Primary hydatidosis of gluteus maximus

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ABSTRACT

Hydatid disease is a parasitic infestation of humans and herbivorous animals, caused by *echinococcus granulosus*. Dogs and some wild carnivores, like foxes, are definitive hosts, harboring worms in their intestines. Eggs are passed in feces and eaten by intermediate hosts and larvae encyst in the liver, lungs and other organs. Primary muscular hydatidosis without involving the thoracic or abdominal organs is extremely rare. A case of intramuscular gluteal hydatid cyst is being reported with the intent of highlighting this atypical localization of the disease. Since the soft tissue tumors may be confused with hydatid cysts, preoperative evaluation of these patients is critical for proper handling during surgery to avoid life-threatening complications. We report a case of a 24-year-old male patient with a cystic gluteal swelling turning out to be hydatid cyst on sonography and computerized scanning. Surgical excision with postoperative antihelmenthics formed the main modality of treatment.

KEY WORDS: Hydatid cyst, muscle, ultrasonography

The flatworm *Echinococcus granulosus* causes the commonest form of hydatid disease. This is usually localized in the liver and the lungs. Although any organ in the body may be involved, primarily muscles are seen to be affected rarely. Musculoskeletal cysts account for 0.7-3% of total cases of hydatidosis.[1,2] In the musculoskeletal system, cysts may grow primarily from direct implantation of oncospheres or secondarily from metastatic dissemination of visceral cyst.[3]

The involvement of the gluteus maximus muscle without the evidence of hepatic or pulmonary disease is quite rare. This atypical localization of primary intramuscular hydatidosis has also been reported by Chiattoni *et al*[4] and Arazi *et al*[5] in their series of 15 patients with echinococcosis of the bone and muscles, noted only one case with the involvement of the gluteus maximus muscle. We report a case of primary hydatid cyst of the gluteus maximus muscle which is a very rare occurrence, thus emphasizing the need for high index of suspicion in cystic swelling of muscles especially in endemic regions of the world.

Case History

A 24-year-old male laborer presented to the surgical outpatient department with a slowly growing painless swelling in the left gluteal region for the past nine months. Physical examination revealed a diffuse, non-tender, cystic swelling of approximately 8x6 cm, fixed to muscle with no evidence of local inflammation. There was no history of trauma, fever or weight loss. There was occasional history of contact with the farm animals. The only significant hematological finding was a raised eosinophil count of 500/cu.mm. X-ray chest and sonography of abdomen were normal. Fine needle aspiration cytology (FNAC) showed clear fluid and microscopic examination revealed acellular smears. X-ray pelvis was also non-contributory. Serological tests were not done. On ultrasonography a well-defined oval cystic lesion was seen in the left gluteus maximus with double wall and internal echoes [Figure 1]. Computerized tomography (CT) scan showed a well-defined oval cystic lesion in left gluteus maximus, with enhancement of its wall on intravenous contrast injection [Figure 2]. Diagnosis of hydatid cyst of the gluteus maximus muscle was made.

Preoperatively albendazole 10 mg/kg/day was given for 15 days. This was followed by surgical excision of the mass. At operation the cyst was found to be surrounded by a thick fibrous capsule and was adherent to the muscles of the gluteal region but a total pericystectomy was possible without any spillage. The cavity was irrigated with hypertonic saline and closed with a suction drain. On microscopic examination it showed three layers: adventitia, laminated and germinal layers. The germinal layers had multiple brooding capsules with many scolices in it. The patient was followed for six months with no evidence of recurrence and had completed a three-month course of albendazole postoperatively.

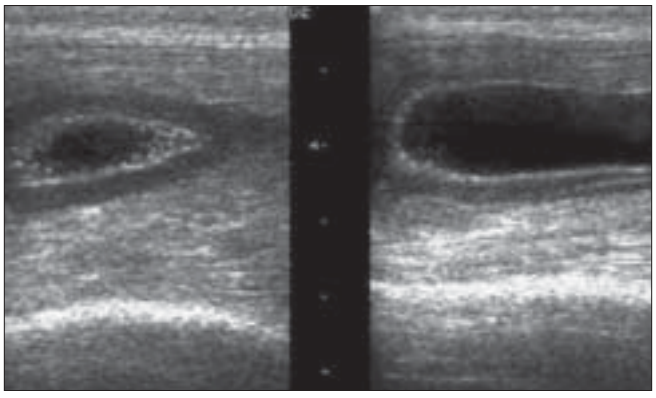

Figure 1: USG - a double-walled cystic lesion with Brodie's capsules in the left gluteus maximus muscle

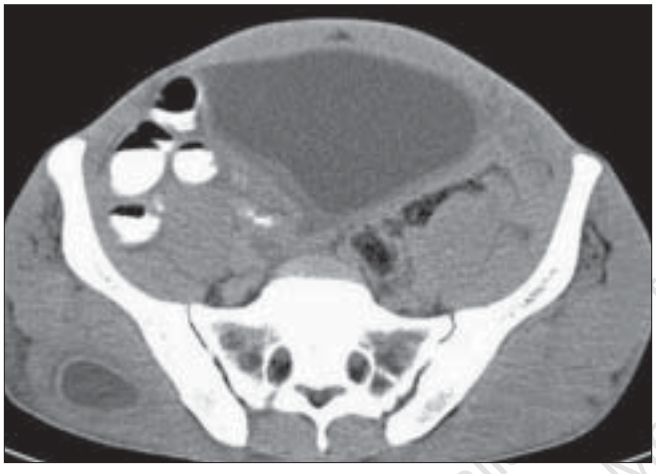

Figure 2: CT scan - a well-defined cystic lesion with wall enhancement in the left gluteus maximus muscle

Discussion

The usual primary sites of hydatid disease are the liver and the lungs with the less common ones being the bone, heart, central nervous system, spleen and muscles. Hydatidosis affecting the muscles without the evidence of the disease in liver or lungs is rare.[2-4] A case of primary hydatid cyst of the vastus lateralis muscle was describe by Kocakusak *et al*.[6] Another case of primary hydatid cyst in the supraspinatus muscle was reported by Tatari *et al*.[2]

Muscular hydatidosis, being rare, may cause a variety of diagnostic problems. Preoperative evaluation of the patients is mandatory in order to prevent rupture, infection, anaphylaxis and recurrence of the cysts. Routine investigations are not effective for diagnosis. Serological tests such as hemaglutination, Compliment fixation test or enzyme linked immunosorbent assay may aid in the diagnosis but are usually not positive in all cases.

Casoni's test which has been used for a long time for the diagnosis of hydatid disease has a high frequency of false positivity due to poor standardization of nitrogen content in the antigen and is no longer recommended as a diagnostic tool.[7]

Pre-operative diagnosis of hydatid disease may be made on ultrasound and confirmed by CT scan. Sonographically they have a thin or thick wall resembling the pericyst with internal echoes. Multiple echogenic foci due to hydatid sand may be evident giving the "snow storm" sign.[8] Simple cysts do not demonstrate internal structure. On CT scan they appear as a well-defined cystic lesion with daughter cysts, may contain septae or debris in it with no enhancement on intravenous contrast. Magnetic resonance imaging typically shows a thin, low intensity rim, probably representing the pericyst which is rich in collagen and is generated by the host.[8] Lewell[9] classified hydatid cysts into three types according to their imaging appearance. Type I is a fluid filled cyst-like structure, which may proceed to a Type II lesion if daughter cysts and/or matrix develop. Type III is mummified, inert calcified lesion.

FNAC is usually done as one of the first modalities for the diagnostic workup of superficial swellings. Although aspiration of hydatid cysts has been discouraged due to the association of various complications, some authors[10] have used aspiration of these cysts for the diagnosis.

The exceptional nature of primary muscle localization concerns diffusion of the infecting embryo; the most reliable hypothesis is that the liver and lungs can be bypassed through precapillary anastomosis between pre- and postparechymal circulation.[11] The muscle environment is not favorable for the growth of hydatid larvae but the volume of the muscle mass and its rich blood supply could explain the exceptional nature of localization in the proximal muscles of the lower limbs.[12]

Surgical excision forms the main modality of treatment. During surgical intervention all precautions like colored packs, antiscolicidal solutions along with meticulous surgical technique go a long way in the prevention of recurrence of this disease.

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