

Available online at www.sciencedirect.com

# **ScienceDirect**

journal homepage: www.elsevier.com/locate/radcr



# Case Report

# Primary intermuscular hydatid cyst: A common disease at unusual location: Case report<sup>☆</sup>

Bemnet Taye Gebregiorigis<sup>a,1</sup>, Yodit Abraham Yaynishet<sup>a,1,\*</sup>, Geletaw Tessema Bekele<sup>b</sup>, Bezawit Teferi Tsegaye<sup>b</sup>, Abdulmejid Suleyman Mume<sup>a</sup>, Lidet Gizachew Amha<sup>c</sup>, Fitsum Solomon Bekele<sup>d</sup>

<sup>a</sup> Department of Radiology, School of Medicine, Addis Ababa University, Addis Ababa, Ethiopia

<sup>b</sup> Department of Orthopedics, School of Medicine, Addis Ababa University, Addis Ababa,Ethiopia

<sup>c</sup> Outpatient Department of Pioneer Diagnostic Center, Addis Ababa, Ethiopia

<sup>d</sup> Department of Surgery, School of Medicine, Addis Ababa University, Addis Ababa, Ethiopia

## ARTICLE INFO

Article history: Received 9 August 2024 Revised 27 August 2024 Accepted 3 September 2024

Keywords: Hydatid disease Intermuscular hydatid Magnetic resonance imaging

# ABSTRACT

Hydatid cyst is a parasitic infection caused by the *Echinococcus granulosus* and *multilocularis* species of tapeworm. An intermuscular hydatid cyst is an unusual location for hydatid cyst disease, even in endemic regions. Therefore, intermuscular hydatid cysts might be mistaken for other benign and malignant soft tissue tumors. Magnetic resonance imaging plays a crucial role in diagnosing intermuscular hydatid cysts by showing the typical imaging characteristics of the disease. Here, we report a case of an isolated intermuscular hydatid cyst disease in a 34-year-old man who presented with left posterior thigh swelling of 5 years duration. The diagnosis was established using magnetic resonance imaging, which showed large, well-encapsulated cystic lesions containing multiple daughter cysts of varying sizes, with some cysts containing detached floating membranes. Surgery was performed, together with preoperative and postoperative albendazole treatment.

© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license

(http://creativecommons.org/licenses/by-nc-nd/4.0/)

## Introduction

Hydatid cyst is a parasitic infection caused by the Echinococcus granulosus and Echinococcus multilocularis species of tapeworm that is a common public health problem in Africa. The cyst commonly affects the liver, followed by the lungs and spleen [1]. Primary intermuscular hydatid cyst is very rare, accounting for 2%-3% of cases [2]. The reason is that muscle contractility and increased lactic acid in muscles make them an inhospitable environment for hydatid cyst development [3]. Patients with intermuscular hydatid cyst commonly present with a slowly enlarging mass, which is a nonspecific finding. Therefore, imaging plays a crucial role in diagnosis. Magnetic

<sup>\*</sup> Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

<sup>\*</sup> Corresponding author.

E-mail address: yodit.abraham@aau.edu.et (Y.A. Yaynishet).

<sup>&</sup>lt;sup>1</sup> Both Bemnet Taye Gebregiorigis and Yodit Abraham Yaynishet are the first co-authors.

https://doi.org/10.1016/j.radcr.2024.09.006

<sup>1930-0433/© 2024</sup> The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 – Thigh MRI images. T1W Coronal (A), T2W Coronal (B), T2W Axial (C) and Postcontrast Axial (D) show large well defined intermuscular cystic masses comprising of multiple different sized daughter cysts with peripheral capsular rim enhancement and few cysts that contain detached floating membranes in the peri cyst or Water Lily sign as shown with the white arrows.

resonance imaging helps in the diagnosis and extent evaluation of the disease. The typical MRI features of an intermuscular hydatid cyst include a mother cyst containing multiple small cysts and membranes inside the cyst, also known as the water lily sign [2,3].

#### **Case presentation**

We present the case of a 34-year-old male patient who came with progressively increasing left posterior thigh swelling of 5 years' duration. He underwent surgery a year after his initial presentation with incomplete removal of cysts, resulting in early recurrence. There was no evidence of histopathologic examination of the excised mass from the initial surgery. He had lived most of his life herding cattle and sheep in a rural area in Ethiopia and had a habit of consuming raw meat. When he presented to our hospital, he had a nontender, fluctuant swelling in his left posterior thigh measuring approximately 30 × 20 cm. There was no neurologic deficit.

Magnetic resonance imaging revealed well-encapsulated cystic lesions comprising multiple daughter cysts of varying sizes, involving the medial and posterior compartments of the left thigh (predominantly involving the hamstring and adductor muscles), measuring 26 (craniocaudal)  $\times$  21 (transverse)  $\times$  16 cm (anteroposterior). The lesions showed mild peripheral enhancement on the postcontrast study. No bone involvement was visualized. Based on the MRI findings, a diagnosis of intramuscular hydatid cyst of the left thigh was established, and surgical excision was planned (Fig. 1). He was put on neoadjuvant chemotherapy with albendazole.

The patient was then taken to the operation theatre. A midline posterior skin incision of 20 cm, avoiding a more lateral skin incision from the previous surgery, was used to open the thigh. There was an extensive superficial cyst layer adhered above the fascia and under the subcutaneous tissue with multiple daughter cysts. Deeper layers of the posterior compart-



Fig. 2 – Surgical specimen of extracted hydatid cyst contents.

ment muscles were extensively engulfed with multiple cysts. The sciatic nerve was also covered in layers of cysts up to its bifurcation distally. The cysts were removed with a portion of muscle fascia, with some adherent cysts rupturing during excision, consisting of clear fluid which made en bloc excision difficult. The wound, with ruptured cyst content, was washed with saline, and the wound was approximated in layers over a drainage tube. The drainage tube was removed after 72 hours. He was then discharged with postoperative albendazole (Fig. 2).

# Discussion

Hydatid disease, caused by the tapeworms *E* granulosus and less commonly *E* multilocularis, involves complex life cycles with dogs as the definitive hosts for the former and foxes or other wild canines for the latter. Humans become accidental intermediate hosts through the ingestion of eggs present in the feces of infected animals. The eggs, once consumed, release larvae that move to different organs and develop into cysts [1,4,5].

Even in endemic locations, intramuscular hydatid cysts represent approximately 1% to 5% of all hydatid cyst cases, making them an extremely unusual symptom of *E granulosus* infection [2,6]. Because of their vascular nature, these cysts are usually found in visceral organs like the liver and lungs; however, they are extremely uncommon in muscle tissue because of the high lactic acid environment and the muscles' continual physical activity, which prevent larval survival [7–9]. Global reports of instances, however, emphasize how crucial it is to rule out hydatid illness in individuals presenting with atypical muscular swellings.

There are 4 main classifications of hydatid cyst based on the morphology. Type I hydatid cysts are unilocular and welldefined with no internal architecture, showing postcontrast enhancement on CT and a hypointense rim on T2-weighted MRI images. Type II cysts contain daughter cysts with various arrangements, visible on MRI as hypointense or isointense relative to the maternal matrix with a T2 hypointense pericyst. Type III hydatid cysts are calcified and appear hyperattenuating on CT and hypointense on MRI, while Type IV cysts are complicated with common issues like rupture and superinfection, making CT and MRI crucial for their imaging [1,10].

The significance of diagnostic problems stems from the fact that intramuscular hydatid cysts might mislead to other benign and malignant soft tissue tumors [9–11]. Ultrasound and MRI are pivotal in the diagnostic process. Ultrasound may reveal the "double-layer wall," the presence of daughter cysts, and hydatid sand, which are echoes within the cyst from the movement of scolices or daughter cysts. MRI offers superior imaging capabilities, detailing cyst morphology and the relationships with surrounding tissues without invasive procedures. Furthermore, serological tests can support the diagnosis but may yield false negatives, necessitating a combination of clinical suspicion and advanced imaging, particularly in patients from endemic regions [10,12,13].

The main treatment strategy for intramuscular hydatid cysts is surgical excision, which aims to remove the cyst entirely without rupturing it to stop any spread and potential anaphylaxis. The complex nature of these cysts sometimes poses challenges in ensuring complete removal, especially when the cysts are located deep within muscle tissue or near vital structures [3,14]. The use of chemotherapy for muscu-loskeletal echinococcosis is debated, with some experts advocating for peri- or postoperative treatment with albendazole (400 mg/day), while others argue that its role should be minimal, recommending it mainly for complex cases and potentially in combination with praziquantel [15,16].

# Conclusion

This case highlights the rare occurrence of an isolated intermuscular hydatid cyst in the posterior thigh, successfully diagnosed through MRI and managed with surgical excision and albendazole therapy. The patient's history of living in a rural area with a habit of raw meat consumption signals the importance of considering hydatid cysts in differential diagnoses, even in atypical locations. The successful management of this case demonstrates the critical role of imaging in diagnosis and the effectiveness of combined surgical and medical treatment in preventing recurrence.

### **Patient consent**

Written informed consent was obtained from the patient for anonymized patient information to be published in this article.

#### REFERENCES

- Mehta P, Prakash M, Khandelwal N. Radiological manifestations of hydatid disease and its complications. Tropical Parasitol 2016;6(2):103–12.
- [2] Khasawneh RA, Mohaidat ZM, Khasawneh RA, Zoghoul SB, Henawi YM. Unusual intramuscular locations as a first presentation of hydatid cyst disease in children: a report of two cases. BMC Pediatr 2021;21(1):371.
- [3] Lakhanpal V, Badgurjar M, Saxena P. An arcane case report of primary intramuscular hydatid cyst of thigh. Int J Surg Case Rep 2021;80:105595.
- [4] Siraj S, Nijrabi MM, Hakimi A, Zaki A, Hares R. Primary intramuscular thigh hydatid cyst: a case report. Int J Surg Case Rep 2023;109:108589.
- [5] Wang Y., Boulic M., Phipps R., Plagmann M., Cunningham C., Guyot G. CMX\_ID= &SIS\_ID= &dgcid= STMJ\_AUTH\_SERV\_ PUBLISHED&utm\_acid= 229490816&utm\_campaign=STMJ\_ AUTH\_SERV\_PUBLISHED&utm\_in= DM368176&utm\_medium= email&utm\_source=AC\_ 2023. https://www.sciencedirect.com/ science/article/pii/S2352710223009816?
- [6] Sreeramulu P, Krishnaprasad, Girish gowda S. Gluteal region musculoskeletal hydatid cyst: case report and review of literature. Indian J Surg 2010;72:302–5.
- [7] Shrestha A, Shrestha AK, Deo A, Sharma GR. Intramuscular hydatid cyst of paraspinal muscle: a diagnostic challenge. Clin Case Rep 2023;11(11):e8105.
- [8] Al-Hakkak SMM. Adductor magnus muscle primary hydatid cyst rare unusual site: a case report. Int J Surg Case Rep 2018;51:379–84.
- [9] Amita K, Rajini T, Sanjay M, Abhishek M, Prashantha K. Intramuscular hydatid cyst of thigh masquerading as a soft tissue tumour diagnosed by fine needle aspiration cytology. Cell Pathol 2021;7(1):15–19.
- [10] Alshoabi SA, Alkalady AH, Almas KM, Magram AO, Algaberi AK, Alareqi AA, et al. Hydatid disease: a radiological pictorial review of a great neoplasms mimicker. Diagnostics 2023;13(6):1127.
- [11] Şimşek S, Hattapoğlu S. Intramuscular hydatid cyst in the lower extremity: report of three cases. Revista da Sociedade Brasileira de Medicina Tropical 2021;54:e0255–2021.
- [12] Pakala T, Molina M, Wu GY. Hepatic echinococcal cysts: a review. J Clin Transl Hepatol 2016;4(1):39.
- [13] Marrone G, Caruso S, Mamone G, Carollo V, Milazzo M, Gruttadauria S, et al. Multidisciplinary imaging of liver hydatidosis. World J Gastroenterol 2012;18(13):1438.
- [14] Yesildag M, Calik M, Calik S. Intramuscular hydatid cyst report of an unusual case. Chest 2015;148(4):150A.

- [15] Argy N, Abou Bacar A, Boeri C, Lohmann C, Pfaff AW, Hansmann Y, et al. Primary musculoskeletal hydatid cyst of the thigh: diagnostic and curative challenge for an unusual localization. Can J Infectious Dis Med Microbiol 2013;24(3):e99–e101.
- [16] Madhar M, Aitsoultana A, Chafik R, Elhaoury H, Saidi H, Fikry T. Primary hydatid cyst of the thigh: on seven cases. Musculoskelet Surg 2013;97:77–9.