CASE REPORT

Renal failure: unusual clinical presentation of an isolated intrarenal hydatid cyst

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drmehwash7@gmail.com**SUMMARY**

Cystic echinococcosis or hydatid disease is a zoonotic parasitic infection caused by larval stage of cestode *Echinococcus* spp. Humans are an accidental intermediate host and present with the involvement of different organs, the liver being the most common while kidney is the rarest site. Here, authors are reporting a case of a 55-year-old woman who presented with renal failure and was found to have a right-sided intrarenal hydatid cyst, diagnosed and treated with nephroscopy without sacrificing the kidney. She had an uneventful postoperative course and her renal function improved dramatically. She is now on oral albendazole therapy and doing well on follow-up. To the best of our knowledge, renal failure has never been reported in the literature as clinical manifestation of intrarenal hydatid cyst in adult population.

BACKGROUND

Cystic echinococcosis is endemic in many parts of the world, especially in developing regions where stock-breeding is a major industry.¹ It is caused by infestation of the larval stage of the cestode *Echinococcus* spp (*Echinococcus granulosus*, *Echinococcus multilocularis* and *Echinococcus vogeli*). Humans are an accidental intermediate host and commonly present with involvement of the liver followed by the lungs (~75% and 15%, respectively). Intrarenal hydatid disease is a rare disease (2%) with a spectrum of clinical presentation.

In this report, we are presenting an intrarenal hydatid cyst as the cause of renal failure in adults that has not been previously reported in the literature to the best of our knowledge.

CASE PRESENTATION

We are presenting a case of a 55-year-old woman, a resident of Baluchistan with no known comorbidities who presented in our clinic with symptoms of right flank pain for the past 10 months, which was non-radiating, mild in intensity and colicky in nature. She was also reporting with loss of appetite with nausea and occasional episodes of vomiting for the past 1 month. There was no associated lower urinary tract symptom but on the day of presentation she reported an episode of painful haematuria without clots. She was a housewife with one child and had a hysterectomy 5 years ago due to uterine fibroids. She had no addiction and no exposure to animals. The rest of the history was unremarkable. On examination she was pale with no gross lymphadenopathy. Her heart rate was 110 bpm, blood pressure 110/70 mm Hg and was afebrile. An abdominal examination was unremarkable except

for the presence of a Pfannenstiel incision. A systemic examination was normal. She was admitted for a workup and further management.

INVESTIGATIONS

Her serum creatinine was 11.9 g/dL (normal value: 0.6–1.1) while haemoglobin was 8 g/dL (normal value: 11.1–14.5 g/dL). Electrolytes were normal, urine detailed report showed pH of 6.5, nitrite: positive, leucocytes >20/high power field (hpf) and red blood cells >20/hpf while a urine culture was positive for growth of *Escherichia coli*, sensitive only to imipenem. X-ray of the kidney, ureter and bladder was normal while an ultrasound showed the left small kidney without signs of obstruction and the right kidney with an obstructing renal pelvic stone of 2 cm causing moderate hydronephrosis (figure 1). Unenhanced CT of the kidney, ureter and bladder (CT KUB) redemonstrated these findings with an addition of a proximal ureteric stone on the left side (figure 2). There was no evidence of any cystic lesion in the kidney on any of these investigations.

DIFFERENTIAL DIAGNOSIS

On the basis of intrarenal density seen on CT KUB, the cysts was initially considered as renal stones, but intraoperatively the presence of cystic structures embedded in the renal parenchyma raises the suspicion of a hydatid cyst. The other differential could be a parapelvic cyst that can cause obstruction and renal failure but has characteristic radiological features unlike in our patient.

TREATMENT

The patient was admitted, started on intravenous antibiotic imipenem 500 mg every eight hourly

Figure 1 Sonographic image of the right kidney with moderate hydronephrosis and posterior acoustic shadowing raising possibility of a stone.

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To cite: Nadeem M, Biyabani SR, Pervez S. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-200616

Figure 2 CT of the kidney, ureter and bladder demonstrating a right renal stone of ~2 cm while the left kidney is small in size with normal parenchyma.

according to culture and sensitivity. Percutaneous nephrostomy (PCN) was placed in the right kidney and purulent urine evacuated from the system was sent for culture and sensitivity. The patient was discharged home with PCN after 3 days and plan for surgery after a week of antibiotics. Creatinine on discharge was 5.9 mg/dL. After a week she underwent left ureterorenoscopy (URS) and right percutaneous nephrolithotomy (PCNL). On retrograde pyelography, filling defects were seen in the left proximal ureter and the right renal pelvis with dilation of the right pelvicalyceal system. Left URS was performed and a soft, rounded material was taken out with a grasper. The right PCN tract was dilated up to 30 Fr and on nephroscopy, the white coloured material was seen in the system (figure 3). An attempt was made to fragment it with pneumatic lithoclast and ultrasound but both failed, so the grasper was introduced to manipulate it. Soft cystic material in abundance came out with ease. There was no vital or haemodynamic instability during the procedure. The right renal pelvic system was completely emptied and the specimen was sent for histopathological evaluation along with blood sample for Indirect haemagglutination (IHA) titres for *Echinococcus* due to intraoperative suspicion of renal hydatidosis. IHA titre was <1:16 (negative) but histopathological findings were suggestive of hydatid cyst (figure 4).

OUTCOME AND FOLLOW-UP

She had an uneventful postoperative course with significant improvement in renal function (Cr=3.2 mg/dL).

On follow-up, she was started on oral albendazole for 3 months which she is tolerating well. Creatinine is now stable at 2 mg/dL at 10 months follow-up.

Figure 3 Nephroscopic view showing intrarenal cysts. The ureteric catheter could be visible along side.

DISCUSSION

Echinococcosis constitutes major health problems² and is tagged by the WHO as one of the five major 'Neglected Zoonotic Diseases' in the 'First WHO report on neglected tropical diseases'.³

The disease involves different organs and presents with various clinical manifestations.⁴ The most common site of involvement is the liver (75%), followed by the lungs (15%) and the peritoneum. Isolated renal involvement is rare (2–3%) with often subtle clinical signs.⁵ It may present with gradual development of waist or

Figure 4 Cyst wall comprising of an avascular eosinophilic refractile chitinous lamellated membrane beneath the germinal layer.

abdominal masses accompanied by lower back pain. Other modes of presentation are ureteropelvic junction obstruction,⁶ haematuria, persistent fever or hydatiduria,⁷ which is caused by rupture of the cyst in the collecting system. It is a pathognomonic sign, though seen in only 10–20% of renal hydatidosis and is usually microscopic.⁸ Renal failure secondary to tubulointerstitial cystitis in the patient with isolated renal hydatid cyst has been reported by Nickavar,⁹ which was treated by a combination of albendazole, corticosteroids and total nephrectomy. In our patient, obstruction due to an intrarenal hydatid cyst was the cause of renal failure, which was successfully managed with renal preservation and albendazole.

Even in endemic areas, preoperative diagnosis of renal hydatid disease is difficult due to lack of specific laboratory findings. Presence of eosinophilia is non-specific while Casoni's skin test is considered as an outdated investigation.¹⁰ Serological and haemagglutination tests offer low reliability, but their positivity confirms the presence of active disease. Counter immunoelectrophoresis against arch-5 has been reported to be highly specific for hydatid disease (79%).¹¹ Plain roentgenogram is often unrevealing but ultrasound may demonstrate 'falling flake sign' which is a pathognomonic sign of renal hydatid disease.¹² A typical CT finding is demonstrated with the presence of a thick-walled multiloculated cyst with mixed internal density, and daughter cysts with lower density than the maternal matrix. It also helps in demonstrating involvement of other organs.⁴ Our patient did not have any of these characteristic findings on the X-ray, ultrasound or CT scan rather presence of an echogenic material with posterior acoustic shadowing on ultrasound leads us to the diagnosis of a renal stone (with supporting findings on CT). Serology was also negative and the diagnosis was made on the basis of intraoperative findings. We have reported a somewhat similar case where the intrarenal hydatid cyst was found incidentally during stone removal in PCNL.¹³ In this case, it was the intrarenal hydatid cyst and not the stone causing the obstruction. Surgery is the mainstay of treatment where ~75% of the cases are managed with renal conserving surgery while remaining with radical surgery.¹⁴

Preoperative diagnosis of hydatid disease is highly desirable. Both preoperative and postoperative therapies with albendazole, which is benzimidazole anthelmintic, is recommended. One month preoperative treatment reduces the larval activity while a 3-month postoperative course reduces the chance of infection.¹⁵

Renal hydatid disease may present with unusual clinical manifestation causing diagnostic difficulty. A high index of suspicion should be maintained while dealing with complex cysts and once renal hydatid disease comes in the list of probable diagnosis, careful dissection with the intraoperative use of hypertonic saline should be performed as complete removal can be achieved with percutaneous treatment in selected patients.

Learning points

- Renal hydatid disease is a rare pathology with a spectrum of clinical presentation requiring a high index of suspicion for appropriate early diagnosis.
- Thorough clinical evaluation with serology and imaging establishes diagnosis.
- Surgery with renal conservation is a valid and successful option in selected patients.

Contributors MN contributed in the writing of case report and literature search. SRB contributed in writing the discussion and proof read the manuscript. SP provided photomicrographs and description of histopathological slides.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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