A rare case of primary renal hydatid cyst presenting with hydatiduria

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Abstract Hydatid disease caused by *Echinococcus granulosus* involves liver in 75 % of cases and lung in 15 % of cases. But primary renal involvement without liver and lung involvement is a very rare event. Among these patients of renal hydatidosis, hydatiduria is seen only in 10–20 % cases, which is considered to be pathognomonic. We report a case of echinococcosis, primarily involving the right kidney, presenting with clinical history of hydatiduria.

Keywords Primary renal hydatid ·
Echinococcus granulosus · Hydatiduria

Introduction

Echinococcosis is a worldwide zoonosis produced by the larval stage of the *Echinococcus granulosus* tapeworm. In humans, hydatid disease involves the liver in approximately 75 % of cases and the lung in 15 %. Secondary involvement due to hematogenous dissemination may be seen in almost any anatomic location (Pedrosa et al. 2000). Kidney involvement in echinococcosis is extremely rare (2–3 % of cases), even in areas where hydatid disease is

endemic (Afsar et al. 1994) and primary involvement of the kidney without the involvement of the liver and lungs is even rarer. Only 10–20 % cases of renal hydatidosis present with hydatiduria, which is usually microscopic (Gogus et al. 1991). Here we present a rare case of primary right renal hydatid cyst which was first suspected from history of hydatiduria.

Case report

A man of 58 years presented with dull aching pain in right flanks for last 5–6 months followed by occasional passage of small, pearly white grape like structure and granular substances per urethra during micturition for last 4 months. He had also experienced pain due to temporary retention of urine and usually got relief on passing granular substance. On examination there was palpable renal mass, hypertension and mild pallor. Initially, blood picture showed marked eosinophilia and USG whole abdomen showed heterogeneous space occupying lesion at the upper pole of right kidney (Fig. 1A), along with echogenic debris within bladder lumen (Fig. 1B). Liver and other organs were normal. Chest X-ray did not show any abnormality. CT scan of abdomen showed a cystic lesion in the right kidney. The whole right kidney was replaced by the cystic mass (Fig. 1C). There was no contrast excretion from the right kidney. Cystic renal mass was removed enbloc. Pre and postoperative albendazole were given. The cystic mass was sent for histopathological examination. Gross pathologic examination showed a huge white cyst (11 × 10.5 × 9 cm) containing translucent fluid and daughter cysts of varying size (Fig. 2). Wet mount preparation of cyst fluid showed presence of larval form of *Echinococcus* containing hooklets and suckers invaginated within a vesicle (Fig. 2, inset).

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Fig. 1 (A) USG whole abdomen showing a heterogenous space occupying lesion at the upper pole of right kidney. (B) Echogenic debris within bladder lumen. (C) CT scan of Abdomen showing a cystic lesion in the right kidney

Fig. 2 Gross examination of nephrectomy specimen showing a huge white cyst containing daughter cysts of varying size within fibrous capsule (renal pelvis indicated by an arrow). Inset: Wet mount preparation of cyst fluid showing larval form of *Echinococcus* containing hooklets and suckers invaginated within a vesicle

Histology revealed a hydatid cyst with three layers: (a) the outer pericyst, composed of modified host cells forming a dense, fibrous protective zone; (b) the middle laminated membrane, which is acellular and allows the passage of

nutrients; and (c) the inner germinal layer, from where the scolices (the larval stage of the parasite) and the laminated membrane are produced (Fig. 3A, B). There was no post-operative complication. Patient is symptom free and doing well 6 months after operation.

Discussion

Echinococcus granulosus adult worm lives in the proximal small bowel of the definitive host, attached by hooklets to the mucosa. Eggs are released into the host's intestine and excreted in the feces. Human may become intermediate host through contact with a definitive host (usually a domesticated dog) or ingestion of contaminated water or vegetables. When embryo passes 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. In human, hydatid disease involves the liver in approximately 75 % of cases and the lung in 15 %. Secondary involvement due to hematogenous dissemination may be seen in almost any anatomic location (Pedrosa et al. 2000).

Kidney involvement in echinococcosis is extremely rare (2–3 % of cases), even in areas where hydatid disease is endemic (Afsar et al. 1994). 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 the liver and retroperitoneal lymphatics (Pedrosa et al. 2000).

Fig. 3 (A) Histopathology of the cyst wall showing outer pericyst, composed of modified host cells forming a dense, fibrous protective zone. (Hematoxylin and Eosin stain, $\times 100$ magnification) (B) Middle laminated membrane, which is acellular and the inner germinal layer with presence of scolices. (Hematoxylin and Eosin stain, $\times 400$ magnification)

Renal 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 (Volders et al. 2001).

The only pathognomonic clinical sign of renal hydatid disease is hydatiduria (Gogus et al. 1991). If all the three layers of the cyst have ruptured, with parenchymal destruction, resulting in free communication with the calyces and pelvis, it is called an open or communicating cyst (Mongha et al. 2008). Rupture into renal pelvis results in acute flank pain, followed by voiding of scolices, hooklets or daughter cysts, with or without hematuria giving rise to gross or microscopic hydatiduria (Unsal et al. 2001). The condition is seen in few cases of renal hydatidosis and is usually microscopic (Mongha et al. 2008). In the present case the history of gross hydatiduria directed the clinicians to investigate the case for renal or bladder hydatid disease. But during hospital stay he did not suffer from renal colic or passed any granular substances in urine. So we could not determine the exact site of obstruction and routine examination of urine failed to document any evidence hydatiduria. In our case the only abnormality in the routine blood examination was eosinophilia, which is reported in 40–50 % of such cases. (Buckley et al. 1985).

Serological tests to diagnosis hydatid disease are immunoelectrophoresis test, immunohaemagglutination test and complement fixation test. Casoni test is unreliable and outdated now. But a combination of serological investigations yields a diagnosis in only 50 % cases (Gharde et al. 2012).

Imaging modalities establish diagnosis in most of cases. Sonography and CT show a uniloculated or a multiloculated

cyst that may have some heterogeneous echoes on sonography or have rim enhancement on CT (Beggs 1985). Daughter vesicles (brood capsules) are small spheres that are formed from rests of the germinal layer and appear as cysts within a cyst. They contain the scolices and hooklets, along with sodium chloride, proteins, glucose, ions, lipids, and polysaccharides (Pedrosa et al. 2000). On changing the patient's position under real time USG there is shifting of hydatid sand gives rise to the 'Falling snow flake' pattern (Gharde et al. 2012). MRI offers no real advantage over CT scan (Volders et al. 2001).

Although safe translumbar puncture of the cyst for diagnostic sampling has been reported, cyst puncture should be avoided, because of the risk of allergic reactions, including anaphylactic shock and possible dissemination (Roylance et al. 1973).

Histopathologically a renal hydatid cyst shows a non-nucleated laminated membrane composed of innumerable delicate laminations and a nucleated inner germinative membrane. High-power photomicrograph of the cyst depicts an *E. granulosus* scolex with shark tooth hooklets floating in proteinaceous cyst fluid (Volders et al. 2001).

In general, surgery is the treatment of choice in renal hydatid cyst. Kidney-sparing surgery (cystectomy with pericystectomy) is possible in most cases (75 %). Nephrectomy (25 % of cases) must be reserved for destroyed kidneys (Volders et al. 2001).

In conclusion, the present report emphasizes that history of passing granular substance in urine in a patient of abdominal colic, without evidence of urolithiasis in plain X-ray should be suspected as hydatiduria, especially in endemic region. As the history of hydatiduria indicates a communicating cyst, there is high chance of rupture, needs

preoperative albendazole therapy to avoid risk of anaphylaxis.

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