

Primary Hydatid Cyst of the Skeletal Muscles: A Rare Presentation of Cystic Echinococcosis

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Abstract

Hydatid cysts of the liver and/or lung are commonly seen in surgical practice in endemic areas like Iraq. Primary or secondary skeletal muscle hydatidosis is rarely reported, even in endemic areas. This is a case report of a 51 year-old, otherwise healthy, man with a firm non-fluctuant cystic swelling in the right thigh that was painless and growing slowly over a period of six months. While ultrasound and magnetic resonance imaging suggested a multilocular cystic lesion, other investigations failed to confirm the diagnosis of muscular hydatid cysts, especially in the absence of a primary cyst in the more common organs. The lesion was initially considered a soft tissue tumor but intraoperative exploration revealed a hydatid cyst with daughter cysts, that was excised and the patient was treated with Albendazole for 1 month postoperatively. The patient developed a seroma that disappeared spontaneously. There was no recurrence for up to 4 months of follow-up. This report illustrates that echinococcal disease should be considered in the differential diagnosis of a cystic mass in any anatomic location, especially when it occurs in endemic areas.

Keywords: Echinococcus granulosus; Muscular hydatid cyst; Thigh swelling.

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INTRODUCTION

In human, three species of Echinococcosis are known to occur: (1) cystic echinococcosis, caused by *Echinococcus granulosus*, (2) alveolar echinococcosis, caused by *Echinococcus multilocularis*, and (3) polycystic echinococcosis due to *Echinococcus vogeli*. *Echinococcus granulosus* (cystic echinococcosis) is the most common species ⁽¹⁾. Human cystic echinococcosis (hydatid cyst disease) is prevalent in the Middle East and Arabic North Africa and is considered hyperendemic in Iraq ⁽²⁾.

Humans are incidental intermediary host in the life cycle of the parasite; becoming infected by ingestion of food contaminated with eggs from the feces of the definitive hosts, mainly dogs ⁽³⁾.

Hydatid cysts are mostly found in the liver and/or in the lungs, but several other sites are possible; bone is included. The musculoskeletal involvement has been registered in only 1–4% of the cases ⁽⁴⁾. Primary skeletal muscle hydatid cyst without liver and lung involvement is rare even in endemic areas ⁽⁵⁾.

growing swelling in the upper part of the right thigh. The patient noticed the swelling 6 months prior to his referral and claimed that it was painless and slowly growing. He had no other local or general symptoms, no history of fever, respiratory or gastrointestinal symptoms in particular.

On examination, the patient had an oval, firm, non-fluctuant, non-tender lump of about 5X15 cm at the anteromedial region of the proximal half of the right thigh. The lump was not fixed to the skin, subcutaneous tissue or to the bone. The overlying skin appeared normal in color and texture and there was no associated lymphadenopathy or peripheral edema. Lower limb arterial pulses and nerve functions were normal.

Blood investigations revealed mild leukocytosis with mild neutrophilia, lymphocytosis and eosinophilia but the ESR was within normal range. Renal and liver function tests and blood glucose levels were all normal.

Ultrasonic examination of the mass using Duplex ultrasound (**figure 1**) revealed an intermuscular multilocular cystic lesion in the adductor group of the right thigh muscles with a 4.6 cm depth. The lesion was avascular under color Doppler ultrasound but it caused anterolateral displacement of the femoral vessels. As the

Case Summary

A 51-year-old male farmer was referred to the orthopedic consultation clinic with a history of a slowly

ultrasonic examination suggested a hydatid cyst, further imaging studies were conducted to detect the presence of such lesion in other regions of the body.

A plain X-ray of the right thigh (**figure 2**) revealed a soft tissue swelling without bony involvement. Plain Chest X-ray (**figure 3**) and liver ultrasound (**figure 4**) were both negative for pulmonary/hepatic hydatid cysts.

MRI of the thigh with and without contrast (**figure 5**) revealed a well-defined large mass (59X48X149 mm) consisting of cystic lesions of variable sizes that appeared hyperintense in T2 and hypointense in T1 weighted images with contrast. The mass was intermuscular in location, the subcutaneous fat and underlying bone were both intact.

From these clinical and radiological findings, the patient was diagnosed with a soft tissue tumor that was to be confirmed by an excisional biopsy. In addition to intermuscular hydatid cyst, lipoma, liposarcoma and multicystic neurofibroma were all suspected.

Intraoperatively, the diagnosis of an intermuscular hydatid cyst was confirmed after visualization of the cyst wall and daughter cysts (**figure 6**).

A longitudinal incision in the skin was made on the medial aspect of the right thigh, beginning about 10 cm distal to the pubic tubercle and extending for 15 cm with undermining of the underlying subcutaneous tissues and on dissection between the gracilis and the adductor longus muscles, the cyst was found to be located between adductor longus and brevis anteriorly and gracilis and adductor magnus posteriorly. Small amount of the cystic fluid was aspirated to confirm the diagnosis, followed by irrigation of cystic cavity with 10% hypertonic saline solution and 10% Povidone iodine for two times (10 minutes each), the cyst was opened and the daughter cysts were evacuated with packing of the surrounding tissues with packs soaked with a scolicedal solution then removal of the laminated membrane with abolishing of the left behind cavity by marsupialization was done (**figure 7**). The incision was closed over a drain which was removed after 48 hours. The patient was put on oral Albendazole 400 mg orally twice a day for 4 weeks followed by a 14-day albendazole-free interval, for a total of 3 cycles to prevent the recurrence.

Two weeks after surgery, the patient developed seroma that was confirmed by clinical examination and MRI (**figure 8**). The seroma resolved gradually over a few weeks and no signs of recurrence could be detected by physical examination and imaging at a 4 month follow-up.



Figure 1: Duplex ultrasound of the thigh mass showing a multilocular cystic lesion.



Figure 2: Plain AP X-ray of the right thigh showing a soft tissue mass and normal bone.

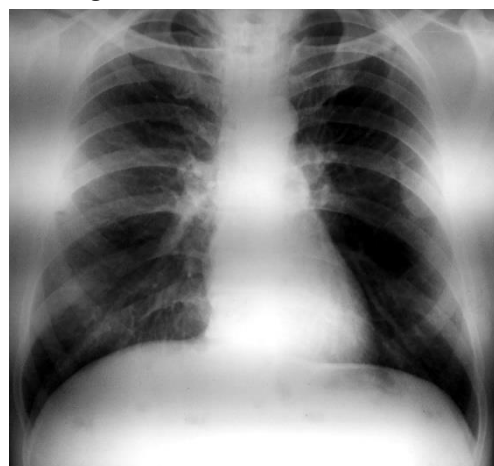


Figure 3: Normal AP Chest X-ray a patient with a primary intermuscular hydatid cyst.

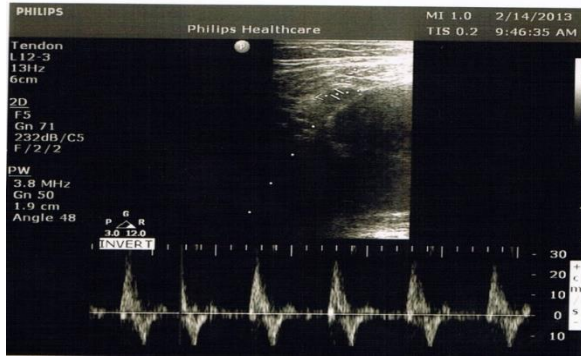


Figure 4: Normal liver ultrasound in a patient with a primary intermuscular hydatid cyst.

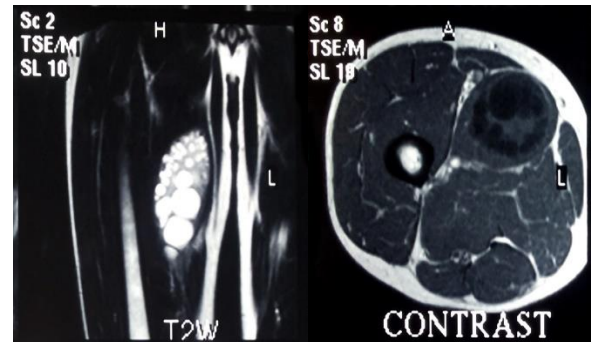


Figure 5: T2W (left) and Contrast (right) MRI of the right thigh swelling showing a multilocular cystic mass



Figure 6: Evacuation of the daughter cysts from an intermuscular hydatid cyst after irrigation with hypertonic saline & povidone iodine.

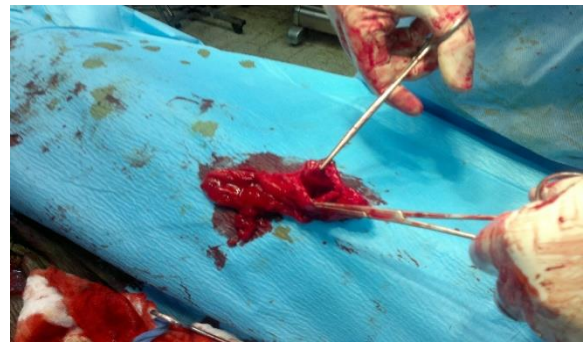


Figure 7: Endocystectomy of the hydatid cyst.



Figure 8: Postoperative MRI showing a seroma at the site of the excised hydatid cyst in the right thigh.

DISCUSSION

Hydatid cyst disease is still a major health problem in developing and agricultural countries including Iraq. Organs affected by hydatid cysts in order of frequency are the Liver (63%), Lungs (25%), Muscle (5%), Bones (3%), Kidney (2%), Brain (1%) and spleen (1%)⁽⁶⁾. The clinical presentation of hydatid disease depends on the size and site of the lesion and the accessibility of the organ involved for clinical examination. Pre-operative diagnosis of hydatid cysts can be made ultrasonically and confirmed by a CT scan

or MRI. Most cases are asymptomatic and are discovered incidentally during radiographic examination.

Muscular hydatid cysts may be primary, but usually occur secondarily when cysts spread from other areas, either spontaneously or after previous operations for hydatidosis, in other regions of the body⁽⁷⁾.

Primary skeletal muscle infection with *E. granulosus* accounts for less than 4% of reported hydatid cases⁽⁸⁾. It is suggested that the low prevalence of this form of disease is due to the physical barriers to the

hematogenous dissemination of cysts created by hepatic sinusoids and pulmonary capillaries⁽⁹⁾. In addition, it is possible that the higher lactic acid concentration in skeletal muscle and mechanical factors, such as contractile activity, may make encystment less likely⁽¹⁰⁾. Nevertheless, some cases of primary muscular hydatidosis at various sites have been reported either as intramuscular or intermuscular lesions⁽¹¹⁾.

The rarity of primary muscular hydatidosis causes difficulty in preoperative diagnosis. In this report, the preoperative diagnosis was that of a soft tissue tumor. Traumatic and developmental lesions were other differential diagnoses that excluded by the history and examination. Percutaneous Fine-needle aspiration cytology (FNAC) was not done in this report because an excisional biopsy of the swelling was planned, in spite that; FNAC is now the standard method of dealing with non complicated hydatid cyst by "PAIR" method, which includes; Puncture, Aspiration, Injection and Re-aspiration under imaging guidance like ultrasound scan⁽¹²⁾. Different types of serological tests like hemagglutination and ELISA tests for Echinococcus; may help in diagnosis⁽¹³⁾. In our case, these tests were not used due to low index of suspicion of primary muscular hydatidosis.

Surgical options for muscular hydatidosis may be: radical (total pericystectomy), partial (endocystectomy) or conservative (open cystostomy and simple tube drainage). The more radical the procedure, lower the risk of relapses but higher the risk of complications⁽¹⁴⁾. Sterilization of the cyst cavity by different scolicial agents provides the best protection with least complications. Pre and Postoperative one month course

of Albendazole should be considered in order to sterilize the cyst and decrease the chance of anaphylaxis, decrease the tension in the cyst wall and to reduce the recurrence rate postoperatively⁽¹⁵⁾.

In our report, the suspicion of a soft tissue tumor other than a hydatid cyst limited the use of Albendazole course preoperatively but intraoperative sterilization with 10% hypertonic saline and 10% Povidone iodine was performed repeatedly in order to prevent the recurrence. However, it seems that the cyst tension was great enough to cause capillary and lymphatic damage, in addition to the extra dissection applied for the removal of the pericyst or ectocyst which result in the development of a seroma postoperatively. However, the seromatous swelling was self-limiting and disappeared with time.

Conclusion

Hydatid cyst of liver and lung is common in endemic areas like Iraq. When the cyst presents in a rare site such as skeletal muscles, it may be confused with a soft tissue tumor as the suspicion of hydatid cyst is unlikely. This is further confusing when the patient does not have any primary hydatid cyst in lung or liver or previous surgical history of cyst excision.

As hydatid cysts can affect any organ of the body, a higher index of suspicion should be adapted in an apparent cystic mass especially in endemic regions.

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