

Hydatid Cyst Treatment and Management in Retroperitoneal Organs; Is Percutaneous Drainage an Option?

Yunus Emre Göger¹, Mehmet Serkan Özkent^{2*}, Mehmet Aykut Yıldırım³, Suleyman Bakdik⁴,
Mehmet Giray Sönmez¹, Cengiz Kadiyoran⁴, Mehmet Balasar¹, Murat Çakır³

Purpose: To evaluate patients who had hydatid cyst (CH) in their retroperitoneal space and organs in order to determine a standard treatment option for CH.

Materials and Methods: The files of 56 patients who were treated for CH in our clinic were evaluated retrospectively. All patients underwent either percutaneous drainage (PD) or surgery. Patients were divided into two groups as PD (Group one) and surgery groups (Group two). Preoperative and postoperative results were compared statistically.

Results: 31 of 56 patients were male. Mean age of the patient was 39.7 (10–85). 16 patients had been treated with PD and 40 with different surgical interventions such as total cystectomy, partial cystectomy, partial nephrectomy, total nephrectomy, surrenalectomy, and laparoscopic partial surrenalectomy. Patients' followed up was 18 months (6-38m). Relapse was seen in 1 patient who underwent PD. On comparing the results, hospitalization period was prolonged in the surgical group with enlarged cyst presence.

Conclusion: CH presence in the retroperitoneal area is rare. PD, a minimally invasive method, has the potential to be the standard treatment option as it can be performed safely in selected patients. However, currently surgical treatment is considered as the first treatment option after CH diagnosis.

Keywords: hydatid cyst; percutaneous drainage; retroperitoneal-placed cyst hydatid; treatment of hydatid cyst disease; minimally invasive surgery

INTRODUCTION

Hydatid cyst (CH) disease is a worldwide zoonosis caused by infection with a small tapeworm parasite called *Echinococcus Granulosus*. Sheep and cattle are intermediate hosts for the pastoral form and humans are an accidental intermediate host⁽¹⁾. Patients with hydatidosis usually present with liver (75%), lung (15%) and other organ involvements (10%)⁽¹⁻³⁾.

CH of kidneys is rare and just 1-5% of all CH diseases in humans involve the kidneys⁽²⁻⁴⁾. It is scarce in the retroperitoneal area⁽²⁾. Patients who refer to clinics with renal CH usually have flank pain, hydraturia, and renal colic^(2,4,5). While pelvic placed small cysts are asymptomatic, bigger pelvic cysts can show symptoms due to the compressions of surrounding organs like bladder and rectum^(6,7). However, cysts placed in retroperitoneal area can be both asymptomatic and can have symptoms such as flank pain^(8,9). Likewise, renal CH also can either be asymptomatic or have symptoms such as hypertension and flank pain⁽⁵⁾.

In treatment of CH disease, treatment options vary according to cyst location, size, and interrelation with surrounding organs. Percutaneous drainage (PD) has an important place in the treatment of liver CH⁽¹⁰⁾. There are small-sized series that show its use in kidneys and the other organs⁽¹⁰⁻¹²⁾.

In cysts of the retroperitoneal area and its organs, despite treatment modalities such as cyst excision, or-

gan-preserving surgery, total excision according to place of the lesions, the first treatment option considered is surgical excision. The present study evaluates CH patients with retroperitoneal area and its organs involvement according to their symptoms, diagnoses, and treatment modalities. Percutaneous drainage in selected patients are compared with surgery.

PATIENTS AND METHODS

Retrospective analyses of the patients referring Necmettin Erbakan University, Meram medicine faculty, urology, general surgery and radiology departments diagnosed with CH with in the past two decades were made. Hereby, cases with unusual cyst location and hepatic involvement were accepted as secondary CH cases and with no hepatic and pulmonary cysts or no history of CH treatment were accepted as a primary cases. Imaging methods such as ultrasonography and computed tomography (CT) were routinely performed in cyst diagnosis. Magnetic resonance imaging (MRI) was performed if considered as necessary. Subsequent to the intervention, computed tomography was performed in the first postoperative month in order to distinguish remaining cyst cavity from recurrent new cysts. Serologic tests were not conducted routinely. The tests were utilized in cases where imaging methods proved to be insufficient in differential diagnosis. Patients were divided into two groups as PD and surgery groups.

¹Department of Urology, Meram Medical Faculty, Necmettin Erbakan University, Konya, Turkey.

²Department of Urology, Konya Education and Research Hospital, Health Sciences University, Konya, Turkey.

³Department of General Surgery, Meram Medical Faculty, Necmettin Erbakan University, Konya, Turkey.

⁴Department of Radiology, Meram Medical Faculty, Necmettin Erbakan University, Konya, Turkey.

*Correspondence: Department of Urology, Konya Education and Research Hospital, Health Sciences University, 42090, Konya, Turkey. Tel: +90 505 9364155. Fax: +90 332 2236416. E-mail: msozkent@gmail.com

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Table 1. Clinical characteristic with hydatid cysts.

Age (mean)		39.7±12.3 (10-85)
Gender	Female	25 (35.5%)
	Male	31 (54.5%)
Contact with animals	No	36 (65.4%)
	Yes	20 (34.6%)
Cyst Localization	Adrenal	3 (9.1%)
	Kidney	31 (50.9%)
	Retroperitoneal	11 (20%)
	Retrovesical	11 (20%)
Clinical symptoms	Back pain	26 (47.2%)
	Upper abdominal pain	19 (34.5%)
	Hematuria	2 (3.6%)
	Hydraturia	4 (7.2%)
	LUTS	5 (10.8%)
	Asymptomatic	8 (14.5%)
Surgery	Hypertension	1 (1.4%)
	Adrenalectomy	1
	Partial Adrenalectomy	1
	Partial Nephrectomy	5
	Nephrectomy	12
	Total Cystectomy	14
	Partial Cystectomy	8
Percutaneous Drainage	16	

Percutaneous Drainage Procedure

The cyst was entered using a 14F (Bioteq, Taipei, Taiwan) trocar with USG or CT-guided trocar method. The contents of the cyst were evacuated as much as possible and irrigated with 0.09% NaCl then aspirated. The vesicles were crushed with negative pressure and aspirated out of the catheter. 20-30% of the aspirated amount was filled with contrast material. The integrity of the cyst and its relationship with the surrounding structure were evaluated by fluoroscopy or CT. The cyst was aspirated again. The cyst was refilled with a mixture of alcohol (98% ethanol, approximately 30-50% aspirated volume) and contrast agent (10% cyst volume). After waiting 20 minutes, the cyst was aspirated again. In large cysts and multiple cysts, the cyst cavity was irrigated with 500 cc saline (0.09% NaCl) in order to prevent the risk of ethanol intoxication in case of not completely draining the given ethanol. It was then left to free drainage. In order not to develop ethanol intoxication, the amount of alcohol was not exceeded 500 cc in any patient during the procedure. Patients treated with PD were hospitalized for 1 day and controlled by USG the next day. If the cyst was completely emptied, the catheter was removed.

Surgical procedure

Hydatid cyst in solid organs such as kidney, adrenal gland, endophytic; If large, multiple and associated with the collecting system, was completely removed by surgery. For exophytic and single cysts, primary

organ-sparing surgery was performed (partial nephrectomy and adrenalectomy). Total cystectomy was performed in all surgically removable cysts. Partial cyst excision was performed if the cyst was adhered to surrounding structures and the total excision could not be performed due to morbidity. During partial cyst excision, serum (20% NaCl) was given into the cyst and it was waited for 10 minutes. Afterwards, the cyst was opened partially, all the contents were aspirated, the cyst wall was removed as far as possible and the procedure was terminated. Preoperative and postoperative results were compared statistically.

The local human research ethics committee approved the protocol 2020/2772 (Necmettin Erbakan University, Meram Medical Faculty Ethics Committee). The analysis and data collection were performed according to the Declaration of Helsinki after written informed consent was obtained from all patients.

Statistical Analysis

SPSS package program was used for statistical analyses of the data obtained from patients' files. Descriptive statistics were used for patient demographic findings. Independent samples t-test was used for parametric data and Mann-Whitney U test was used for non-parametric data with *P* values considered significant at < 0.05.

RESULTS

A total of 56 patients were divided into two groups.

Table 2. Evaluations of patient groups

Characteristics	PD Group (N:16)	Surgical Group (N:40)	<i>p</i> value
Female	6	15	0.18
Male	12	25	
Age	38.8±12.6 (18-55)	40.1±10.3 (10-80)	0.09
Diameter of the cyst	8.6±1.8 (4-16)	15.4±2.3 (4-25)	0.002
Hospital time	1.1±0.9 (1-3)	4.7±1.6 (3-10 days)	0.003

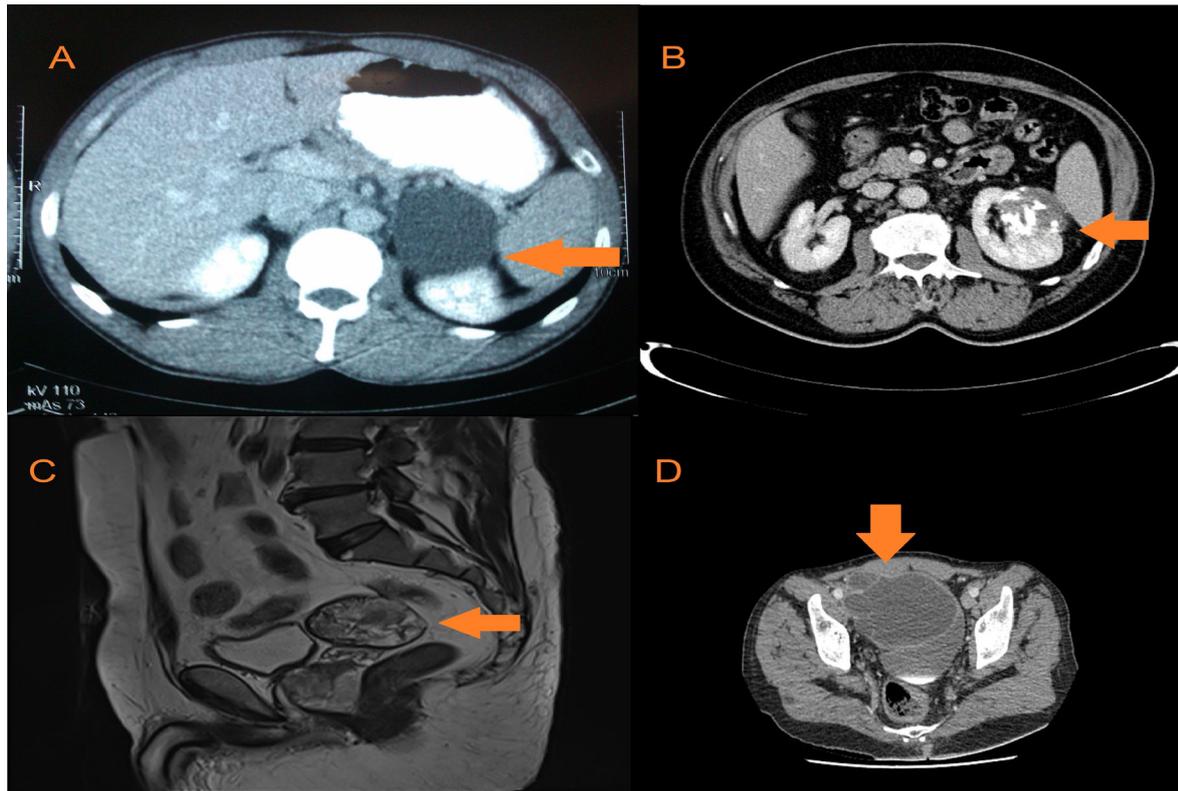


Figure 1. Retroperitoneal organs hydatid cyst (CH). **A:** Adrenal hydatid cyst; **B:** Renal hydatid cyst; **C:** Retrovesical hydatid cyst (MRI image of the cyst from the back wall of the bladder and seminal vesicles); **D:** Retrovesical hydatid cyst (CT image of the cyst from compressing bladder anterior wall and the patient had hydraturia)

16 patients (Group 1) had been treated with PD and 40 with different surgical interventions (Group 2). Out of the 56 patients, 31 were male and 25 female. Patients' mean age was 39.7 ± 12.3 years (10–85 years). Of the 56 patients, 51.9% (31 patients) had renal CH, 9.1% (3 patients) surrenal CH, 20% (11 patients) retroperitoneal-placed CH, and another 20% (11 patients) retrovesical-placed CH. Retroperitoneal organs hydatid cyst (CH) are shown in **Figure 1**. Renal Hydatid cyst different imaging methods, and surgery specimen are shown in **Figure 2**. Out of the renal CH cases, in 17 patients, cyst location was on the left kidney and in 14 on the right. Simultaneous cyst presence on both kidneys could not be determined in any of the patients. Whereas 9 patients with kidney CH, 1 with retroperitoneal-placed CH, 1 with retrovesical-placed CH, 2 with surrenal CH were considered as primary CH, all the others as secondary cyst. Amongst the renal CH patients, simultaneous hepatic CH was determined in 16 patients, CH in liver and lungs in 4 patients. In retroperitoneal CH patients, simultaneous CH in the liver in 8 patients and in the spleen of 1 patient was determined. In retrovesical CH patients' simultaneous spleen and peritoneum placed CH was determined in 1 patient, spleen and liver CH in 1 patient, and CH in the liver of 7 patients. Simultaneous CH in the liver of 2 patients with surrenal CH was also determined.

Among the symptoms present was the most common one back pain with 26 (47.2%) ratio, followed by upper abdominal pain 19 (34.5%), hematuria 2 (3.6%), hydraturia 4 (7.2%), lower urinary tract symptoms 5 (10.8%), palpable mass 8 (14.5%), and hypertension 1

(1.4%) (**Table 1**).

41 patients' indirect hemagglutination (IHA) values were preoperatively controlled and the results were found to be significant in 31 of these patients.

7 patients with renal CH, 4 patients with retrovesical-placed CH, 4 patients with retroperitoneal-placed CH, 1 patient with adrenal CH underwent PD. PD in Retroperitoneal Hydatid cyst is shown in **Figure 3**. Also, PD in Retrovesical hydatid cyst is shown in **Figure 4**. Among the patients who underwent a surgical intervention, total cystectomy was performed in 14 patients, partial cystectomy in 8 patients, partial nephrectomy in 5 patients, nephrectomy in 10 patients, surrenalectomy in 1 patient, and laparoscopic partial surrenalectomy in 1 patient (**Table 2**).

All patients who underwent PD had secondary CH. Medical treatment was administered 1 month before PD and completed to 3 months in total after PD.

7 patients who were operated due to kidney CH and 2 patients who were performed surrenalectomy had primary CH and surgical intervention was decided subsequent to pathology results. In these patients, a 3 month postoperative medical treatment in total was administered.

Mean hospitalization period in Group 2 was 4.7 ± 1.6 days (3-10 days), prolonged ileus emerged in 2 patients who had undergone nephrectomy and in 2 patients who had undergone total cystectomy, and fewer in 6 patients. These patients were discharged after they were followed-up with medical treatment.

Patients of Group 1 drainage were followed-up for only 1 day for control purposes discharged. Drainage cath-

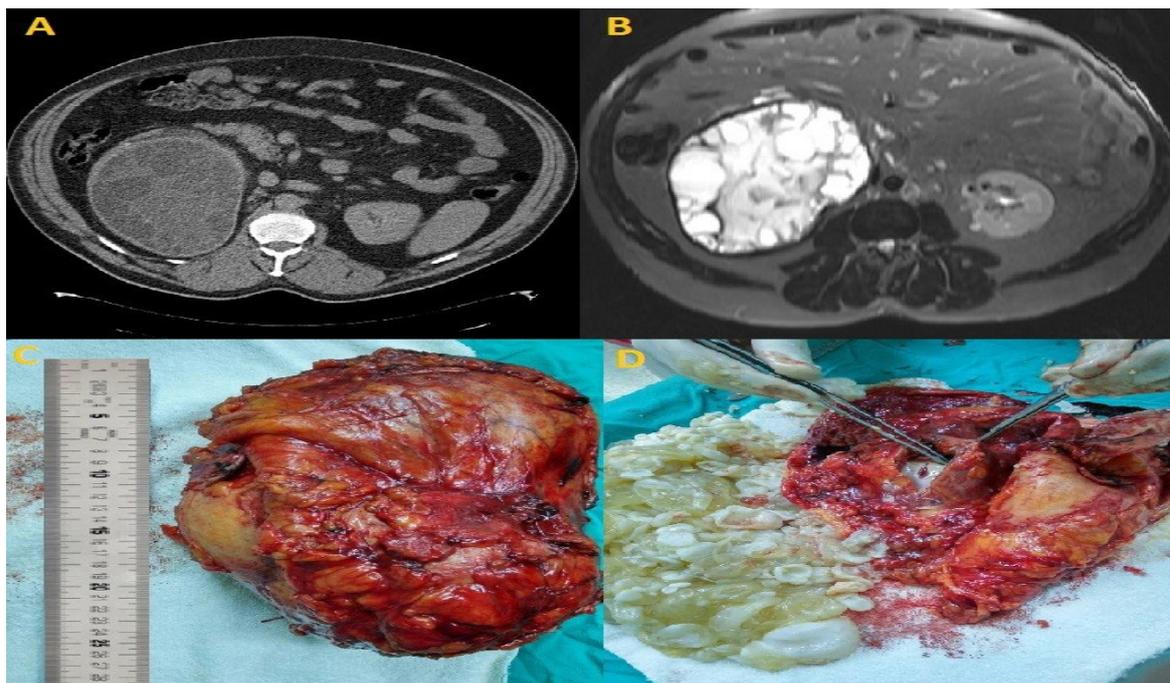


Figure 2. Renal Hydatid cyst different imaging methods and surgery material. **A:** Huge renal hydatid follicle cyst image CT image; **B:** Huge renal hydatid follicle cyst image MRI image; **C-D:** Nephrectomy material and macroscopic follicle image

ter was removed when the drainage stopped after 2nd or 3rd day. Out of Group 1, 1 patient had fever and was discharged with medical treatment after 3 days.

A mean follow up period of 19 months (10-38 months) was determined. Total nephrectomy was performed in renal CH patient subsequent to PD due to relapse detected in 6th post op month.

Comparing Group 1 (PD Group) and Group 2 (surgery group), cyst diameter was longer in the group that underwent Group 2 with a statistically significant difference. Group 1 had a shorter hospitalization period. There was no statistically difference the other parameters.

DISCUSSION

CH is endemic in places where cattle and pig breeding are widely practiced such as South Africa, Australia, New Zealand, South America and a number of Mediterranean countries including Turkey. Location of CH is in the liver with 60-70% ratio, in the lungs 20-25%, in the serosal surfaces 6%, in the kidneys 2.9%, in the spleen and other organs 0.5%, according to the literature⁽¹⁻⁴⁾. Adult *Echinococcus Granulosus* worm lives in the proximal small intestine of the definitive host, attaching by their hooks to the mucosa. Eggs are released into intestine of host and are excreted in the feces. Humans may become intermediate host through contact with a definitive host (usually a domestic dog) or ingestion of contaminated water or vegetables. When embryo passed through the intestinal wall to reach the portal venous system or lymphatic system, the liver acts as the first line of defense and is therefore the most frequently involved organ. It is not clear how hydatid embryo reaches the kidney, retroperitoneum and pelvis in cases of primary hydatid disease but it was postulated that it must pass through the portal system into the liver and retroperitoneal lymphatics⁽¹³⁾. Etiopathogenic

mechanism in CH of pelvis is possible with the primitive blood-graft of oncosphere or hexacanth and a graft in the pelvic pouch of Douglas of protoscolices which come from the cracking of abdominal hydatid cysts^(6, 14). The oncospheres in blood pass through the liver and lungs without seeding and develop an implant in retroperitoneum. Other hypothesis including disseminating through intestinal lymph vessel and thoracic duct⁽¹⁵⁾. Renal and adrenal hydatid cysts usually remain asymptomatic for many years. Patients usually present with complaints of dull flank pain, hematuria, palpable flank mass, hypertension, and renal colic⁽⁵⁾. Our cases applied to our clinic with similar symptoms. Even, there was complaint of hydraturia in 4 patients.

Göğüş C. et al reported 20 patients with renal hydatids in 14 cases on the left and in 6 cases on the right side⁽¹⁶⁾. Rexiati et al noted cyst presence in the left kidneys of 22 patients and in the right kidneys of 9 patients, in their study⁽¹⁷⁾. Likewise, the present study also determined higher cyst sequence in the left kidney.

Retroperitoneal masses are considered secondary to previous rupture of hepatic cysts or spillage of cystic fluid during surgery or puncture, most of the abdominal or pelvic cystic hydatid disease⁽⁸⁾. Primary retroperitoneal hydatid cyst means cysts presented in retroperitoneum independent from any peritoneal viscera, and no simultaneous or previous hydatid disease of other organs⁽⁹⁾. The occurrence of primary retroperitoneal hydatid cyst is extremely rare. Retroperitoneal hydatid cysts can present as abdominal or back pain (31%), palpable mass (65.1%) or urinary tract symptoms (13.9%) depending on the size^(9,18). 8 of 11 patients who had retroperitoneal CH referred to hospital with complaints of back pain and palpable mass. There was no symptom in 3 patients.

Incidence of hydatid disease in surrenal is about 0.5%.

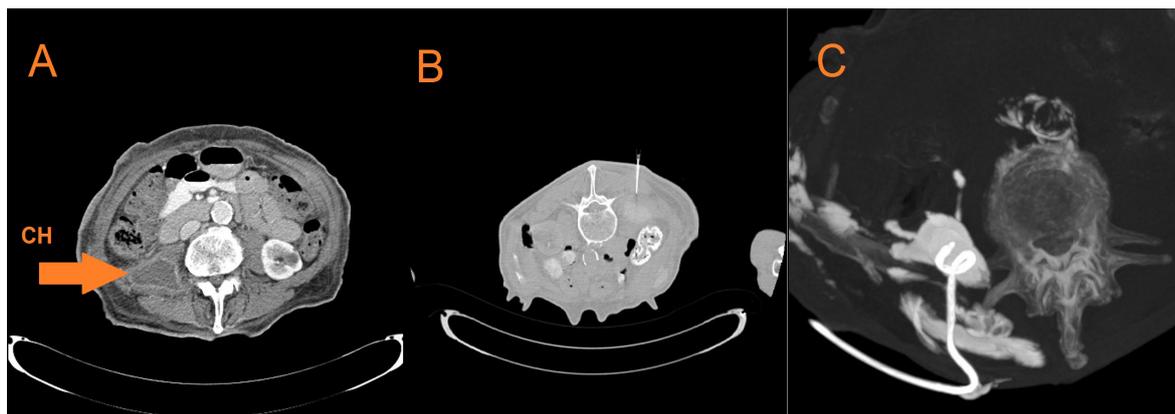


Figure 3. PD in Retroperitoneal Hydatid cyst. **A:** After the operation, the placement of the catheter into the cyst; **B:** Retroperitoneal hydatid cyst accessing needle CT image; **C:** After the PD operation, the placement of the catheter into the cyst

Although they are usually asymptomatic, they can show symptoms such as hypertension, flank pain due to compression of the mass⁽¹⁹⁾. In the present study, one of the 3 patients had hypertension, the other 2 patients had pain. The retrovesical location of hydatid cyst is very rare^(6, 20). It represents 1-2% of Tunisian series and less than 1% of the European series⁽⁶⁾. Patients can apply to hospital with palpable mass in abdomen and dysfunction due to compression on pelvic organs such as the bladder and rectum^(7,14,18). 5 of the 11 patients in the present study applied with lower urinary tract symptoms (LUTS), 3 with abdominal pain, and one of them with vesical fistula.

Serological tests can be utilized in the follow-up of operated cases. Kuru et al. stated that the IHA test had less sensitivity in the pulmonary hydatid cysts and might show false negativities in inactive or calcified cysts and therefore should be supported by another serological method⁽²¹⁾. Some researchers consider IHA sensitivity and specificity of above 90%⁽²²⁾. The IHA values of the 41 patients in the present study were determined and were positive in 31 (75.6%) cases.

Imaging modalities enable differential diagnosis in most of cases. Sonography and CT show a uniloculated or a multiloculated cyst that may have some heteroge-

neous echoes on sonography or have rim enhancement on CT. Daughter vesicles (brood capsules) are small spheres that are formed from rests of the germinal layer and appear as cysts within a cyst. It was stated that MRI in cyst hydatid diagnosis can be helpful for suspected cases because of it shows soft tissue better in solid organ invasions especially⁽²³⁾. Therefore, tomography was widely used for the diagnosis of secondary cases. CT and MRI failed in the differential diagnosis of 12 cyst hydatid patients diagnosed as primary and hence differential diagnosis was verified with pathological results in 5 patients of the present study.

Previous to PD, albendazole treatment is recommended in CH of the liver and lungs, peritoneal CH, and presence of multiple cysts in 1 or more organs, as it reduces cyst tension, contributes to dying and decreasing activity of Echinococcus larvae and in needle aspiration it reduces the risk of planting⁽²⁴⁾. There are studies reporting albendazole treatment efficient success in combined treatments and combined treatment even as more efficient than surgery^(25,26). Likewise albendazole treatment was initiated to 18 patients 10 days before PD performance. A total of 3 months treatment was administered in total continuing after PD. No recurrence, except in 1 case, was seen.

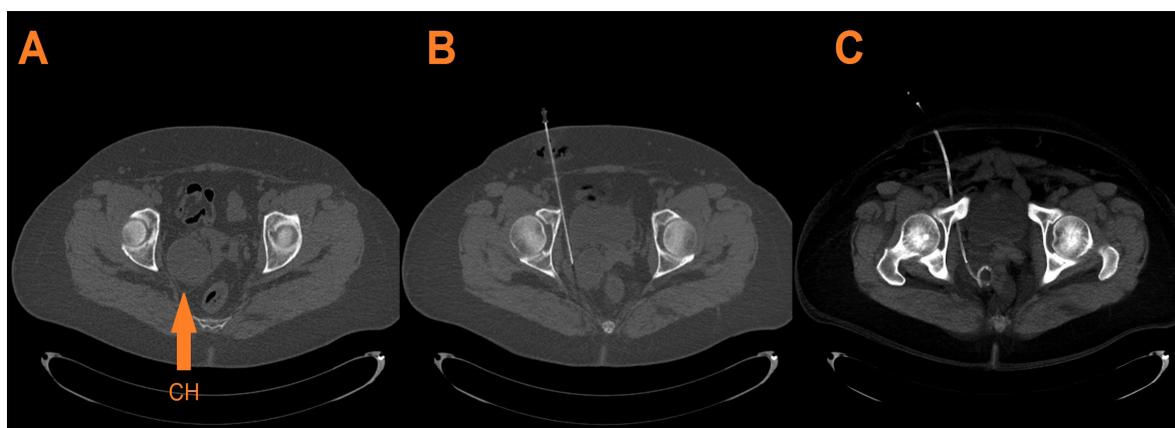


Figure 4. PD in Retrovesical hydatid cyst. **A:** Retrovesical hydatid cyst CT image; **B:** Retrovesical hydatid cyst accessing needle CT image; **C:** After the PD operation, the placement of the catheter into the cyst

Surgical intervention is more on the fore in the treatment of renal cyst hydatids according to the review of the current literature^(16,17,27). However, recent publications show that renal cyst hydatid follow-up with PD is on the increase. Göğüş et al. performed nephrectomy to 13 out of 20 renal cyst hydatid patients and cystectomy to the rest⁽¹⁶⁾. In the study with a patient series of 30, Raxiati et al. performed nephrectomy for renal CH treatment in 1, partial nephrectomy in 1, external capsule excision in 5, internal capsule excision in the remaining 23 cases⁽¹⁷⁾. Ödev et al. performed nephrectomy in 8, partial nephrectomy in 4, cystectomy in 3 of their patients in their seventeen-case study⁽²⁷⁾. In the present study, nephrectomy was performed in 9 out of 30 patients, partial nephrectomy in 5, and total cyst excision in 6 of cases. There were renal cyst hydatid patients who were performed elective PD. PD was performed by Akhan et al to 5 cases, El Kady et al to 4 cases, Goel et al to 4 patients, Baijal et al to 2 cases and they did not report any complications neither during the intervention nor during the follow-up^(11,28-30). Zerem et al. performed PD to 7 renal CH patients in their 72 patient series. 29 patients have developed short-term complications such as urticaria, hypotension, and fever in the aftermath of the intervention; however, no long term complications emerged⁽¹²⁾. In the present study, nephrectomy was performed in 1 case due to relapse during the follow-up of 8 cases who had undergone PD. Although adrenal gland CH was represented generally in the relevant literature as case reports, with surgical excision and within surrenalectomy, but only in 1 case it was followed-up subsequent to draining with puncture^(19,31,32). In the present study, surrenalectomy was preferred in 1 of CH patients, partial surrenalectomy in another 1, and PD treatment to patient with accompanying liver CH. PD drained cyst of patient was 6 cm as it was unilocular and easily differentiated from surrounding tissues. Partial adrenalectomy was decided subsequent to pathological evaluation.

Total cystectomy was performed in 14 patients who have had retroperitoneal, pelvic and kidney cysts that weren't related with surrounding tissue and without neighboring pancreas, duodenum, ureter, bladder and major vessels. Partial cystectomy was performed in the remaining 8 patients. For PD process, into the cyst an injection of a scolical solution (hypertonic saline) for 10 minutes is made and the cyst than drained. Saadi et al mentioned difficulties of extravascular dissection stemming from relation with surrounding organs (ureter, bladder, and gastrointestinal system), and from the relation of cyst with the peritoneum, in their 4-case study⁽⁷⁾. They performed partial cystectomy in 3 of their cases. Khouaja et al performed total cystectomy in 5 and partial cystectomy in 3 cases in their 8-case series⁽¹⁴⁾. Akbulut et al performed total excision to 55.8% of the patients with retroperitoneal CH, partial cystectomy to 44.2%⁽⁹⁾. Ben Abdullah et al performed partial cystectomy in their 9-case retroperitoneal and retrovesical hydatid cyst series and reported no complications⁽¹⁸⁾. Unlike the current literature in the present study PD was applied to 8 cases with retroperitoneal and retrovesical CH. The patients we had undergone PD were all secondary cases. No complications emerged during or after the intervention.

In the present retrospective study, the clinical approach to CH at various sites in the human body except the

liver is evaluated. In CH endemic geographies, Turkey included, CH is to be considered as a differential diagnosis in patients referring to hospitals with C, abdominal pain, and cystic mass in the retroperitoneal area. Moreover, in locations where CH is widespread, organs unlikely to be affected are also to be considered. Cyst hydatid is not a common widespread phenomenon in our clinic; therefore, the first preference is surgery. While organ-preserving surgery (partial nephrectomy, total cystectomy etc.) is commonly preferred, in cases where this is beyond possibility, total excision is performed. Another fact to be considered is that CH can be confused with mass, tumor or cysts during the radiological examination of the cases. The most important underlying reason is the lack of experience because CH presence in retroperitoneal organs is not as common as CH in the liver or lungs.

PD is a minimal invasive method for patients and has advantages such as shorter hospitalization period and decreased postoperative complications emergence (bleedings, fever, ileus etc.). Therefore, PD has the potential to be an efficient treatment modality that can be employed with frequent patient control. The retrospective analyses of patient files revealed shorter hospitalization in PD compared to surgery. Moreover, it can be assumed that first PD and if still necessary subsequently surgery can be practiced in urological CH treatments as currently conducted in liver CH treatment in time with the increase in patient series as the routine treatment modality.

CONCLUSIONS

In spite of rare CH presence in the retroperitoneal area and organs, these can lead to important symptoms and severe disorders in patients. CH should be considered in a differential diagnosis in endemic regions.

Although currently surgical treatment is the first line treatment option considered, PD as a minimal invasive method can be performed in suitable cases. PD comes to the fore due to advantages such as shorter hospitalization and diminished possibility of surgery related complications. The present study is a retrospective study favoring PD to become a standard treatment modality; however, studies with larger patient series are required to determine the first choice treatment modality.

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CONFLICT OF INTEREST

The authors report no conflict of interest.

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