#### **Case Report**



# A Primary Hydatid Cyst of Thigh Mimicking as Soft Tissue Tumour: Diagnostic Dilemma

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Received 17 April 2024;

Accepted 12 May 2024;

Published 14 May 2024

#### Abstract

Hydatid disease is one of the most prevalent parasitic infections caused by *Echinococcus granulosus*. It is likely to affect all organs. However, primary hydatid cysts of muscle tissue are so uncommon. A timely and precise diagnosis is critical to avoiding an unnecessary biopsy. Here, we present a case of a 29-year-old female presented with soft tissue swelling over the anterior aspect of the right thigh. The diagnosis of a hydatid cyst was made based on ultrasonography (USG) and magnetic resonance imaging (MRI). A complete surgical excision of the cyst was done with an uneventful postoperative course.

Keywords: Hydatid disease, Echinococcus granulosus, intramuscular hydatid, Magnetic Resonance Imaging.

### Introduction

Hydatid cysts are parasitic infections transmitted to people by dog faeces, and they are common in places where agriculture and livestock breeding are practiced. The most common causative agent is *Echinococcus granulosus*<sup>[1]</sup>. The liver and lungs are the two most often affected organs in humans, as ingested eggs hatch in the intestines and go to the liver via the portal system. Embryos that escape the hepatic filter are delivered to the right heart via venous circulation and then to the lungs. If embryos exit the pulmonary circulation and enter the left heart chamber, the arterial circulation can transport them to any part of the body. Echinococcosis may also affect the brain, heart, kidney, ureter, spleen, uterus, fallopian tube, mesentery, pancreas, diaphragm, and muscles <sup>[2]</sup>. Musculoskeletal involvement is uncommon, accounting for just 1-5.4% of musculoskeletal hydatid cyst cases. Because of the low prevalence and similarity of these lesions to soft tissue masses, pre-operative diagnosis is challenging in this condition. In many cases, the diagnosis is confirmed intraoperatively or after histological examination<sup>[3]</sup>.

#### **Case report**

A 29-year-old female patient presented with swelling over anterior aspect of the right thigh for 1 year. On local examination, there was 20x15 cm intramuscular swelling present over anterior aspect of right thigh, and firm in consistency (Figure 1). All biochemical parameters were within normal limits. In clinical suspicion of soft tissue tumour of right thigh ultrasonography was done and it showed a large oval shaped hypoechoic lesion of size 14.8x8.3x4.5 cm with multiple thin walled, variable sized clear cysts in intramuscular plane (Figure 2). Contrast-enhanced MRI revealed a 24×10x8 cm cystic lesion with multiple daughter cyst in vastus muscle of right thigh (Figure 3). Ultrasonography of abdomen and chest was done to see the presence of hydatid cyst in peritoneal and pleural cavity, which was normal. Pre-operatively patient was started tab albendazole 400 mg BD for 15 days. Excision of entire cyst with hydatid sand was done (Figure 4). Intra-operative and postoperative course was uneventful. Postoperatively patient was again started on tab albendazole 400 mg BD.



Figure 1: Clinical photograph showing 20x15 cm intramuscular swelling present over anterior aspect of right thigh

Figure 2: Ultrasonography of right thigh showed a large oval shaped hypoechoic lesion of size 14.8x8.3x4.5 cm with multiple thin walled, variable sized clear cysts in intramuscular plane



with multiple daughter cyst in vastus muscle of right thigh

# sand (Total excision of cyst with removal of hydatid sand was done)

# Discussion

Hydatid disease is frequent in rural regions, with carnivores as primary hosts and herbivores as intermediate hosts. Over 90% of these instances involve the liver, lungs, or both. However, musculoskeletal involvement is uncommon, accounting for just 1% to 4% of all cases. Humans become infected accidentally by consuming contaminated water or vegetables. As the larvae enter the host's body, they travel via the circulation, penetrating the intestinal mucosa, and get lodged in the capillary bed of the liver, resulting in liver cysts in approximately 75% of cases. However, some enter the systemic circulation, forming cysts in the lungs, muscles, soft tissues, brain, or bones. Around 15% of cysts occur in the lung, 8-10% in unusual places, and 1-4% in the musculoskeletal region <sup>[4]</sup>.

due to the presence of high lactic acid levels, muscle contractions, and blood filtration activities in the liver and lungs. The proximal muscles of the lower leg could be one of the sites due to the strong blood supply and the presence of muscular mass <sup>[4,5]</sup>.

Primary hydatid cysts in muscle tissue are extremely rare, with only a few cases reported in the literature. Khanna *et al.* identified 24 atypical hydatid cyst sites in 110 patients with cystic echinococcosis during a 23-year period. These areas were in decreasing order of frequency: spleen, skin, and soft tissue <sup>[6]</sup>. Mseddi *et al.* found 11 intramuscular hydatid cysts over a period of 17 years <sup>[7]</sup>.

Clinically, muscle hydatid disease is asymptomatic in its early stages. The symptoms and signs of hydatid cyst disease vary depending on the organ affected, the site of localization, the effect on adjacent tissue, complications following rupture, immunological reactions, and secondary infection. A palpable mass is the most common clinical finding of hydatid disease in soft tissues, and clinical symptoms are caused by organ compression. Muscle hydatid illness is typically characterized by a slow-growing soft-tissue tumour that can resemble myositis or a calcified hematoma. However, complications such as nerve compression or infection can reveal some cysts, mimicking an acute abscess or a malignant tumour. Peripheral nerves may be squeezed, resulting in neurological impairments <sup>[5,8,9]</sup>. Our patient typically presented with a painless, progressive-developing swelling over the anterior aspect of the right thigh mimicking a soft tissue tumor without neurological symptoms in an afflicted area of the body.

A muscular hydatid cyst is diagnosed based on symptoms and imaging studies. Examining the fluids aspirated from the cyst can help to make a specific diagnosis, although it is not recommended routinely due to the risk of leakage, disease transmission, and anaphylactic shock. USG is an effective diagnostic tool for determining the size, position, and type of cysts. USG has a sensitivity of 95%, and if vesicular fibrils are present, the sensitivity rises to 100%. The snowstorm sign may be characterized by several echogenic foci caused by hydatid sand. A computed tomography (CT) scan shows a well-defined cystic lesion with daughter cysts that may contain wall calcification, septae, or debris. Aside from calcifications, MR imaging can accurately depict the majority of hydatid cyst characteristics in relation to surrounding structures. Furthermore, MR imaging often reveals a thin, low-intensity rim, which most likely represents the pericyst, a collagen-rich tissue produced by the host. A low-intensity rim (rim sign), more visible on T2-weighted images, has been noted as a typical sign in muscle hydatid <sup>[10]</sup>.

Hydatid cysts can be diagnosed by serological techniques such as ELISA, Western blot, immunodiffusion, immunofluorescence, indirect hemagglutination (IHA), specific immunoglobulin E (IgE), specific IgE, and complement fixation. The IgG ELISA and IHA are the two most commonly done techniques for detecting hydatid cysts. The sensitivity and specificity may vary based on the location of the cyst, its viability, and the parasite type <sup>[9]</sup>.

Differential diagnosis of intramuscular hydatid cysts includes soft tissue tumours, abscesses, lipomas, sebaceous cysts, helminthic cysts, neurofibromatosis, tuberculosis, fibrocystic illness, and necrotic soft tissue tumor <sup>[9]</sup>. Togral *et al.* found that it can imitate osteomyelitis and sarcomas in bone involvement <sup>[11]</sup>. According to Selahi *et al.* the differential diagnosis could include myositis or a calcified hematoma <sup>[12]</sup>.

Before surgery, the diagnosis must be confirmed. However, accidental cyst opening during needle biopsy or excision increases the possibility of anaphylactic shock or dissemination. Anaphylactic shock, danger of spread, and recurrence have all been recorded in the literature as complications of perforation or unintentional cyst opening during resection <sup>[9]</sup>. Our case was treated with albendazole for two weeks, and we attribute the absence of systemic anaphylactic symptoms to that. We recommend neoadjuvant albendazole for muscle hydatid cysts because it lowers preoperative intracystic pressure and parasite burden, which is helpful for preventing anaphylaxis and dispersion during and after surgery.

Surgery is the most effective treatment option for hydatid cysts. It should be removed completely whenever feasible. Spillage of cyst content during surgery should be avoided because it can induce serious anaphylaxis and spread. Irrigation with 0.5% cetrimide, 15% hypertonic saline, and 0.5% silver nitrate solution can be performed intra-operatively to kill the daughter cysts and lessen the likelihood of an allergic reaction. The World Health

Organization (WHO) recommends aggressive surgery and antihelmintic treatment to avoid the recurrence of localized lesions. Percutaneous aspiration, infusion of scolicidal drugs, and reaspiration (PAIR) can be used as an alternative to surgery <sup>[9]</sup>.

### Conclusion

Hydatid disease in muscle tissue is uncommon. Blood and imaging investigations should be done to rule out other differential diagnosis. Pre and post-operative chemotherapy and total surgical excision is recommended to produce successful results.

# Abbreviations

USG: Ultrasonography MRI: Magnetic Resonance Imaging CT: Computed Tomography MR: Magnetic resonance ELISA: Enzyme linked immunosorbent assay IHA: Indirect Hemagglutination WHO: World health organization PAIR: Puncture, aspiration, injection, re-aspiration

# **Data Availability**

The authors confirm that the data supporting the findings of this article are available within the article and its supplementary materials. Detail data regarding the participant is available with the authors.

# **Conflicts of interest**

Authors declare that they have no conflicts of interest.

# Ethical Approval and Consent to participate:

Ethical Approval: Not applicable.

Written and Informed Consent was obtained from the patient regarding her data to be shared for scientific research purpose and publicly.

# **Consent for publication**

Written and Informed Consent was obtained from the patient before submission of an article.

# **Funding Statement**

Not applicable

# Authors' contributions

**Dr. Nitin Sherkar;** Writing - original draft /Conceptualization/ Data curation / Resources

**Dr. Bhupendra Mehra;** Supervision/ Validation /Writing - review & editing/ Project administration

Dr. Siddharth P. Dubhashi; Supervision/ Validation /Writing -

review & editing/ Project administration

Dr. K. Senthilkumaran; Data Curation/ Resources

Dr. Vinod Pusdekar; Data Curation/ Resources

Dr. Ateet Fulmali; Data Curation/ Review of Literature

# References

[1] Sadullah S, Hattapoglu S. Intramuscular hydatid cyst in the lower extremity: report of three cases. Rev Soc Bras Med Trop. 2021;54:e0255-2021.

- [2] Alimehmeti R, Seferi A, Rroji A, Alimehmeti M. Saphenous neuropathy due to large hydatid cyst within long adductor muscle: case report and literature review. J Infect Dev Ctries. 2012 Jun 15;6(6):531-535.
- [3] Soltany S, Hemmati HR, Toussy JA, Nazifi H, Alibakhshi A, Toosi PA. Evaluation of Musculoskeletal Hydatid Cyst Cases in Terms of Clinical Manifestations, Method of Dealing, Treatment, and Recurrence. Open Access Maced J Med Sci. 2020 Mar 25;8:e99-104.
- [4] Shah E, Shah S. Journal of Clinical and Diagnostic Research. 2019 Feb;13(2): 01-02.
- [5] Tekin R, Avci A, Tekin RC, Gem M, Cevik R. Hydatid cysts in muscles: clinical manifestations, diagnosis, and management of this atypical presentation. Rev Soc Bras Med Trop.2015; 48(5):594-598.
- [6] Khanna AK, Prasanna GV, Khanna R, Khanna A. Unusual sites of hydatid cysts in India. Trop Doct 2005;5:233-235.
- [7] Mseddi M, Mtaoumi M, Dahmene J, Ben Hamida R, Siala A, Moula T, et al. Hydatid cysts in muscles: eleven cases. Rev Chir Orthop Reparatrice Appar Mot. 2005; 91:267-271.
- [8] Jia Q, Wu S, Guo J, Alimujiang A, Zheng H, Zhang J, et al. Giant muscle hydatid in lower extremity: a rare case with neurological symptoms as the first manifestation. BMC Infectious Diseases,2023;23:645.
- [9] Orhan O, Kaptan AY, Percin A, Tekpınar I, Sepetci O, Cetin VB, et al. Don't miss it: Extremity-located cyst hydatid may mimic soft tissue tumors. Jt Dis Relat Surg 2023;34(3):687-693.

- [10] Atalar MH, Cankorkmaz L, Koyluoglu G, Şalk I. Kafkas J Med Sci.2012; 2(2):74–77.
- [11] Togral G, Arıkan SM, Ekiz T, Kekec AF, Eksioglu MF. Musculoskeletal hydatid cysts resembling tumors: A report of five cases. Orthop Surg 2016;8:246-252.
- [12] Selahi O, Kuru Bektaşoglu P, Somay A, Çelikoglu E, Gurer B. Primary hydatid cyst of lumbar paravertebral muscle. Asian J Neurosurg.2022;17:661-663.

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