Case Report

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A rare presentation of hydatid cyst disease at the thigh: a case report from Syria

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Introduction and importance: Hydatid cyst disease is a zoonotic disease caused by *Echinococcus granulosus* and is recognized as a significant health issue in many countries, particularly Mediterranean countries. Hydatid cysts in the musculoskeletal system are rare, with only a few reported cases. These usually occur as secondary cysts resulting from the hematogenous dissemination from primary sites.
Case presentation: This paper reports a case of a 77-year-old man with a mass in his thigh that had been increasing dramatically in size for 4 months with no signs of local inflammation, fever, or any other symptoms. Findings from an MRI were consistent with a large abscess, so a true-cut biopsy was taken before referral. A pathological study after surgery revealed the lesion was a hydatid cyst, and a previously performed biopsy caused a rupture of the cyst.
Clinical discussion: Misdiagnosing hydatid cysts, especially in non-usual areas, may lead to an unwelcome biopsy and consequences.
Conclusion: This case report highlights the importance of considering hydatid cysts when encountering any enlarging mass, regardless of its location or rate of growth.

Keywords: case report, hydatid cyst, musculoskeletal system, Syria

Introduction

Hydatid cyst disease is a parasitic infection mainly caused by *Echinococcus granulosus*. It is a significant disease in certain parts of the world, especially South America, Southern Europe, Australia, New Zealand, Africa, Turkey, India, and the Middle East. This distribution was almost the same for the last 20 years. People become infected by ingesting *E. granulosus* eggs orally. The disease manifests with multiple cysts affecting various organs, particularly the liver and lungs[1–8].

Hydatid cysts can be asymptomatic or cause symptoms such as pain or mass effects. They are most commonly found in the liver (68.8–80%), followed by lungs (10–22.4%), with remaining cases occurring in other parts of the body[2,9]. Other organs like spleen, heart, and brain are rarely affected[9]. Soft tissue hydatidosis is rare, and musculoskeletal involvement is considered extremely uncommon[10].

HIGHLIGHTS

- Hydatid cysts in the musculoskeletal system are rare, with only a few reported cases. These usually occur as secondary cysts resulting from the hematogenous dissemination of cysts from primary sites.
- Misdiagnosing hydatid cysts, especially in non-usual areas, may lead to an unwelcome biopsy and consequences.
- This case report highlights the importance of considering hydatid cysts when encountering any enlarging mass, regardless of its location or rate of growth.
- Hydatid cysts may come with a vague presentation, and serological tests are not enough to exclude the diagnosis.

Hydatid cysts are usually asymptomatic until complications arise from rupture or infection. After many years of growth, they may cause symptoms due to the compression of surrounding tissues. Some cysts may spontaneously collapse or calcify, while others continue to grow in size and compress nearby healthy tissues and organs[9,10]. This case report presents a rare occurrence of a hydatid cyst within the thigh muscle. Since this presentation is highly unusual, this case report helps to expand the range of possible diagnoses for soft tissue masses in this location, particularly in areas where the disease is endemic. The case has been reported in line with the SCARE 2023 criteria[11].

Presentation of the case

A 77-year-old man presented to our tertiary university hospital with a gradually increasing swelling in the left thigh for about 4 months.

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Before referral, his primary physician ordered an ultrasound, which showed a 7.3 × 5.9 cm cystic mass with thick walls and irregular edges, containing multiple cystic cavities of different sizes, the largest of which measures 2 cm. This cystic lesion was located within the soft tissue in the middle of the left thigh. Another accompanying cystic mass above and next to the previous one, measuring 4 × 2.5 cm, was observed.

The primary physician regarded the lesion as a soft tissue mass, so an MRI was ordered, which showed a wide, heterogeneous signal area with unclear borders occupying the lateral aspect of the thigh, measuring 25 × 9 × 13 cm (Fig. 1). The lesion had a low signal on the first-time sequence and a heterogeneous high signal on the second and flare sequences, with multiple cavities. After a gadolinium injection, it was enhanced peripherally. The wall thickness was up to 10 mm. No damage to the periosteum or adjacent bone tissue was revealed. MRI findings suggested a large thigh abscess.

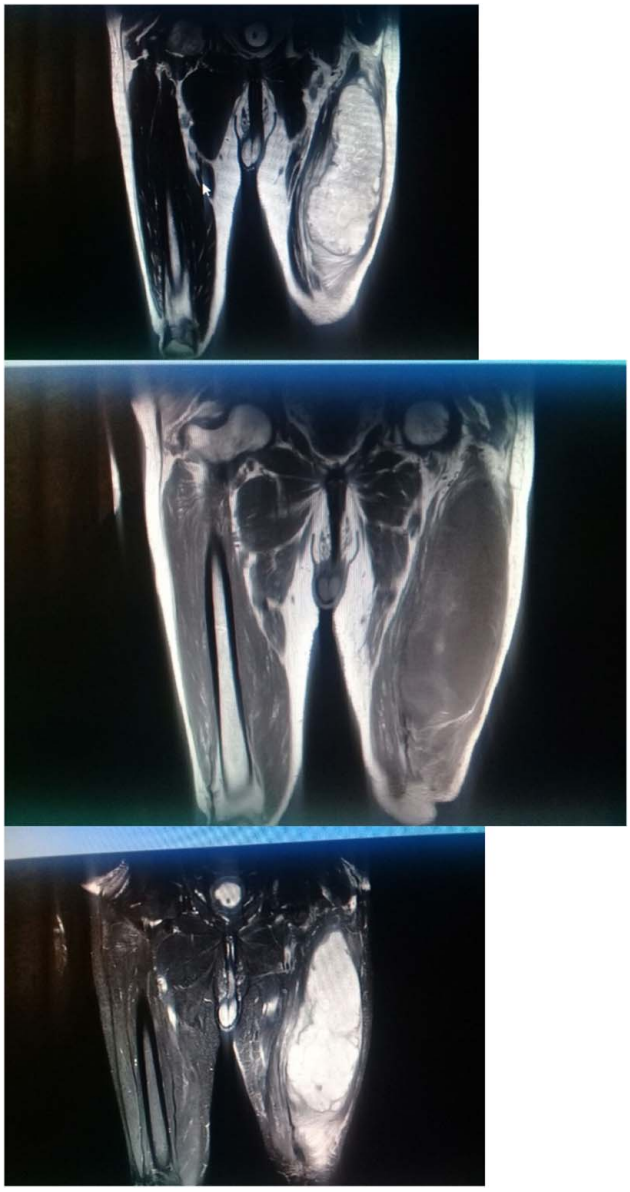

Figure 1. An MRI showed a wide, heterogeneous signal area with unclear borders occupying the lateral aspect of the thigh measuring 25 × 9 × 13 cm.

Unfortunately, based on these findings, a decision was made before referral to our tertiary university hospital to perform a true-cut biopsy of the lesion. The biopsy revealed pathological features consistent with a hydatid cyst and excluded malignancy.

On admission to our hospital, the patient's vital signs were within normal ranges. A physical examination revealed a cystic mass located on the lateral aspect of the left thigh with no signs of inflammation.

Laboratory tests showed an increased WBC count and neutrophil percentage, eosinophils 0.9%, international normalized ratio (INR) 1.43, glucose 120, urea 61, albumin 2.8, total protein 6, alanine aminotransferase (ALT) 29, aspartate aminotransferase (AST) 20, total bilirubin (TB) 0.57, and direct bilirubin (DB) 0.17.

Chest and abdominal computed tomography (CT) revealed no other cysts in the liver or lungs, excluding any other cysts that may need intervention before the thigh lesion.

Pulmonary and anesthesia consultations were done, and the patient was being prepared for surgery.

During surgery, epidural anesthesia was administered, and a longitudinal anterolateral incision was made. An abscess cavity was found full of daughter cysts. Good drainage was performed and the cysts were removed. And a drain was inserted.

The patient recovered well in the surgical ward with no complications and was discharged from the hospital after 2 days with no reported signs or symptoms. The patient was followed up for about 3 months with no complications.

Final pathological analysis confirmed the diagnosis of hydatid cysts (Fig. 2).

Discussion

Hydatid cysts were first diagnosed during Hippocrates' time. It is an infectious disease caused by the parasite *Echinococcus granulosus* and recognized as a significant health issue in many countries, particularly Mediterranean countries[4,5,10,12]. Wolves and dogs are primary carriers, while sheep, cattle, and deer are secondary hosts. Humans usually become infected by consuming vegetables contaminated with the feces of infected dogs. As a result, humans are considered accidental hosts[4,9]. Hydatid cysts are almost always found in the liver and lungs[3,7]. This is because the eggs hatch in the intestines and are then transported to the liver through the portal system[4,5,13,14].

In the musculoskeletal system, hydatid cysts are considered rare with only a few reported cases in literature. This may be due to the presence of lactic acid in muscles, which creates an environment that inhibits the growth of hydatid disease. Muscular hydatid cysts usually occur as secondary cysts resulting from the hematogenous dissemination of Echinococcus cysts from primary sites such as the liver and lungs[10,14].

Symptoms and signs vary depending on the affected organ and its surrounding tissues. Complications can include cyst rupture, immunological reactions, and secondary infections. However, hydatid cysts grow slowly and remain asymptomatic for several years until they rupture or become complicated. The most consistent clinical finding for cysts affecting soft tissues is a palpable mass, and clinical manifestations occur due to the compression of the affected organ[5,9,10,13]. Muscle hydatid disease can be

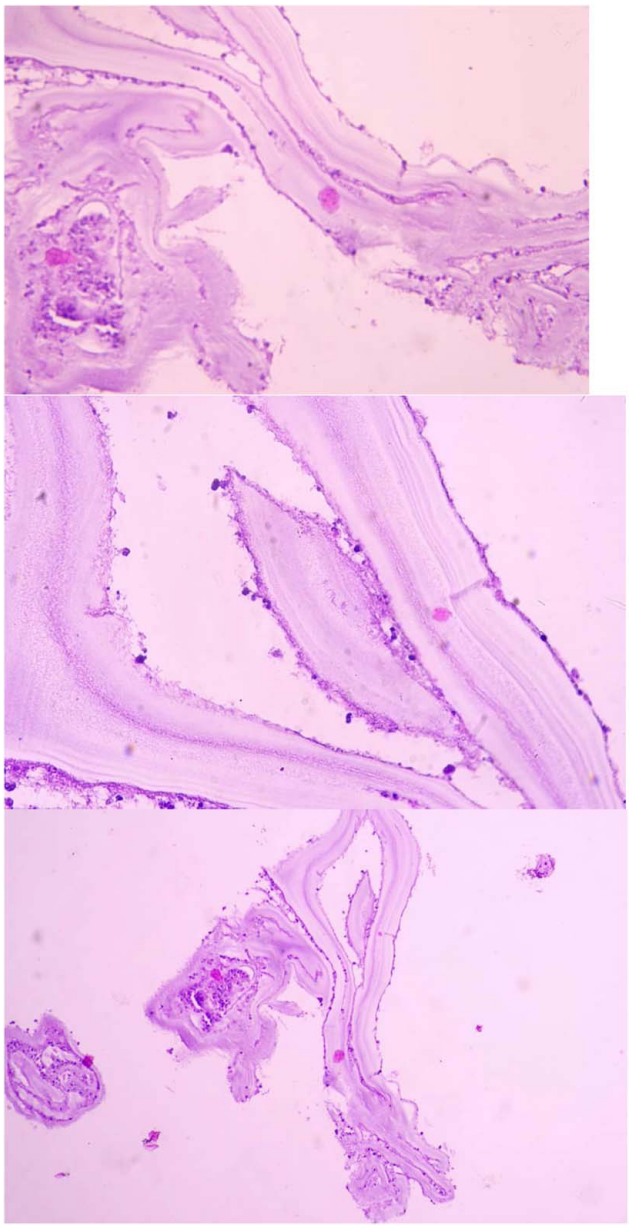

Figure 2. Pathological analysis of biopsies obtained during surgery confirmed the diagnosis of hydatid cysts.

mistaken for other conditions such as myositis or calcified hematoma. Although hydatid cysts can remain dormant for years, some may present with complications such as nerve compression or infection, mimicking abscesses or tumors[5].

There are many ways to establish the diagnosis, one of which is serological testing; however, it is not sufficient to exclude the disease if there is no accompanying organ involvement – as local muscle involvement only may not be detected by it. Furthermore, eosinophilia is usually not detected in healthy cystic echinococcosis – as in our case. As a result, diagnosis of a primary skeletal muscle hydatid cyst is challenging and needs a lot of suspicion[5].

Imaging modalities are more critical in diagnosing hydatid cysts. Ultrasound plays a major role with a sensitivity and specificity up to 100%. A CT scan can give clear information about many characteristics, such as location, number, size, and relationship to the surroundings. Moreover, MRI is considered an extremely helpful diagnostic tool for identifying muscular hydatid cysts[14]. Imaging studies have almost 100% specificity and sensitivity and could give valuable information regarding the morphological characteristics of the cyst, differentiating it from other lesions such as abscesses or tumors[14].

The treatment of choice for hydatid cysts, wherever its location, is total cystectomy. However, under specific circumstances, irrigation with scolicidal agents and tube drainage may be carried out as necessary suboptimal treatments[9,13,15].

It is worth mentioning that pre-operative and post-operative 1-month courses of albendazole and 2 weeks of praziquantel should be taken into consideration as this reduces the risk of spillage during surgery. Moreover, these drugs help in sterilizing the cyst, decreasing the chance of anaphylaxis[16].

The patient reported in this paper presented to a physician complaining of a palpable mass in his left thigh that had been increasing in size over 4 months with no reported signs of inflammation. Echography showed two cystic lesions with irregular edges. MRI revealed a cavity within the soft tissues of the thigh with multiple cystic formations inside. These findings suggested an abscess; thus, a true-cut biopsy was taken by his physician, which revealed features of a hydatid cyst and caused its perforation. A CT scan was then performed, excluding other affected organs. As a result, the patient was diagnosed with a primary hydatid cyst within the soft tissues of the thigh. Total cystectomy was performed to achieve good drainage for the entire cavity and to remove daughter cysts that had spilled.

Khan *et al.*[14] reported a case of a 32-year-old female who had an enlarging mass on her left thigh, which was diagnosed as a hydatid cyst after final pathology. Cankorkmaz *et al.*[10] also reported a case of a 4-year-old girl who had a hydatid cyst in the left thigh. There were some reported cases of hydatid cysts at the axilla, as Yagmur and Akbulut[4] reported in their paper about a 73-year-old man with an axilla mass that was confirmed to be a hydatid cyst after surgery. Cardiac hydatid cysts are also reported, as Fiengo *et al.*[2] reported about a giant cardiac hydatid cyst.

What is unique about this case report is that primary hydatid cysts within the soft tissues of the thigh with no involvement of other organs are extremely rare[14]. Moreover, this case discusses misleading findings that led to performing a true-cut biopsy. To our knowledge, this has not been discussed before. Additionally, the period of 4 months during which the mass has been enlarging is too short to consider hydatid cysts as a differential diagnosis. This is another unique feature of this case report that informs practitioners that they should not rely solely on the speed of enlargement when faced with a soft tissue mass to exclude hydatid cysts.

Conclusion

Although primary cysts in soft tissues including muscles are extremely rare findings, they should be considered for differential diagnosis in any patient presenting with a palpable mass, especially if they live in an endemic country.

Ethical approval

This is a case report and there is no need for ethical committee approval. All informed consent is taken from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

A.Ahmad, O.J., M.B., and A.Alnasan: gathering information, literature review, and writing of the manuscript; B.A.: gathering information, literature review, writing and review of the manuscript; M.A.: performing the procedure, writing and review of the manuscript, and supervising the whole work.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

Research registration unique identifying number (UIN)

1. Name of the registry: not applicable.
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Guarantor

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Data availability statement

The datasets used during the current study are available from the corresponding author on reasonable request.

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