Case report: Primary Subcutaneous Cyst Hydatic disease in proximal thigh : an unusual localisation

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Abstract

**Background:** Musculoskeletal hydatidosis is very rare and represents 1%- 5.4% of all cases of all echinococcosis. On clinical basis, infection mimics a soft-tissue tumor, and the preoperative radiologic diagnosis is very important to avoid biopsy.

**Patients and methods:** We report an unusual case of primary subcutaneous hydatidosis in proximity to vastus lateralis muscle, was diagnosed according to the computed tomography appearance, clinical and pathological findings. A 43 year old female patient was admitted with history of pain at proximal thigh for last 30 days. On examination, a mass which was 4x5 cm in diameter, painful and erythematous, was palpated over greater trochanter. Sedimentation rate was 40 mm in the 1st hour. CT (Computed Tomography) scan demonstrated cystic structure in subcutaneous tissue near vastus lateralis muscle.

**Results:** Histopathological examination of specimen revealed a pericystic structure, which consisted of connective tissue and scattered hyaline cells showing a necrotic basophilic structure which resembles cuticular membrane. Treatment with high dose albendazole was conducted for 4 weeks.

**Conclusions:** This case illustrates that echinococcal disease should be considered in the differential diagnosis of every cystic mass in every anatomic location, especially when they occur in areas where the disease is endemic.

**Keywords:** Cyst Hydatic, Subcutaneous, Vastus lateralis muscle.
Background

Hydatidosis is known since Hippocrates. Echinococcosis or hydatid disease is caused by the tape worm, E. granulosus. The life cycle of E. granulosus is well described [1]. Echinococcosis has its highest prevalence in countries where the common intermediate hosts, sheep and cattle, are raised such as the Middle East, Central Europe, Australia and South America [2]. Hydatid disease mainly affects the pulmonary and digestive systems [3]. The liver is the most frequently involved organ (75%), followed by the lung (15%) and the rest of the body (10%) [2]. Primary involvement of muscle is very rare and has been reported in approximately 3% of all patients with hydatidosis [3]. It has been suggested that muscle provides a poor environment for the parasite because of the presence of lactic acid [4]. A review revealed cases involving the musculature of the chest wall [5], pectoralis major, sartorius [3], biceps brachii [4], and Sartorius [6]. Soft tissue hydatidosis also is very rare. There are a few reports about primary subcutaneous hydatidosis in the extremities [7,8]. To our knowledge, subcutaneous involvement of echinococcal disease near the vastus lateralis muscle in the thigh has not been reported earlier.
CASE REPORT

A 43 year old female patient was admitted with history of pain at proximal thigh for the last 30 days. There was no history of trauma in the proximal thigh region. No history of fever and weight loss was described.

On examination, a mass was palpated over greater trochanter. The mass was 4X5 cm in diameter, painful, and erythematous. Range of motion of left hip was full and painless.

Investigations revealed that CBC (complete blood count), electrolytes, alkaline and acid phosphatase levels were within normal limits. Sedimentation rate was 40 mm in the 1st hour.

X-ray of left hip showed neither calcification nor bone destruction. Abdominal ultrasonography did not reveal any organ involvement. CT scan demonstrated cystic structure close to vastus lateralis muscle. (Figure 1). CT showed no involvement of lung and brain.

Ultrasonographic examination of the mass showed a cystic structure. Hemoagglutination test for echinococcosis and ELISA were negative. The patient underwent surgery for excision of the cyst.

The diagnosis of Echinococcus granulosus infection was confirmed peroperatively after visualization of the cyst wall. Following irrigation of cystic cavity with hypertonic saline solution, we excised the cyst wall along with a portion of the vastus lateralis muscle.

Histopathological examination of the specimen revealed fibrosis, necrosis, histiocyte cell clusters, mixed type of inflammatory cells, fibroblastic activity and vascular proliferations in adipose and connective tissues. A pericystic structure which consisted of connective tissue and scattered hyaline cells was showing a necrotic basophilic structure which resembles cuticular membrane detected in the pericyst. Scoleces were detected. Fresh bleeding parts were seen external to the cyst. (Figure 2).

A high dose of albendazole was initiated for 4
weeks. Respond to albendazole treatment was obtained. During the follow-up period, radiologic examination and blood work were performed. This consisted of chest radiography, abdominal and proximal thigh ultra sonography, blood routine biochemistry (glucose, ALT, AST, alkaline phosphatase, blood urea nitrogen, serum creatinine, total protein, albumine and indirect hemoglutination test.

Discussion

Incidence of musculoskeletal hydatidosis is not clear due to various reports. According to authors, incidence of musculoskeletal Echinococcosis including subcutaneous tissue is 1%-5.4% among all cases of all echinococcosis [9,10]. Soft-tissue hydatid cysts occur in 2.3% of cases reported from endemic areas. They are usually associated with involvement of other structures.

Diagnosis of echinococcosis must be considered when slowly growing soft tissue is present in patients from rural areas and especially endemic countries with close proximity to dogs. Before surgical excision, biopsy and extirpation of cyst, the diagnosis of echinococcosis must be made to avoid leakage of cyst contents and accompanying risks of anaphylaxis. Ultrasound is useful in diagnosis, detecting dimension, localisation and type of cyst. The sensitivity of US (Ultrasonography) is 95%, and if vesicular fibrils are present, the sensitivity of US increases to 100%. CT scan should be performed in suspicious cases, in order to decide the technique of surgery and to demonstrate the relationship to adjacent organs [11].

Preoperative diagnosis of musculoskeletal E. granulosis infection is difficult clinically and radiological. It may resemble any soft tissue tumor. MRI is capable of adequately demonstrating most features of hydatid disease, with the exception of calcifications [12]. Garcia-Diez reported MRI examinations of 7 patients with musculoskeletal
hydatidosis involving soft tissue. Typical signs of hydatidosis were multivesicular lesions with or without hypointense peripheral ring (“rim sign”). According to the presence of viable daughter cysts MRI conveyed as high signal intensity or low signal intensity on T2-weighted images[13]. There is controversy about the value of MRI in diagnosing the vitality of the cyst. Hypointense daughter cysts compared with the matrix of the mother cyst on T2-weighted images are associated with the death of the parasite[14,15,16]. Proton density-weighted images generated by gradient echo sequences as a sign of biological activity was suggested by Tekkok et al[15].

Echinococcosis resembles a benign neoplasm. In order to prevent serious complications, the diagnosis must be made before any manipulation. The diagnosis is based on the history of exposure in an endemic area and US, CT findings [17]. The diagnosis can be made by specific Ig G, complement fixation, indirect fluorescent, and ELISA tests. The sensitivity of various serological tests used for hydatid disease vary from 64 % to 87 %. False positivity of Casoni skin test was reported in infestations of tenia saginata and other helmints because of cross reactions. The specificity of Casoni skin test is in doubt because of this high, 40% false positivity. ELISA/ Western blood serology is 80-100 % sensitive and 88-96 % specific for liver cyst infection, but less sensitive for lung (50-56 %) or other organ involvement (25-26 %) [18]. After surgical excision of the cyst, titers of reagenic antibody (Ig E) decreases and becomes negative after 1-2 years. If titers do not decrease, one must think about a recurrence of echinococcosis. Hydatic serology is only valuable when it is positive, negative serologic test does not exclude the diagnosis.

If the cyst is not complicated, serology is less sensitive but it may be useful for controlling the recurrence of cyst.

Cure depends on total or subtotal surgical excision of cysts. Echinococcosis can be prevented by eradication programs.
E. granulosis is best treated by complete excision of the intact cyst. If it is impossible to excise a large cyst en bloc, the cyst has to be drained intraoperatively, irrigated with a scolecidal agent such as hypertonic saline, and then excised [4]. The likelihood of recurrent infestations increases after rupture of the parent cyst. In addition, leakage of the cyst contents may cause anaphylactic shock [4, 19]. The propensity for recurrence, dissemination mortality is about 70% [20]. During the follow-up for 3 years, recurrence of hydatidosis has not been found. This case illustrates that echinococcal disease should be considered in the differential diagnosis of every cystic mass in every anatomic location, especially when they occur in areas where the disease is endemic.

Acknowledgement: Written consent was obtained from the patient for publication of study.

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Figure 1.Computed Tomographic view of hydatic cyst in vastus lateralis muscle.

Figure 2. The scoleces are seen in histopathologic section of hydatic cyst of vastus lateralis muscle.