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Hydatid Cyst of Soft Tissues / Muscles - A Case Report of Multiple Hydatid Cysts Thigh

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Abstract

Introduction: Hydatid diseases are a cystic parasitic infestation caused by *Echinococcus granulosus* which are endemic to parts of India and commonly occur in agricultural workers. Primary intramuscular hydatidosis is a rare manifestation of hydatid disease.

Presentation of Case: Our case report describes a rare case of primary intramuscular hydatidosis in an agricultural worker with no involvement of any other viscera. This was diagnosed with clinical evaluation, radiological modalities and managed surgically.

Discussion: Hydatid disease is endemic in parts of India. Primary intramuscular hydatidosis is uncommon because the lactic acid in the muscle and muscle contractility hinders the development of cysts making it improbable diagnosis on first presentation. Hydatid cyst demonstrates a wide variety of imaging features, which can vary according to growth stage, associated complications and affected tissue. The treatment of choice is complete surgical excision of the cyst along with thorough irrigation of the surrounding soft tissues combined with the use of systemic antiparasitic drugs after surgery.

Conclusion: In a patient of an agricultural background of cattle rearing living in an endemic region and presenting with a swelling in the musculoskeletal system, a suspicion of hydatidosis should always be kept in the clinician's mind, so that it can be diagnosed using a multimodal approach and managed properly in a timely manner.

Keywords: ***Hydatid cyst; Echinococcus; Ultrasound; Magnetic resonance imaging; Thigh; Muscle; Surgery***

1. Introduction

Hydatid cyst is a zoonosis caused by larval or cyst stage of the tapeworm Echinococcus granulosus. These are commonly encountered in farm workers dealing with agriculture and livestock [1]. Transmission to humans occurs by eggs in the feces of the infected dogs, contaminated food and drinking water, and oral ingestion of eggs that are too small to be visible. This

parasitic disease can affect any organ of the body, however, liver (55%-70%) followed by the lung (18%-35%) are the most commonly affected organs. The heart, brain, vertebral column, ovaries, pancreas, gall- bladder, thyroid gland, breast, and bones have lower incidence rate of involvement. Primary soft tissue involvement such as thigh muscles are extremely rare (0.5%-4.7%) [2]. Intramuscular hydatid cyst cases usually occur with the spread of the cyst in another area or secondary to the surgery of the cyst in a different area. Primarily, cases of hydatid cysts in the thigh muscles are extremely rare and are generally in the form of case reports. The patient could present with a slow growing large swelling in the extremities, which on clinical examination may give the appearance of a soft tissue tumour such as a sarcoma, liposarcoma or a lipoma. The preoperative radiological diagnosis, provided by ultrasound and MRI is of utmost importance to take precautions and avoid certain procedures that could cause fatal complications. This case report presents a case of primary isolated hydatid cyst in the thigh, with a successful surgical cure.

2. Case Description

2.1 Clinical findings

A 37-year-old male working as a farmer in rural India, presented with complaints of swelling over left thigh region. Disease onset was 6 years ago when patient first noticed a small swelling over left thigh. Swelling was insidious in onset, spontaneous in origin and gradually progressive in nature. No period of rapid increase or regression in size of swelling, no history of pain over swelling, no history of trauma or fever, No history of any co-morbidities. General examination of the patient showed no irregularities. Physical examination showed a firm, non-tender mass of the upper two third of the left thigh. Overlying skin was normal. No other visible or palpable swelling was present in the rest of the body. Chest X-ray and Contrast Enhanced Computed Tomography (CECT) abdomen pelvis and chest did not reveal any other organ involvement (FIG. 1).

FIG. 1. **Pre and post operative image of left thigh.**

2.2 Diagnostic assessment

All routine blood investigations were normal. Serology for echinococcus was positive 2.61 (reference <0.9). Ultrasonography (USG) revealed large multi-loculated complex cystic mass measuring 12.1× 8.5 × 8.7 cm in the anterior and medial aspect of the upper two third of left thigh. Magnetic Resonance Imaging (MRI) revealed a large ill defined, multilobulated solid-cystic lesion is seen in the anterior, medial and posterior compartment of left thigh in the inter - and intra-muscular planes. The lesions together measure approx. 22 × 20 × 17 cm in size. Posteriorly part of lesion is seen displacing the sciatic nerve postero-medially. Similar lesion is also seen in the left inguino-femoral region. It is seen displacing the femoral vessels antero-medially. The lesion measures approx. 21 × 7 × 3 cm in size. No skeletal involvement was seen (FIG. 2).

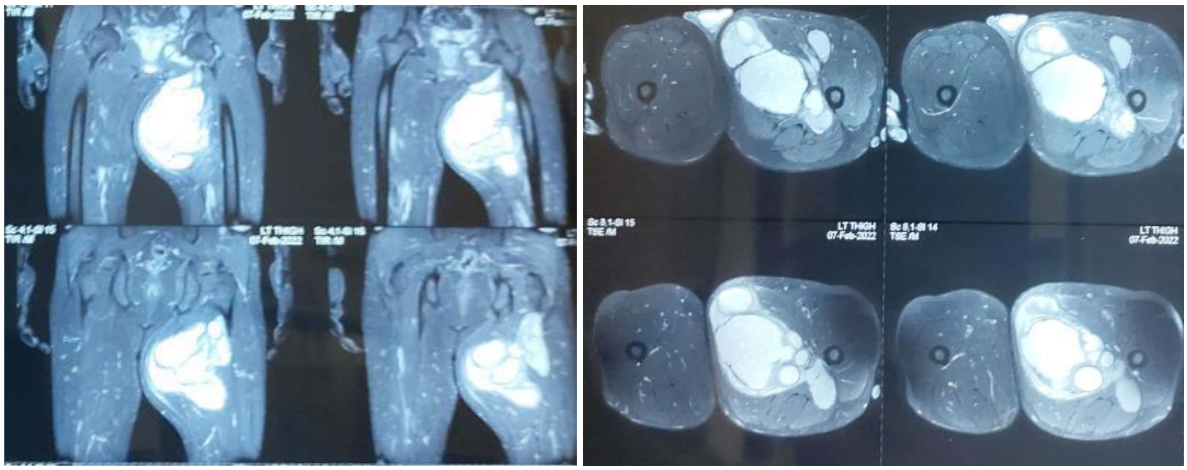

FIG. 2. **MRI showing inter- & intra-muscular hydatid cyst with multiple daughter cysts in the left thigh (a) Coronal section (b) Axial section.**

2.3 Therapeutic intervention

The patient was treated with Albendazole 400 mg twice daily for a week before surgery was done. Under general anesthesia patient in supine position with left knee in flexion and left hip is abducted. Single oblique incision was given over left thigh medially (13 cm). After securing the wound cysts were injected with a 2% solution of cetrimide-chlorhexidine combination to reduce the risk of recurrence and cyst with all its layers along with daughter cysts were resected without any rupture or puncture. Pericystectomy was done for all cysts along with evacuation of daughter cysts. Thorough wash was given with 2% solution of cetrimide-chlorhexidine. Romo vac drain (16Fr) was inserted in the antero medial intramuscular plane, and the wound was closed primarily. Post-operative period was uneventful. Histopathological examination confirmed the diagnosis of muscular hydatid cyst. Drain was removed on the fifth day and discharged on eigth day (FIG. 3).

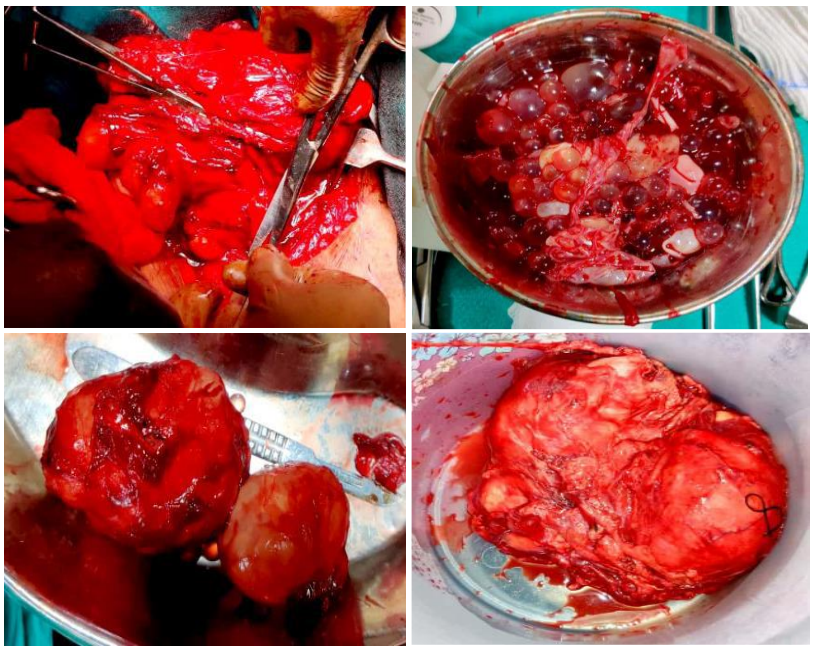

FIG. 3. **Intra-op specimens.**

3. Follow-Up and Outcome

On discharge, patient was advised Albendazole 400 mg twice daily and follow up done weekly. Patient had no local or systemic complications. There were no clinical signs of recurrence. The patient was followed up for three months, USG thigh was repeated no residual tumour was detected.

4. Discussion

Hydatid disease is an endemic zoonosis caused by echinococcus granulosus prevalent mainly in countries where sheep raising is practiced; however, due to increased travel and tourism around the world, it can be found anywhere, even in developed countries. In India, hydatid disease is commonly seen in the southern states of Andhra Pradesh and Tamil Nadu [3].

Hydatid disease can be primary or secondary. Primary hydatid disease is spread by the ingestion of the ova and may develop in almost any organ. Liver and lungs are the most frequently affected organs. In secondary hydatid disease, larval tissue proliferates after spreading from the primary site. However, there is a primary site of hydatid disease situated in the liver, lung, or spleen [4].

Differential diagnosis of hydatid disease should be considered for every soft cystic mass in any anatomical location, especially in endemic areas of the disease [5].

Musculoskeletal involvement as a primary location, is rarely seen even in endemic countries with a frequency that does not exceed 5% [6]. This low rate is due to the liver detoxification function, to muscle contractions preventing larvae fixation, and to the secretion of lactic acid with a toxic action. Among the reported muscle locations, proximal muscles seem to be the most affected probably due to a richer vascularization [7,8].

Clinical presentation depends on the site, the effect on adjacent structures and complications. Initially the course of disease is asymptomatic, oftenly painless swelling of the soft tissues is observed. However, complications such as superinfection, nerve compression, rupture and allergic reactions lead to an inflammatory expression which can simulate clinically as an abscess or a malignant tumor, a source of diagnostic delay [9]. During muscle invasion, biology reveals occasionally a non-specific eosinophilia and serology is inconstantly positive with many false positives up to 80% of cases [10].

Imaging techniques are essential to raise the possibility of hydatid disease and to prevent inappropriate biopsy or aspiration. Imaging findings allow one to take intraoperative pre-cautions in order to prevent any cracking or rupture causing dissemination which may result in severe anaphylactic reactions [11]. USG, MRI, and CT are widely used imaging methods in the diagnosis of hydatid cyst, confirmation of the diagnosis, and investigation of adjacent organ involvement.

MRI is the examination of choice in case of suspicion of intra-muscular hydatid disease. This is due to its ability to adequately demonstrate most features of hydatid disease, with the exception of calcifications. MRI images may identify various patterns of intra-muscular hydatidosis such as its peripheral rim (also known as the rim sign), the membranes inside the cyst, peripheral oedema and peripheral enhancement with gadolinium which occurs due to the vascularization of the peri cyst.

In the treatment of hydatid cyst, surgical excision and preoperative and postoperative anthelmintic chemotherapy are the ideal treatments. Patient in our study was treated with wide surgical excision and anthelmintic chemotherapy both preoperatively and postoperatively. After intraoperative excision, washing 2% solution of cetrimide-chlorhexidine should definitely be used to prevent the scoleces from spreading [12]. We recommend the use of anthelmintic chemotherapy to reduce preoperative intra-cystic pressure and parasitic load, and to prevent anaphylaxis and dissemination in the intraoperative-postoperative period. Albendazole or Mebendazole can be used in anthelmintic treatment. In our study, we used Albendazole in the treatment because it reaches a higher concentration in the cyst content and has better intestinal absorption [13]. Hydatid cyst should be kept in mind especially in masses that grow slowly in soft tissue in endemic areas. Despite high complication rates and recurrence rates up to 70%-80%, the risk of recurrence in primary hydatid cyst of the thigh can be reduced by careful preoperative planning, delicate surgery avoiding cyst rupture, and neoadjuvant and adjuvant anthelmintic chemotherapy. As our study shows, this combination of surgical method and chemotherapy seems to be an effective and gold standard method in the treatment of primary hydatid cyst of the thigh. The treatment of choice at present is complete surgical excision of the cyst along with thorough irrigation of the surrounding soft tissues with hypertonic saline to prevent recurrence. These procedures should be combined with the use of systemic antiparasitic drugs after surgery [14]. The patient must be kept under long-term clinical evaluation after surgery, to check for recurrence.

5. Conclusion

Primary intramuscular hydatidosis is an extremely rare manifestation of Hydatid disease which is endemic in India. The diagnosis of this disease proves challenging and requires a multimodal approach. The patient presented to us with a large swelling over his left thigh and a diagnosis was established with detailed history, clinical examination along with radio imaging. MRI considered the best technique in the diagnosis. Excision of the hydatid cyst using Pericystectomy technique is the first choice of treatment along with evacuation of daughter cysts, followed by Albendazole therapy.

6. Conflicts of Interest

There is no conflict to be declared.

7. Funding

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