# A Rare Case of Hydatid Cyst in the Inguinal Area: A Case Report

A. Dalili (MD)<sup>1</sup>, A. Ebrahimi (MSc)<sup>2</sup>, A. Keshavarz (MSc)<sup>3</sup>, H. Khosravi (MSc)<sup>4</sup>, M. Sadeghi (MSc)<sup>\*5</sup>

1.Surgical Oncology Research Center, Mashhad University of Medical Sciences, Mashhad, I.R.Iran

2.Department of Surgical Technology, School of Paramedical Sciences, Bushehr University of Medical Sciences, Bushehr, I.R.Iran

3. School of Allied Medical Sciences, Mazandaran University of Medical Sciences, Sari, I.R. Iran

4. Faculty of Nursing & Midwifery, Mashhad University of Medical Sciences, Mashhad, I.R. Iran

5. Montaserie Organ Transplantation Hospital, Mashhad University of Medical Sciences, Mashhad, I.R.Iran

## J Babol Univ Med Sci; 22; 2020; PP: 236-240

Received: Nov 9th 2019, Revised: Jan 7th 2020, Accepted: Feb 10th 2020.

## ABSTRACT

**BACKGROUND AND OBJECTIVE:** Hydatid disease is a common health problem in developing countries. The liver and lungs are the most common organs affected by this infection. Hydatid cyst is very rare in the inguinal area. The aim of this case report was to introduce a patient with multiple hydatid cysts in the right inguinal area who developed symptoms of inguinal hernia.

**CASE REPORT:** The patient is a 92-year-old man who was referred to Imam Reza Hospital in Mashhad due to pain, swelling and redness in the right inguinal area. The patient's blood tests showed an increase in neutrophils, and a decrease in eosinophils, basophil and monocyte. Ultrasound examinations of the patient revealed right inguinal hernia and hydatid cysts in the liver, abdomen and pelvic cavity. Finally, the patient was operated with a diagnosis of incarcerated inguinal hernia. During the operation, multiple cysts were observed in the inguinal area; the cysts were removed and sent to pathology. Pathological findings confirmed the presence of hydatid cyst. After 2 days, the patient was discharged in relatively good general condition and with albendazole (400 mg) twice daily for 6 months. At the end of the treatment period, no signs of recurrence of hydatid cyst were observed in the patient.

**CONCLUSION:** According to the cases reported in patients diagnosed with hydatid cyst with inguinal pain, the possibility of migration or hernia of hydatid cyst should be considered, because knowing its presence is useful for preoperative planning and reduction of complications.

KEY WORDS: Hydatid Cyst, Inguinal Canal, Echinococcus.

#### Please cite this article as follows:

Dalili A, Ebrahimi A, Keshavarz A, Khosravi H, Sadeghi M. A Rare Case of Hydatid Cyst in the Inguinal Area: A Case Report. J Babol Univ Med Sci. 2020; 22: 236-40.

## Introduction

Hydatid Disease (HD) is a parasitic infection mainly caused by Echinococcus granulosus, which is endemic in different parts of the world (1). Hydatid disease is found in almost every part of the body, however, it often affects the liver and has a variety of complications. Radiological and serological findings usually diagnose the disease (2). The most common sites are liver (63%), and lungs (25%) followed by muscles (5%), bones (3%), kidneys (2%), spleen (1%) and others (1%) (3). The presence of hydatid cysts in the inguinal canal is very rare (4).

In one study, iliopsoas muscle hydatid cyst was reported in a 35-year-old man who had symptoms such as pain and swelling in the right groin (5). In another study, mesenteric hydatid cyst was presented as inguinal hernia in a 5-year-old boy (6). In another study, a rare case of inguinal hydatid cyst was reported in a 54-yearold man with symptoms of cystic swelling in the right inguinal area (7). Another report also examined a 45year-old man who had a history of pain in the right inguinal hernia for 2 years and was eventually diagnosed with hydatid cyst in the right inguinal region (8).

Consumption of contaminated food by humans leads to spawning in the gastrointestinal tract. The larvae are transferred to the liver through circulation of the portal. The liver is the first filter involved in trapping larvae. Following that, the lungs act as a second filter. Some larvae escape these filters and nest in other organs (9). Hydatid cysts may cause rare complications such as migration, invasion of the trans-diaphragmatic abdominal wall, and hernia from the anatomical cavities of the body (10). Primary treatment of hydatid cyst is surgery. In cases of diffuse cyst or impossibility of surgery, drug treatment with mebendazole and albendazole is performed. Rupture of the parent cyst increases the likelihood of recurrence and may also cause anaphylactic shock in the patient (11, 12).

How echinococcosis larvae reach the inguinal region is not available. However, considering the anatomy of the groin and the abundant lymphatic tissues in this area, there is a hypothesis about the transmission of larvae through the lymph flow. Currently, the most effective treatment for hydatid disease in soft tissue is surgery. The main purpose of surgery is to prevent complications such as pressure on surrounding structures, infection and rupture of the cyst. With complete removal of the cyst (cystectomy) along with removal of the surrounding adventitia, all parasitic elements are removed without leaking the contents of the cyst (13, 14). In organs with cystic edema, especially in endemic areas, the presence of hydatid cyst should be considered as a differential diagnosis. Since preoperative diagnosis of hydatid cyst is very important to prevent anaphylactic shock and local recurrence, it is difficult to diagnose hydatid cyst in unusual places. Therefore, familiarity with the uncommon manifestations of this infection is necessary for timely diagnosis and treatment and reduction of complications. Therefore, a rare case of inguinal hydatid cyst has been reported.

#### **Case Report**

The patient is a 92-year-old man who referred to Imam Reza Hospital in Mashhad due to pain, swelling and redness of the right inguinal area. The patient had been suffering from pain in the right inguinal area for 6 months. He underwent an unsuccessful taxis, which resulted in increased pain and tenderness in the right inguinal area. Differential diagnoses included iliopsoas abscess, spermatic cord hydrocele, and enlarged lymph nodes. Finally, the patient was diagnosed with surgery with a diagnosis of right inguinal hernia.

Based on the ultrasound of the patient, the liver has a normal size and parenchymal echo is increased slightly, which indicates grade 1 fatty liver. The image of the cystic foci with a size of  $98 \times 71$  mm in the hepatic segment shows hydatid cyst, while the image of another similar foci with dimensions of  $45 \times 55$  mm shows the hydatid cyst in the left lobe and the segment IV of liver. Furthermore, the image of multiple cystic areas in the abdominal and pelvic cavity with the largest dimensions of  $85 \times 66$  mm was seen in the midline of the abdomen and above the umbilicus and with dimensions of  $70 \times 92$ mm inside the pelvis and the left side of the bladder, which suggests hydatid cyst. Bile ducts were normal without expansion, and spleen was normal.

Ultrasound of the kidneys showed that the right and left kidneys were of normal size with increased parenchymal echo and no hydronephrosis. The right kidney had several cysts with the largest diameter of 24 mm in the right upper pole and in the left kidney, several cysts with the largest diameter of 4 mm were observed in the upper pole. Ultrasound of the testicles showed that the testicles on both sides have a natural echo and a symmetrical axis, a slight hydrocele image was observed in both testicles. The image of edema was also seen in the soft tissue of the scrotum. Ultrasound of the inguinal area showed a multi-cystic focal image with an approximate diameter of 58×44 mm in the right inguinal canal, which may indicate a strangulated inguinal hernia. Radiographic examination of the patient's chest showed a prominent aortic arch, the cardiothoracic ratio was normal, opacity in the lower lung area suggested effusion, and opacity was observed in the left retrocardiac mass. Moreover, the left diaphragm was seen higher than normal (Figure 1).

In the patient's blood test, urea, creatinine, sodium, and potassium levels, and white blood cell, red blood cells and platelets counts were normal. Abnormal cases of blood test were Hb= 10.1, Hct= 33.7, MCV= 70.5, MCH= 21.1, MCHC= 30.0, RDW-CV= 18.5, Neut= 83.9%, Lym= 11.6%, and Mixed (Mono+Eos+Baso)= 4.5%. For surgery, the patient underwent general anesthesia in the supine. A surgical incision was made in the inguinal area. After incision, anatomical cavities containing multiple cysts with a hydatid-like appearance appeared, the cysts were emptied and sent for pathological examination (Figure 2).

To prevent recurrence, hypertonic saline serum (15) was left in the cavity for 10 minutes, then inner part of the cavity was sucked and the surgical site was sutured and bandaged. In addition, the patient did not report any

previous history of hydatid cyst. In pathological examination of the right inguinal cyst sample, several white cysts containing clear fluid were the smallest with a diameter of 0.3 and the largest with a diameter of 1 cm. On microscopic examination of the sample, germinal and cuticular layers of hydatid cyst without pre-cystic tissue were seen. After 2 days, the patient was discharged in relatively good general condition with albendazole (400 mg) twice daily for 6 months. At the end of the treatment period, no signs of hydatid cyst recurrence were observed in the patient.



Figure 1. Chest radiographic image



Figure 2. Cysts removed from the operation site

#### Discussion

In our study, the patient had symptoms of pain, swelling, and redness in the right inguinal area for 6 months. Differential diagnosis included inguinal hernia, spermatic cord hydrocele and spermatic cord lipoma, spermatic cord hydrocele, iliopsoas abscess, enlarged lymph nodes, and external iliac artery aneurysm. Due to the rarity of this disease in this unusual location, hydatid cyst disease was not diagnosed in the inguinal canal before surgery. Imaging findings have different characteristics according to hydatid disease and the growth stage of hydatid cysts and damaged tissue (16, 17). Imaging findings in our study confirmed the presence of hydatid cysts in the liver, abdominal cavity and pelvis. However, strangulated hernia was diagnosed in the right inguinal area. Preoperative diagnosis of hydatid cyst is essential for successful surgery and reduction of complications. Cross-sectional imaging techniques can be used to identify sites where hydatid cysts are present and even places where cysts occur infrequently (10). Because in our study, only ultrasound was used to examine the abdominal cavity and inguinal area, the accuracy of cyst diagnosis, especially in the inguinal area, was less than when CT scan imaging was used. Preoperative diagnosis of cystic echinococcus is mandatory to prevent anaphylaxis or local recurrence. Emergency ultrasound should be the first choice of imaging in abdominal hydatid cysts with a sensitivity of 93% to 97% (3). In the present study, hydatid cysts in the abdominal and pelvic cavities were confirmed by ultrasound findings. Computed tomography should be performed in rare cases of the disease (3).

In our study, only ultrasound was used to diagnose inguinal lesion, which was not successful in diagnosing hydatid cyst. Routine tests such as total leukocyte count and hemoglobin percentage should also be performed. An average eosinophilia of 6% or more is usually present (3). Although eosinophilia is expected to be present in parasitic infected patients, it may not always be present (18). In our study, the total percentage of eosinophils, basophils and monocytes was 4.5%, which shows a decrease in these cells in the blood, indicating that the above study is not consistent with other studies (19).

Treatment of hydatid cyst is basically surgery (3). Our patient has no previous history of hydatid cyst surgery. The possibility of inguinal hydatid disease in this patient may be due to the shedding of abdominal hydatid fluid in the hernia sac with the formation of inguinal hydatid cyst. In the study of Yurtçu et al., a hydatid cyst in the spermatic cord of a 9-year-old boy was reported with an increase in blood eosinophils, which was surgically removed and albendazole was started for the patient (19). The treatment method was in line with our study, but the number of eosinophils in our laboratory findings was less than normal, which in this respect was not in line with the above study. In the study of Wani et al., hydatid cyst was reported in the inguinal canal of a 4-year-old boy with no previous history and was surgically removed (4). In the study of Imani et al., a 4 cm hydatid cyst below the inguinal canal was reported in a 31-year-old woman with no previous history (20).

In the study of Bagga et al., a subcutaneous hydatid cyst was reported in the inguinal canal of a 33 –year–old man without previous history, which was removed by surgery, and albendazole was started for the patient (21). In our study, the cysts were surgically removed and albendazole treatment was considered for the patient. In the study of Singh et al., hydatid cyst of the right inguinal area was reported with the initial diagnosis of inguinal hernia in a 54-year-old man with a previous history of hepatic hydatid cyst. Finally, the cyst was surgically removed and the patient was treated with albendazole (7).

Cystic echinococcosis may cause cystic lesions anywhere on the body. Active hydatid cysts may migrate due to pressure differences between anatomical cavities and show signs of hernia. Therefore, cystic echinococcosis should be considered as a differential diagnosis in patients with cystic edema anywhere in the body in endemic areas, unless proven otherwise. Previous surgery may also play a role in migration. In patients with hydatid cyst, the possibility of migration or hernia should be considered, as awareness of its presence is useful for preoperative planning and reduction of complications.

#### Acknowledgment

We would like to thank the patient and his family who provided the necessary information in all stages of the study, as well as the hardworking staff of Montaserieh Transplant Hospital (Mashhad, Iran) who sympathetically assisted us in conducting this study.

## References

1.Butt A, Khan J. The Maverick Disease: Cystic Echinococcosis in Unusual Locations: A Ten Year Experience from an Endemic Region. Cureus. 2019;11(10):e5939.

2.Ramteke P, Phulware RH, Shende T, Sahoo B, Barwad A. Hydatid cyst of femur, radiologically mimicking a sarcoma. Diagn Cytopathol. 2019;47(10):1045-8.

3.Usharani A, Deepica G, Aruna S, kulkarni S, Sai Kamal Kumar GS, Balamuralikrishna P. Case reports of hydatid disease. J Epidemiol Glob Health. 2013;3(2):63-6.

4.Wani SA, Baba AA, Bhat NA, Hamid R, Mufti GN. Inguinal Hydatid cyst in a child: A rare case report. Int J Surg Case Rep. 2015;10:236-7.

5.Galani K, Trivedi B, Vaghela J. A rare case presentation of hydatid cyst in ilio-psoas muscle. Int J Res Med Sci. 2018;6(9):3182-5.

6.Kerkeni Y, Sahli S, Gasmi M, Sghairoun N, Hamzaoui M. A rare cause of recurrent vaginal hydrocele: Herniating mesenteric hydatid cyst. Iran J Parasitol. 2017;12(3):461-5.

7.Singh A, Soni ML, Khandelwal RG, Gora N. Secondary inguinal hydatidosis mimicking irreducible inguinal hernia: report of a rare case. Hernia. 2016;20(3):489-91.

8.Malekpour-Alamdari N, Gholizadeh B, Kimia F. Inguinal Canal Hydatidosis Presenting as Irreducible Inguinal Hernia: A Case Report. Acad J Surg. 2017;4(3):87-9.

9.Ahmad QA, Ahmed MS. Splenic hydatid, a rare presentation of hydatid disease. Ann King Edward Med Univ. 2010;16(2):129-31. Available from: <u>https://annalskemu.org/journal/index.php/annals/article/view/198/173</u>

10.Koc Z, Ezer A. Migrating and herniating hydatid cysts. Eur J Radiol. 2008;65(1):120-4.

11.Pathak TK, Roy S, Das S, Achar A, Biswas AK. Solitary hydatid cyst in thigh without any detectable primary site. J Pak Med Assoc. 2011;61(12):1244-5.

12.Yagmur Y, Akbulut S. Unusual location of hydatid cysts: a case report and literature review. Int Surg. 2012;97(1):23-6.

13.Canda MŞ, Güray M, Canda T, Astarcio/Lu H. The Pathology of Echinococcosis and the Current Echinococcosis Problem in Western Turkey (A Report of Pathologic Features in 80 Cases. Turk J Med Sci. 2003;33(6):369-74.

14.Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev. 2004;17(1):107-35.

15.Arer IM, Yabanoglu H, Akdur A, Caliskan K. Inguinal hydatid cyst. J Turgut Ozal Med Cent. 2016;23(3):342-3.

16.Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A. Hydatid disease from head to toe. Radiographics. 2003;23(2):475-94.

17.Engin G, Acunaș B, Rozanes I, Acunaș G. Hydatid disease with unusual localization. Eur Radiol. 2000;10(12):1904-12.

18. Arora V, Nijjar IS, Gill KS, Singh G. Case report: Primary hydatid cyst of muscle-a rare site. Indian J Radiol Imaging. 2006;16(2):239-41.

19.Yurtçu M, Gündüz M, Toy H, Günel E. Spermatic cord hydatid cyst: an unusual localization. J Pediatr Surg. 2007;42(12):e15-6.

20.Imani MR, Tizmaghz A, Salmasi MA. Rare Lower Limb hydatid cysts presenting as mass. J Arak Uni Med Sci. 2014;17(3):82-7. [In Persian]

21.Bagga PK, Bhargava SK, Aggarwal N, Chander Y. Primary subcutaneous inguinal hydatid cyst: diagnosis by fine needle aspiration cytology. J Clin Diagn Res. 2014;8(8):FD11-FD13.