

Case Report

A Rare Case of Hydatid Cyst in the Thigh of 28-Year-Old Male

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Submitted: 26 October 2015

Accepted: 13 November 2015

Published: 14 November 2015

ISSN: 2373-9282

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Abstract

Hydatid cyst is a benign tumor-like growth caused by the *Echinococcus* tapeworm parasite that can cause high morbidity and mortality. It is rare in the United States, with about 200 cases presenting per year, mostly within the immigrant population. An estimated 80 to 90 percent of hydatid cysts develop in the liver or lungs. Today we present the unusual case of a 28-year-old male immigrant from Serbia with a 20-cm hydatid cyst in his left quadriceps muscle. Further, the soft tissue surrounding the cyst demonstrated granuloma formation, which rarely has been reported in hydatid cyst.

CASE DESCRIPTION

A 28-year-old male immigrant from Serbia with no significant past medical history presented with an indurate, swollen, growing mass on his left medial thigh for about one year. The mass was painless and did not affect his knee or leg movement. The patient did not report recent trauma to his left thigh. Physical exam demonstrated a well-appearing male with a 20-cm firm, non-tender, non-moveable mass of his left anterior thigh. Clinically, the mass was presumed to be a sarcoma and the patient was referred to medical oncology for possible neo adjuvant therapy.

An MRI demonstrated a large intramuscular cystic mass within the quadriceps muscle with multiple intralesional cystic lesions. That architecture that "would be highly unusual for a soft tissue sarcoma," according to the radiologist, who further noted that "atypical parasitic infections such as echinococcal infections with daughter cyst formation can have a cyst within a cyst appearance."

A biopsy showed a cystic lesion with fragments of fibro muscular tissue with granulation tissue, lymphohistiocytic infiltrates, granuloma formation, eosinophils and detached lamellar material and hook lets, suggestive of cystic echinococcus.

The patient began treatment with the anti-parasitic albendazole and underwent resection of the mass a month later. Surgical pathology received a 29.5 x 11.2 x 10.8 cm tan to pink-red fragment of muscle which revealed an exposed necrotic sac on one aspect (8.8 x 5.5 cm) and a multiloculated cystic cut surface with abundant (approximately 150 cc) yellow tan, hemorrhagic purulent material with multiple translucent saccular structures within (up to 5.0 x 4.0 cm each) and a cyst lining that was yellow-red roughened and granular. A final diagnosis was made of cystic echinococcus (hydatid cyst) with extensive necrosis,

inflammatory response and chronic myositis with foreign body giant cells.

On follow-up two months following the excision, the patient reported feeling better and demonstrated a healing scars with two 1.0 cm eschars. The patient indefinitely was continuing treatment with albendazole.

DISCUSSION

Hydatid cyst is a disease caused by *Echinococcus granulosus* that has worldwide distribution. It has been recognized since

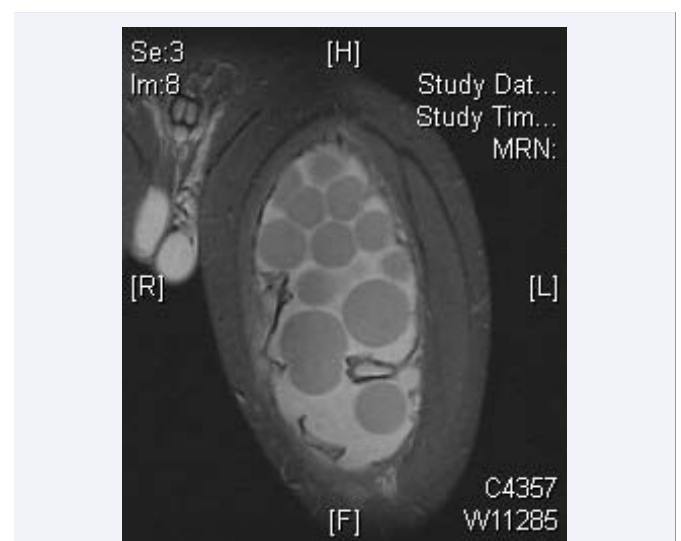


Figure 1 MRI demonstrated a large intramuscular cystic mass within the quadriceps muscle with multiple intralesional cystic lesions, characteristic of *Echinococcus granulosus*.

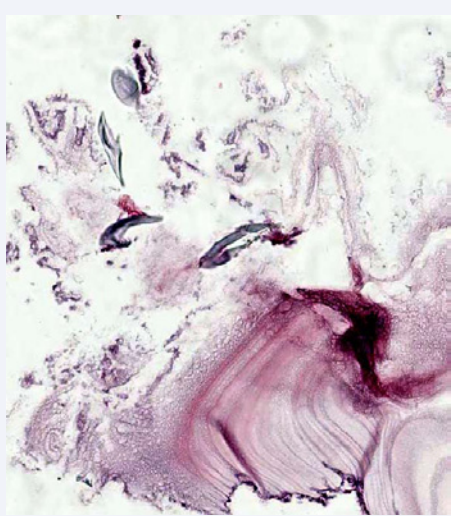


Figure 2 Echinococcus hooklets identified in the initial biopsy.

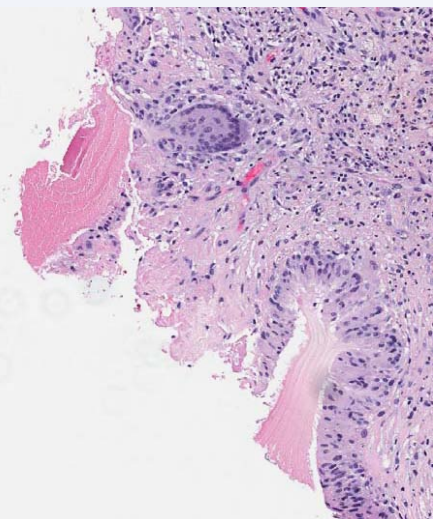


Figure 3 Initial biopsy showed a cystic lesion with fragments of fibro muscular tissue with granulation tissue, granuloma formation, lymphohistiocytic infiltrates and detached lamellar material, suggestive of cystic Echinococcus.

ancient times and was first described by Hippocrates who called it “liver filled with water.” Though benign, the disease can cause high morbidity and mortality.

Echinococcus infection is rare in the United States with approximately 200 cases presenting per year, mostly within the immigrant population [1]. A study of U.S. death records from 1990 to 2007 identified 41 echinococcus-associated deaths, with 73 percent occurring in foreign-born persons [2].

Human infection with *E. granulosus*, the most common Echinococcus species to cause disease, is directly correlated with raising sheep in many countries in Europe, South America, Africa, Asia, Australia and New Zealand [3]. The patient in our report later reported on an infectious diseases consult that while in Serbia he had lived in close contact with farm animals such as sheep, cattle, goats and dogs.

Humans are a dead-end intermediate host for *E. granulosus* – adult tapeworms do not develop in humans or herbivores, such as sheep, cattle, pigs and goats. In nature, adult *E. granulosus* tapeworms, which are about 2-7 mm long and hermaphroditic, develop in the intestines of canines (dogs, foxes, wolves, coyotes, jackals, dingoes). Canines are exposed to the parasite when they ingest organs from animals containing the hydatid cyst. Adult tapeworms in canine intestines produce infective eggs that pass in feces. Human infection is due to hand-to-mouth transmission of water, soil or vegetation contaminated by canine stool which carries the infected eggs [4].

In humans, an immature oncosphere with six hooklet hatches in the intestines, penetrates the intestinal wall and enters the circulation, where it is dispersed to various organs and tissue sites where it embeds and forms a cyst. The first organ it encounters in the circulation is the liver, making it the most likely site in which it forms a cyst.

Though hydatid cysts can occur in almost any part of the body, primary cyst formation in the musculature is very rare (0.5-3%) [5]. A retrospective survey of 244 patients at Sheri Kashmir Institute of Medical Sciences in India from January 2005 to December 2009 demonstrated that approximately 92 percent of the cases involved either the liver or the lungs or both, with just 8 percent of the infections occurring in atypical locations including the gall bladder, peritoneum, spleen, ovary, subcutaneous, seminal vesicle, spinal cord, pancreas, kidney, mediastinum, muscle and brain [6]. A survey of 1,802 patients with hydatid cyst in Australia in 1976 found that just 5 percent of the cases involved muscles [7]. The patient in our case had no further lesions that were detected other than the muscle cyst.

In humans, the larvae form a unilocular hydatid cyst which is slow-growing, tumor-like and space-occupying. Daughter cysts may develop in the mother cyst. The mother cysts typically are about 5 cm in diameter, but they have been reported as large as 20 cm. The cysts accumulate fluid as they grow – up to 2L, according to reports – that can cause anaphylactic shock and death if spilled into body cavities.³ Spillage also can result in cysts developing in other body sites. Along the membrane wall, brood capsules are produced in which an immature stage of the Echinococcus known as protoscolices develops. When brood capsules and daughter cysts disintegrate in the mother cysts, they release accumulated protoscolices known as hydatid sand. The unilocular cyst grows slowly. Five to 20 years may pass until symptoms appear [3]. The patient in our case likely had been infected many years before he presented for medical management.

Pressure of the expanding cysts on adjacent organs or pain is usually the first sign of infection. The cyst can mimic malignant or benign nerve sheath tumors, aneurysmal bone cysts, hemangiomas or pseudomeningoceles, resulting in symptoms similar to those caused by chondrosarcomas and tumor metastasis, including brain damage and bone destruction [8].

Diagnosis of hydatid cyst is difficult and depends on clinical, radiological, serologic and pathologic findings. Aspiration may demonstrate presence of protoscolices, but there is risk of anaphylaxis. Serological testing (indirect hemagglutination and IgG ELISA) is useful, but negative in 10 to 40 percent of infections

[3]. Routine labs will reveal only eosinophilia. On histopathology, the cyst is bilayered, consisting of an opaque, eosinophilic, acellular, avascular laminated layer of varying thickness that is strongly PAS and GMS positive and an inner nucleated germinal layer of epithelium. The cyst typically is surrounded by fibroblasts, mononuclear cells, chronic inflammatory cells, eosinophils, multinucleated giant cells and variable calcification after five or more years. Granuloma formation rarely has been reported [9]. Also, protoscolices with a double row of refractile, birefringent, acid-fast hooklets and round suckers that comprise "hydatid sand" may be identifiable on histopathology.

Surgery is the treatment of choice for hydatid cyst and the best option for a complete cure. Chemotherapy with high-dose albendazole, mebendazole or praziquantel can be considered if the cyst is inoperable due to its location. Prevention is key, including education regarding the means of transmission. Personal hygiene and hand-washing is critical in rural areas inhabited by dogs and livestock. Dogs should not be fed the viscera of slaughtered animals.

Although hydatid cysts are rare in the United States, this case demonstrates that hydatid cyst should be in the differential in large soft tissue masses, particularly in patients from the immigrant population.

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Cite this article

Biemer J, Song A, Nystrom L, Borys D (2015) A Rare Case of Hydatid Cyst in the Thigh of 28-Year-Old Male. *Ann Clin Pathol* 3(3): 1055.