

ROMANIAN  
NEUROSURGERY

Vol. XXXV | No. 4      December 2021

Giant, primary, hydatid cyst of the  
gluteal region. Case report

G. Iacob,  
R. Rosiu,  
Mihaela Luchian,  
G. Simion



# Giant, primary, hydatid cyst of the gluteal region. Case report

G. Iacob<sup>1</sup>, R. Rosiu<sup>2</sup>, Mihaela Luchian<sup>2</sup>, G. Simion<sup>3</sup>

<sup>1</sup> Professor of Neurosurgery "Carol Davila" University of Medicine and Pharmacy, Bucharest, ROMANIA

<sup>2</sup> Department of Neurosurgery, Emergency University Hospital, Bucharest, ROMANIA

<sup>3</sup> Department of Histopathology, Emergency University Hospital, Bucharest, ROMANIA

## ABSTRACT

**Background:** Primary hydatid disease in the muscle is extremely rare, resulting in either the spread of cysts spontaneously or after operations for hydatidosis in distant regions.

**Methods:** Report of an unusual case of primary hydatid cyst in the gluteus muscle, behaving as enlarging soft-tissue tumour, with a review of the literature.

**Results:** Magnetic resonance imaging revealed an intramuscular cyst in the gluteus muscle; and no cyst existed in any other location. The cyst was excised totally, and the diagnosis of muscular cystic hydatidosis was confirmed by histopathologic examination. In follow-up of two years after the operation, there has been no recurrence in either patient.

**Conclusion:** The involvement of the gluteus maximus muscle without the evidence of hepatic or pulmonary disease is rare, characterized by slow development, but a major local extension must be considered in the differential diagnosis of a cystic mass with well-defined margins in the extremities of individuals from endemic regions. Physical examination, serology results and radiological findings should be interpreted with care, especially in hydatid cyst endemic countries. Surgery is still the treatment of choice, with total cyst excision.

## INTRODUCTION

Human echinococcosis is a zoonotic disease, caused by ingestion of ova of the cestode *Echinococcus granulosus* (Eg) and formation of the larval stage in different organs and tissues. Cystic echinococcosis (CE) has become an important burden on human population in endemic area and no endemic countries, by infected people migration and livestock exchange (1). CE is responsible for the loss of 1–3 million disability-adjusted life years per annum and severe economic losses to the livestock industry probably amounts to US\$ 2 billion (2). Romania was listed in 1995 among the countries with the highest prevalence of CE worldwide. Nevertheless, latest study Heracles FP7 EU Project oct 2013 - sept 2018 confirm that CE has a prevalence of 0.41%000 in

## Keywords

cystic echinococcosis,  
primary musculoskeletal  
location hydatid cyst,  
gluteal region hydatid cyst



Corresponding author:  
**Iacob Gabriel**

"Carol Davila" University of Medicine  
and Pharmacy Romania

gbr\_cb@yahoo.com

**Copyright and usage.** This is an Open Access article, distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives License (<https://creativecommons.org/licenses/by-nc-nd/4.0/>) which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is unaltered and is properly cited.

The written permission of the Romanian Society of Neurosurgery must be obtained for commercial re-use or in order to create a derivative work.

ISSN online 2344-4959  
© Romanian Society of  
Neurosurgery



First published  
December 2021 by  
London Academic Publishing  
[www.lapub.co.uk](http://www.lapub.co.uk)

Romania (3). Molecular characterization belonged to the G1-G3 complex (*E. granulosus*), which contains the most widespread and infective strains of the parasite, also a non-G1-G3 genotype of *E. granulosus*, likelihood, a G7 genotype, which is often found in pigs and dogs in most countries of eastern and south-eastern Europe (4). Primary musculoskeletal location hydatid cyst are rare: only 2 to 3%, possibly most affected sites are the neck, thigh and the paravertebral region, the psoas muscle, the thoracic wall muscles, sartorius, and quadriceps muscles. On the gluteal region hydatid cyst are very rare (5-19). Clinical, diagnosis and therapeutic considerations are discussed.

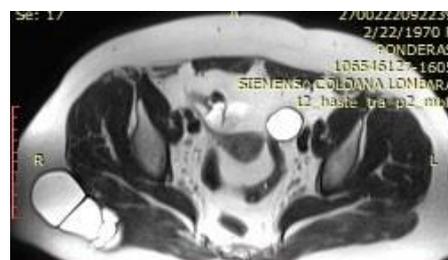
#### CASE PRESENTATION

A 53 years old woman, seamstress in town, presents to his primary care provider with a 6 months history of lower back pain, stiffness. The pain was intermittent, improves after rest without medication, worse with activity and in the morning when changing the position of the body from supine position to orthostatism, at the effort of coughing, sneezing, defecation, sometimes with right irradiation on the back of the buttocks, thighs, left calves to the heels; but she was able to lean forward. Also, the patient has been reporting for about 4 months a painful mass installed insidiously at the right gluteal region, firm consistency, normal skin, which has progressively increased in the last 2 weeks before admission. She does not have any known chronic medical conditions, also she has never smoked, but she does drink an occasional glass of wine with dinner. No significant family history of disease is reported. She has not noted any other masses on his body with self-examination, no history of rash, fever, malaise, anorexia, no weight loss, neither she nor other family members came in contact with pets.

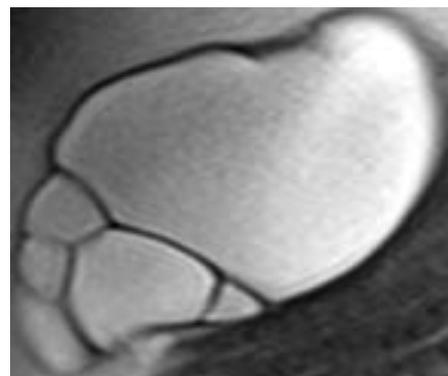
Upon physical examination, this patient has a blood pressure of 131/75 mm Hg and a heart rate of 65 beats/min. His respiratory rate is 7 breaths/min and his temperature is normal at 98°F (36.7°C). His cardiovascular and respiratory findings are normal; specifically, no murmurs or rubs are detected. The patient has no photophobia, eye redness, or decreased visual acuity. Upon examination of the back, flexion of the lumbar spine is possible, without limited range of motion with rotation and lateral flexion at the lumbar spine. The right buttock is

painful to the touch. On clinical examination we found a painful mass in the posterior lateral region of the upper third of the right thigh, with normal looking skin on top, of firm consistency, 30 cm long axis, 18 transversal axis, fixed to muscle with no evidence of local inflammation. No motor, sensitive deficit or abnormal hip mobility were encountered. The remainder of the physical examination is within normal limits.

The biological assessment was unremarkable: it reveals a white blood cell count of  $4.6 \times 10^3$  cells/ $\mu$ L (reference range,  $4.5-11 \times 10^3$  cells/ $\mu$ L), a hemoglobin level of 13.7 g/dL (reference range, 13.5-17.5 g/dL), a hematocrit of 43% (reference range, 41% to 50%), eosinophilic count of 2.8 (reference range, 0.8-8.1 %), a platelet count of  $120 \times 10^3$  cells/ $\mu$ L (reference range,  $150-450 \times 10^3$  cells/ $\mu$ L); electrolyte values are within normal limits, only the erythrocyte sedimentation rate was 35 mm/hr (reference range, < 10 mm/h). The diagnosis was made by lumbar and pelvic magnetic resonance (MRI) which brought into evidence of lumbar disc protrusions at L2-L5, without root conflict, lumbar vertebral marginal osteophytes; multiple cystic formations, single or partitioned cysts, on the posterior and lateral surfaces of the root of the right thigh and the gluteal region, mainly occupying the gluteus maximus and sub- aponeurotic fat. They had a thin wall in enhanced T2 hypo-intense after injection of gadolinium with variable signal fluid content, the largest measured 13 x 12 cm (Fig.1).



A.



B.



C.

**Figure 1.** Primary, multiple hydatid cysts of the right gluteal region MRI, enhanced T2 hypo-intense after injection of gadolinium: A axial section, B & C coronal sections.

A total resection under general anesthesia was performed by an extended posterior lateral approach of the thigh. Intraoperative finding highlighted a very thick hydatid pericyst with tight adhesion to the gluteus maximus muscle bundles, composed of inflammatory tissue, the hydatid exocyst and the hydatid endocyst with irregularly shaped daughter cysts of different sizes and hydatid fluid. Despite our anatomical exposure and delicate dissection of the cyst, a minimal breach with hydatid fluid exteriorization was found. The cyst content was appropriately evacuated and the surgical field was soaked with hypertonic saline (Figure 2).



A

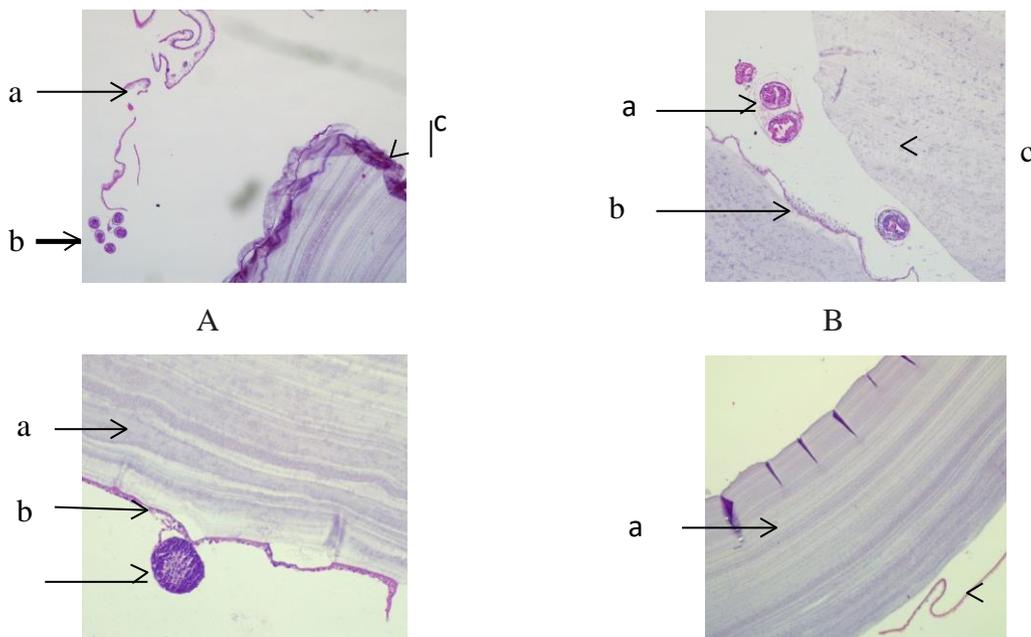
B

C

D

E

**Figure 2.** Intraoperative pictures: A. General aspect of the hydatid cyst during dissection, B&C several irregularly shaped daughter cysts of different sizes, D the hydatid exocyst, E thick hydatid pericyst with tight adhesion to the gluteus maximus muscle bundles completely dissected.



**Figure 3.** The histopathologic examination in our case:

A H.E col. obj. 20 X 10, a. the inner germinal layer, b. protoscolices, c. the middle-laminated membrane;

B H.E col. obj. 20 X 20, a. protoscolices, b. the inner germinal layer, c. the middle-laminated membrane;

C H.E col. obj. 20 X 20, a. the middle-laminated membrane, b. the inner germinal layer, c. daughter vesicle, attached by a pedicle to the germinal layer;

D H.E col. obj. 20 X 10, a. the middle-laminated membrane, b. the inner germinal layer.

The serology was positive for E.g., and the extension assessment: cerebral MRI, pulmonary and abdominal CT were normal.

The histopathologic examination (Figure 3) confirmed the diagnosis of hydatid cyst.

## RESULTS

The post-operative evolution was uneventful. The patient had completed a chemotherapy with an anti-helminthic agent Albendazole (Zentel) 15 mg / kg /day, for the next 3 months postoperatively; 6 months after, there was no evidence of recurrence or complications.

## DISCUSSION

Hydatid cysts result from infection by the *Echinococcus* tapeworm species and can result in cyst formation anywhere in the body. Typically, the primary cyst locations hydatid are the liver and lungs. This can be explained by the significant vascularization of these structures that can be filters. Rarely, the hydatid cyst occurs at the musculoskeletal tissues, with slow and relatively asymptomatic evolution, without hepatic involvement, as in our case. Primary hydatid disease in the muscle is extremely rare, resulting either from the spread of cysts spontaneously or after operations for hydatidosis in distant regions. The most reported sites are the neck, thigh and the paravertebral region, very rare at the gluteal region (5-7)(10-20).

In general recognition of hydatid disease is possible, sometimes even when serologic tests had been non-conclusive (9). In endemic areas the etiological diagnosis of the cyst can be difficult in some cases. Accurate diagnosis could be made using pelvic radiograph, the USG exam, the first-line radiological examination - in our case could have highlighted multiple hydatid cysts, within the stage 2 - septated cystic lesion and stage 3 - cystic lesion with the daughter lesion, see the Gharbi ultrasound classification (9). CT scan rarely shows typical characteristics of the hydatid cyst, possible wall calcifications, other hydatid cyst localizations and MRI - the reference examination as in our case is useful, especially in the case of giant cysts, assessing the size of the mass, relationships with neighboring

tissues and nervous and vascular structures (8), recognizing complications (e.g., rupture, infection of cysts)(6), correlated to macroscopic and microscopic pathology (20). Also MRI should be considered for differential diagnosis between slow-growing tumors, hematomas, myositis, and abscesses (19). Total cyst excision is the primary treatment method, avoiding in suspected hydatid cysts biopsy and aspiration to prevent rupture and spread (8)(14)(20). In case of the hydatid cyst rupture hypertonic saline irrigation of the surgical field is the best method to prevent recurrences by antigens spreads to the muscles, inflammatory reaction complicated by a secondary bacterial infection (6)(20). As a particularity of the presented case there was a concomitant lesion generated by lumbar protrusions with right root irritation phenomena and buttock hydatid cyst.

## CONCLUSIONS

The involvement of the gluteus maximus muscle without the evidence of hepatic or pulmonary disease is quite rare, characterized by a slow development, but a major local extension must be considered in the differential diagnosis of a cystic mass with well-defined margins in the extremities of individuals from endemic regions. Physical examination, serology results and radiological findings should be interpreted with care, especially in endemic areas. Surgery is still the treatment of choice, with total cyst excision, followed by chemotherapy with an anti-helminthic agent.

## ABBREVIATIONS

*Echinococcus granulosus* (Eg)  
cystic echinococcosis (CE)  
ultrasonography (USG)  
tomograph computer (CT)  
magnetic nuclear resonance (MRI)

## CONFLICTS OF INTEREST

There are no conflicts of interest.

## REFERENCES

1. Mamuti W., Yamasaki H., et al. - Usefulness of hydatid cyst fluid of *Echinococcus granulosus* developed in mice

- with secondary infection for serodiagnosis of cystic Echinococcosis in humans, *Clinical and Diagnostic Laboratory Immunology* 2002, 9, 3, 573–576.
2. Torgerson P.R., Craig P. - "Updated global burden of cystic and alveolar echinococcosis," Report of the WHO Informal Working Group on Cystic and Alveolar Echinococcosis Surveillance, Prevention and Control, with the Participation of the Food and Agriculture Organization of the United Nations and the World Organisation for Animal Health, Department of Control of Neglected Tropical Diseases WHO, Geneva, Switzerland, 2011.
  3. Cretu C.M., Banu T. et al. - Situation of cystic echinococcosis in humans and animals: a real alarming concern. Heracles FP7 EU Project oct 2013- sept 2018.
  4. Piccoli L., Bazzocchi C. et al. - Molecular characterization of *Echinococcus granulosus* in southeastern Romania: evidence of G1–G3 and G6–G10 complexes in humans, *Clinical Microbiology and Infection* 2013, 19(6):578-582.
  5. Lamine A, Fikry T, et al. - Primary hydatidosis of the peripheral muscles: 7 case reports. *Acta Orthop Belg.* 1993;59(2):184-188.
  6. Aloui.Y, Saadana J, et al.- Kyste hydatique géant de la région glutéale : A propos d'un cas, STPI & SPILF Congress 9-11.09.2021, [www.infectiologie.org.tn](http://www.infectiologie.org.tn).
  7. Jerbi O.S, Abid F. et al. - Primary hydatid disease of the thigh: a rare location. *Orthop Traumatol Surg Res.* 2010;96(1):90-93.
  8. von Sinner WN. - New diagnostic signs in hydatid disease; radiography, ultrasound, CT and MRI correlated to pathology. *Eur J. Radiol.* 1991; 12 (2): 150-159.
  9. Stojkovic M, Rosenberger K, et al. - Diagnosing and staging of cystic echinococcosis: how do CT and MRI perform in comparison to ultrasound. *PLoS Negl Too Say,* 2012; 6 (10): e1880.
  10. Haque F., Harris S.H. et al. - Primary hydatidosis of gluteus maximus, *J Postgrad Med* 2006;52(4):300-301.
  11. Ates M., Karakaplan M. - Hydatid Cyst in the Biceps and Gluteus Muscles: Case Report, *Surgical Infections,* 2007, 8.4.20.
  12. Kaplan M., Aktoz T. et al. - Atypical Renal and Gluteal Hydatid Cysts: Report of Two Cases, *Firat Tıp. Dergisi* 2009;14(4): 297-299.
  13. Eryilmaz MA, Eroglu C, et al. - Gluteal hydatid cyst: Case report. *Türkiye Klinikleri J Med Sci.* 2010;30:1380–1383.
  14. Mushtaque M., Mohammad F. Mir et al. - Solitary subcutaneous gluteal hydatid cyst: a case report, *Eastern Journal of Medicine* 2010, 15, 2010, 76-79.
  15. Zeren S, Kesici U, et al. - Gluteal Hydatid Cyst: Report of a Case. *Acta Med Iran,* 2015;53(6):389–391.
  16. Aksakal N., Kement M., et al - Unusually located primary hydatid cysts, *Ulus Cerrahi Derg.* 2016; 32(2): 130–133.
  17. Hona BN, Emami Y.F. - Gluteal Hydatid Cyst: Report of a Case from Iran, *Iran J Parasitol* 2017;12(2):305–308.
  18. Seyedsadeghi M. Ghobadi J. t al. - Gluteal Hydatid Cyst: A Case Report, *Iran J Parasitol* 2019;14(3):487- 491.
  19. 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).
  20. Rabhi S., Saadana J. et al. - Primary giant hydatid cysts of the thigh and the gluteal region: a case report. *Pan Afr Med J* 2021, 5, 39:15, eCollection 2021.