

In the name of God

Shiraz E-Medical Journal
Vol. 12, No. 3, July 2011

<http://semj.sums.ac.ir/vol12/jul2011/89021.htm>

Hydatid Cyst of the Biceps Femoris Muscle (a Rare Case in Orthopedic Surgery).

Karimi A*, Asadi K*, Mohseni F**, Hossein Akbar M±.

* Assistant Professor, Department of Orthopaedic Surgery, Guilan University of Medical Sciences, Rasht, Iran, ** General Surgeon, Golsar Hospital, Rasht, Iran, ± Research Assistant, Heshmat Hospital, Rasht, Iran.

Correspondence: Dr. A. Karimi, Trauma Research Center, Guilan University of Medical Sciences, Rasht, Iran, Telephone: +98(131)323-8373, Fax: +98+98(131) 323-8373, Email: karimi_ortho@yahoo.com.

Received for Publication: July 19, 2010, Accepted for Publication: April 30, 2011.

Abstract:

Hydatid cysts are endemic zoonosis in Iran. They may involve various organs of body. Hydatid cyst in musculoskeletal system is rare and constitutes 1% to 4.5% of all cases. Usually, the hydatid cyst in muscle present as a benign soft tissue tumor. The commonest clinical presentation is an asymptomatic slow growing mass.

We report this rare case from orthopedic surgery center in a university teaching hospital at North Iran. The literature review and mode of diagnosis has been discussed through this case report.

Introduction:

Cystic hydatidosis as the larval stage of Echinococcosis is a zoonosis and is a significant public health problem. It is prevalent in different areas of world including the Middle East.⁽¹⁾ Domestic dogs, foxes and wolves serve as definitive hosts, whereas sheep act as intermediate hosts.⁽²⁾ Lungs and liver are

most frequently involved, but other organs such as kidneys, intestines brain, genitourinary and bones may also be involved.⁽³⁾ Hydatid cyst within the inguinal canal has also been reported.⁽⁴⁾ Bone involvement is usually seen in less than 1% of cases.⁽⁵⁾ Hydatid cyst in musculoskeletal system is rare and constitutes 1% to 4.5% of all cases.⁽⁶⁾ Usually the Hydatid cyst in muscle present as a be-

nign soft tissue tumor. The commonest clinical presentation is an asymptomatic slow growing mass

We report this rare case from orthopedic surgery center in a university teaching hospital at North Iran. The literature review and mode of diagnosis has been discussed through this case report.

Case Report:

A 50 year old woman from rural area around Rasht city at southern Caspian region complained of a painless mass in the back of her left thigh. There was no history of trauma, fever, or weight loss. She described this progressively growing mass in her left thigh for last two years.

On physical examination, there was a diffuse non-tender cystic swelling which occupied medial and posterior aspects of thigh fixed with the Hamstring muscles. There was no sign of pressure effect on the Sciatic nerve.

Distal part femoral artery was palpable, with good pulse at popliteal and dorsalis pedis artery while no other organ was involved. Lungs and Livers were normal on physical examination as well as radiographic and sonographic study. There was no cystic mass any where else in the body. Results of routine blood tests and serology were within normal range.

Radiography and ultrasonographic study revealed a cystic mass within the lateral hamstring muscle (Biceps Femoris, Fig.1). A routine MRI examination showed a large multi-loculated cystic lesion in the posterior thigh occupying Hamstring muscles. Multiple small daughter cysts and a free floating membrane were seen.

The patient was selected for a routine surgical excision of Hydatid cyst. After spinal nerve block anesthesia, on prone position, a longitudinal incision was made over the protrusion on the back of left thigh. Medial and lateral Hamstring muscles were exposed and the mass was removed in complete, as a white cystic ovoid mass (Fig. 2). Pathological examination of cyst confirmed our preoperative diagnosis of Hydatid cyst. The patient was discharged uneventful with a post-operative Albendazole drug therapy to prevent recurrence. Figure 3 shows the site of Hydatid cyst removal on completion of surgery.

Discussion:

Hydatid cyst is a zoonotic infection involving larval forms of small tapeworms of *Echinococcus granulosus*. This parasite inhabits the small intestine of carnivores such as dogs and wolves as definitive hosts in the cycle. Humans become infested by ingesting ova from the feces of these carnivorous animals. The liver and lung are primarily affected in this disease. This disease is distributed with more frequent occurrence in the Mediterranean countries, East Africa, South America, Russia and Australia. Primary involvement of the muscles is very rare and has been reported in only 1–4% of all patients with hydatidosis.⁽⁶⁾ Intramuscular Hydatid cysts grow gradually and may mimic a soft tissue tumor.⁽⁷⁾ Thus, the diagnosis of soft-tissue Hydatid cysts needs a high index of suspicion. However, some case reports of intra muscular Hydatid cysts have been published until now.^(8, 9)

It has been suggested that muscles provide a poor environment for the parasite because of the presence of lactic acid. To our knowledge and literature review, sub-fascial and muscular involvement of echinococcal disease in the posterior compartment of the thigh have been reported.^(10,11, 12, 13)

Diagnosis of Echinococcosis (E) should be considered when slowly growing soft tissue is present in patients from rural areas, especially in endemic countries. Pre-operative diagnosis of musculoskeletal E. granulosus is sometimes difficult clinically and radiologically.⁽¹³⁾ It may resemble any soft tissue tumor such as abscess, chronic hematoma, synovial cyst, and necrotic malignant tumor. Before surgical excision or biopsy of cyst, diagnosis of echinococcosis should be excluded to avoid leakage of cyst contents and the accompanying risks of anaphylaxis.

Hydatid cyst resembles a benign neoplasm in many ways. In order to prevent serious complications, it should be diagnosed before any therapeutic intervention. The diagnosis is based on the history of exposure in an endemic area and Sonography, CT imaging and MRI findings.^(13, 14, 15)

Imaging is a good tool for diagnosis in Hydatid cysts.⁽¹⁴⁾ Plain film shows rounded masses of uniform density. In endemic area, such as Iran, such lesions are mostly the Hydatid Cyst. MRI, CT, and ultrasound reveal well defined cyst with thick or thin wall. The most pathognomic finding is daughter cyst or laminated membranes and septation in Hydatid Cyst cavity.^(14, 15)

The Hydatid cyst of hamstring muscles in our patient was diagnosed by Ultrasono-

graphic, MRI and CT-scan. Finally, Pathological report confirmed the pre-operative diagnosis. Presence of daughter cyst, eggshell or mural calcification on CT is indicative of Agranulosis infection and helps to distinguish the cyst from carcinoma, bacterial or amebic abscesses and hemangioma. Diagnostic aspiration is not usually recommended because of the risk of dissemination and anaphylaxis.⁽¹⁶⁾

Preoperative radiological diagnosis is very important to avoid biopsy. Diagnosis of Echinococcosis should be excluded to avoid leakage of cyst contents and the accompanying risks of anaphylaxis and even death⁽¹⁶⁾

Primary involvement of other viscera is rare and in endemic areas surgeons do such an experience rarely. In developed countries with high grade public health care and excellent hygienic condition, Echinococcosis infestation and Hydatid disease is uncommon. Besides, physicians in endemic areas of underdeveloped countries and surgeons dealing with rural and cattle grazing areas should be aware of Hydatid cysts. Furthermore, in the case of soft tissue tumor probability, biopsy should be avoided to prevent the risk of dissemination and anaphylaxis.^(16,17)

The conventional treatment of muscular hydatid cysts is surgical; however, it may require an extensive surgical resection, and need of general anaesthesia is inevitable. Preoperative medical treatment may sterilize the cyst cavity and might decrease the intra-operative complication of spillage and consequential anaphylaxis. Intra-operative irrigation of 0.5% Cetrimide, 15% hypertonic saline and 0.5% silver nitrate solution before open-

ing the cyst, may kill the daughter cysts and further reduces the risk of dissemination and anaphylactic reaction.^(16,17) Albendazole was the choice treatment we used to prevent and avoid recurrence.^(18,19,20)

In conclusion, hydatid disease can affect any organ in the body; the infestation

may mimic a soft tissue tumor in, or around the muscle and therefore, a high suspicion of this disease is justified in any cystic neoplasm of each organ.



Figure 1. Ultrasonographic picture of a cystic mass within the lateral hamstring muscle

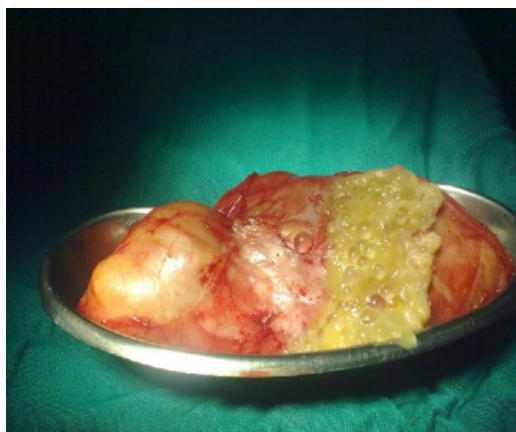


Figure 2. Hydatid cyst removed as a whole from Biceps Femoris muscle



Figure 3. Site of Hydatid cyst removal on completion of surgery.

References:

1. Wen H., Yang W.G. Public health importance of cystic echinococcus in China. *Acta Trop* 1997; 67: 133-145.
2. Saidi, F: *Surgery of Hydatid Disease* Saunders, Philadelphia 1976, 3: 3115. (S)
3. Jeffrey A Park; *Infectious Arthritis*; In: *Campbells Operative Orthopedics*; 11th ed. 2008 Vol 1; 767-768.
- 4- Kouhsari, MR; Akbar, MH: Inguinal Hydatid Cyst; Report of a rare case: *MJIRI*, Vol.12; No.2; 177-179; 1998.
5. Rask, MR & Lattig, GJ: Primary intramuscular hydatidosis of the sartorius. *J Bone Joint Surg Am* 1970, 52: 582-584.
6. S. Kumar, S. Kumar, P. Ramakanat, A. Ahmad & Ramakant: Intramuscular Hydatid Cyst: A Rare Presentation. *The Internet Journal of Surgery*. 2008 Volume 16 Number 1.
7. McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *Lancet* 2003; 362: 1295-304.
8. Merkle E, Schulte M, Vogel T. Musculoskeletal involvement in cystic echinococcosis. Report of eight cases and review of literature. 1997; 168: 1531.
9. Duncan, GJ & Tooke, SMT: Echinococcus infestation of the biceps brachii *Clin Orthop* 1990, 261: 247-250.
10. K. Khatami SM: Acute lower limb ischemia due to femoropopliteal embolism of hydatid cyst. *MJIRI* 1998; 12 (2); 163-165.
11. Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary Subcutaneous cyst hydatid disease in proximal thigh an unusual localisation: a case report. *BMC Musculoskeletal Disord* 2003; 4: 25.
12. G. Ben M'Rad S, Mathlouthi A, Merai S, Ghrairi H, Mestiri I, BenMi:edM'RadK, Djenayah FJ : Multiple hydatid cysts of the thigh: the role of magnetic resonance imaging. *Radiol* 1998; 79 (9); 877-879.
13. Hamamci EO, Besim H, Korkmaz A. Unusual locations of hydatid disease and surgical approach. *ANZ J Surg* 2004; 74: 356-60.
14. Torricelli P, Martinelli C, Biagini R, Ruggieri P, De Cristofaro R. Radiographic and computed tomographic findings in hydatid disease of bone. *Skeletal Radiol* 1990; 19: 435-9. (S)
15. Comert RB, Aydingoz U, Ucaner A, Arıkan M. Waterlily sign on MR imaging of primary intramuscular hydatidosis of sartorius muscle. *Skeletal Radiol* 2003; 32: 420-423. (S)
16. Saenz de San Pedro B, Cazana JL, Cobo J, et al. Anaphylactic shock by rupture of hydatid hepatic cyst. Follow-up by specific IgE serum antibodies. *Allergy* 1992; 47: 568-7011.
17. Senyuz OF, Eyail L; Zvebil F; Stamler B; Anaphylactic shock due to rupture of a hepatic hydatid cyst. *J Ped Surg* 260; 217-218; 1991.
18. Yesildag E, Celayir S. Albendazole therapy in the treatment of hydatid liver disease. *Surg. Today* 2001; 31: 487-91.
19. Schatz PM: Effective medical treatment of hydatid diseases; *JAMA* 253; 2095: 1985 *Medical Parasitology, Hydatid Cyst: Philadelphia* 2008.
- 20- Horton R.J. Albendazole in treatment of human cystic echinococcosis: 12 years experience. *Acta Tropica* 1997; 64: 79-93.
21. Kiresi DA, Karabacakoglu A, Odev K, Karakose S. Uncommon locations of hydatid cysts. *Acta Radiol* 2003; 44: 622-36.