

## Case Report

# An Unusual Location of Hydatid Cyst, Cause of Severe Pollakiuria

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Hydatidosis is an important zoonotic parasite disease in several herbivorous mammals. Human cystic echinococcosis (CE) caused by infection with larval stage of *Echinococcus granulosus* has been frequently reported from different organs. Here, we report the first case of a urinary echinococcosis in Iran with a pain in the lower left abdominal quadrant and severe frequent urination (pollakiuria). We detected a cyst 120 × 15 mm dimensions with heterogeneous mass contain fluid in the back of the urinary bladder neck between the umbilical region and external urethral sphincter. The patient was candidate for open-abdomen surgery and cysts were resected. The isolated cysts from liver and urinary bladder were referred to pathology laboratory, and the tissue sections were stained with Tri-chrome and Hematoxylin/eosin staining methods. Microscopic examination of prepared tissue sections showed protoscolecocytes of *E. granulosus* with specific and thick laminated hyaline layer (non-cellular membrane), with covers the thin activate germinal epithelium, which revealed the diagnosis of a hydatid cyst.

This rare case illustrates that CE is the necessity of considering in the differential diagnosis from cysts, abscesses, malignant and benign tumors, especially is essential in endemic areas of CE.

**Keywords:** Cystic echinococcosis, Hydatid cyst, Pollakiuria

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**Introduction**

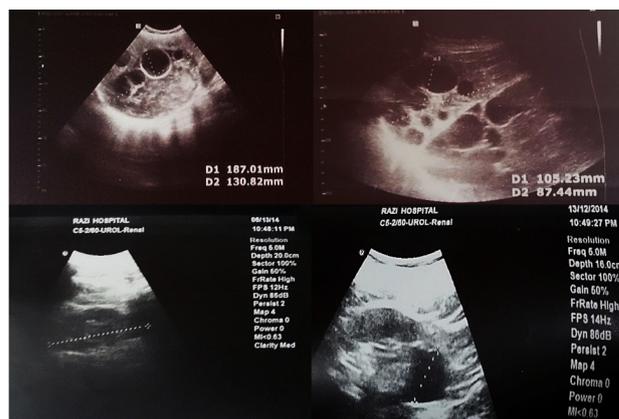
Hydatidosis also known as cystic echinococcosis (CE) is an infectious global zoonotic disease due to parasites that belonged to class of Cestoda and family of Taeniidae and *Echinococcus* genus. The parasite life cycle is completed between wild or domesticated carnivores and different mammalian herbivores. In many countries dogs are the main definitive host and adult parasites are located in their small intestine. The parasite eggs that are excreted by dog feces are immediately contaminant for livestock animals and humans.<sup>1</sup> Humans are infected due to accidental ingestion of parasite eggs in contaminated food, water and edible vegetables or through direct contact with infected animal and soil.<sup>1</sup> The incubation period can vary between 5 to 20 years, according to the location, type and severity of the lesions.<sup>1</sup> Hydatidosis is medically important and very common in small ruminants as intermediate host in Iran, with a high infection rate and economic losses on livestock breeding industry.<sup>2-5</sup> Echinococcosis scattering is widespread throughout the world. Commonly endemic areas for this disease are the Mediterranean countries, the Middle East, southern part of South America, New Zealand, Australia, and southern parts of Africa, central Asia and particularly China, Iran, southern Russia and

some parts of Europe.<sup>6</sup> Echinococcosis is not endemic in the United States, but transcontinental transport and immigration patterns over the last fifty years, have caused the relative increase of this unusual disease throughout the North America.<sup>6,7</sup> According to the world hydatidosis reports, incidence of CE in endemic areas ranges 1-220/100 000, and it is estimated that the frequency of this disease (cystic form, 2015) was 1.4 million and numbers of related deaths was 1.200 in the world.<sup>8</sup> 5% to 49% of stray dogs in various regions of Iran are infected and human infection prevalence is 0.6-1.2/100 000.<sup>9</sup> Infections persist asymptomatic in most cases proceed, until symptoms including the cholestatic jaundice, epigastric pains, fatigue, weight loss and hepatomegaly appear. CE is most commonly diagnosed in younger people between the ages of 30 to 40, but in high prevalence areas, it is often observed in the elderly with an average age above 50 years.<sup>1</sup> Disease in dogs is usually asymptomatic.<sup>1</sup> The symptoms of this disease depend on the location, size and position of the cysts. For example, it quickly signs up in the brain and the eyes, while in the liver it takes many years to make a mark. About 90% of the cysts are formed in the lungs and liver and can be enlarged if there are soft tissues surrounding the cysts.<sup>1</sup> The cysts mostly

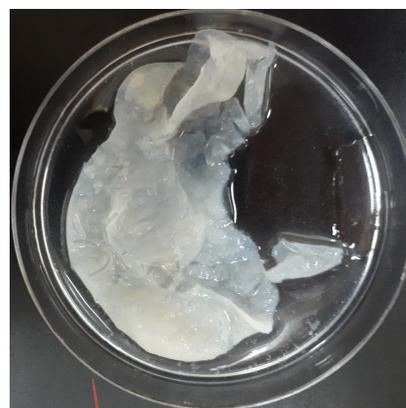
located in the lungs, liver, spleen, bone, kidney, heart, and brain, although it could be found in any organs. Lung and liver cysts are more common. Symptoms of this disease include extended abdominal pain and shortness of breath, dry coughing, secondary infections after cyst tearing in the internal organs and anaphylaxis. Sometimes, these cysts increase to a degree in the liver, which can lead to acute liver failure,<sup>1</sup> and if left untreated, leads to death. Surgery of hydatid cysts is the only basic treating way for this disease in humans but sometimes due to the spread of cysts in different organs of the body or the presence of cysts in a sensitive and dangerous area, surgery is difficult.<sup>1</sup>

### Case Report

A 77 years-old male farmer with left lower-quadrant abdominal pain and tenesmus was referred to the department of surgery at Koh Kamari Marand hospital. The patient was residing in the village near Marand, East Azerbaijan province, Iran. In physical examinations, the weight was 64 kg, height; 167 cm, temperature; 37°C, heart pulse; 64 per minute and blood pressure was 110/77 mm Hg. The patient had nausea, vomiting, abdominal pain and diarrhea, without conversion and feeling of incomplete urination during last 2 years with frequent urination since 15 years that more problems have been reported the last four months. The patient complained of temporary retention of urine and severe pelvic pain with a feeling of bladder fullness and difficulty passing urine. The patient had a history of direct contact with dogs and soil. General physical examination revealed tender on palpation and dullness on percussion. In laboratory examinations, WBC count was 19400, platelets; 189000, and RBC was  $5.21 \times 10^3$ . In addition liver enzymes SGOT and SGPT, were 41 and 81, respectively. The total bilirubin was 32.1 and direct bilirubin was 0.73. Hematocrit; 6.8%, hemoglobin 13 and blood sodium level reported lower than normal. Elisa was used for detection IgG hydatid cyst antibody (EUROIMMUN AG, Germany) and abdomen ultrasonography showed a cyst with heterogeneous mass in the back of urinary bladder neck between the umbilical region and external urethral sphincter with the dimensions of  $120 \times 15$  mm. In right lower quadrant, the upper right lobe of the liver an unilocular cystic mass ( $50 \times 35$  mm) with regular borders in eight segment, adheres to the upper part of spleen and a calcified cyst with the size of  $10 \times 8$  mm in the right part of the liver were found (Figure 1). The patient was candidate of open-abdomen surgery. A measuring 12 cm uniloculated white cyst contain clear fluid was detected inside the umbilical region and external urethral sphincter of urinary bladder neck was observed (Figure 2). Left hepatic and behind bladder cysts were resected. During surgery, prostate hyperplasia was not observed. The right hepatic hydatid cyst was drainage. The isolated cysts from liver and urinary bladder were referred to pathology laboratory. Tissue sections were stained with



**Figure 1.** Ultrasound of the Liver and the Urinary System. In the above images multiple hydatid cysts are seen. In below images, a large hydatid cyst in the bladder neck of the patient's is visible.

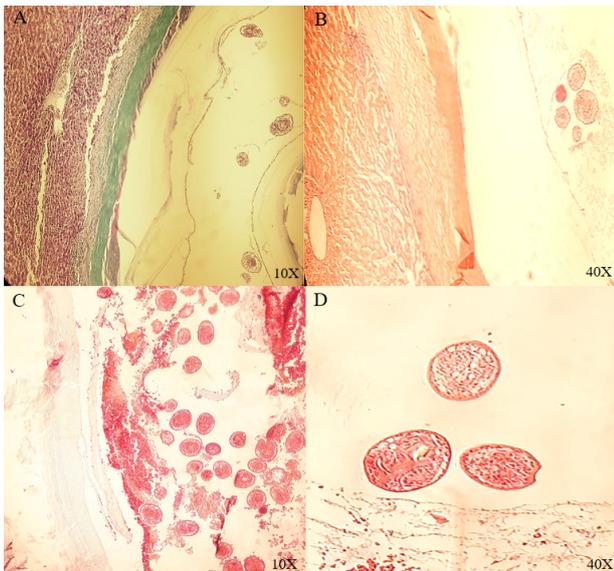


**Figure 2.** Removed Hydatid Cyst From Urinary Bladder of Patient.

Tri-chrome and Hematoxylin/eosin staining methods. Microscopic examination of prepared tissue sections showed *Echinococcus granulosus* protoscoleces with thick outer laminated hyaline layer (non-cellular membrane) which covers the thin activate germinal epithelium which represents the hydatid cyst diagnosis (Figure 3). In the follow-up, frequent urination problems were improved. For treatment, ciprofloxacin (500 mg) twice a day for three days, Albendazole (10 mg/kg/d) for a period of 12-weeks, and mebendazole (100 mg/kg/d) for 6 months were used. Sonography was recommended 1 month later. After surgery, the patient has recovered and discharged from hospital with a good general condition.

### Discussion

Human CE (hydatidosis) is a most important cosmopolitan parasitic zoonosis. According to the studies more than 1 million people are at risk of contamination with echinococcosis at any one time. These disease has a worldwide distribution but the prevalence is much higher in countries with high animal husbandry industry. Echinococcosis is often expensive and complicated to



**Figure 3.** A) Tissue Section of Hydatid Cyst From Liver by Trichrome Staining (10×). B) Section of liver hydatid cyst stained with hematoxylin/eosin staining (40×). C) Cross section of removal hydatid cyst from neck of urinary bladder (10×). D) Hydatid cyst protoscolex isolated from neck of bladder stained with hematoxylin/eosin staining (40×).

treat, and may need invasive surgery and/or prolonged drug therapy. WHO is trying to validate of efficient and executable strategies for controlling the CE by 2020.<sup>8</sup> Common locations of involvement are liver (59%–75%), lung (27%), kidney (3%), bone (1%–4%), and brain in low frequency (1%–2%).<sup>6,7</sup> Pelvic and urinary bladder echinococcosis are extremely rare, with an incidence of only 0.2%–2.25%.<sup>10</sup> Therefore, liver and lungs are the main organs involved for hydatid cysts but in rare cases, the heart, brain, appendix, cervix, mesenteric, pancreas, spinal canal, parathyroid, pelvic, small intestine, bone, muscle, kidney, spleen and breast can be involved.<sup>6,7</sup> The most reports of hydatid cyst and published papers in Iran, are based on postoperative experiences. The results from previous studies in Iran showed that the lung, liver, central nervous system, muscular system, cardiovascular system, kidney, urinary tract, spleen, uterus, ovary, pancreas, salivary glands, breast, adrenal, appendix, mediastinum, omentum, diaphragm, peritoneal, maxillary and mandible bones were involved in most reported cases.<sup>11–14</sup> Numerous studies have reported hydatid cyst in urinary system from different area. Izol et al in 2012 reported a case of hydatidosis in a 21-year-old man with symptoms of frequency and reduced force in urinary stream, urinary retention due to CE.<sup>15</sup> Zargar-Shoshtari et al in 2006 followed-up 11 patients with urinary tract hydatid cyst. Radiological, clinical pathology findings and surgical outcomes were reviewed.<sup>16</sup> Another case study in Turkey, reported a 34-year old patient with lower urinary tract symptoms and primary huge hydatid cyst with lesion adjacent to the posterior wall of the bladder between

sigmoid colon and bladder.<sup>17</sup> The common differential diagnosis of urinary cysts in an adult male patient includes seminal vesicle cyst, prostatic abscess or hyperplasia, ureterocele, ureteral obstruction bladder cysts, bladder tumors, mesenteric cysts, and other cystic intra pelvic benign masses or neoplasms.<sup>17</sup> A 52-year-old Tunisian patient, was another case of this disease, with dysuria and gross haematuria who referred to the urology department. In transrectal ultrasonography, an intraprostatic hydatid cyst was observed. Transurethral incision of the prostate, allowing wide communication of the bladder with the intraprostatic cystic cavity and pathological examination of prostate confirmed a calcified hydatid cyst in the prostatic tissue.<sup>18</sup> A 48-year-old-male, with hydatiduria, diffuse lower abdominal pain, temporary retention of urine and incomplete bladder evacuation with increasing frequency of micturition and feeling of incomplete urination for last 2 years which had history of liver hydatid cyst surgery reported by Ganie et al in 2013.<sup>19</sup>

Primary urinary bladder hydatid cyst is rare. Although it should be considered in differential diagnosis of cystic masses and tumoral positions, especially in endemic areas. CE in urinary system is not occur commonly and in such cases the embryo of hydatid is trapped in pelvic cavity organs by hematogenous dissemination (bypassing the hepatic or pulmonary filters) or lymphatic vessels pathway. Peritoneum connective tissue is the usual origin site of pelvic hydatidosis, and from here it can be extend to the other organs such as uterus, ovaries, fallopian tubes, urinary bladder or to the intestines. The choice of treatment method for this patient is a complete surgical excision, which can be done either by open or laparoscopic surgery. Sometimes these large hydatid cysts may adherent to the other lateral organs, where complete surgical extirpation is dangerous and endocystectomy would be done. Preventive programs focus on treat of dogs and removal of sheep at an early age. Some measures including food inspection, improved slaughterhouse hygiene, and public health education campaigns as well as raising the level of community health can be useful in controlling the CE.

#### Authors' Contribution

JK; Data collection. FA, HSH; Data analysis. MKH; Drafting the manuscript. MKH; Intellectual input and manuscript revision. All authors have read the manuscript and approved its final version.

#### Conflict of Interest Disclosures

The authors declare no conflict of interest.

#### Ethical Statement

As this manuscript is an unusual case report, and did not done anything against principles of ethics, there was no necessity any ethical certification. Therefore, in accordance with the principle of confidentiality, the results of information was published.

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