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Echinococcosis in an Unusual Site: A Rare Case Report of a Primary Hydatid Cyst in the Thigh

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Background: Hydatid disease, caused by the *Echinococcus granulosus* parasite, primarily affects the liver and lungs. Muscular involvement is extremely rare. A primary hydatid cyst in the thigh is an unusual and rare presentation, as hydatid disease typically affects the liver (70%) and lungs (20%), with muscle involvement being extremely uncommon (0.5–5%). This report presents a case of a primary hydatid cyst in the thigh.
Case Presentation: A 30-year-old male patient presented with a swelling in the anterior thigh, which had been gradually enlarging. The patient reported no pain, fever, or trauma. On the basis of local examination, suspected of small cystic swelling, patient was planned for excision. Intra-operatively the swelling was diagnosed as a case of hydatid cyst with multiple cysts in the thigh.

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This case report marks the significance of preoperative radiological investigations in patients with rare clinical presentation of hydatid disease.
Conclusion: Primary hydatid cyst of the thigh is rare but should be considered in differential diagnoses of soft tissue masses, especially in endemic areas.

Keywords: Hydatid disease; Echinococcus granulosus; scolicidal; albendazole.

1. INTRODUCTION

Hydatid disease, also known as cystic echinococcosis, is a parasitic infection caused by the larval stage of *Echinococcus granulosus*, a tapeworm endemic to many regions worldwide, particularly the Mediterranean, Middle East, South America, sub-Saharan Africa, and parts of Asia [1]. Humans are accidental intermediate hosts, typically through ingestion of eggs from contaminated food or contact with animals [2]. The liver and lungs are the primary organs affected, with muscular involvement being very rare. In the musculoskeletal system, the incidence of hydatid cysts is low, with muscle involvement making up less than 5% of cases. However, primary hydatid cysts in muscle, meaning no other organs are affected, are exceedingly uncommon and have only been documented in a small number of cases worldwide [3,4]. The thigh is an especially rare location for primary hydatid cyst formation, possibly due to the high vascularity and dynamic contractions of the muscle, which might prevent larvae from embedding and maturing [4,5]. However, when hydatid cysts do present in this location, they pose a diagnostic challenge due to their nonspecific presentation. Clinically, a thigh hydatid cyst may resemble other soft tissue tumors, such as lipomas, abscesses, or malignant sarcomas, making accurate diagnosis critical but challenging. The rarity of muscle hydatidosis and the nonspecific presentation underscore the importance of considering hydatid disease as a differential diagnosis for any unusual cystic lesions, particularly in patients from endemic regions or with a history of exposure to risk factors [2,6]. Treatment typically involves surgical excision, often supplemented by anti-parasitic therapy to minimize the risk of recurrence or dissemination. Proper surgical handling to prevent cyst rupture and spillage is crucial to avoid secondary echinococcosis [7,8].

2. CASE PRESENTATION

A 30-year-old male patient presented with a swelling in the anterior thigh, which had been gradually enlarging over. The patient reported no pain, fever, or trauma to the area and had no significant past medical or travel history. There were no systemic symptoms. On examination, the swelling was non-tender, soft to firm, with no signs of inflammation. There was no restriction in the range of motion, and distal neurovascular status was intact. On the basis of local examination and suspected small cystic swelling was planned for excision. No preoperative radiological investigation was done as it was suspected to be single, small cystic swelling. Intra-operatively, it was found to be hydatid cyst with multiple cysts in the subcutaneous space of the thigh, and incision over the thigh was extended to explore the area for more cysts with extreme care taken to avoid rupture and dissemination. More cysts were found in the subcutaneous region of the anterior thigh over the thigh muscle which were carefully dissected and removed intact (Figs. 1 and 2). Surrounding muscle tissue was irrigated with povidone-iodine (scolicidal agent) to prevent recurrence or secondary infection. The closure of wound was done after placing the suction drain in the subcutaneous space. The patient recovered well, with no postoperative complications. Drain was removed on 5th postoperative day and sutures were removed on 14th postoperative day. Albendazole was administered for one month to reduce the risk of recurrence.

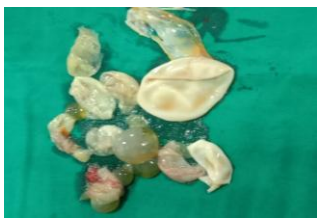

Fig. 1. Multiple hydatid cysts removed from the thigh

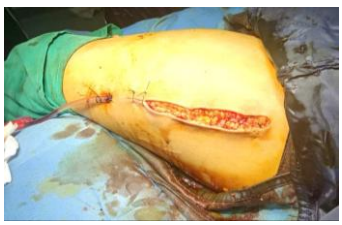

Fig. 2. Wound closure with suction drain

3. DISCUSSION

Primary hydatid disease involving skeletal muscle is exceptionally rare, especially in the thigh. Muscular involvement occurs in only 0.5–5% of all hydatid cases, largely due to the muscle tissue's unique physiology [2]. Factors such as the constant movement of muscles, blood supply characteristics, and local immune defense mechanisms are thought to create an environment hostile to Echinococcus larvae, making muscle an unusual site for cystic development. In addition, the lactic acid produced by muscle metabolism may inhibit the survival of the larvae, thus reducing the likelihood of cyst formation in muscle tissue [9]. When hydatid cysts do occur in the muscle, they are more often secondary, originating from a primary cyst in the liver or lungs that spreads hematogenously. In this case, however, no evidence of primary cysts in other organs was found, suggesting that this is a true primary hydatid cyst localized to the thigh. Cases of primary musculoskeletal hydatidosis have been documented in other muscles, such as the psoas, biceps, and gluteal muscles, but thigh involvement remains especially uncommon. The pathophysiology behind primary hydatid disease of muscle remains a topic of discussion. It is hypothesized that the parasite's oncospheres (embryos) might bypass the liver and lung, where they typically lodge, due to anatomical variations or an unusual immune response, and travel to muscles via the systemic circulation. In this case, it is conceivable that the larvae settled directly in the thigh muscle after penetrating the gastrointestinal barrier, leading to the formation of a primary cyst in the muscle. The presentation of hydatid cysts in thigh is generally nonspecific, with patients often reporting a painless, slowly enlarging mass that may go unnoticed for months or even years. Clinical symptoms usually arise only when the cyst reaches a considerable size, causing compression of surrounding structures [10]. In this case, the patient presented with a painless swelling in the thigh, which, given its slow growth, was initially not alarming. Due to the atypical location, differential diagnoses included benign and malignant soft tissue tumors, abscesses, hematomas, or other parasitic infections [11]. The nonspecific nature of the symptoms makes imaging and serology crucial for accurate diagnosis.

Imaging modalities such as ultrasound, CT, and MRI are essential for diagnosing hydatid disease. Ultrasound is often the first-line investigation, revealing the cystic structure and characteristic internal septations. MRI, however, offers superior soft-tissue contrast, allowing for a more detailed view of cyst wall, daughter cysts, and septations, which are highly suggestive of hydatid disease [5,12]. In our case, no preoperative radiological investigation was performed as it was suspected to be single, small swelling. In such cases and where this disease is endemic, diagnosis of hydatid should be kept in mind. It is advised to have preoperative diagnosis with radiological investigations before proceding for intervention in such patients. Kurz K et al, in the study also reported the significance of preoperative radiological investigation to establish the diagnosis in patients with atypical presentations [13]. Positive serology for Echinococcus antibodies further support the diagnosis, though serology may occasionally yield false negatives, particularly in cases with isolated muscle involvement [4]. The mainstay of treatment for muscular hydatid cysts is complete surgical excision. During surgery, it is crucial to avoid cyst rupture to prevent the dissemination of viable protoscolices, which could lead to secondary echinococcosis or anaphylactic reactions. For this reason, surgeons often employ techniques such as "pericystectomy" (excision of the cyst and its surrounding capsule) and may irrigate the surgical field with hypertonic saline or other scolicidal agents to kill any potentially spilled protoscolices [8,14,15]. In this case, careful dissection allowed the cysts to be removed intact, and the surrounding tissue was treated with povidone iodine as a preventive measure. Postoperative antiparasitic therapy, typically with albendazole, is recommended to minimize the risk of recurrence or any potential missed microscopic disease. Albendazole works by inhibiting the uptake of glucose by the parasite, thereby disrupting its metabolism and growth. The usual course of treatment varies depending on the size and location of the cyst, but therapy generally continues for several weeks postoperatively [8,14]. This case underscores the importance of considering hydatid disease as a differential diagnosis for unusual cystic masses in patients from endemic areas or with known risk factors for echinococcosis. The rarity of primary hydatid cysts in muscle and their variable presentation poses a diagnostic challenge, and cases are often misdiagnosed or identified late. Awareness of such unusual presentations can lead to earlier diagnosis, appropriate management, and better outcomes for patients. Furthermore, there is a role of multidisciplinary management, including radiology, surgery, and

infectious disease consultation, in achieving successful treatment outcomes. Hydatid cyst of the thigh is the rare presentation and diagnosis of hydatid should also be kept in mind in patients of endemic areas. This case report marks the significance of preoperative radiological investigations in patients with rare clinical presentation of hydatid disease.

4. CONCLUSION

Primary hydatid cysts of the thigh, though rare, are possible, and this diagnosis should be considered in the differential for soft tissue masses in endemic areas. Accurate diagnosis relies on imaging and serology, with MRI providing detailed visualization of the cystic structure. Surgical excision is the preferred treatment, and careful handling during surgery is essential to prevent rupture and subsequent complications. Postoperative antiparasitic therapy is beneficial in reducing the risk of recurrence. Increased awareness of atypical presentations of hydatid disease may improve diagnosis and management, ultimately leading to better patient outcomes.

CONSENT

Written informed consent was taken from the patient.

ETHICAL APPROVAL

Exempted from ethical approval from institutional ethics committee as it's a case report.

DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Authors hereby declare that no generative AI technology and text to image generator have been used during the writing or editing of this manuscript.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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