

Contribution of Imaging in the Hydatid Cyst of Soft Tissues in the Thigh: A Case Report and Literature Review

M. El Hassnaoui^{1*}, H. Loukili¹, Y. Bouktib¹, A. El Hajjami¹, B. Boutakioute¹, M. Ouali Idrissi¹, N. Cherif Idrissi Gannouni¹¹Radiology Department, ARRAZI Hospital, Mohammed VI University Hospital, FMPM, Cadi Ayad University, Marrakech, MoroccoDOI: [10.36347/sjmc.2024.v12i05.033](https://doi.org/10.36347/sjmc.2024.v12i05.033)

| Received: 22.03.2024 | Accepted: 30.04.2024 | Published: 16.05.2024

***Corresponding author:** M. El Hassnaoui

Radiology Department, ARRAZI Hospital, Mohammed VI University Hospital, FMPM, Cadi Ayad University, Marrakech, Morocco

Abstract**Case Report**

Hydatid cysts in soft tissues remain a rare occurrence, even in endemic regions. This study underscores the crucial importance of imaging modalities for positive diagnosis and topography, providing optimal guidance for effective management strategies. In this article, we present a case of soft tissue hydatidosis in the thigh, directly explored through magnetic resonance imaging (MRI), complemented by soft tissue ultrasound. The observed mass contained vesicles, localized in the thigh. The diagnosis of hydatidosis suggested by ultrasound was confirmed by MRI. A curative surgical intervention was performed on our patient. The diagnosis of soft tissue hydatidosis can be established when ultrasound and/or MRI reveal a multivesicular appearance. MRI proves to be the most useful imaging modality in cases of atypical or pseudo-solid lesions. The peri-cystic enhancement serves as an indicative feature of soft tissue hydatidosis compared to other locations. The study emphasizes the rarity of muscular localization of hydatid cysts and highlights the crucial importance of radiological, surgical, and preventive diagnostic approaches in managing this pathology. The heightened sensitivity of MRI in cases of atypical ultrasound features underscores its central role in the contemporary management of this complex condition, enhancing both diagnostic precision and treatment planning.

Keywords: Hydatid cysts, soft tissue hydatidosis, Radiology, topography.

Copyright © 2024 The Author(s): This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC BY-NC 4.0) which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use provided the original author and source are credited.

INTRODUCTION

The parasitic infestation caused by *Echinococcus granulosus*, known as hydatid cyst, poses a significant public health problem, especially prevalent in Mediterranean countries. The dog serves as the primary host for the parasite, while humans can contract the infection accidentally. Although the liver is the organ most frequently affected, with a prevalence of 60 to 70%, followed by the lungs, affected in 5 to 27% of cases, it is noteworthy that hydatid localization in soft tissues remains relatively rare, even in endemic countries, with an estimated frequency of less than 3% [1, 2].

Due to its often asymptomatic nature and slow progression, the diagnosis of hydatid cyst in soft tissues is frequently established late. Practitioners face significant challenges in choosing paraclinical investigations and implementing appropriate therapeutic management. To better understand the radiological diagnostic characteristics of peripheral muscular hydatidosis, this study relies on an observation conducted at the radiology department of Arrazi Hospital at the Mohamed VI University Hospital in Marrakech. This observation is complemented by an in-depth

bibliographic review, aiming to consolidate our knowledge of this pathology and provide crucial information to guide clinical management.

OBSERVATION

A 51-year-old patient, of rural origin with no notable medical history, presented to a traumatologist with a significant, painless swelling in the right thigh that had been present for several months and gradually increasing in size.

The patient was afebrile, and their general condition was well-preserved. Clinical examination revealed a large mass occupying the anteromedial compartment of the upper third of the right thigh, with normal overlying skin and no signs of neural or vascular compression.

The patient was referred to our facility for an MRI of the soft tissues to obtain a positive, topographical, and etiological diagnosis, as well as a possible preoperative mapping. The examination included T1-weighted FSE sequences with fat saturation, both pre- and post-Gadolinium injection in axial,

Citation: M. El Hassnaoui, H. Loukili, Y. Bouktib, A. El Hajjami, B. Boutakioute, M. Ouali Idrissi, N. Cherif Idrissi Gannouni. Contribution of Imaging in the Hydatid Cyst of Soft Tissues in the Thigh: A Case Report and Literature Review. Sch J Med Case Rep, 2024 May 12(5): 714-718.

coronal, and sagittal planes, as well as T2-weighted sequences without and with fat saturation (STIR).

In the medial compartment of the right thigh, the MRI revealed, at the upper third, multiple confluent intermuscular cystic formations of variable size and shape. These included a unilocular cyst and several multilocular ones grouped in a grape-like cluster, with the largest measuring 8.5 x 9.5 x 12 cm and 5.5 x 5.5 x 11 cm. The signal characteristics of the cystic formations showed isosignal on T1 and hypersignal on T2 and STIR, enhanced peripherally after contrast injection (Fig 3), enclosing daughter vesicles with a clear hypointense signal on T1 (Fig 1). Most cysts were surrounded by a thin hypointense peripheral wall in all sequences (Fig 2).

This cystic formation developed in the medial compartment of the thigh, displacing but not invading the superficial femoral vessels superiorly and medially and

the sciatic nerve posteriorly. The patient underwent a monobloc excision of the cyst (Fig 3), and the postoperative course was uneventful. Histological examination confirmed the diagnosis of muscular hydatid cyst.

A subsequent ultrasound of the right thigh, performed after the MRI, showed an intramuscular, hypoechoic, heterogeneous formation on the inner side of the thigh root, with regular contours and containing multiple vesicles. This supported the diagnosis of a multivesicular muscular hydatid cyst (Fig 4). Chest X-ray and abdominal ultrasound were normal. Hydatid serology was negative.

The patient underwent a monobloc excision of the cyst, and the postoperative course was uneventful. Histological examination confirmed the diagnosis of muscular hydatid cyst.

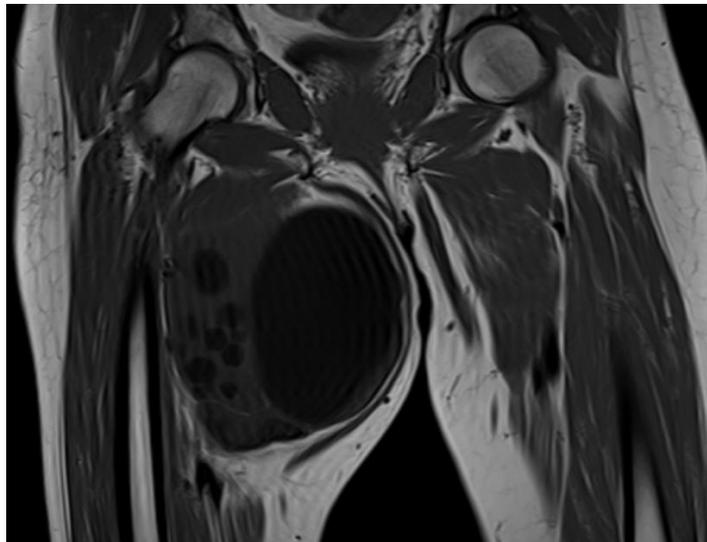


Figure 1: MRI. Coronal T1-weighted image: well-defined, multivesicular lesion located in the medial compartment of the thigh. Vesicles with distinctly hypointense signal compared to the rest of the cystic fluid

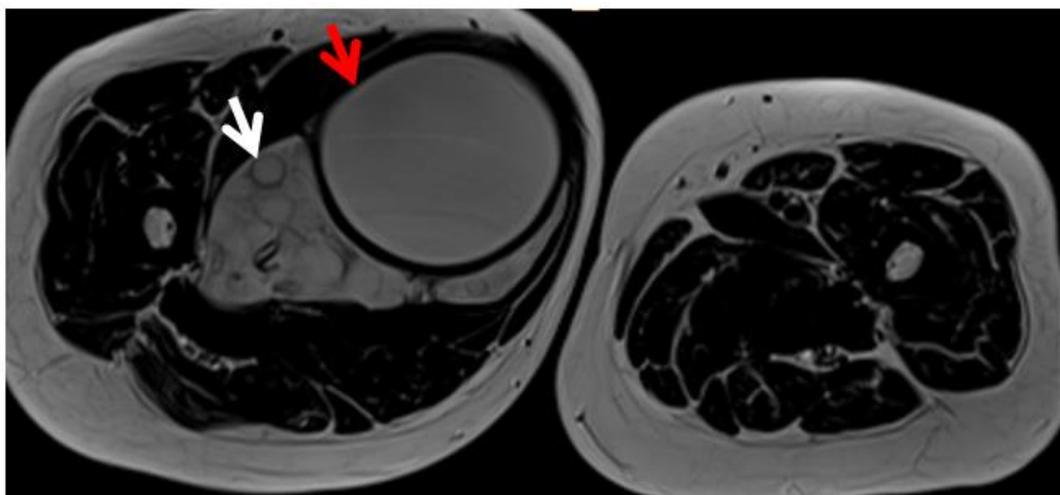


Figure 2: MRI. Axial T2-weighted image: cystic formation containing multiple daughter vesicles with hyperintensity (white arrow) and a hypointense pericystic rim (arrow)

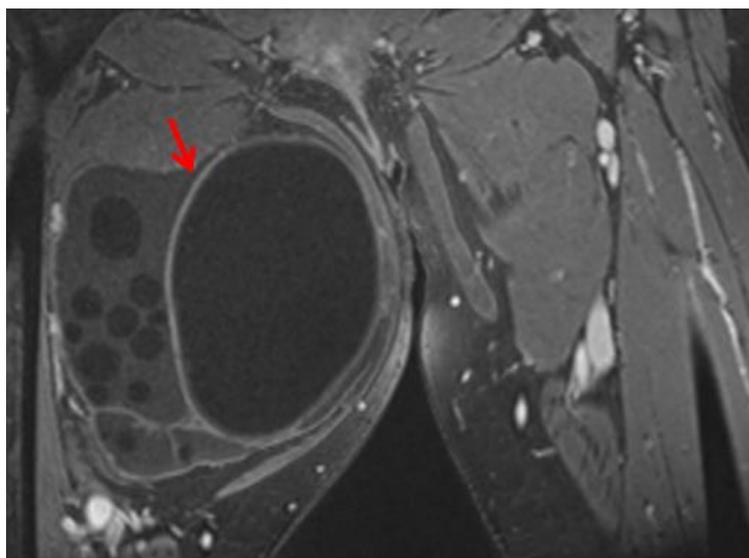


Figure 3: MRI. Coronal section of the left thigh in T1-weighted imaging with fat saturation after gadolinium injection: diffuse pericystic enhancement (arrows)

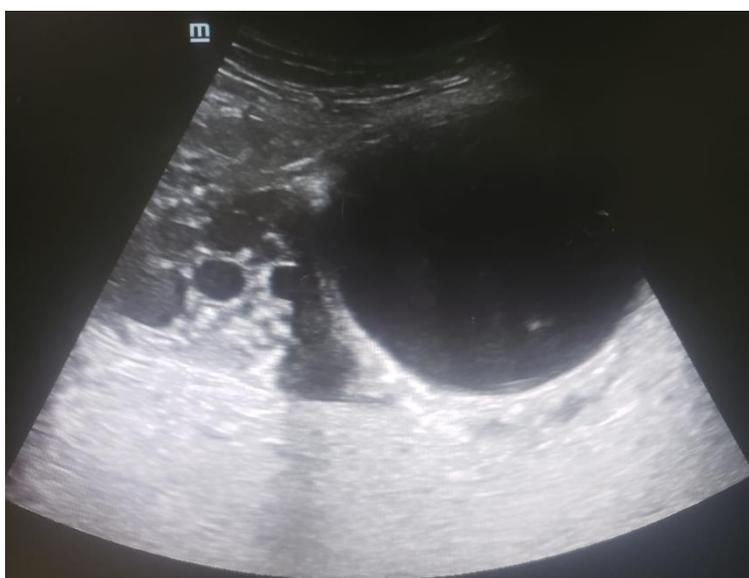


Figure 4: Ultrasound of the Right Thigh: A well-defined multivesicular cystic formation in the medial compartment of the thigh

DISCUSSION

Echinococcosis, a cosmopolitan anthroponosis, poses a significant health challenge globally, manifesting endemically primarily in regions where sheep farming is predominant, particularly in the Mediterranean, the Middle East, and South America [3]. The definitive host of this parasite is the dog, while the sheep acts as an intermediate host. Humans accidentally become infested and serve as intermediate hosts through the ingestion of food contaminated with the parasite's eggs or through direct contact with an infected dog. The ingested eggs traverse the intestinal wall, utilizing the portal system to disseminate throughout the body, thereby explaining the prevalence of hepatic involvement (60 to 70% of cases) and pulmonary involvement (5-27% of cases) [4].

It is noteworthy that these two organs, the liver and the lungs, function as a dual physiological filter, limiting the parasite's dissemination. This anatomical configuration explains the lower frequency of extrahepatic and extrapulmonary localizations. These organs act synergistically to restrict the migration of the parasite, creating a physiological environment that contributes to limiting the extrahepatic and extrapulmonary localizations of echinococcosis.

Hydatid localization in muscles is rare, with an estimated frequency between 1.75% and 2.42%, including locations in subcutaneous tissues [1, 2]. This rarity is primarily explained by the parasite's life cycle, which, utilizing the portal system, is interrupted in 80% of cases at the level of the liver and lungs [5]. Furthermore, local phenomena specific to muscles

hinder the nesting and growth of the parasite. The contractility of muscle fibers and the production of lactic acid, a toxic element for the parasite, limit its development. The parasite's particular tropism for muscles in the neck, trunk, and limb roots is attributed to the high blood flow and relatively low contractility of these muscle groups [2]. The primitive and isolated nature represents another peculiarity of this type of localization in most published case series, including the case we report.

There is no gender or age preference for this condition [6]. The clinical presentation is nonspecific, characterized by a painless mass without inflammatory signs and with preserved general health. In case of rupture or infection, the cyst can be complicated by a hot abscess or a malignant tumor [5, 6]. Serological tests for hydatidosis are often negative [5, 7].

Medical imaging plays a crucial role in the positive diagnosis of muscular hydatidosis, utilizing various modalities to assess this pathology. Standard radiography is an important initial step for excluding any bone involvement and can highlight calcifications in the case of an advanced cyst [6, 7].

Ultrasound typically serves as a key diagnostic tool. It confirms the cystic nature of the lesion, determines its size, location (above or below the aponeurosis), and classifies it according to the Gharbi classification. Its diagnostic sensitivity is high when proligamentous membranes and daughter cysts are visible, creating a multilocular "honeycomb" appearance [5], as was the case in our patient. However, atypical forms may present, where the lesion is either mixed or pseudo-tumoral solid, with or without rounded anechoic images, potentially making the diagnosis more challenging [6].

MRI presents itself as an advantageous alternative to CT, which has been traditionally utilized, to specify the number, size, topography, and relationships with vascular pedicles in hydatidosis. CT faces similar diagnostic challenges as ultrasound when the mass is not distinctly liquid and when vesicles and/or membranous structures are not patently identified.

Due to its high resolution, MRI allows for better local anatomical study and precise analysis of cystic wall structures [1, 8]. It proves to be a valuable tool for characterizing muscular hydatid cysts.

In soft tissues, the hydatid cyst typically contains multiple daughter cysts arising from the proliferation of the endocyst (proligamentous membrane), giving the appearance of "cysts within the cyst." These daughter cysts exhibit a distinct hyposignal compared to the rest of the cystic fluid on T1-weighted sequences and hypo- or hypersignal on T2-weighted

sequences [2, 3] depending on the presence or absence of scolex.

This semiology is perfectly illustrated in our first observation (Fig 1b-c). The peri-cystic hypo-intense rim, deemed characteristic, is more prominent on T2-weighted sequences, representing the hydatid membrane or endocyst, and a second collagen-rich membrane resulting from the host's reaction to parasitic infection, known as the peri-cyst.

The detached proligamentous membrane may give rise to a linear or ribbon-like hyposignal inside the cyst, described as the "serpent sign" by Anglo-Saxons. It serves as evidence of hydatid cyst involution, representing the detached and collapsed parasitic membrane that appears as a hyposignal on all sequences [2]. After Gadolinium injection, cysts may exhibit moderate peripheral enhancement due to vascularization of the peri-cyst. In T2-weighted imaging, Menis [1] described a wall consisting of two layers, internal in hyposignal and external in relative hyper signal, with the latter enhancing with contrast. However, in the case of cystic overinfection, more pronounced enhancement is noted due to both the hyper-vascularization of the peri-cyst and the edema and acute inflammation of surrounding soft tissues [1, 2].

This intense and relatively poorly defined contrast uptake in the surrounding soft tissues was observed in our case (Fig. 1e and 3d). This manifestation is quite unusual, as in most other locations, the complete absence of enhancement is more characteristic of a hydatid origin whenever the cyst is uncomplicated. It is noteworthy that, unlike computed tomography (CT), this type of enhancement is not clearly visible, and its absence is even considered supportive of the diagnosis. In contrast, magnetic resonance imaging (MRI) often demonstrates this enhancement [10-12].

Thus, hydatidosis in soft tissues can present various imaging aspects, and it is crucial to be familiar with them to establish a precise diagnosis before surgery. In cases of atypical, pseudosolid, infected, and/or ruptured cysts, MRI is preferred, especially when ultrasound has not been able to suggest or confirm the diagnosis of hydatidosis. Another advantage of MRI lies in its ability, through its multiplanar approach, to provide a precise topographic analysis of the cyst with surrounding structures, which is particularly useful before surgical resection [13].

Hydatidosis in soft tissues can manifest in various imaging aspects, and it is essential to be aware of them to establish a precise diagnosis before surgical intervention. In the presence of cysts with atypical features such as pseudosolid aspects, signs of infection and/or rupture, MRI is considered the preferred method, especially when ultrasound has not been able to suggest or confirm the diagnosis of hydatidosis. The presence of

daughter cysts and/or intracystic membranes helps establish the diagnosis, as does the visualization of a peripheral hypo-intense rim surrounded by a relative hyper-intense signal (on T2-weighted sequences), which may enhance after the administration of a contrast agent.

The treatment of muscular hydatid cyst is primarily surgical. Surgery must adhere to certain principles; it should be cautious, avoiding cyst opening during dissection [9-11]. Additionally, the surgical field should be protected with a hypertonic saline solution and/or hydrogen peroxide from the onset of surgery to prevent dissemination during the intervention, which can lead to perioperative anaphylactic shock [11, 12]. Monobloc excision with total pericystectomy is the ideal procedure but may not always be feasible, especially if the cyst is large, deep, and in contact with nearby vasculo-nerve elements [9, 11, 12]. In deep and large cysts, cyst aspiration followed by pericyst resection can be performed, preventing accidental rupture and content dissemination [11].

Medical treatment relies on the use of Albendazole [3]. However, the most effective means to combat hydatid disease, regardless of its location, remains prevention [7].

Combination drug therapies (albendazole, praziquantel) have demonstrated their effectiveness but come with adverse side effects, especially on the lymphatic system, and require a prolonged treatment duration [14].

Long-term patient monitoring is necessary to detect local or distant recurrence. This monitoring is based on clinical.

CONCLUSION

The muscular localization of hydatid cysts remains rare, even in endemic countries. A routine and inexpensive ultrasound scan can aid in the diagnosis, especially in typical cases. Currently, MRI stands out as the most effective tool for detecting and characterizing soft tissue masses, serving as the most valuable imaging modality in hydatid pathology of soft tissues, particularly when the ultrasound appearance is atypical.

Surgical intervention is the primary treatment, but the most effective approach to combat hydatid disease, regardless of its location, remains prevention.

REFERENCE

- Memis, A., Arkun, R., Bilgen, I., & Ustun, E. E. (1999). Primary soft tissue hydatid disease: report of two cases with MRI characteristics. *European Radiology*, 9, 1101-1103.
- Garcia-Diez, A. I., Ros Mendoza, L. H., Villacampa, V. M., Cozar, M., & Fuertes, M. I. (2000). MRI evaluation of soft tissue hydatid disease. *European radiology*, 10, 462-466.
- Dawson, J. L., Stamatakis, J. D., Stringer, M. D., & Williams, R. (1988). Surgical treatment of hepatic hydatid disease. *British journal of surgery*, 75(10), 946-950.
- Dirican, A., Unal, B., Kayaalp, C., & Kirimlioglu, V. (2008). Subcutaneous hydatid cysts occurring in the palm and the thigh: two case reports. *Journal of Medical Case Reports*, 2, 1-4.
- Cissé, A. M., Nassar, I., Hammani, L., Dafiri, R., & Imani, F. (2002). Hydatidose primitive et étendue de la cuisse: aspect radiologique inhabituel. *Journal de radiologie*, 83(11), 1778-1780.
- Dali M, Hssaida R. L'hydatidose musculaire. *Presse med* 2000;29:1166-9.
- Ben M'rad, S., Mathlouthi, A., Merai, S., Ghrairi, H., MESTIRI, I., & BEN MLIED M'RAD, K. (1998). Hydatidose multiple de la cuisse: Place de l'imagerie par résonance magnétique. *Journal de radiologie (Paris)*, 79(9), 877-879.
- Markonis, A., Tavernaraki, A., & Papaevangelou, M. (2001). Humeral hydatid cyst complicated with extraosseous involvement: a case of unusual location of echinococcosis. *European journal of radiology*, 37(2), 130-133.
- Bourée, P., & Bisaro, F. (2007). Hydatidose: aspects épidémiologique et diagnostique. *Antibiotiques*, 9(4), 237-245.
- Durakbasa, M. O., Kose, O., Islam, N. C., & Kilicoglu, G. (2007). A primary hydatid cyst of the gracilis: a case report. *Journal of Orthopaedic Surgery*, 15(1), 118-120.
- Filice, C., & Brunetti, E. (1998). Percutaneous drainage of hydatid cysts. *The New England Journal of Medicine*, 338(6), 392.
- Thursky, K., & Torresi, J. (2001). Primary muscle hydatidosis of the thigh: management of a complicated case with combination adjunctive albendazole and praziquantel chemotherapy. *Clinical Infectious Diseases*, 32(3), e65-e68.
- Örmeci, N., Idilman, R., Akyar, S., Palabıyıkoglu, M., Çoban, S., Erdem, H., & Ekiz, F. (2007). Hydatid cysts in muscle: a modified percutaneous treatment approach. *International journal of infectious diseases*, 11(3), 204-208.
- McManus, D. P., Gray, D. J., Zhang, W., & Yang, Y. (2012). Diagnosis, treatment, and management of echinococcosis. *Bmj*, 344, e3866.
- Rask, M. H., & Lattig, G. J. (1970). Primary intramuscular hydatidosis of the sartorius: report of a case. *JBJS*, 52(3), 582-584.