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Research article

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Retrospective analysis of isolated renal hydatid cysts: A single-center study

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Harran Universıty Clinical Studies Ethics Committee (Decision No.HRU/21.11.04). All procedures in this study involving human participants were performed in accordance with the 1964 Helsinki Declaration and its later amendments.

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No conflict of interest was declared by the authors.

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Abstract

Background/Aim: Hydatid disease is a parasitic infection caused by Echinococcus granulosus larvae settling in organs through blood and lymph circulation. Hydatid cysts are often observed in the liver and lungs, and isolated renal involvement is extremely rare. In this study, we aimed to evaluate patients who were diagnosed with an isolated renal hydatid cyst and treated in our clinic.

Methods: In this case series, the data of patients with renal hydatid cysts, who presented to our clinic between January 2010 and April 2021, were retrospectively reviewed. Only patients with an isolated renal hydatid cyst were included in the study. Demographic data, clinical findings, laboratory findings, indirect hemagglutination test results, radiological findings, treatment methods, complications and follow-up processes of the patients were evaluated.

Results: Eighteen patients (11 males and 7 females) were included in the study. The mean age and cyst diameter of the patients were 29.6 years and 9.2 cm, respectively. Reasons for hospital admission included flank pain (38.9%), palpable mass (22.2%), incidental (16.6%), abdominal pain (11.1%), nausea (5.5%) and hematuria (5.5%). Seropositivity in the indirect hemagglutination test was detected in was detected in 61.1%. Fourteen and four of the patients underwent cystectomy and nephrectomy, respectively. No major postoperative complications were observed in any of the patients. The mean follow-up period was 27 months, during which no local and renal recurrence were observed.

Conclusion: Although hydatid cysts are most observed in the liver and lungs, they may also be observed in other organs, especially the kidney. The primary treatment for a renal hydatid cyst should be kidney-sparing surgery. Additionally, albendazole treatment is should be recommended pre- and post-operatively to reduce the risk of inoculation and recurrence.

Keywords: Echinococcus granulosus, Hydatid cyst, Kidney, Cystectomy, Nephrectomy

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Introduction

Hydatid disease is a parasitic infection transmitted by oral ingestion of Echinococcus granulosus eggs. Dogs are the definitive hosts of the parasite, and sheep and cattle are the intermediate hosts. Humans may also be intermediate hosts [1]. The disease is endemic in the Mediterranean Basin, including Turkey, and some parts of Eastern Europe, South America, the Middle East, Australia and South Africa [2]. Liver, lung and kidney involvement occurs in 70%, 25% and 2%–4% of the patients, respectively. As most parasites are retained by hepatic and pulmonary filters during circulation, isolated renal involvement is extremely rare (1.9%) [3, 4]. Renal involvement is usually in the form of a single cyst in the renal cortex. Clinical signs vary depending on the size, location and extension of the cyst. As the growth of the cyst takes years, patients may remain asymptomatic for long periods. Sometimes, patients may present with symptoms of flank pain, hematuria, hydatiduria or abdominal mass [5, 6]. In addition, they may be mistaken as tumors because they can mimic cystic kidney tumors clinically and radiologically [7]. In this study, we aimed to evaluate the rare cases of isolated renal hydatid cysts (HCs).

Materials and methods

For this study, the hospital records of patients who were treated for renal HCs between January 2010 and April 2021 were retrospectively reviewed after approval was obtained from Harran University Clinical Studies Ethics Committee (Decision No.HRU/21.11.04). All procedures in this study were conducted in accordance with the guidelines of Helsinki Declaration. All patients with isolated renal HC were included in the study. Patients' age, gender, reason for hospital admission, physical examination findings, serum creatinine levels, complete blood count, urinalysis, indirect hemagglutination test (IHA) results, cyst localization, cyst size, treatment methods, hospital stay, surgical complications and follow-up findings were analyzed. Abdominal ultrasonography (US), abdominal computed tomography (CT) (Figure 1) and IHA test results aided in the diagnosis of HCs. An IHA test result of ≥1/160 was considered positive. Patients underwent open or laparoscopic cystectomy or nephrectomy based on the cyst size and location as well as kidney function. Albendazole (10mg/kg/day) was initiated 3 weeks before the operation in all patients who were scheduled for surgery with a preliminary diagnosis of HC. Postoperative albendazole treatment was continued for 3 months in patients who underwent cystectomy and discontinued in those who underwent nephrectomy. Preoperative retrograde pyelography was performed in patients with a large cyst presenting with colic pain, and whether the cyst opened up to the collecting system was investigated. Povidone-iodine gauze pads were placed on the surrounding tissues to prevent intraoperative spread in patients who underwent cystectomy. After the cyst content was aspirated, hypertonic sodium chloride, a scolicidal agent, was injected into the cyst. After 10 min, the cyst was opened, daughter vesicles and germinative membrane were removed and cyst wall was excised (Figure 2). Nephrectomy was performed in patients who could not be conclusively diagnosed as having either malignancy or HCs by radiological and clinical data and in patients with non-functional kidneys. A non-functioning kidney was diagnosed using Tc-99m dimercaptosuccinic acid (DMSA) scintigraphy. Postoperative complications were classified according to the Clavien classification. The patients were followed up with an abdominal ultrasound (US) in the first and third months postoperatively. Subsequently, the patients were followed up every 6 months.

Statistical analysis

Mean, lowest, highest, frequency and ratio values were included in descriptive statistics. SPSS 27.0 software was used in the analysis.

Figure 1: Abdominal CT; 124x112 mm hydatid cyst located in the left kidney upper pole

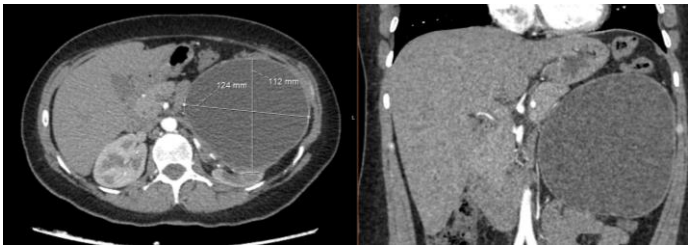
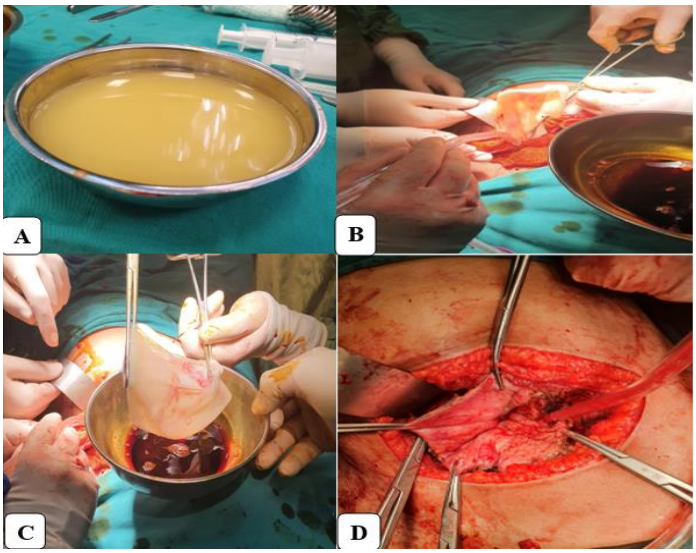


Figure 2: Intraoperative cystectomy images: A; aspirated cyst content, B and C; removal of germinative membrane, D; after cystectomy



Results

Among the 18 patients included in the study, 11 were male and 7 were female. The mean age and cyst diameter of the patients were 29.6 years and 9.2 cm, respectively (Table 1).

Table 1: Characteristics and clinical findings of the patients

		Mean (Min-Max) n (%)
Age (year)		29.6 (6-58)
Cyst diameter (cm)		9.2 (5-16)
Gender	Male	7 (38.9%)
	Female	11 (61.1%)
Side	Right	9 (50.0%)
	Left	9 (50.0%)
Symptoms	Flank pain	7 (38.9%)
	Palpable mass	4 (22.2%)
	Abdominal pain	2 (11.1%)
	Sickness	1 (5.5%)
	Hematuria	1 (5.5%)
	İncidental	3 (16.6%)
Indirect hemagglutination test	Positive	11 (61.1%)
	Negative	7 (38.9%)
Surgical complication	Yes	3 (16.6%)
	No	15 (83.4%)
Duration of hospitalization (days)		5.1 (3-8)
Duration of follow-up (months)		27 (6-60)

The reasons for admission to the hospital included flank pain (38.9%), palpable mass (22.2%), incidental (16.6%), abdominal pain (11.1%), nausea (5.5%) and hematuria (5.5%).

Seropositivity was detected in 61.1% of the patients in the IHA test (Table 1).

Cyst excision was performed in 14 of the patients with an isolated renal HC. A total of 4 patients underwent nephrectomy as 2 patients had non-functional kidneys and 2 were suspected to have malignancies (Table 2). Retrograde pyelography was performed in 4 patients with large cysts, and the connection of the cyst with the collecting system was evaluated (Table 2). There was no opening to the collecting system in these patients. The postoperative complication rate was 16.6%, and no major complications were observed in any of the patients. Three of the patients were found to have postoperative fever, wound infection and transient creatinine elevation, each. The complications were Grade 1 according to the Clavien classification.

Table 2: Cyst localization and treatment methods

		n	%
Cyst location	Upper Pole	8	44.4%
	Middle Pole	6	33.3%
	Lower Pole	4	22.2%
Treatment	Open cystectomy	13	72.2%
	Laparoscopic cystectomy	1	5.5%
	Open nephrectomy	2	11.1%
	Laparoscopic nephrectomy	2	11.1%
Retrograde pyelography	Yes	4	22.2%
	No	14	77.8%

The mean hospital stay and follow-up was 5.1 days and 27 months, respectively (Table 1). During the follow-up period, no recurrence was observed in any patient, whereas a new cyst was detected in the ovary of one patient.

Discussion

Although the kidney is the most frequently involved organ in the urogenital system, renal involvement is observed in only 2%–4% of all HC cases. More than half of those with kidney involvement simultaneously have cysts in other organs, and an isolated renal HC is very rare [3, 4, 8]. Although renal involvement is unilateral in 85% of the patients, it is bilateral in 15% [3]. A renal HC can cause serious complications, such as vascular compression, cyst infection, shock, sepsis and death [9]. A renal HC is difficult to diagnose. The cyst gradually grows and remains asymptomatic for a long time. Hence, it is usually diagnosed in adults, with an average age at diagnosis of 30 years. The most common complaint is flank pain due to cyst compression. In enlarged cysts, the reason for hospital admission may be a palpable mass in the abdomen [3, 4, 10]. In our study, the most common reason for admission was flank pain (38.8%), followed by palpable mass (22.2%), abdominal pain (11.1%), nausea (5.5%), and hematuria (5.5%). There were no clinical symptoms in 16.6% of the patients, and incidental cysts were detected. These findings were consistent with literature.

Although hydatiduria is a pathognomonic finding for an HC, it is very rare. The opening of the cyst to the collecting system occurs when the scolexes pass into the urine. Hydatiduriais is usually associated with renal colic [9, 10]. There are different incidences of hydatiduria in literature; a study by Horchani et al. reported it to be 28% [11] and a study by Göğüş et al. reported it as 5% [4]. Hydaturia was not observed in any of the patients in our study.

There is no laboratory finding that can aid in making a definitive diagnosis of a renal HC. Although IHA and enzyme-linked immunosorbent assay used in the diagnosis are the most sensitive tests, they may provide false positive or negative results. Positive serological tests confirm the diagnosis, whereas negative ones do not rule it out [12]. In a study conducted by Efesoy et al. [6] in isolated renal HC patients, the rate of IHA positivity was 71.4%, and a study conducted by Rexiati et al. [10] stated that the positivity rate of serological tests was 74%. In our study, the IHA positivity rate was 61.1%.

Although imaging plays an important role in diagnosis, there is no specific finding that may aid in making a definitive diagnosis [10]. On US, kidney HCs can be seen as unilocular, multiseptal or calcified cysts, and daughter vesicles can be detected [13]. CT has the highest specificity and sensitivity among imaging methods. Multiple internal septations in the cyst and hypodense areas of daughter vesicles compared with the fluid in the cyst (rosette pattern), calcification in the cyst wall and increased density of the germinative membrane after intravenous contrast agent injection can be detected on CT [13, 14]. US and CT were used as imaging modalities in the diagnosis of renal HCs in all cases in our study.

Renal HC management options include medical treatment, percutaneous intervention and surgical treatments. Mebendazole and albendazole are used in medical treatment. Although these drugs reduce the size of the cyst, they have a low rate of effectiveness. In addition, serious side effects of these drugs, such as hepatotoxicity, allergic reaction, leukopenia and alopecia may be observed. There is not enough data on the effectiveness of medical treatment in renal HC cases [15]. Hence, medical treatment before and after surgical intervention, instead of as primary treatment, is advocated to prevent disease spread [16]. Thus, albendazole 10–15 mg/kg/day administration 1–4 weeks before surgery and its continuation for 1–3 months after the procedure is recommended [4, 17]. We do not recommend medical treatment as primary treatment in our clinic, but we recommend it perioperatively to reduce the risk of recurrence and need for a potential transplant.

The primary treatment for renal HC is surgical removal of the cyst. Here, the aim is to remove the cyst without contaminating the patient. Cystectomy and total or partial nephrectomy are among the surgical options. Anaphylactic shock and death due to allergic reactions may occur in case of HC rupture [18, 19]. Therefore, irrespective of which surgical technique is followed, the surgery should be performed with utmost care. Evacuating the cyst contents and washing the cyst with a scolicidal (hypertonic sodium chloride, 0.5% silver nitrate, 2% formalin and 1% iodine) solution before cystectomy reduces the risk of allergic reaction and implantation [20]. Hence, we used hypertonic sodium chloride as a scolicidal in all patients who underwent cystectomy. We did not observe an allergic reaction intraoperatively in any of our patients.

Renal-sparing surgery should be the first line of treatment in patients with an isolated renal HC. However, nephrectomy is recommended in dysfunctional kidneys, large cysts thought to be associated with the collecting system and cysts suspected to be tumors [3, 4, 21]. Today, kidney loss, the rate of which is 25%, has been reduced due to factors such as an increase in the rate of incidental diagnosis and the ease of access to health services [22]. In the literature, nephrectomy was performed in 13 patients in a series of 20 patients according to a

study published in 2003, whereas it was performed in 1 patient in another series of 30 patients, as reported in 2014 [4, 10]. In our series consisting of 18 patients, 14 kidneys were preserved, 2 patients had non-functional kidneys, and 2 patients underwent nephrectomy due to suspected malignancy. There is no study in literature reporting local recurrence. In our study, which had a mean follow-up period of 27 months, no local recurrence was observed in any patient; however, a new cyst was detected in the ovary of one patient.

Conclusion

Although the HCs are commonly observed in the liver and lungs, they may also occur in other organs, especially the kidney. Isolated renal involvement, although rare, has been observed. In areas where hydatid disease is endemic, HCs should be considered as a diagnosis in patients with renal cysts, even if serological tests are negative. The primary treatment in a renal HC should be kidney-sparing surgery. Nephrectomy should be performed in cases of large cysts opening into the collecting system, non-functioning kidneys and suspected malignancy. To reduce the risk of recurrence and inoculation, albendazole treatment should be recommended before and after surgery.

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