual concept of the article by HUO; statistical analysis and review of the article by ÖK; data analysis and writing of the article by CY; data collection by KY; and review of the article by ÜT.

Consent for publication

Written consent to publish patient identifiable information and data was obtained from patients.

Ethics approval and consent to participate

The ethical research committee approved study protocol (AŞEAH/10.04.2019-426). Written consent from the patients next-of-kin has been consent.

Declaration of conflicting interests

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