CASE REPORT

Primary hydatid disease in adductor muscles

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Abstract

Primary muscular *Echinococcus* infection is very rare without involvement of thoracic and abdominal organs. In this case a 31-year-old man who had a growing mass in the postero-medial part of his right thigh was examined. The mass was diagnosed as hydatid cyst using ultrasound, magnetic resonance imaging (MRI) and serological tests. It was removed surgically and there has been no recurrence one year after the surgery.

The MRI imaging characteristics may differ depending on the life cycle stage of the parasite. In this case report, we discuss the imaging characteristics of the muscular hydatid cyst with special emphasis on the MRI findings. In regions where hydatidosis is endemic, a mass found in body muscles should be considered as a muscular hydatid cyst.

Echinococcosis (hydatid disease) caused by the tapeworm *Echinococcus granulosus* is a worldwide problem especially in sheep and cattle among developing countries. It is endemic in some regions, including the Mediterranean, the Middle East, the South America and Far East regions.^{2,3} The annual incidence in Turkey is 12 per 100.000.¹ When humans ingest the eggs of the tapeworm, the embryos that emerge penetrate the intestinal mucosa and are transported via the circulation to various organs. Most commonly they reach the liver (28 % of cases) and lungs (64%). Other or-

gans are rarely affected. Primary hydatid cyst of skeletal muscle is rare, occurring in 1-3% of all cases.^{2,3}

The diagnostic procedures include obtaining history data with special reference to the patient's occupation and residence, clinical examination, ultrasonography, computed tomography, magnetic resonance imaging (MRI), and serology.^{3,4} MRI findings may change depending on the stage of the disease and concomitant infection.

After written informed patient consent was obtained, we present a rare case of intramuscular hydatid cyst and includes serology tests and MRI findings.

Case report

A 31-year-old man was admitted to the orthopaedic clinic with eight months history of a slowly growing mass in the medial aspect of the right thigh. Physical examination revealed a non-tender fixed mass deep in the poster-medial part of the right thigh measuring 9x16 cm. There was no erythema, ecchymosis or fluctuation to suggest inflammation. There was no evidence of bone destruction or effect on adjacent bone. Hip and knee joints exhibited normal ranges of motion. Peripheral pulses were equal in both limbs.

A. T1-weighted coronal image. The small cyst is hypointense (arrow). The large medial cyst is more hyperintense due to high protein content than the small one.

B. T2 weighted axial image : The membrane detachment is apparent in the medial large cyst (arrow). Note the hypo-intense rim surrounding both cysts.

C. T1- weighted contrast enhanced image. Note the large cyst which contains detached membrane had enhanced prominently with intravenous contrast administration (arrow heads) whereas the small cyst had no enhancement (arrow).

Complete blood count, electrolytes, alkaline and acid phosphatase values were within normal limits. Soft tissue swelling was present on the medial and posterior side of the distal side. Total IgE concentration was high (294 kU/L). After evaluation of clinical, radiological and serological findings, the diagnosis of hydatid disease was made. Serological tests included indirect hemagglutination (IHA, Dade Behring, Germany) indirect immunofluorescence (IFAT, Euroimmun, Luebeck, Germany) and enzyme linked immunosorbent assay (ELISA, Euroimmun, Luebeck, Germany). The tests were positive. Titers for IHA and IFAT were 1/ 8192 and 1/1000, respectively. ELISA for echinococcosis resulted 180 RU/ml. Postoperative treatment was with albendazole 200 mg bid for six weeks after which specific antibody titers were 1/256 for IHA, 1/32 for IFAT and 48RU/ml for ELISA. The cut-off titers for IFAT and the IHA and cut-off values for ELISA were 1/100, 1/16, 22RU/mL, respectively.

Ultrasonography revealed an intramuscular cystic mass with multilocular daughter cysts. The extent and characteristics of the mass were detailed with MRI. In

T2-weighted images the mass consisted of two different cystic components; a medial large component and a lateral small cyst. The large cyst had the appearance of a detached membrane within. Both cysts had a thick hypo-intense rim resembling a fibrous capsular pericyst layer in all sequences. The inner content of the large medial cystic component showed more hyperintensity than the smaller lateral part in T1-weighted images. This sign was attributed to the high protein content of the large cyst. With intravenous contrast administration prominent contrast enhancement was seen in the wall of the large cyst but not in the wall of the small cyst.

Preoperatively, albendazole (10 mg/kg per day) was given for 10 days. Under general anaesthesia, the lesion with adductor muscles was excised en bloc with a wide margin without destroying the cyst wall which was irrigated with 3% hypertonic saline. The patient tolerated surgery well and postoperative recovery was uneventful. Albendazole (10 mg/kg twice daily) was given for 3 months postoperatively. After a follow up of 12 months, ultrasonography and serology (IFAT

and ELISA) were normal and no recurrence was noted.

Histological examination of the specimen revealed daughter cysts and fragments of the lamellar membrane of the hydatid cyst. Scolices were also detected within the surgical specimen. No superimposing infection was detected with histopathological analysis.

Discussion

Primary intramuscular hydatidosis is rare because intramuscular growth of the cysts is hindered by the muscle contractility and by the high level of lactic acid. Cysts develop very slowly and act as space occupying lesions, producing symptoms related to pressure on the surrounding tissues. Other possible primary locations of the disease must be excluded by careful clinical and radiological examination. Several serological tests are available but may give negative results because of encapsulation. Ultrasonography helps to prove the cystic character of the muscular mass while CT and MRI findings show the location, size and appearance of the cyst.

As the embryos are growing in particular organs they form large cysts which are composed of some parts and layers. The outer layer of the cyst, pericyst, is a layer composed of modified host cells that form a dense, fibrous zone. The middle layer is a laminated acellular membrane. The inner layer is the germinative membrane which the protoscoleces and laminated membrane are produced. The cyst may contain smaller cysts named daughter cysts which are generated by the germinative membrane nests.

The classic MRI findings include multiple cysts within the cyst, a low-intensity rim on T2-weighted images or a detached membrane.⁵ The hypo-intense rim is thought to represent the pericyst and this rim enhances after intravenous contrast administration because of high vascularity of the cyst. The degree of the enhancement is much higher in infected cysts. The rim is showed to be less developed in muscle cyst hydatid lesions.^{5,7}

The present case had two adjacent cysts with different internal composition as seen by MRI. The large cyst had a detached germinative membrane and a prominent contrast fixation in the pericyst layer. The detachment of the endocyst from the pericyst is thought to be related to decreased intracystic pressure, inflammation, host response or response to therapy.^{5,6,7} Inflammatory signal changes are apparent around the cysts in the soft tissues of the thigh. The small cyst is homogenous and faint contrast enhancement in the outer rim. The unenhanced T1-weighted images show the large cyst to have a more hyper-intense than the small cyst. This is concordant with high protein content of the large cyst. On T2-weighted images, both cysts had hyper-intense inner content with a hypo-intense rim.^{8,9} Collapse of the germinative membrane may bring the cyst content in contact with the host tissues. In this case the rim of the larger cyst with detached membrane was enhanced much more with contrast than the small cyst with intact inner layer which may be due to the inflammatory response to cyst content by the host tissues. This shows that strong pericyst enhancement could exist without coincident infection.

Surgery is the most effective way to treat hydatid cysts. Medical treatment alone is not effective, complete surgical resection plus medical therapy is the preferred treatment for the isolated echinococcus. Rupture or spillage of the cysts should be avoided to prevent local or distant dissemination and anaphylaxis.

In conclusion, preoperative recognition and characterization of muscular or soft tissue hydatid lesions are essential and have vital roles in patient outcome. Avoidance of preoperative biopsy is important due to the high possibility of an anaphylactic response. At present, MRI is the modality of choice for thorough lesion characterization. The use of contrast agents may reveal additional information about the host response.

References

1. Republic of Turkey Ministry of Health. Health Statistics. Ankara: Research Planning and Coordination Council; 1997.
2. King HC. Cestodes. Mandell GL, Bennett JP, Dolin R (eds). Principles and Practice of Infectious Disease. New York: Churchill Livingstone 2005; 3290.
3. John DT. William A. P. Markell and Voge's Medical Parasitology 9th Edition. Saunders Company, 2006;139-48.
4. Zhang W, McManus DP. Concepts of immunology and diagnosis of hydatid disease. Clin Microbiol Rev 2003; 16:18-36.
5. Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa C. Radiographics 2000;20:795 -817.
6. Comert RB, Aydingoz U, Ucaner A, Arikan M. Skeletal Radiol. 2003;32:420- 3
7. Tarhan NC, Tuncay IC, Barutcu O, Demiros H, Agildere AM. Skeletal Radiol 2002; 31: 608-11.
8. Meddeb N, Bachrouh N, Elleuch M, et al. Hydatid cyst of the adductor muscles. MRI aspect, Apropos of 1 case. Bull Soc Pathol Exot. 2001; 94:106 8.
9. Lühr T, Junginger T. Muscular echinococcal cysts. A rare differential diagnosis of a soft tissue swelling. Chirurg. 1995;66:1275-6.

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