

HYDATID DISEASE IN MUSCULOSKELETAL TISSUES: DIAGNOSIS AND TREATMENT

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Abstract

Musculoskeletal hydatidosis is a rare occurrence of hydatid disease. Anatomical and clinical changes are, however, peculiar to this localization. Musculoskeletal hydatid disease is defined as development of multiple cysts within bone or soft tissue, that are the larval form of the tapeworm *Echinococcus granulosus*. Bone cysts account for only 0.5–2.5% of all hydatid cysts in humans.

We present a case of 39-years-old male, presented at our clinic with one month duration of pain in his right calf. Previously, there was asymptomatic swelling that existed one year, but as it got painful, the patient decided to seek medical help. Bone tumors, tumor-like conditions, and specific and nonspecific infections should be considered as a possible differential diagnosis.

The final diagnosis depends on histology. The treatment depends on surgical excision. Recurrence is common, particularly at sites that are difficult to access.

Keywords: Echinococcosis, Hidatid bone disease, tumor-like conditions.

Introduction

Echinococcosis is a parasitic disease, disease well known since the time of Hippocrates. It is widespread in countries where sheep are raised. Hepatic (75%) and pulmonary (15%) forms are the most common forms of the disease[1].

Cystic and alveolar echinococcosis are caused by *Echinococcus granulosus* and *Echinococcus multilocularis*. Only 10% of lesions are found in the bony human body. Musculoskeletal lesions present with serious problems, without visceral involvement. Nevertheless, hydatid skeletal occurrence is present in only 0.5 to 2.5% of cases.[2,3]

The spine is the most common skeletal localization (35-50%). Adult tapeworm is found in the intestines of dogs. The eggs are released into their stool, and are introduced into the intermediate hosts, usually sheep, where they pass through their intestinal mucosa and enter into their portal circulation.

After being released into the circulation, they pass through the filters of the liver and lungs before being transferred to the bones. The bony form usually begins in the bone metaphysis, which then spreads through its diaphysis. The daughter cysts extend into the bone tissue, replacing the medulla. Articular affection is usually due to secondary expansion from adjacent bone. Characteristic of this disease is that it may remain asymptomatic for a long period of time. It can sometimes take up to 10 to 20 years for it to become clinically manifest. It most often appears in the age group of 30 to 50 years.

There is no pathognomonic clinical sign or symptom. It can manifest as a pathological fracture, a secondary infection of the cystic mass, or just the appearance of constant local pain. Therefore, hydatid disease should be included in the differential diagnosis of any osteolytic bone lesion or soft tissue mass, especially in endemic areas, thus facilitating early accurate diagnosis, and therapy and treatment.

Because of the sometimes difficult definitive diagnosis, it is often made intraoperatively. Differential diagnosis includes a wide range of pyogenic infections, tuberculosis, fibrous dysplasia, bone tumors, benign cystic changes, multiple myeloma, as well as secondary deposits or metastases.

Radiographically, osteolysis is seen, and sometimes invasion of the bone cortex. CT is somewhat more specific, due to a more accurate assessment of bone destruction as well as in

determining extra-osseous spread. MRI is a superior and useful method in assessing extra-bone invasion of disease or extraskelletal soft tissue form of the disease[2,3]. A skeletal scan with Tc99m is often an unnecessary method when it comes to the appearance of this disease.

Case report

S.B., 39-years-old male, presented at our clinic with one month duration of pain in his right calf. Previously, there was asymptomatic swelling that existed one year, but as it got painful, the patient decided to seek medical help. It started in an outpatient orthopedic clinic, where ultrasonography was made, indicating a cystic lesion.

The patient was referred at our clinic from the University Clinic for Plastic Surgery. (Figure I) the biopsy specimen indicated an inflammatory process.

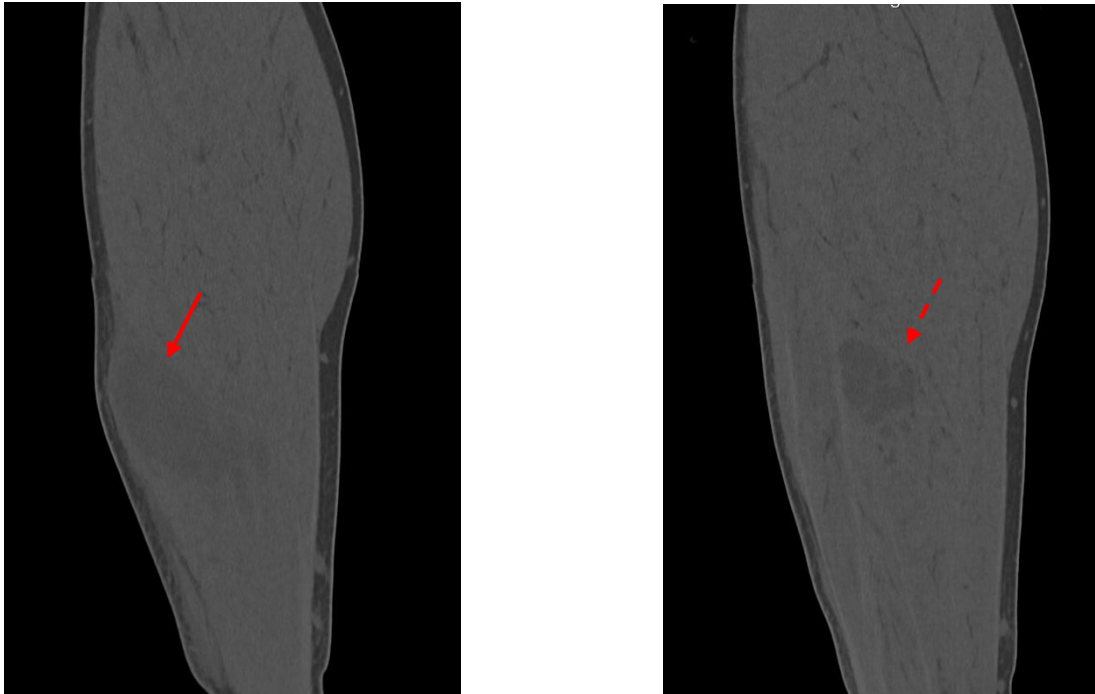


Figure I: a.

b.

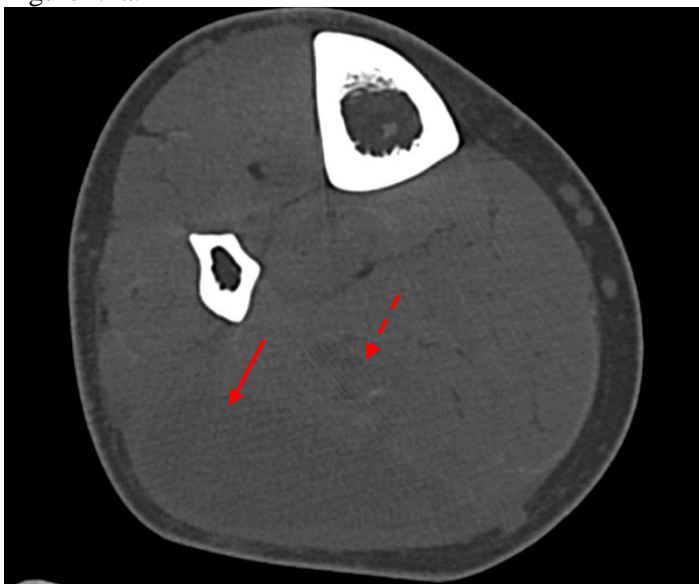


Figure I (a, b, c). CT scan of right calf in sagittal (a, b) and transversal plane (c) revealing two soft tissue changes resembling myositis, the first one unitary cystic (a, c; solid arrow), the other one lobulated cystic (b, c; dashed arrow).

At our clinic, MRI, wide laboratory analysis and CT angiography were performed.

The MRI revealed two lesions, the first one, homogenous and clearly demarked from the surrounding was abutting the lateral head of the gastrocnemius muscle and with dimensions 59,8 x 26 x 65 mm, showing hypersignal in T1 that was more accentuated in T2. The second lesion was multilocular, resembling a grape fruit, not demarcated clearly from the surrounding, giving a hypersignal in T2 and iso and hyposignal in T1. It was located in soleus muscle (figure II). Laboratory and CT angiography findings were normal (no pathologic vascularization was noticed).

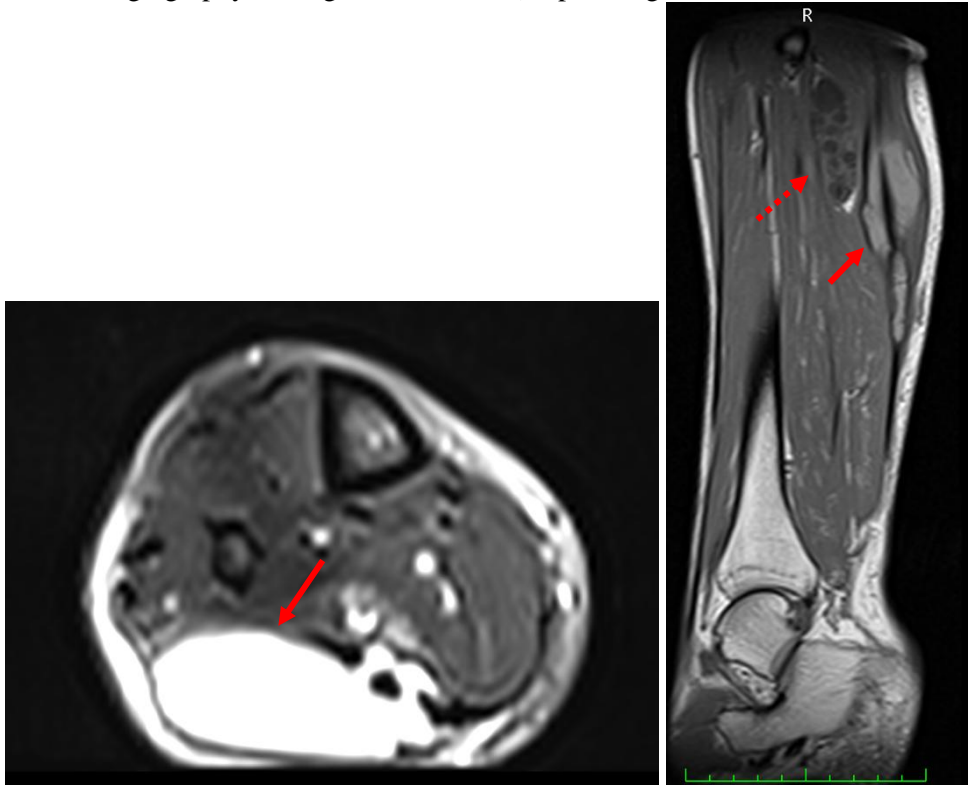


Figure II a.

b.



c.

Figure II (a, b, c). Two lesions revealing at the MRI finding, transverse (a; T2 weighted image) and sagittal cuts (b, c; T1 weighted images), the first one being unitary (solid arrow) and the other lobulated (dashed arrow).

All findings indicated benign lesion, so we decided to perform marginal excision of the painful lesion. Gross appearance of the first lesion was a large cyst filled with grey liquid material. The pathohistological finding revealed hydatid cyst. Postoperatively serologic and imaging tests were performed in order to assess the extent of the echinococcosis.

The serology test came back positive for IgG antibodies against *E. granulosus*, the imaging tests were negative (radiograph of the lungs and liver ultrasound). Also, the patient was sent to infectiology specialist. Two treatment cycles with albendazole were performed (albendazole administered per orally with meals in a dose of 400 mg twice daily for 28 days) [1-3].

The first year follow up was performed and one in the second year consisting of clinical examination and ultrasound of the calf.

The patient remains asymptomatic and the ultrasound finding unchanged.

Discussion

Hydatid disease is a well-known entity in the human parasitology and the most commonly affected organs are the liver and lungs[1,2].

The primary, musculoskeletal form of this disease, without any visceral affection, is uncommon and less well known, at about 0,5-2.5%, and is usually delayed in presentation because it is often misdiagnosed, especially if isolated on bone tissue alone[2,3].

Presence of hydatid disease should be taken into consideration, especially when talking about differential diagnosis of osteolytic bone and soft tissue changes. The presence of musculo-skeletal hydatid disease is mainly in absence of other systemic disease. Main diagnostic test is the ELISA serological test for presence of IgG antibodies against Echinococci.

The best possible treatment is surgical extirpation, with help from local and systemic chemotherapy. Long-term survival is expected, but the difficulty that appears with this disease is that systemic eradication and is hard to achieve and sometimes impossible to cure. Local recurrence is present in 40% of the cases, despite the use of the aggressive treatment.

Recurrence is described, even up to 28 months. Use of wide local surgical excision is not always the best possible solution. The intra-operative seeding of the disease is almost impossible to avoid. Mortality rates vary from 0-3%. The presence of hydatid cysts in other organs should be always excluded at first, but until now, the musculo-skeletal form is always an isolated form.

In summary, primary musculo-skeletal form of presence of hydatid cysts is an uncommon entity, and thus, can sometimes be misdiagnosed in the primary clinical and radiological evaluation. Liang et al reports nine cases of osseous hydatidosis involving the bone.

Patients were treated through different surgical options, including simple debridement, bone cement filling with or without internal fixation. Patients must be treated at the early stages of the disease. Internal fixation should not be used when surgical eradication is not clear.

For final diagnosis Liang et all used plain radiography, computed tomography (CT), and magnetic resonance imaging (MRI) Laboratory testing for echinococcosis was performed using eight immunodiagnosis tests, with ELISA and the gold-label method [4].

From 1964, Alldred and Nisbet *et al* advocated wide surgical excision when treating disease in long bones but followed a more conservative approach when treating disease in the axial skeleton. Booz, Hooper and McLean and Duran *et al* advocated thorough mechanical curettage to remove macroscopic cysts and “chemical” sterilisation of the scolices using formalin, 0.5% silver nitrate or hypertonic saline, any major defect being filled with autogenous bone graft [5].

Conclusion

Osseous hydatid disease is defined as development of multiple cysts within bone that are the larval form of the tapeworm *Echinococcus granulosus*

The imaging is nonspecific, even a CT and MRI-evaluation. MRI scan can sometimes identify specific features, and can lead to correct diagnosis. Although MRI and CT scan aren't 100% specific and sensitive methods of evaluation, they can help make the definitive diagnosis[3].

Clinical and serological correlation can also help to achieve the final diagnosis, when the imaging methods aren't sufficient and also non-specific. Most important, we must not forget this entity and always include as a possible diagnosis, when evaluation of bony and soft tissue pathologic entities is necessary.

The final diagnosis depends on histology. The treatment rests on surgical excision. Recurrence is common, particularly at sites that are difficult to access[3,6].

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