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CASE REPORT

Gluteal region musculoskeletal hydatid cyst: Case report and review of literature

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Abstract Hydatid disease is a parasitic infestation of humans and herbivorous animals, caused by *Echinococcu granulosus*. Dogs and some wild carnivores, like foxes, are definitive hosts, harboring worms in their intestines. Musculoskeletal cysts account for 0.7–3% of total cases of hydatidosis. Primarymuscular hydatidosis without involving the thoracic or abdominal organs is extremely rare. Intramuscular infestation may mimic a soft tissue tumor leading to inappropriate cyst rupture with the attendant risks of anaphylaxis and dissemination to other organs. So preoperative evaluation is critical to avoid life-threatening complications. We report a case of 34-year-old male patient with a cystic gluteal swelling turning out to be hydatid cyst on surgical exploration. Surgical excision with a pharmacology complementary treatment with antihelementhics is necessary to achieve the complete healing.

Keywords Echinococcosis · Hydatid disease · Musculoskeletal hydatid

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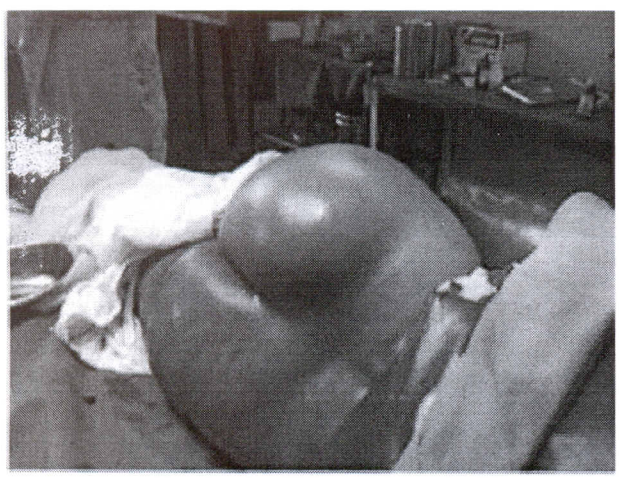
Introduction

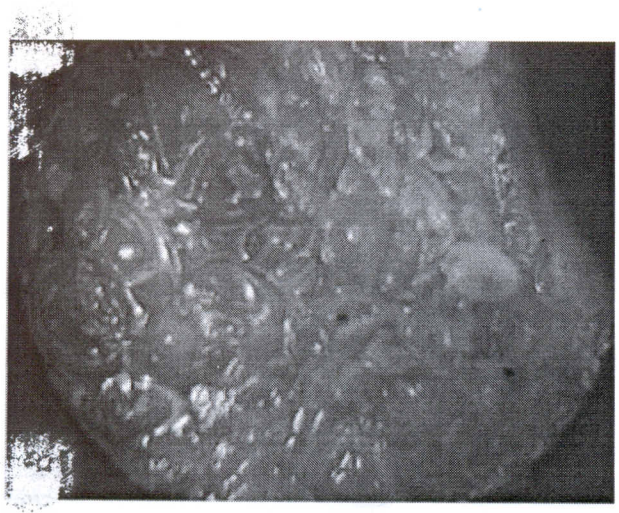
Echinococcosis (hydatid cyst disease) is a zoonotic infection most commonly caused by the larvae of *E. granulosus*, and the larvae of *Echinococcus multilocularis*, *Echinococcus vogeli*, and *Echinococcus oligarthrus* which inhabits in the small intestine of carnivores [1]. The adult worms produce eggs that are released with the feces and spread in various ways, such as through the wind, water or flies [2]. After ingestion by the host, the embryos migrate through the intestinal wall and are either arrested in the capillary bed of the liver developing into liver cysts, or manage to penetrate into systemic circulation thus ending up in remote organs. The lung, the brain, and the muscles or bones are the more frequently involved distant organs. Due to their physiologic role as capillary filters and their vast capillary volume, the liver and lung are most often affected. Other manifestations are found in 15% of the patients, with the skeletal system making up for 1–4% of all cases [3]. Voluntary muscles are a very rare site of infection, counting for <1% of total [4]. Hydatid cyst of muscle is not commonly encountered, as the presence of lactic acid creates an unfavorable milieu for growth [5]. In this report, we describe a case of cystic gluteal swelling turning out to be hydatid cyst on surgical exploration 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 34-year-old male labourer presented to the surgical outpatient department with a slowly growing painless swelling in the left gluteal region for the past 6 months.

Physical examination (Fig. 1) revealed a diffuse, tender, cystic swelling of approximately 12 × 10 cm, fixed to muscle with evidence of local inflammation. There was no history of trauma or weight loss. There was occasional histoy of contact with the farm animals. The only significant hematological finding was a raised eosinophil count of 500/cu mm. X-ray chest was normal. Needle aspiration showed pus. Serological tests were not done. A differential diagnosis of gluteal abscess, soft tissue tumour, and parasitic cyst were considered. But aspiration revealed pus. Diagnosis of gluteal abscess was done. 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 not possible without any spillage (Fig. 2). 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 6 months with no evidence of recurrence and had completed a 3-month course of albendazole postoperatively.





Discussion

Hydatid cyst disease is widespread throughout sheep-producing regions of southern Europe, Asia, Australia, Africa, and the Middle East. In humans, the liver is the most common site of cyst development (60%), followed by the lungs (20%), and much less commonly, the kidney, spleen, brain, and soft tissue. The incubation period may last 5–20 years, and symptoms may present several years after exposure. Infections involving bony sites, including the spine, pelvis, and hip, also have been reported [6].

Hydatidosis affecting the muscles without the evidence of the disease in liver or lungs is rare [7, 8, 9]. The literature mentions a very few cases worldwide, as per our knowledge (Table 1). 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. However, serology alone is insufficient to diagnose hydatid cyst disease. False negative test results occur in as many as 50% of patients with solitary lung cysts [1]. The sensitivity and specificity of serologic tests in patients with soft tissue hydatid cyst disease is not known. 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 [19].

Aspiration 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 [20] have used aspiration of these cysts for the diagnosis. Concerns over microscopic spillage at the time of needle biopsy do not appear warranted, especially if patients receive subsequent medical treatment and biopsy tracts are resected at the time of surgery [21].

The diagnosis of hydatid cyst disease usually is based on the identification of a hydatid cyst in tissue. 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. Simple cysts do not demonstrate internal structure. On computed tomography 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 [22]. MRI findings of hydatid cysts in the liver are well described

Table 1 Reported sites of Intramuscular infection and bone, liver or lung involvement [2, 10]

Author	No	Site of infection	Liver/lung/bone involvement
Merkele et al. [3]	8	Iliopsoas, left adductor musculature, left femur, left medial gluteal muscle, musculature of right upper leg	Yes
Jeber et al. [11]	1	Paravertebral structures	Yes
	1	Infratemporal	Yes
Von sinner [12]	1	Pelvic	Yes
Torricelli et al. [13]	14	Bone infection with adjacent soft tissue involvement in 12 cases	
in et al. [14]	1	Cerebral	Yes
	1	Biceps brachii	No
	7	5 bone infections without softtissue involvement, 2 primary intramuscular (left shoulder, rectus femoris and vastus lateralis)	No
Sanjay Marwa . [15]	1	Quadriceps muscle	No
	1	Sartorius	No
Adnan Hasanoglu [17]	1	Left gluteal region	No
Arazi et al. [18]	15	Gluteus maximus muscle	Yes
Panagiotis G Drimousis et al. [10]	1	Left gluteal muscle and left iliopsoas muscle	No

but the diagnosis is more difficult to make in soft tissues because the findings are not well described [5, 23, 24]. A low-intensity rim can be seen on both Tl and T2-weighted images, but is more prominent on T2-weighted ones, this rim is less developed in muscle [5]. A high-intensity signal on T2-weighted images without significant central uptake of paramagnetic contrast, suggest a cystic component. Computed tomography or ultrasound-guided needle biopsies also are helpful in diagnosis [25], although some authors do not recommend it because of the risk of cyst rupture and anaphylactic reaction could occur [26]. Lewell [27] 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.

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 [28]. 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 [29].

The treatment of muscle hydatid cyst disease is a combination of chemotherapy and surgery, with wide excision of the presenting swelling from adjacent muscle. Albendazole has been found to be more useful in treating this lesions. The fluid of *E. granulosus* cysts contains significant amounts of foreign protein and is extremely toxic to the host. Cyst rupture can cause anaphylactic shock or may release a large number of viable scoleces that implant elsewhere and produce secondary cysts [30]. Albendazole sulfoxide, albendazole sulfone and combined albendazole sulfoxide and albendazole sulfone mixed together in concentrations of 50 μg/ml of 4%, NaCl 20%, and 1.5% cetrimide – 0.15% chlorhexidine (10%) did not provide a marked result as scolicidal agents [31].

In this case incision and drainage was planned initially, but on table procedure done was incision and drainage of the pus and cyst in the cavity and complete excision of the wall of the swelling [cyst wall] from the adjacent muscle. Hypertonic saline irrigation was given and wound closed with suction drain *in-situ*.

In summary, muscular Echinococcus infestation is a rare but important entity, more frequently seen in rural areas. 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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