JIDC | THE JOURNAL OF INFECTION IN DEVELOPING COUNTRIES

Case Report

A rare case of intravesical and urethral cystic Echinococcosis with multi-organ involvement from Türkiye

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Abstract

Introduction: Cystic echinococcosis is a parasitic disease recognized as a global public health problem in countries engaged in agriculture and animal husbandry. In its natural life cycle, ungulates are intermediate hosts, and canids are definitive hosts. It could be accidentally transmitted to humans by the fecal-oral route and migrate to the liver and other visceral organs to form cystic echinococcosis. It spreads hematogenously, lymphatically, and locally. The majority of initially asymptomatic patients develop liver and lung involvement. Involvement of other organs is rare. Cystic echinococcosis is a disease diagnosed by radiologic imaging and confirmed by histopathology, serologic, and molecular tests that can be used for diagnosis and follow-up of primary and secondary infections. In this case report, the involvement of multiple and rare organs of cystic echinococcosis is presented.

Case Presentation: An 82-year-old patient was admitted to the hospital with an inability to urinate. A glob vesicle was detected during the examination, and urine output was achieved through urinary catheterization. Imaging techniques revealed multiple cysts in the abdomen, ureter, and bladder. Urine flow in the ureter orifice was obstructed by cysts. Cystectomy was performed for the intraabdominal, intravesical, and ureteral cysts. Perioperative albendazole treatment was started as adjuvant and antiparasitic treatment was completed for one month. The case was confirmed histopathologically, and no secondary infection or complication was detected in the one-year follow-up.

Conclusions: By a multidisciplinary approach, the 82-year-old patient was diagnosed with Cystic echinococcosis, with a rare and multi-organ involvement, given the high risk of infection due to the environmental contamination in this country.

Key words: Cystic echinococcosis; anuria; intravesical.

J Infect Dev Ctries 2025; 19(10):1566-1569. doi:10.3855/jidc.18947

(Received 25 July 2023 – Accepted 02 November 2023)

Introduction

Echinococcus is a zoonotic infection caused by the larval stages (metacestodes) of the cestode species of the genus Echinococcus widely found in nature and can be fatal if left untreated [1]. It is a serious public health problem, especially in countries where agriculture and animal husbandry are practiced, including Turkey [2]. In their life cycle in nature, dogs and other canids play a role as definitive hosts, and mammals such as sheep, goats, horses, and cattle as intermediate hosts. Humans are incidental intermediate hosts and have no part in the transmission cycle. Thousands of parasites infecting the definitive host survive in the small intestine as adult tapeworms 2-7 mm in length. It produces thousands of 30-40 micron-sized eggs containing oncospheres from its proglottids; the eggs are dispersed into the environment with feces and could infect intermediate and incidental hosts if ingested with contaminated food [1]. Following fecal-oral transmission of the eggs, the oncospheres hatch and migrate from the intestinal mucosa to the liver and other visceral organs by translocation through the blood and lymphatic system. Cystic echinococcosis (CE) formation develops in the organ [3].

In humans, primary liver infection and secondary infection with direct expansion or hematogenous spread to other organs can be observed [4]. *E. granulosus sensu lato (s.l.)* infections give symptoms according to the cyst site, cyst size, and complications such as cyst rupture [5]. Approximately two-thirds of patients have liver involvement, and one-fourth have lung involvement. Other organs are rarely infected [6]. Renal involvement develops in only 2-4% of CE cases, and most of these cases are not isolated primary cases [7]. Renal cysts may present with hematuria, flank pain, nephrotic syndrome, and secondary amyloidosis. This report presented a case of intravesical CE secondary to primary liver infection.

Case Report

An 82-year-old male patient engaged in animal husbandry was admitted to the emergency department

with the complaint of inability to urinate. In his anamnesis, it was learned that he had intermittent dysuria and nocturia for the last 2-3 months. There was no history of previous genitourinary operations or kidney stones. Physical examination revealed stable vitals and an abdominal glob vesicle. A urethral catheter was inserted. There was urine output under the catheter. Abdominal ultrasonography revealed a type 5 CE with a diameter of 30 mm in the right lobe posterior superior of the liver; a type 5 CE with a diameter of 53 mm between the liver and the anterior abdominal wall in the epigastric region; multiple cortical cysts in the right and left kidneys; a type 5 CE [8] with a diameter of 68 mm in the left kidney; and multiple vesicular CE at different stages in and around the left kidney. Cystoscopy, procedure performed with a flexible cystoscope under sedoanalgesia after urethral instillation with a glycerin-based lubricant containing the local anesthetic agent lidocaine, performed because of anuria showed a ruptured cyst expulsed into the bladder, obstructing the ureteral orifice (Figure 1), cyst membrane in the bladder (Figure 2), multiple membranous gray and white sliding cysts in the ureteral lumen (Figure 3). Intravesical and ureteral cysts were surgically removed using normal saline irrigation. Parasite material collected from the intra-abdominal region was immediately fixed in 10% neutral buffered formalin and embedded in paraffin. Glass slides prepared by serially cutting the paraffin block into several 3-4 µm sections were stained with hematoxylin and eosin and examined under an optical microscope.

Figure 3. Multiple membranous gray and white sliding cysts in the ureteral lumen seen on cystoscopy.

Figure 1. A ruptured cyst expulsed into the bladder, obstructing the ureteral orifice, seen on cystoscopy.

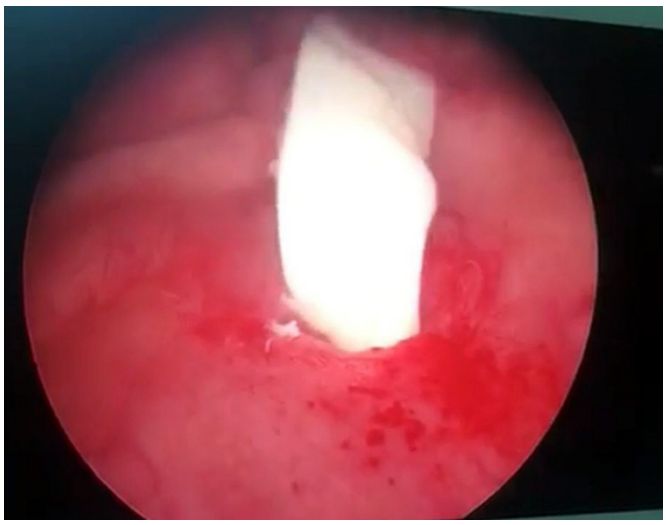


Figure 2. Cyst membrane in the bladder seen on cystoscopy.

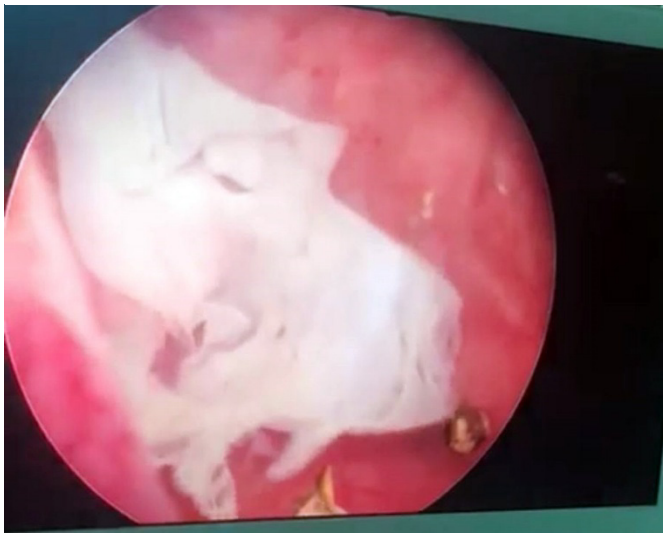
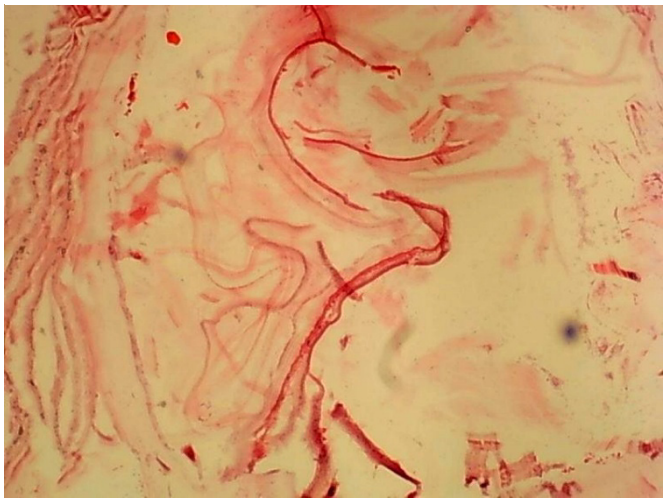


Figure 4. Histopathological image of cyst membrane (x400).



The ruptured cyst membrane was confirmed by histopathology (Figure 4). Perioperative albendazole 400 mg oral treatment (twice 400 mg/day, one week before the perioperative and for 1 month after) was started, and cystostomy was performed for all intraabdominal cysts, and multiple cyst foci were irrigated with saline during the procedure. Intracystic fluid material was aspirated, and membranes were removed. The patient was discharged on the 5th postoperative day with continued antiparasitic treatment. It was learned that the patient had no new symptoms after one month of treatment. No recurrence and complications were observed in the one-year follow-up.

Discussion

CE is a parasitic disease affecting all organs, especially the liver and lungs. Since the liver is the first site of hepatic portal flow, primary infections are most commonly detected in the liver. It reaches the lung by joining the venous return through the microvascular structures in the liver. However, an infection can be seen in any tissue in the body, such as the heart, kidney, spleen, pancreas, intraperitoneal space, prostate, thyroid, bladder, brain, breast, and axillary space, with the participation of hydatid larvae in the systemic circulation [9,10]. The larvae passing transmural through the intestinal wall via venous mesenteric lymphatic pathways may cause CE formation in intra-abdominal organs [10]. Since the reported patient presented multiple cysts adjacent to the anterior abdominal wall and around the kidney, we think that transmission was via mesenteric lymph nodes.

Although the diagnosis of cystic echinococcosis is based on radiological techniques, other investigations, such as serology histopathology and molecular biology including a latex agglutination test, indirect hemagglutination test (IHA), enzyme-linked immunosorbent assay (ELISA), and a Western blot and polymerase chain reaction are useful confirmation tools, in particular in challenging human cases [11,12]. On imaging, a cystic structure with an irregular contour and calcification on the wall, accompanied by necrosis in the center, is detected. The general condition and history of the patient should be taken into consideration in the differential diagnosis with malignancy [13]. ELISA and IHA are also used in the follow-up of recurrence in the postoperative period. Although ELISA has high sensitivity and specificity, IHA is preferred because it is cheap and practical [12]. In our case, those who underwent cystoscopy due to anuria, histopathologic confirmation was obtained from the cysts for diagnosis.

An antiparasitic treatment should be applied as an adjunct to surgical and percutaneous treatments. The primary antiparasitic treatment is albendazole. In cysts with a single compartment and a diameter of less than 5 cm, a maximum of 400 mg albendazole daily should be administered for one to three months, depending on clinical factors [14]. The development of symptoms and complications is expected based on high age, cyst size, number of cysts, and location of the cysts [15]. Surgery should be preferred for multiple liver cysts smaller than 5 cm, cysts larger than 5 cm, and cysts with more than one compartment. Surgery should be planned to prevent complications such as secondary infection, cyst rupture, and circumferential compression. Total cystectomy is recommended to prevent recurrence by avoiding cyst rupture [9]. Every possible precaution must be taken to prevent spillage of the contents into the peritoneum; otherwise, recurrence, dissemination, and severe anaphylactic shock may develop iatrogenically [16]. In this case, the cyst was sterilized by injecting 20% hypertonic saline into the cyst for its protoscolicidal properties before removing the contents, followed by cystectomy. Perioperative antiparasitic treatment reduces recurrence [17]. With the combination of pharmacologic therapy and surgery, a challenging case of human hydatidosis can be treated safely and effectively [18]. In this case, perioperative albendazole treatment was initiated to prevent secondary cysts.

Conclusions

Since the most commonly involved organ is the liver, the most common complication is spontaneous rupture into the biliary tract. However, in our patient, anuria due to renal and intravesical CE accompanying primary hepatic CE, which is rare, was observed. Therefore, it should be kept in mind that CE may have secondary localization, may present with associated symptoms, and reliable treatment of CE is medical therapy accompanied by surgery.

Authors contributions

All authors contributed to the study conception and design. Conceptualization, data curation were performed by Ayşegül Tuna and Emrah Topbaş. Visualization and supervision were performed by Emrah Topbaş. The first draft of the manuscript was written by Ayşegül Tuna, and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Conflict of interest
No conflict of interest is declared.

References
1. Eckert J, Deplazes P (2004). Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev 17: 107–135. doi: 10.1128/CMR.17.1.107-135.2004.
2. Çelik T, Akçora B, Tutanç M, Yetim TD, Karazincir S, Akın MM, Çelikkaya ME, Kurtoğlu A (2012) Ruptured pulmonary hydatid cyst: a case report. Turkiye Parazitol Derg 36: 45-7. doi: 10.5152/tpd.2012.11. [Article in Turkish]
3. Jenkins DJ, Romig T, Thompson RC (2005) Emergence/re-emergence of *Echinococcus* spp.--a global update. Int J Parasitol 35: 1205-1219. doi: 10.1016/j.ijpara.2005.07.014.
4. Schantz PM, Kern P, Brunetti E (2011) Echinococcosis. In: Tropical infectious diseases: principles, pathogens and practice, 3rd ed, Guerrant R, Walker DH, Weller PF (Eds), Saunders Elsevier, Philadelphia 2011. p.824.
5. Moro PL, Gilman RH, Verastegui M, Bern C, Silva B, Bonilla JJ (1999) Human hydatidosis in the central Andes of Peru: evolution of the disease over 3 years. Clin Infect Dis 29: 807-812. doi: 10.1086/520440.
6. Bektaş M, Dökmeci A, Cinar K, Halici I, Oztas E, Karayalcin S, Idilman R, Sarioglu M, Ustun Y, Nazligul Y, Ormeci N, Ozkan H, Bozkaya H, Yurdaydin C (2010) Endoscopic management of biliary parasitic diseases. Dig Dis Sci 55: 1472-1478. doi: 10.1007/s10620-009-0850-0.
7. Göğüş C, Safak M, Baltaci S, Türkölmez K (2003) Isolated renal hydatidosis: experience with 20 cases. J Urol 169: 186-189. doi: 10.1016/S0022-5347(05)64064-5
8. Tévez-Craise L, Daiana-Vaccaro R, De Luca PA, Vásquez-Guillén ME, Calaramo OA, Logioco F (2022) Hydatidosis: Clinical-imaging classification according to Gharbi and the World Health Organization. Rev. Argent. Radiol 86: 41-48.
9. Unalp H.R, Kamer E, Rezanko T, Kilic O, Tunakan M, Onal MA (2011) Primary hydatid cyst of the axillary region: a case report. Balkan Med J 28: 209–211.
10. Versaci A, Scuderi G, Rosato A, Angiò LG, Oliva G, Sfuncia G, Saladino E, Macrì A (2005) Rare localizations of echinococcosis: personal experience. ANZ J Surg 75: 986–991. doi:10.1111/j.1445-2197.2005.03588.x
11. Bal N, Kocer N.E, Arpaci R, Ezer A, Kayaselcuk F (2008) Uncommon locations of hydatid cyst. Saudi Med J 29: 1004–1008.
12. Sbihi Y, Rmiqui A, Rodriguez-Cabezas MN, Orduña A, Rodriguez-Torres A, Osuna A (2001) Comparative sensitivity of six serological tests and diagnostic value of ELISA using purified antigen in hydatidosis. J Clin Lab Anal 15: 14-8. doi: 10.1002/1098-2825(2001)15:1<14::aid-jcla3>3.0.co;2-7.
13. Clavel A, Varea M, Doiz O, López L, Quílez J, Castillo FJ, Rubio C, Gómez-Lus R (1999) Visualization of hydatid elements: comparison of several techniques. J Clin Microbiol 37: 1561-3. doi: 10.1128/JCM.37.5.1561-1563.1999.
14. Brunetti E, Kern P, Vuitton DA, Writing Panel for the WHO-IWGE (2014) Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop 114: 1-16. doi: 10.1016/j.actatropica.2009.11.001.
15. Gomez I, Gavara C, López-Andújar R, Belda Ibáñez T, Ramia Ángel JM, Moya Herraiz Á, Orbis Castellanos F, Pareja Ibars E, San Juan Rodríguez F (2015) Review of the treatment of liver hydatid cysts. World J Gastroenterol 21: 124-31. doi: 10.3748/wjg.v21.i1.124.
16. Hailu SS, Gebremariam M, Nigussie T, Girma K, Misgea A, Arega G (2023) Isolated renal hydatid cyst in a ten-year-old female child: A rare case report. Urol Case Rep 50: 102482. doi: 10.1016/j.eucr.2023.102482.
17. Erzurumlu K, Hökelek M, Gönlüsen L, Tas K, Amanvermez R (2000) The effect of albendazole on the prevention of secondary hydatidosis. Hepatogastroenterology 47: 247-50.
18. Santucciu C, Ferrari PA, Grimaldi G, Murenu A, Nemolato S, Bonelli P, Masala G, Porcu GS, Cherchi R (2023) Environmental influence on the occurrence of multi-organ cystic echinococcosis infection in a patient from Sardinia, Italy. Diseases 11: 90. doi: 10.3390/diseases11030090.