Case Report

DOI: 10.4274/haseki.galenos.2023.9518
Med Bull Haseki 2023;61:379-383

Giant Hydatid Cyst Originating from Psoas Muscle Extending to the Iliac Bone, Inguinal, and Femoral Canals: A Case Report and Current Literature Review

Kagan Gokce*, Demet Dogan**, Fatma Mehlika Ilhan***, Ahmet Midi****

*Okan University Faculty of Medicine, Department of General Surgery, Surgical Oncology Unit, Istanbul, Turkey

**Okan University Faculty of Medicine, Department of Radiology, Istanbul, Turkey

***Okan University Faculty of Medicine, 4th Grade Student, Istanbul, Turkey

****Bahcesehir University Faculty of Medicine, Department of Pathology, Istanbul, Turkey

Abstract

Retroperitoneal hydatid cysts are extremely rare and difficult to distinguish from other intra-abdominal pathologies, such as synovial sarcoma. In this study, we present a rare case of a complicated retroperitoneal hydatid cyst originating from the psoas muscle without any other focus. A 59-year-old male patient presented to the outpatient clinic with complaints of constipation and a feeling of gradually increasing swelling in the left lower quadrant of the abdomen and left groin area, progressing toward the left leg. In the examinations performed, a multiloculated giant hydatid cyst that filled the left pelvis in the retroperitoneal region, deviated the intra-abdominal organs to the right side, and extended to the left femoral and inguinal canals was detected. The patient underwent surgical excision and was followed up during the postoperative period. No evidence of recurrence was found at the patient's 3rd and 6th month follow-ups. Primary muscle hydatid cysts necessitate a distinct approach to treatment and management when compared with hydatid cysts in other bodily organs. While recurrences remain a potential concern after resection, it is noteworthy that the window for the formation of fertile cysts typically spans up to 10 months. Thus, it is advisable to conduct regular postoperative follow-up examinations during the first year following surgery to ensure comprehensive monitoring and care.

Keywords: Echinococcosis, cysts, psoas muscles, pelvis, retroperiton, bone

Introduction

Hydatid cyst is a parasitosis caused by Echinococcus granulosus helminth. It is endemic in many countries of the world, such as New Zealand, Africa, and Turkey, and maintains its importance for public health. It is a very difficult disease to diagnose and treat (1). Diagnosis is made by radiological imaging appropriate to the anatomical region where the disease is believed to be present and by performing an indirect hemagglutination test (IHA). Surgery is the mainstay of treatment. When a hydatid cyst is not treated, it can lead to complications such as anaphylactic shock, infection of existing cysts, and compression symptoms (2). In terms of location, the liver comes first with a rate of 50-70%, followed by the lung with a rate of 11-17%. The rate of localization in muscle and subcutaneous tissues is between 0.5 and 4.7%. In cases of hydatid cysts with retroperitoneal localization and muscle origin, i.v. contrast-enhanced magnetic resonance imaging (MRI) and computed tomography (CT) imaging are valuable in terms of differential diagnosis. In cases of hydatid cysts with atypical localization, the liver and lung should be investigated in terms of primary focus (3,4). In this article, we aim to underline the atypical localization of a hydatid cyst by presenting a case of a giant multiloculated cyst originating from the psoas muscle, located retroperitoneally, eroding the left iliac bone, and extending to the gluteal muscles.

Case Presentation

A 59-year-old male patient was admitted to the outpatient clinic with complaints of pain in the left lower

Address for Correspondence: Kagan Gokce, Okan University Faculty of Medicine, Department of General Surgery, Surgical Oncology Unit, Istanbul, Turkey
Phone: +90 532 581 05 90 E-mail: kagangokce2023@gmail.com ORCID: orcid.org/0000-0003-4712-0512
Received: 13.09.2023 **Accepted:** 24.11.2023

quadrant of the abdomen, abdominal fullness, and constipation. He also complained of swelling and tingling in his left leg. In his history, it was learned that the swelling in the abdomen gradually increased and progressed from the left inguinal region to the leg. During the examination, a hard-consistent mass lesion starting from the upper flank region of the left abdomen and extending to the midline was palpated. On palpation, the mass lesion was pushing up the anterior abdominal wall and was immediately felt under the skin. The mass lesion was progressing toward the pelvis. The mass filled the femoral and inguinal canals on the lower side and progressed from the thigh region to the left lower extremity.

In the abdominal ultrasonography (USG), a multiloculated cystic lesion progressed toward the femoral and inguinal canals and filled both canals. In the left lower quadrant of the abdomen, anterior to the iliac muscle, a cystic lesion with a size of approximately 77x90 mm, with macrolobule contours, containing multiloculated cystic foci, and with no significant blood flow in Doppler USG, with dense content, was detected (Figure 1). Initially, the pre-diagnosis of retroperitoneal sarcoma and hydatid cyst was considered, and an i.v. contrast-enhanced CT of the entire abdomen and pelvis was performed (Figure 2A). In the MRI examination, hypointense on T1-weighted images, hyperintense on T2-A images, and non-contrast-enhancing, multilocus cystic lesions on post-contrast images were observed (Figure 2B). In the results, hydatid disease was thought to be more prominent, and an IHA test was performed. The IHA titer was found to be 1/2560 positive in blood tests, and surgery was decided.

Considering the pelvic extent of the mass, the transperitoneal approach was preferred for this type of surgery. During laparotomy, it was observed that the giant mass started from the retroperitoneum and pushed the sigmoid colon and rectosigmoid junction up to the anterior abdominal wall, causing partial obstruction. In the dissection, it was observed that the hydatid cyst lesions originated from the psoas muscle. The psoas muscle was dissected up to the paravertebral area, and the mass lesion was separated from the psoas muscle and paravertebral area. The mass filled the pelvis on the iliac bone and entered the gluteal muscle group by eroding the bone from the posterior. The mass lesion was separated by posterior dissection and removed from the gluteal muscle group. Multiple cystic lesions were detected in the inguinal and femoral canals of the patient with a left inguinal hernia. Cystic lesions were excised, preserving the femoral vein, artery, and nerve. The inguinal canal was dissected,

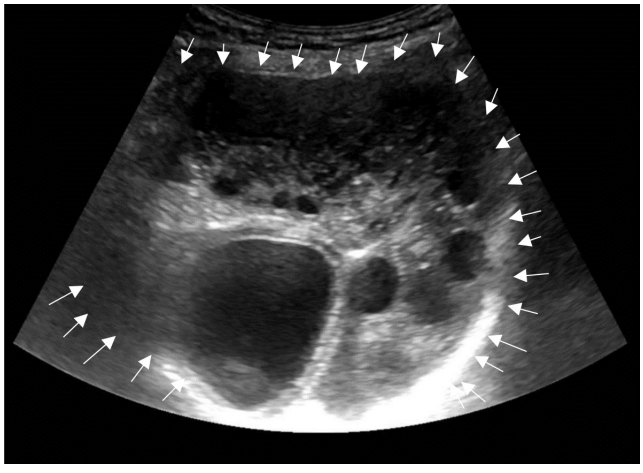

Figure 1. Multiloculated cystic lesion on USG (little white arrows)
USG: Ultrasonography

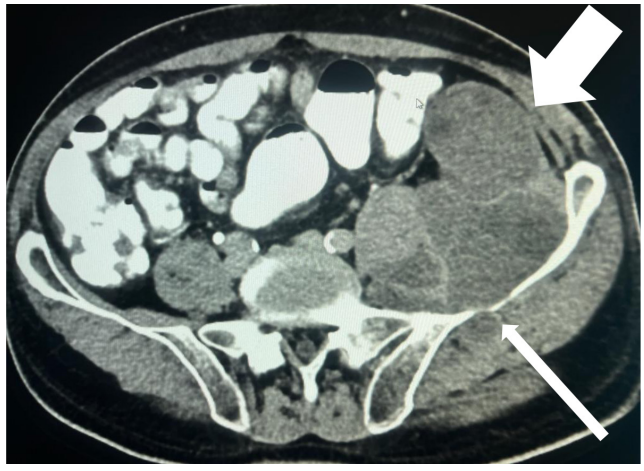

Figure 2A. Septal cystic mass (thick white arrow) that erodes the bone structure within the iliacus muscle (thin white arrow) on axial CT
CT: Computed tomography

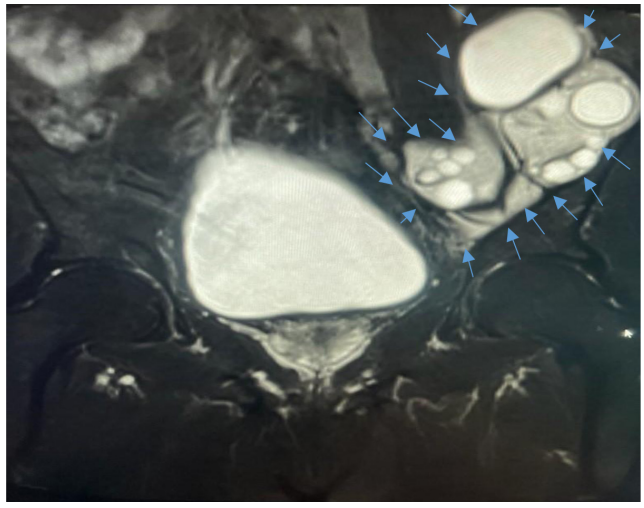

Figure 2B. Multiloculated septal cystic mass in the iliac muscle on coronal section T2-A MRI (little blue arrows)
MRI: Magnetic resonance imaging

the cord and vascular structures were preserved, and cystic lesions were excised from the inguinal canal into the abdomen. Because of the diagnosis of a hydatid cyst, each cystic lesion was filled with hypertonic 3% serum saline, and after waiting for 5 minutes, excision was initiated. All cystic masses were resected without perforation. The operation area was washed with 5 L of body-temperature isotonic fluid, and a drain was placed in the retroperitoneal area. The left and sigmoid colons were brought to their anatomical positions, and the peritoneum was repaired to ensure retroperitoneal integrity and isolation.

The resection specimen was examined under a microscope in the pathology laboratory using hematoxylin/eosin and Periodic acid-Schiff (PAS) dye. A diagnosis of a hydatid cyst originating from Echinococcus granulosus was made. Histopathological examination revealed that the cysts consisted of a cellular germinal membrane, a thick PAS-positive cell-free homogeneous pink-colored laminated layer, and an outer adventitial layer (Figures 3A and 3B). Germinative vesicles were observed in the cysts. An increase in connective tissue and inflammatory cells was observed around the membrane of some cysts (Figure 3C).

No lesion was detected in the early postoperative control abdominal CT (Figure 4). Benzimidazole group Albendazole 400 mg 2x1 treatment was started in the patient in the postoperative period. It was observed that the swelling in the left leg of the patient regressed, and the tingling complaint of the patient disappeared in the first week postoperatively. The patient did not have any constipation complaints anymore.

In the 3rd postoperative month, the entire abdomen of the patient was checked with USG, and no cystic lesion was found. In the 6th postoperative month, the patient

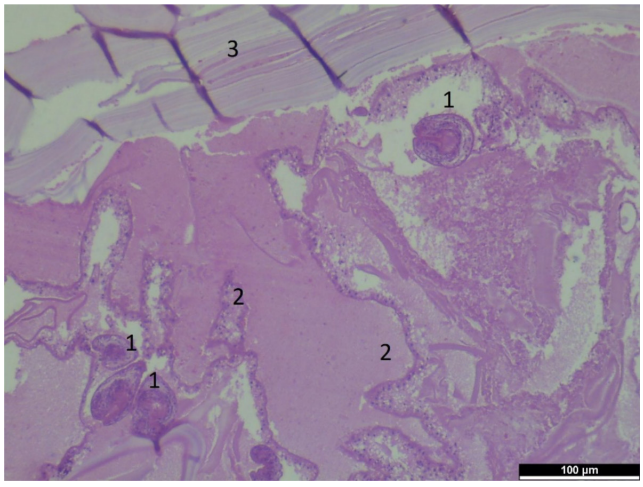

Figure 3A. Microscopic image of hydatid cyst, protoscolices (1), germinal layer (2) followed by laminated layer (3) (H&E-Original magnification ×40)

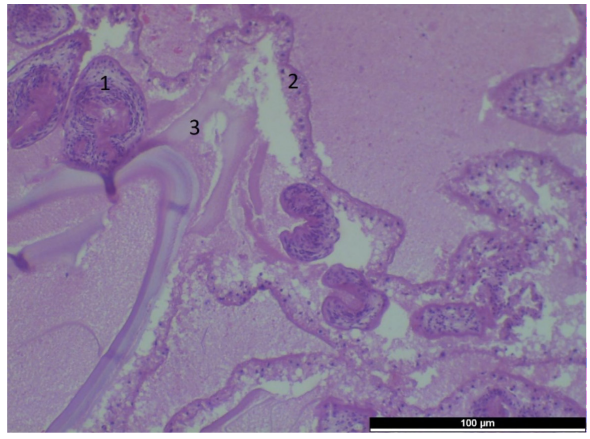

Figure 3B. Protoscolices (1), germinal layer (2), laminated layer (3) (H&E-Original magnification ×100)

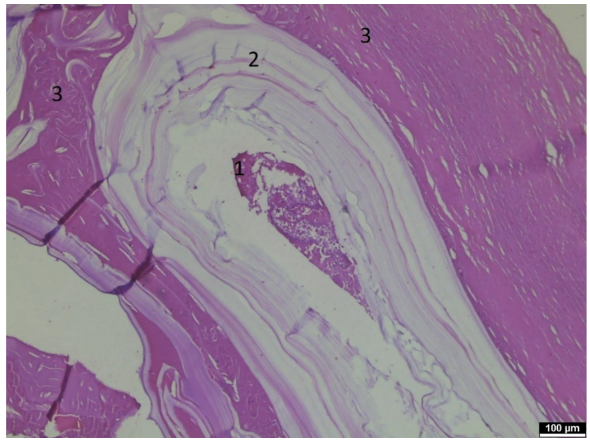

Figure 3C. Germinal layer (1), laminated layer (2), adventitia (1) (H&E-Original magnification ×40)

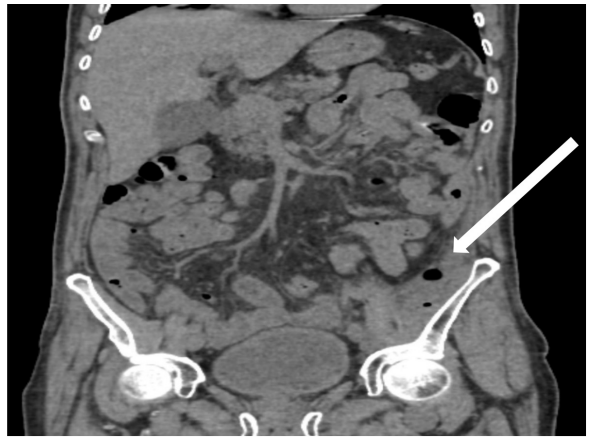

Figure 4. No postoperative residue was observed in the coronal section CT examination (white arrow)
CT: Computed tomography

was evaluated with an ultrasound of the entire abdomen, i.v. contrast-enhanced abdominal and pelvic CT, and chest X-ray examinations. No cystic lesions were found in the examinations. Because of the high risk of recurrence in the first 10 months, clinical follow-up was planned for the patient every 3 months for the first year.

Discussion

Hydatid cysts are usually located first in the liver, second in the lungs, and rarely in the brain (5). Muscle involvement in hydatid cysts is rare and has rarely been reported in the literature as case reports. It is thought that the reason why muscle involvement of hydatid cysts is rare is due to the amount of lactic acid in the muscle and the contraction of the muscles, preventing the growth of the cysts in the striated muscles (6). However, primary hydatid cysts of muscle origin are known to involve the pectoralis major, sartorius, quadriceps, and gluteus muscles (7-9). Cases of hydatid cysts in the retroperitoneum often have another primary focus. Therefore, when a hydatid cyst is detected in the retroperitoneum, it is useful to seek another focus with radiological examinations showing liver, lung, and brain tissue. Cases of primary hydatid cysts originating from the retroperitoneal muscles are even rarer (10,11). Such lesions may progress in the inguinal canal and mimic an irreducible inguinal hernia (12). In the case we are presenting, another focus was investigated preoperatively with cranial MRI and whole abdomen, pelvic, and thorax CT scans, and no focus was found other than the retroperitoneum. In this study, we observed that the hydatid cyst filled the inguinal canal and could not be reduced from the inguinal canal in the testicular examination. At the same time, the situation was the same in the femoral canal.

The components of the hydatid cyst are formed by an inner, thin germinal layer and an outer, thick laminated layer. The outermost layer is the adventitia. The adventitia layer forms as a fibroinflammatory response and attempts to limit the cyst. Degeneration and calcification can be observed in long-lasting cases. Cysts filled with protoscolex may be observed, indicating that the cyst attached to the germinal layer is fertile. In this study, the hydatid cyst showed the characteristics of a fertile cyst, and calcification was observed in some cysts in the resection material. While the probability of fertilization of lung hydatid cysts is 55%, it is 45% for liver hydatid cysts (13).

Hydatid cyst treatment usually includes surgery, medication, or a combination of both. For hydatid cysts with muscle involvement, the treatment process may differ from cysts in other organs. The treatment of hydatid cysts with muscle involvement may vary depending on the size and location of the cyst and the general health status of the patient. In cases where surgical intervention is not appropriate or to prevent postoperative recurrence, antiparasitic drugs such as albendazole or mebendazole are used. These medications can help shrink the cyst by preventing the parasite from growing and reproducing. Muscle cysts that are small and suitable for placement can be surgically removed. The operation can be performed with open or laparoscopic surgery, depending on the region and size of the cyst (14,15).

In this study, midline incision and transperitoneal approaches were preferred because of the localization of hydatid cysts. Albendazole treatment was started from the moment the patient was diagnosed in the pre-operative period, and it continued to be used in the postoperative period.

In conclusion, primary muscle hydatid cysts present a unique clinical challenge, necessitating a tailored approach to treatment and management compared with hydatid cysts in other organs. Although the risk of recurrence exists following surgical resection, it is important to emphasize that the fertile cyst formation period typically spans up to 10 months. Consequently, conducting regular postoperative follow-up examinations within the first year is paramount to ensuring diligent monitoring and comprehensive care for patients with primary muscle hydatid cysts.

Ethics

Informed Consent: Informed consent was obtained.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Concept: K.G., D.D., A.M., Design: K.G., D.D., A.M., Data Collection or Processing: K.G., D.D., A.M., F.M.I., Literature Search: K.G., Writing: K.G., D.D., A.M., F.M.I.

Conflict of Interest: The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article. All authors have read and confirmed that they meet the ICMJE criteria for authorship.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Bhalla VP, Paul S, Klar E. Hydatid disease of the liver. Visc Med 2023;39:112-20.
2. Khalili N, Iranpour P, Khalili N, Haseli S. Hydatid disease: a pictorial review of uncommon locations. Iran J Med Sci 2023;48:118-29.
3. Alshoabi SA, Alkalady AH, Almas KM, et al. Hydatid disease: a radiological pictorial review of a great neoplasms mimicker. Diagnostics (Basel) 2023;13:1127.

4. Laasri K, Naggar A, Bahlouli N, et al. Osseous hydatid disease: A mimic of other skeletal pathologies. Radiol Case Rep 2023;18:3145-51.
5. Dietrich CF, Douira-Khomsi W, Gharbi H, et al. Cystic echinococcosis, review and illustration of non-hepatic manifestations. Med Ultrason 2020;22:319-24.
6. Nath K, Prabhakar G, Nagar RC. Primary hydatid cyst of neck muscles. Indian J Pediatr 2002;69:997-8.
7. 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.
8. Shrestha A, Shrestha AK, Deo A, Sharma GR. Intramuscular hydatid cyst of paraspinal muscle: A diagnostic challenge. Clin Case Rep 2023;11:e8105.
9. Sachar S, Goyal S, Sangwan S. Uncommon locations and presentations of hydatid cyst. Ann Med Health Sci Res 2014;4:447-52.
10. Tekin R, Avci A, Tekin RC, et al. Hydatid cysts in muscles: clinical manifestations, diagnosis, and management of this atypical presentation. Rev Soc Bras Med Trop 2015;48:594-8.
11. Hatipoglu AR, Coskun I, Karakaya K, Ibis C. Retroperitoneal localization of hydatid cyst disease. Hepatogastroenterology 2001;48:1037-9.
12. Özkardeş AB, Tokaç M, Yetişir F, et al. Retroperitoneal hydatid cyst simulating irreducible inguinal hernia: case report. J Clin Anal Med 2014;5:243-5.
13. Beigh AB, Darzi MM, Bashir S, et al. Gross and histopathological alterations associated with cystic echinococcosis in small ruminants. J Parasit Dis 2017;41:1028-33.
14. Berto CG, Liou P, Coyle CM, Emond JC. Surgical management of cystic echinococcosis of the liver. Curr Opin Infect Dis 2023;36:348-52.
15. Parlak E, Kerget F, Demirdal T, et al. The epidemiology, clinical manifestations, radiology, microbiology, treatment, and prognosis of echinococcosis: results of NENEHATUN study. Vector Borne Zoonotic Dis 2021;21:948-54.