# Isolated Renal Hydatid Cysts: A New Series from an Endemic Area

## Niema H. Al-Heeti

## **ABSTRACT:**

## **BACKGROUND:**

Hydatid disease is a zoonotic infection caused by the larval stage of a tapeworm particularly Echinococcus granulosus. It is considered an important public health problem in many areas of the world. It is one of a serious endemic parasitic diseases in Iraq.

## **OBJECTIVE:**

We report our experience in management of 29 case of isolated hydatid cysts of the kidney over an 18-year period in Anbar Governorate- West of Iraq.

## **PATIENTS AND METHODS:**

Twenty- nine patients with isolated renal hydatid cyst were surgically treated from 1994 to 2011 in Al-Ramadi teaching Hospital, Anbar / Iraq. Clinical, laboratory, radiological data, and surgical findings are analyzed.

## **RESULTS:**

Out of the 29 patients included in this series;15 were males and 14 were females. Mean age was 22.5years. The main clinical symptom was flank pain in 24patients (82.7%), hydatidurea in 3 patients (10.3%), and palpable flank mass only in one (3.5%). The cyst was asymptomatic in one (3.5%). All patients were treated by open surgery. Nephrectomy was performed in one patient, partial nephrectomy in 2, and total cystopericystectomy in 2. All others 24 cases were treated by modified partial cystopericystectomy. No recurrence reported during the follow up period from 19 to 28 months (mean :23 months) in 18 patients, and 11 patients were lost from follow up after 6 months of surgery.

# **CONCLUSION:**

The exact incidence of isolated renal hydatid cyst is unknown in Iraq. It is of equal incidence in both sexes, and more in young age .Preoperative diagnosis could be done depending on ultrasonography ,intravenous urography ,and computerized tomography. The pressure difference between the renal pelvis and inside the cyst will determined the fluid movement at site of communication. Subtotal cystopericystectomy (Modified partial cystopericystectoy) is kidney-sparing surgery with minimal morbidity.

KEY WORDS: cystopericystectomy, renal hydatid cysts.

# INTRODUCTION:

Hydatid disease is a zoonotic infection caused by the larval stage of a tapeworm Echinococcus particularly Echinococcus granulosus<sup>(1)</sup>. The adult form of the parasite lives in the intestinal tract of the canines, where sheep ,cattle ,and human are intermediate hosts<sup>(1,2)</sup>. The disease is relatively frequent in countries where sheep , dogs ,and humans live in close contact<sup>(2)</sup>. Hydatid disease is one of the most serious endemic parasitic diseases in Iraq <sup>(3,4)</sup>. Al-Anber

Governorate in the West of Iraq was thought to be one of the highest endemic area with hydatid disease<sup>(5)</sup>.Despite its high frequency and morbidity, little has yet been done to prevent it in man and eradicate it in animals.

Hydatidosis can be found in all parts of the body .The most affected organs are liver and lungs, and renal involvement develops only in 2% to 4% of all cases (6,7). Kidneys are the most commonly affected organs in the urogential tract (6). Echinococcal larvae may reach the kidney through the blood stream "lymphatics or by direct inoculation (1,7).Renal invasion is mostly due to secondary manifestation of the disease

Department of Surgery ,College of Medicine, Anbar University, Anbar Governorate-Iraq.

usually with involvement of other viscera<sup>(8)</sup>.Isolated renal involvement is extremely rare even in endemic areas (up to 2% of patients)<sup>(9)</sup> .In this series we present our experience with 29patients with isolated renal hydatidosis in endemic area during 18years.

## **PATIENTS & METHODS:**

A total of 29 patients with isolated renal hydatid disease were treated between 1994 and 2011 in Al-Ramadi ,Anbar **Teaching** Hospital Governorate / Iraq. The medical records of these patients were retrospectively reviewed .Investigations included a history, physical examination, urinalysis, complete blood count, blood biochemistry,. No serology tests were done because they are not available .All patients underwent radiologic evaluation with plain xultrasonography(US), intravenous urography(IVU),and computed tomography (CT). Magnetic resonance imaging (MRI) study was done in 3 patients only.

All patients were treated by open surgery through retroperitoneal approach by lumber incision. We performed nephrectomy in one patient, partial nephrectomy in 2 and total cystopericystectomy in 2 patients while all others 24 cases were treated by modified partial cystopericystectomy. This procedure includes: the kidney specially near the cyst was mobilized, packing of surounding area with 10% povidone iodine soaked sponges ,injection with 10% povidone iodine and aspiration of cyst ,opening of the cyst, evacuation of all daughter cysts and inner germinal layer of cyst, excised all pericyst outside the renal tissue with the level of renal parenchyma leaving its part which only adheres to renal parenchyma, and scrap the surface of this part by surgical knife to remove most of its thickness specially at site of connection to calvseal system to refresh its edges .Closure the site of connection was done in 14 cases using catgut suture ,and left unclosed in 2. A closed tube drain was placed for 7days except in one patient who had a continuous urinary leakage .He was treated with a double-J stent that improved leakage within one day.

## **RESULTS:**

Out of the 29 patients included in this series;15 were males and 14 were females(male to female ratio 1.07:1). Mean patients age was 22.5 years (range 12 to 51). Twenty-two patients (75.9%) lived in rural areas and were farmers, while the

others 7 patients (24.1%) lived in the centers of the cities and had various occupations.

The main clinical symptom was flank pain which is found in 24patients(82.7%); of them 17 patients had only flank pain, and 2patients had fever, 2patients had palpable mass, 2 patients had generalize ill heath and anemia, and one had macroscopic hematuria with flank pain. Three patients were presented with hydatidurea(10.3%), and one with palpable flank mass only(3.5%). In one patients the cysts were asymptomatic (3.5%) and was discovered during ultrasonographic examination for other disease.

Eosinophilia was detected in 5 patients (17.2%).Plain abdominal x-ray revealed a peripheral curvilinear calcification in only 3 patients(10.3%). Intravenous urography revealed a space occupying lesion and caliceal distortion or caliceal ectasia in 26 patients, pushing of kidney down ward in 2patients due to rather small upper pole cyst caused little calyceal distortion and nonfunctioning kidney in one case due to communicated cyst with pyonephrosis. US showed a complex cyst in 28 patients and unilocular cyst with thick wall in one. C T showed multivesicular cysts in 25cases complex cyst in 3 cases and simple cyst with thick wall in one .MRI was done in above 3patients with complex cysts and did not add any special findings to CT to aid in diagnosis. Diagnosis of hydatid cyst was made in all cases preoperatively with high suspicion.

All cysts were unilateral and solitary .The hydatid cyst was on left side in 17(58.6%) patients and on right in 12(41.4%). Upper pole of kidney was involved in 7patients, lower pole in 12, and middle part in 9cases; 4 cases mainly at anterior surface and 5 at posterior surface. The entire kidney was involved in one case. Communicated cysts were seen in 19 patients and in all the cyst connected to minor calyex. In 3cases of them there are daughter cysts in the pelvio-calyseal system and presented with hydatidurea ,and In 2 of them the cyst was complicated by pyogenic infection .In other 16 cysts, there were changes of daughter cysts inside the parent cyst to more deep yellow to light green color and be shrink and more viscous specially in area near the communication site, while the daughter cysts away from this site remain white to light yellow color.

Three patients had superficial wound infection. No mortality was reported. Hospital stay ranged from 3 to7days with average (4.6 days). All of the patients were treated with oral albendazole,400mg twice a day for 3 months postoperatively although 13 patients did not taking the drug regularly.

Pathological examination confirmed the diagnosis of hydatid cyst in all patients. Radiological assessment showed improvement in all cases after 3 to 6 months. No recurrence reported during the follow up period from 19 to 28 months(mean :23 months) in18 patients, and 11 patients were lost from follow up after 6months of surgery.

## **DISCUSSION:**

Hydatid cyst is parasitic infection caused mainly by the larval form of Echinococcus granulosus. The adult worm of is present in the dog's small intestine. Humans can be accidental intermediate host<sup>(1)</sup>·Humans may ingest echinococcal eggs excreted in the feces of dogs contaminate grass ,farmland and water. The larvae hatch, penetrate venules in the wall of the duodenum and are carried by the portal circulation to the liver, which becomes infected most frequently (70%). The larvae that enter the systemic circulation may lodge in lung(25%),kidney(2-4%) or other  $\operatorname{organ}^{(10,11)}$ .It is not clear how the hydatid embryo reaches the kidney in cases of primary hydatid disease but it is postulated that it must pass through the portal system into liver and retroperitoneal lymphatics. Secondary involvement due to hematogenous dissemination may be seen from other organ<sup>(12)</sup>. In Iraq; the exact incidence of isolated renal hydatid cyst is unknown because of lack of epidemiological survey, accurate statistic and unreliable hospital records. In world literature the incidence ranges from 1.28% to 4%<sup>(10,13)</sup>.It is usually encountered between the ages of 30 and 50 years, and rarely seen in children. (11) In most literature from endemic areas the age tend to be lower (7,14). In our series mean patients age was 22.5 years (range 12 to 51). Equal incidence of renal hydatid cyst in both sexes was seen in most of literature from both endemic<sup>(7)</sup> and non-endemic<sup>(11)</sup>areas, similar to our results ,although some literature showed increase cases in male significantly (14,15) and few literature with increase incidence in female. (16) Really in the rural areas of Iraq, humans from both sexes and all age groups had

chances to get infection with this disease equally specially younger age groups ,as they are the main employers in the farms with poor health hygiene.

The clinical presentation of our cases is not specific to suggest the diagnosis of renal hydatid disease except hydatiduria as in other published series. The main presentations are flank pain palpable mass ,hematuria ,malaise ,fever and hydatiduria. (2,10,15) Complications of hydatid cyst such as infection, hemorrhage, and obstruction of pelvicaliceal system are also present in some cases<sup>(10)</sup>.In our series the main clinical presentation was lumber pain which was present in 24 patients(82.7%). Hydatiduria is urinary excretion of microscopic scolexes macroscopic membranes and / or daughter cysts, (17) typically passage of a grape-like material in the urine. If all three layers of the cvst have ruptured resulting in free communication with the calyces, it is called an opened or communicated cyst<sup>(18)</sup>. Hydatiduria is seen in only 10-20% of renal hydatidosis and is usually microscopic (19). Gross passage of daughter cysts is rather rare dramatic presentation(20) ,and has tremendous diagnostic utility<sup>(21)</sup>.Uncommonly hydatiduria causes acute renal pain or ureteric colic (19). Hydatiduria was seen only in 3patients(10.3%) in our study, none of them with acute renal pain.

In our series, esinophilia was detected in 5 patients(17.2%). It is detected in 20% to 50% of patients with renal hydatidosis ,but its detection cannot help much since false positive results may develop in other parasitic diseases<sup>(22)</sup>. Many immunodiagnostic tests have overall sensitivity against the hydatid crude antigen. Negative or positive results do not exclude or confirm the diagnosis and do not correlate with pathological stage of renal hydatid disease (22). Positive tests may suggest further workup for hydatid disease. Radiological investigations have a more important role in the preoperative diagnosis of renal hydatid disease. On plain x-ray peripheral curvilinear , multilaminated or amorphous calcifications can be observed (10). They are nonspecific, (7) and could be seen in 62% of patients. (10) in our series calcifications were seen only in 3 (10.3%). The IVU findings in our series were similar to results in all previous studies. A nonfunctioning kidney or a filling defect in the renal pelvis may rarely be evident on IVU<sup>(10)</sup>.Ultrasonography is usually the

primary radiological investigation and usually demonstrates the typical appearance of a hydatid cyst<sup>(23)</sup>or the daughter cysts<sup>(7)</sup>. The cyst may be unilocular or multivesicular<sup>(10)</sup>.CT is more accurate and sensitive<sup>(10,15)</sup>.It may demonstrate a cyst with a thick or calcified wall, a unilocular cyst with detached membrane, a multiloculated cyst with mixed internal density, and daughter cysts with lower density than the mother cvst<sup>(12,23,24)</sup>.The MRI shows the cysts adequately ,but MRI offers no real advantage over CT scan<sup>(25)</sup>.Our results in this series did not differ from these facts. However ,hydatid cysts with simple cyst appearance may cause serious diagnostic problems<sup>(26)</sup>. The disease may also mimic benign or malignant tumors, abscesses, infected cyst, and other renal space occupying lesion<sup>(17)</sup>. Our series shows that, in endemic country preoperative provisional diagnosis of isolated hydatid cyst could be achieved with high suspicion depending on ultrasonography, IVU, and CT. So most of patients went into surgery with primary diagnosis of renal hydatid cyst. Nevertheless in non-endemic areas ,half of the patients went into surgery with primary diagnosis of renal tumors (11)

Although the middle portion of kidney is not frequently involved (17), our study showed that no part of the kidney was immune against this

In 16 communicated cysts, there are physical changes of daughter cysts inside the parent cyst in area near the connection site . These findings could be explained depending on pressure difference between the renal pelvis and inside the cyst. This difference are determined mainly by osmotic pressure in hydatid cyst fluid and urine ,and presence or absence of obstruction distal to the site of communication. The hydatid fluid is produced by the germinal layer and it is under pressure varying from organ to organ<sup>(3)</sup>. This fluid is having a specific gravity containing 1.007-1.015 and albumin<sup>(3)</sup>, while the specific gravity for urine is 1.002-1035 and the osmolalitry of urine may reach 1400mosm/kg of water (27). The hydatid fluid contains some protein<sup>(3)</sup>. The osmotic pressure inside the cyst is determined by protein particle and electrolytes mainly sodium. This pressure difference between the renal pelvis and inside the cyst will determined the fluid movement at site of communication. This fluid movements was responsible for the physical

changes noticed in the daughter cysts at site of communication. This may explain that not all communicated hydatid cyst presented with hydatidurea,( in our study only 3 out of 19 communicated renal cysts).

Treatment of renal hydatid cysts is essentially surgical<sup>(11)</sup>.Kidney-sparing surgery is performed whenever possible<sup>(28-30)</sup>.The surgeon must be careful to totally remove the cyst and avoid spilling its contents, as this Spilling may cause severe anaphylactic reaction even death, (10) and dissemination of the disease. Partial or total nephrectomy is required if a cystectomy is not feasible, (11) or preoperative diagnosis is not settled<sup>(7)</sup> (like suspicion of malignancy<sup>(11)</sup>),and if function of the kidney affected<sup>(14)</sup>.Destruction of renal parenchyma may caused by pressure from the cyst, (31) or infection with obstruction by daughter cysts<sup>(17)</sup>. For the last reason we did nephrectomy in one case only in this series. Partial nephrectomy was done in two patients, both with presence of daughter cysts inside the collecting system. In two cases total cystopericystectomy was done as a safely cleavage plane has been obtained between cyst and normal renal tissue without opening the cyst. All others 24 cases were treated by partial cystopericystectomy (modified procedure). This procedure beside it is kidney-sparing surgery, it was included nearly removal of all diseased tissues and provided chance to close communication between the cyst and collecting system.

This will decrease the possibilities of; post operative deep infection ,prolonged urine leak, and the need for drains for long time. In two patients the connection couldnot be closed due to techniqual difficulties from adherence of upper pole cyst to diaphragm. One of them had continues urine leak which was treated by double J ureteric stenting successfully. This procedure could be named subtotal cystopericystectomy rather than partial cystopericystectomy as nearly all cyst related tissues were removed.

Although there is a risk of dissemination and anaphylactic reaction, the technique percutaneous aspiration injection and aspiration has been described as safe and effective treatment for renal hydatid cyst<sup>(32)</sup>.Recently few reports of laparoscopic treatment of hydatid cyst were published. Rabii et al<sup>(33)</sup> reported first case of laparoscopic hydatid cystectomy. Basiri et al<sup>(34)</sup>reported first

case of laparoscopic partial nephrectomy and laparoscopic retroperitoneal or transperitoneal nephrectomy also had been reported for treatment of renal hydatid cyst. Laparoscopic treatment is feasible safe and as effective as its open counterpart safe.

## **REFERENCES:**

- White AC Jr, Weller PF. Helminthic infections. Cestodes in :Kasper DL, Braunwald E, Fauci AS, Hauser SL, Longo DL, Jamesom AL, editors. Harrison's principles of internal medicine. 16th ed. New York:McGrow-Hill;2005:1253-72.
- **2.** Horchani A, Nouira Y, Kbaier I, Attyaoui F, and Zribi AS. Hydatid cyst of the kidney. A report of 147 controlled cases. Eur Urol 2000;38:461-67.
- **3.** AL-Hashimi HM .Hydatid disease in abdominal organs.1st ed. Amman, Jordan ,A1-Essraa Hospital, 2007; chapter2:8 and chapter4:26.
- **4.** Nazar B.Elhassani, The neglected disease, Journal of Kurdistan medical society-Kirkuk 2004;1:6-7.
- **5.** Al-Hadithi T.S. Hydatid disease in Western region of Iraq , J Comm Med 1993;6:11-18
- **6.** Kilciler M, Bedir S, Erdemir F, Coban H, Sahan B, Ozgok Y. Isolated unilocular renal hydatid cyst: a rare diagnostic difficulty with simple cyst. Urol Int. 2006;77:371-74.
- **7.** Gogus C ,Safak M, Baltaci S, Turkolmez K. Isolated renal hydatidosis: experience with 20 cases. J Urol ;2003;169:186-89.
- **8.** Handa R, Harjal MM. Hydatid cyst of the renal pelvis. Pediatr Surg Int ,2005;21:410-12
- **9.** Zayed AM, Abdel Ruhman AH. Isolated hydatid cyst of the kidney. Diagn Surg Treat AJU 2005;3:6-12.
- **10.** Angulo JC, Sanchez-Chapado M ,Diego A, Escribano J, Tamayo JC, Martin L. Renal echinococcosis: clinical study of 34cases.1997; J Urol:157:787-94.
- **11.** Mokhtar AA,Sayyah AA, Al-Hindi H,Seyam RM, AlKhdair W. Isolated renal hydatid disease in a non-endemic country:a single center experience.Can Urol Assoc J. 2011.DOI:10.5489/ cuaj.10049.

- **12.** Pedrosa I, Saiz A, Arrazola L, Ferreiros J, Pedrosa CS. Hydatid disease: Radiologic and pathologic features and complication. Radiographics 2000;20:795-817.
- 13. Pezeshki A,Kia EB, Gholizadeh A,Koohzare A.An analysis of hydatid cyst surgeries in Tehran Milad hospital ,Iran, during 2001-2004.Pak J Med Sci 2007;23:138-40.
- **14.** Kalinova K, Usunov k. Primary renal Echinococcosis –experience with 14 cases. Journal of IMAB,2007;1:5-7.
- **15.** Zargar-Shoshtari M, Shadpour P, Robat-Moradi N, Moslemi M. Hydatid cyst of urinary tract:Eleven cases at a single center.2007;Urol J:4:41-45.
- **16.** Efesoy O, Tek M, Erdem E, Bozlu M. Treatment of isolated renal hydatid cysts with cyst excision and omentoplasty.2010;Turk J Urol:36:176-81.
- 17. Ciftci H, Ozgonul A, Savas M, Verit A, Yeni E .The rupture of hydatid cyst into the renal pelvis:macroscopic hydaturia. Harran Universitesi Tip Fakultesi Dergisi.2008;5:61-63.
- **18.** Mongha R, Narayan S,Kundu AK. Primary hydatid cyst of the kidney and ureter with gross hydatiduria: A case report and evaluation of radiological features. Indian J Urol;2008;24:116-17.
- **19.** Unsal A, Cimentepe E, Dilmen G, Yenidunya S, Saglam R .An unusual cause of renal colic: Hydatiduria. Int J Urol 2001;8:319-21.
- **20.** Loiwal V, Gupta P .Hydatiduria . Indian Pediatr 2000; 37:111-2.
- **21.** Mudholkar VG, Suwarnkar SV, Deshpande SA, Ksdam PN .Isolated renal hydatid disease with gross hydatiduria. Indian J Pathol Microbiol 2011;54:640-1.
- 22. Shukla S,Singh SK,Pujani M. Multiple disseminated abdominal hydatidosis presenting with gross hydatiduria: a rare case report. Indian J Pathol Microbiol;2009:52:213-4.
- **23.** Turgut AT, Altin L, Topcu S et al .Unusual imaging characteristics of complicated hydatid disease . Eur J Radiol 2007;63:84-93.
- **24.** Turgut AT, Odev K, Kabaalioglu A, et al .Multitechnique evaluation of renal hydatid disease. AJR Am J Roentgenol 2009;192:462-67.

- **25.** Volders WK, Gelin G, Stessens RC. Hydatid cyst of the kidney: Radiologic-pathologic correlation. Radiographics 2001;21:S255-60.
- **26.** Engin G, Acunas B, Rozanes I, Acunas G. Hydatid disease with unusual localization. Eur Radiol 2000;10:1904-12.
- 27. Sylvia S.Mader.urinary system and excreation, chapter 10; in Human Biology ,10th ed, New York: MacGraw Hill.2008:196.
- **28.** Ameur A ,Lezrek M, Boumdin H, et al. Hydatid cyst of the kidney based on a series of 34cases. sProg Urol 2002;12:409-14.
- **29.** Zmerli S, Ayed M, Horchani A, Chami I, EI Ouakdi M, Ben Slama MR. Hydatid cyst of the kidney: diagnosis and treatment. World J Surg 2001;25:68-74.
- **30.** Fazeli F, Narouie B, Firoozabadi MD,et al. Isolated hydatid cyst of kidney. Urology 2009;73:999-1001.
- **31.** Yilmaz Y, Kosem M, Ceylan K, et al. Our experience in eight cases with urinary hydatid disease: a series of 372 cases held in nine different clinic. Int J Urol 2006;13:1162-65.
- **32.** Akhan O, Ustunsoz B, Somuncu I. Percutaneous renal hydatid treatment: long term results. Abdom Imaging 1998;23:209-
- **33.** Rabii R, Mezzour MH, Essaki H, Fekak H, Joual A, Meziane F, Laparoscopic treatment for renal hydatid cyst .J Endourol. 2006;20:199-201.
- **34.** Basiri A, Nadjafi-Semnani M, Nooralizadeh A. Laparoscopic partial nephrectomy for isolated renal hydatid disease .J Endourol. 2006;20:24-26.
- **35.** Khan M , s Nazir S , Ahangar S, Farooq Qadri S, Ahmad Salroo N. Retroperitoneal laparoscopy for the management of renal hydatid cyst .Int J Surg. 2010;8:266-68.
- **36.** Kartik J. Shah, Arvind P. Ganpule, Mahesh R. Desai. Isolated renal hydatid cyst managed by laparoscopic transperitoneal nephrectomy. Indian J Urol.2009;25:531-33.