

# Muscular hydatidosis. An atypical location.

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**Abstract:** Introduction: Muscular hydatidosis is an extremely rare entity. Intramuscular hydatid cysts are usually secondary, resulting from spontaneous dissemination of the cysts or after operations for hydatid disease in distant regions. Material and methods: A case of multiple muscular hydatidosis is presented, which behaves like a soft tissue tumor that increases in size, with a review of the pertinent literature. Results: MRI images showed a mass in the medial intermuscular septum of the thigh, adjacent and anterior to the femoral vascular bundle. In turn, several satellite lesions are visualized. The extension study shows another lesion between the right lumbar psoas and paravertebral musculature in the posterior pararenal space. The diagnosis of hydatidosis was confirmed with BAG. The patient underwent surgery at another center for one of the lesions with poor evolution, finally opting for treatment with Abendazol. Conclusion: In geographic regions where hydatidosis is endemic, the hydatid cyst should be included in the differential diagnosis of a cystic mass in the muscle for its correct treatment.

**Key words:** hydatid cyst. Echinococcus. Musculoskeletal system. Atypical location. Muscular hydatidosis.

## 1. Introduction.

Hydatidosis is a zoonotic infection caused by the Echinococcus cestode. In our environment, the biological cycle of Echinococcus is usually maintained between canids (mainly dogs) and domestic ungulates (mainly sheep), with the possible accidental intervention of man by ingesting the larval stages with contaminated water or food, or more frequently, by direct contact with infested dogs [1] (Figure 1).

The Echinococcus species that most commonly affects humans is *E. granulosus*; Less common species are *E. multilocularis* and *E. vogeli*. It is endemic in many areas, such as the Mediterranean countries, the Middle East, South America, Australia, and New Zealand. Spain is among the European countries with the highest incidence of the disease in humans, with variable rates, but especially relevant in rural areas dedicated to raising sheep [2].

When humans ingest cestode eggs, the embryos enter the intestinal mucosa and are transported via the bloodstream to various organs. They commonly reach the liver (70%) and the lungs (20%); other organs such as the spleen, heart, and brain are rarely affected. Osteomuscular hydatidosis is rare, represents



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approximately 1 to 5% of all cases, with the most frequent locations being the neck, trunk, and roots of the extremities [2]. It has been suggested that muscle provides a poor environment for the parasite due to the presence of lactic acid [3].

We present below the case of a 69-year-old man with muscular hydatid disease who was referred to our center.

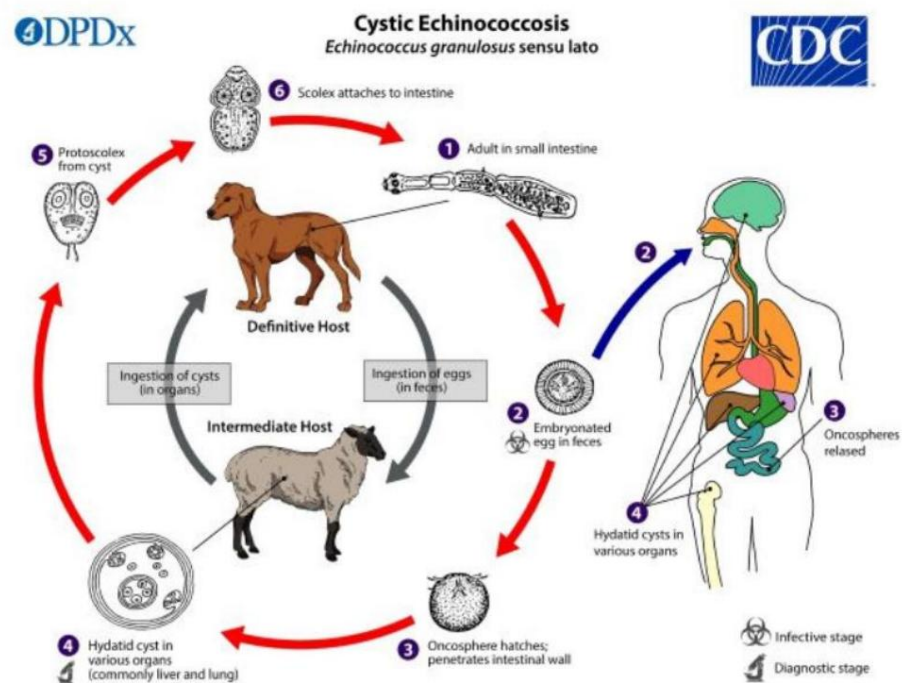


Figure 1. Echinococcus cycle. Image taken from the Centers for Disease Control and Prevention Image Library.

## 2. Clinical case.

A 69-year-old male with no relevant history, except being a caretaker for several dogs. He was evaluated at the Hospital Clínico de San Juan for suspected sarcoma due to a rapidly growing mass in the anterior compartment of the right thigh of about 15cm. During its study, an MRI was performed in which a large solid mass of 19x9x8cm was visualized, centered in the medial intermuscular septum of the thigh, adjacent and anterior to the femoral vascular bundle. In turn, several satellite lesions in the subcutaneous cellular tissue, popliteal fossa, and muscle bellies of the sartorius and vastus medialis are visualized.

Due to the suspicion of soft tissue sarcoma, an extension study was carried out using thoraco-abdomino-pelvic CT in which a homogeneous retroperitoneal mass without enhancement was observed located between the right lumbar psoas and paravertebral musculature in the posterior pararenal space (Figure 2). An extension study with PET-CT is completed, where the same lesions described in the CT and MRI can be seen, most of the lesions being ametabolic with the exception of the lesion present in the right thigh musculature.



Figure 4. CT image with pararenal lesion between psoas muscle and right paravertebral musculature.

After these studies, the diagnosis of metastatic sarcoma was considered and it was decided to perform ultrasound-guided AGB of the thigh lesion. The histopathological study reveals "chronic inflammation with the presence of multinucleated giant cells, with the presence of PAS-positive laminated remains compatible with the wall of a probable hydatid cyst." With the results of the anatomopathological study, the diagnosis of multiple muscle dose hidati was reached.

In June 2021, radical excision of a hydatid cyst in the vastus intermedius was decided medial and subcutaneous region of right thigh in anterior and posterior compartments.

Subsequently reoperated on several occasions due to bacterial superinfection of surgical wound. In suppressive treatment with Albendazole since the first surgery.

The patient was referred to our center from the Hospital Universitario de Torre Vieja for evaluation of multiple hydatidosis with a mother cyst in the peritoneal region between the psoas muscle and the right paravertebral musculature and a cystic lesion at the level of the iliotibial band of the right thigh on its distal slope due to spontaneous drainage. from the last batch. At the present time, the patient has no cutaneous signs of infection or pain. A new MRI of the thigh was performed to monitor the lesions (figure 3). After evaluation by the "Infectious Diseases Committee" and given the patient's refusal to undergo surgical treatment, it was decided to maintain suppressive treatment with albendazole and follow-up in outpatient clinics for clinical and radiological evaluation of the lesions.

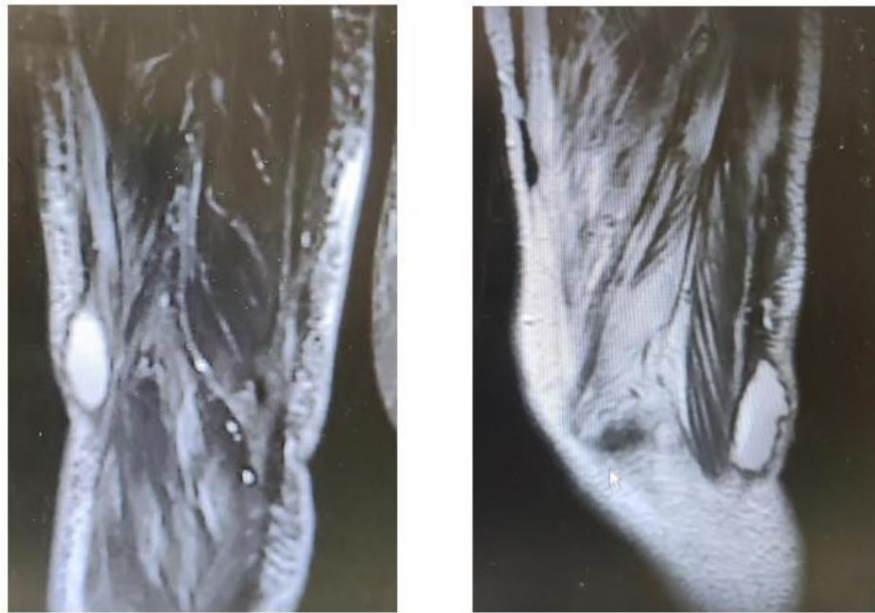


Figure 3. Last MRI control of a hydatid cyst in the thigh.

### 3. Discussion.

Hydatidosis is an infection caused in humans by the larval stage of basket two belonging to the Echinococcus genus of the Taeniidae family. Four species of Echinococcus responsible for the disease are recognized: *E. granulosus*, cosmopolitan, responsible for cystic echinococcosis; *E. multilocularis*, rare, responsible for alveolar echinococcosis; and *E. oligarthrus* and *E. vogeli*, very rare, responsible for multicystic equine coccosis.

While the other forms of echinococcosis are rare but serious or sometimes life-threatening diseases, cystic echinococcosis is endemic and treatable. The adult *E. granulosus* tapeworm lives in the small intestine of canids (definitive hosts). The gravid proglottids (eggs) are shed in the feces. After ingestion of the eggs by herbivores (intermediate hosts), the larvae are released into the small intestine (oncospheres). Once they reach the circulatory system, they can potentially land anywhere in the body, although they generally stop at the first two filters: the liver (70%) and the lungs (20%). As soon as the migration stops, the oncosphere turns into a slowly growing hydatid cyst (metacestode). The cyst capsule consists of a pericyst, from the host tissue, which envelops the endocyst of metacestode origin. The outer wall of the endocyst is the chitinous layer, the presence of which is characteristic on histopathological examination of *E. granulosus* cysts, while the inner wall is a germ membrane, which proliferates and produces a large number of small daughter capsules filled with protoscolices. . The life cycle is perpetuated when dogs eat parasitized organs of herbivores that contain protoscolices. Humans can accidentally become intermediate hosts by ingesting eggs in different ways. Eggs can remain viable in grass, fomites, sand or water surviving for months and can be eaten in contaminated food or water. They can also adhere to hands when a human pets an infested dog or cat, or touches contaminated soil or vegetation [4].

Considering that *E. granulosus* is ubiquitous, hydatid cysts have been diagnosed in almost all regions. Endemicity, however, particularly involves countries in the Mediterranean basin like ours. Life in the countryside and agriculture represent exposures with a higher risk for parasitic infection, since the possibility of ingesting echinococcus eggs is more likely to occur.

Isolated primary hydatidosis of skeletal muscle is rare and has been reported in only 1-5% of all patients with hydatidosis and in 2.3% of cases in endemic areas [4]. Muscle is not a favorable environment for larval growth due to the presence of lactic acid and mechanical factors, such as contractile activity, which make encapsulation less likely.

The preoperative diagnosis of muscular hydatidosis is clinically and radiologically difficult. The muscular Hydatid Cyst (QH) is an insidious infection without specific symptoms that grows gradually and can mimic any soft tissue tumor such as abscesses, chronic hematomas, synovial cysts and malignant tumors. Therefore, the diagnosis of soft tissue HC needs a high index of suspicion.

Imaging tests are always used as the first approach when it is necessary to investigate a subcutaneous mass, and it is advisable to start with ultrasound. The international classification proposed by the WHO [5] on ultrasound images in cystic echinococcosis makes it possible to distinguish the activity of the cysts (Figure 4). MRI is also a very sensitive test for the diagnosis of this disease. In particular, magnetic resonance imaging can identify various patterns of intramuscular hydatid disease, such as its peripheral border (known as the "ring sign"), membranes within the cyst, peripheral edema, and peripheral gadolinium enhancement (which is related to the vascularization of the pericyst) [2].

Tipo de lesión	Actividad	Ecografía	Esquema
Lesión quística (CL)	Activo (fértil)	Lesión quística unilocular sin pared visible	
Quiste hidatídico tipo I (CE 1)		Lesión unilocular con pared quística visible, arenilla hidatídica	
Quiste hidatídico tipo II (CE 2)		Lesión multivesicular, multiseptada, vesículas hijas visibles	
Quiste hidatídico tipo III (CE 3)	Transicional	Lesión unilocular con desprendimiento de la membrana laminar dentro del quiste	
Quiste hidatídico tipo IV (CE 4)	Inactivo	Lesión heterogénea hiper o hipocogénica sin vesículas hijas, con contenido degenerativo	
Quiste hidatídico tipo V (CE 5)		Calcificación quística de la pared quística, total o parcial	

Figure 4. Ultrasound classification of hydatid cysts according to the WHO [5].

If hydatidosis is suspected, serological confirmation should be obtained. Currently, it is performed through a combination of serum determinations; the most widely used are indirect haemagglutination (IHA), detection of indirect fluorescent antibodies (IFA) and enzyme immunoassays. The overall sensitivity of these tests in patients with cystic disease varies between 64-87% [1]. Needle biopsy should be avoided due to the risk of cyst rupture.

Once the diagnosis is confirmed, considering that pulmonary and hepatic locations are the most frequent, an extension study with at least one chest x-ray and an upper abdominal ultrasound is always necessary to define whether we are dealing with primary or secondary hydatidosis. In our patient, given the suspicion of sarcoma, the extension study was completed with a thoracoabdominopelvic CT scan.

Surgery is the treatment of choice for muscular hydatidosis and the best option for a complete cure. Chemotherapy with high doses of albendazole, mebendazole, or praziquantel may be considered if the cyst is inoperable due to its location. Prevention is key in this pathology, including education about the means of transmission. Personal hygiene and hand washing is essential in rural areas inhabited by dogs and cattle [6].

#### 4. Conclusions

In the differential diagnosis of soft tissue masses, the presence of hydatid disease should be suspected, especially in endemic areas. After collecting the clinical history, ultrasonography should be the first imaging approach and, if positive, an MRI should be accompanied. The history and physical examination together with imaging are usually sufficient to achieve a correct diagnosis.

Solitary subcutaneous localization should be confirmed by negative liver ultrasound and chest X-ray. Once the diagnosis has been made, the treatment of choice is pericystectomy and administration of anthelmintic drugs such as albendazole for prophylaxis of possible recurrences. In inoperable cases or patients who reject surgery, only pharmacological treatment can be performed with follow-up of the lesions.

**Funding:** This study has not received external funding.

**Conflicts of Interest:** The author declares there is no conflict of interest.

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