Research article

Series of Cases of Hydatid Disease Which Presented at an Exclusive Nephro –Urology Center from South India

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Abstract

Echinococcus granulosus and Echinococcus multilocularis are a cause for hydatid disease which is universal in distribution. This is an original review article of a series of four cases which presented as primary Echinococcus granulosus, 3 of which presented to us as Echinococcus multilocularis primary in the kidney and co-incidentally they were all in the left kidney, one presented as retrovesical hydatid that was the 4th case. About 25% of the cases of Echinococcus granulosus usually pass of life as unnoticed and symptom free referred to as tape worm infestation in lay mans terminology. It is the rest 75% which become symptomatic as in infected hydatid, suppuration and abscess formation. When the hydatid fluid gets into circulation allergic symptoms such as urticaria, angioneurotic oedema, eosinophilia, they may develop in various stages. All of our renal hydatid cysts presented with a history of flank pain lasting over a period of 1-2 years. Associated symptoms of the latter were that of low grade fever on and off and dysuria. All the cases were found to be diagnosed while evaluating for flank pain or dysuria. The case of retro vesical hydatid presented as acute urinary retention of short duration 72 hours, mass per abdomen on evaluation. Their ages ranged from 35-46 years in the case of renal hydatid disease and the retro vesicular hydatid disease patient was of 60 years. All the cases have been followed up for a minimum of 1 year duration.

Keywords: Hydatid Disease; Retrovesical; Kidney; Echinococcus granulosus/multilocularis

Introduction

Echinococcal infection of the genital tract is a form of parasitism. Parasitism means a temporary or permanent reciprocal association between two species, where one species depends upon another for its existence [1]. It is the second most frequent and important parasitic diseases involving the genitourinary system in humans. Hydatid disease (HD) is a parasitic infestation caused by tape worms of genus Echinococcus [2,3]. HD per se is universal in distribution [1] HD of the urinary tract is uncommon accounting for only 2-4% of all the cases [4-6] Retrovesical hydatid is even less frequent [4-7]. HD caused by E. granulosus is the most common variant prevalent world over, followed by E. multilocularis, E. oligarthrus and E. vogeli [2,3,10,11]. Genitourinary hydatid patients are usually asymptomatic on the rupture of the cyst and this probably explains the varied clinical features and presentation [2,3,10,12]. The disease is caused when food and water that contains eggs of the parasite are consumed or due to close proximity to the infected animal. The eggs are released in the stool of animals that infected by the parasite, commonly dogs, foxes and wolves. E. granulosus is endemic in India, Middle East, Asia, Australia, Africa, especially northern parts like Tunisia, Southern Europe, South America, Mediterranean countries, New Zealand and Alaska where sheep and cattle rearing is a common occupation. Reservoir of the disease is maintained by allowing dogs to have free access to ingest organs of infected animals, slaughter of animals from farms or villages or from communities that practice ritual sacrifice [13]. The surgical incidence is 50 per 100,000 person-years in endemic areas [14]. Very few countries are considered to be completely free of E. granulosus. Endemicity of the disease varies by virtue of change in occupational practices of the locals, nomadic migration of shepherds and their herds and various other such incidental factors. The resurgence of the disease as an endemic is a constant threat to certain countries. Echinococcosis is an infection caused by the larval stage of a cestode (tapeworm) called Echinococcus [4,5]. HD caused by E. vogeli is known as human polycystic hydatid disease or neotropical echinococcosis, characterised by unicocular and multicocular cyst due to invagination of the wall [4-7]. Genitourinary HD patients are usually asymptomatic even on rupture of the cyst [2,3,10,12]. Hence the importance of individual experience in clinical presentation and approach towards these
cases per se. This is a review of a series of cases that presented to our outpatient department during the last 5 years at our institute.

**Materials and Methods**

A series of 4 cases presenting at our outpatient as primary HD were seen during the years 2011 to 2015, their modes of presentation, relevant clinical and diagnostic findings and corroborative histopathology findings. We had three patients with renal HD. The first was a 46 year old lady with left flank pain and on and off low grade fever for 2 years. She was a house wife. Clinical examination was non-contributory. Ultrasound (US) showed enlarged left kidney with multiple cysts. Contrast enhanced computed tomography (CECT) of the abdomen showed an enlarged left kidney (12.5 cm) with non-enhancing cystic lesions, suggestive of HD (Figure 1).

Liver was normal and no other abdominal cysts were seen. As kidney was completely involved she underwent left simple nephrectomy after packing the retroperitoneum with 10% povidone soaked mops. Postoperative histopathology revealed renal hydatidosis. We also had 2 male patients, one patient aged 39 and tailor by occupation and the other aged 35 years, an agricultural laborer by occupation. They presented with left flank pain for 6 months and 1 year respectively. They also were preoperatively diagnosed to have primary left renal Echinococcosis on US and CECT and underwent left simple nephrectomy in view of the entire kidney being replaced by the disease intraoperatively. All three patients received albendazole 15mg/kg for 2 weeks preoperatively. A 60 year old gentleman, agricultural laborer by occupation, presented with acute urinary retention and was catheterized elsewhere. He had lower urinary tract symptoms for 6 months. He had a non-tender firm intraperitoneal mass with limited intrinsic mobility and dull note on percussion. US showed a huge retrovesical mass with cyst in cyst appearance (Figure 2).

CECT abdomen and pelvis showed an isolated large non-enhancing cystic lesion in the retrovesical area (800cc) with multiple daughter cysts, suggesting primary pelvic hydatid cyst (Figure 3).

Laparotomy was done after 2 weeks of preoperative albendazole. Intraoperatively he had a large retrovesical hydatid cyst adherent to posterior wall of bladder; there were no cysts in liver or abdominal cavity (Figure 4).
Total pericystectomy was done and scolicidal agent was injected. Gross and microscopy revealed daughter cyst and germinal layer, confirming hydatid cyst (Figure 5, 6).

Serology was not done due to economic constraints in all patients the Wet mount preparations were done and daughter protoscolices were well appreciated in the hydatid sand, one of these cases has been mounted in our institute museum, work contributed from the Department of Pathology of the Institute (Figure 7, 8).

These patients were followed up for a 1-3 years and all are doing well with no recurrence to date.

**Results**

These cases presented to our outpatient department in the last 4 years, all the renal HD presented to us with involvement of the left kidney. In the era of nephron sparing surgery simple nephrectomy is a choice when the entire kidney is involved that is in 25% of the cases and 75% cases where the kidney can be spared nephron
sparing surgery is done. All cases received the preoperative dose of standard medical line of management in accordance to the parasite load for the prerequisite time and duration. Simple nephrectomy was done in all the 3 cases of renal HD and a point of interest as a common denominator that all the cases were confined to the left kidney in the renal cases which is a finding of significance. The last case which was retro vesicular hydatid, presented with obstructive symptoms of short duration where pericystectomy was done with adjunct therapy in the form of scolicidal injection was done. A point of relevance is that ours is a tertiary care referral center with technical compatibility to meet the needs of such patients and feasibility of such patients, for most of the cases are active, expanding and symptomatic and the infected cysts are best treated with surgery and installation of scolicidal agents. Recurrent cysts caused due to seeding during any of these procedures usually present over long period of time well above 5 years. Currently all are patients are being followed up at our center itself.

Discussion
Renal HD is rare as the liver and lungs are the first to be attacked by the parasitic larvae /embryos of E.granulosus [15]. All of our cases at the institute are from a background of low socio economic strata where quality of water and sanitation are questionable and open air defecation is practiced as an easy alternative to going to long distances in search of bathrooms due to its unavailability. The latter is one of the main target points in all preventive programs towards control of the disease process. Embryo enters the blood stream and lymphatics and spreads to the other regions [2,10]. Clinical symptoms include flank pain or abdominal mass, hematuria, malaise and fever [16]. It presents between the third and the fifth decade of life, the left renal involvement is more common than the right owing to the shorter renal artery [10,17]. The latter is in concurrence of our findings in the present study. Apart from the interventional modalities we need to look at public health aspect or perspective as well. The reason being that this anthropozoonosis is caused by the larval stage tape worm seen in dogs, wolves and foxes [1]. The adult tapeworm resides in the latter mentioned carnivores. Humans constitute intermediate hosts and are thus accidental dead end hosts [2,10]. When these embryos get passively transported by hematogenous and lymphatic means these embryos which do not get destroyed by phagocytosis develop into hydatid cysts (greek: hydatitis means drop of water) [2,18]. The other rather interesting and baffling part is its clinical features seldom give the clinician an inkling of the disease, this is a slow growing lesion which takes anywhere between 5-20 years to clinically manifest [2,3,10,19]. Due to economic constraints, we could not do serology in our patients. The second reason was that most of the patients test seronegative and serology associated confirmation is only in 50% of the cases. Diagnosis is by a combination of immunoelctrophoresis, immuno-haemagglutination tests and complement fixation test; Casino's test is at its best unreliable and outdated. Enzyme Linked Immuno Sorbent Assay (ELISA) is the most widely used [10,20,21]. In seropositive cases, the confirmation is by arc electrophoresis. Polymerase chain reaction (PCR) using recombinant DNA antigen is useful in sub classifying the *Echinococcus* species. Routine blood and urine investigations are non-contributory. We would also like to end by saying that there is no pathognomonic serological and immunological test despite the fact that several serological tests have evolved over a period of time, but are of low sensitivity [22]. From the view of management, nephrectomy should be avoided due to the benign nature of its presentation. Nephrectomy is indicated when the entire kidney is replaced by a hydatid cyst or in poorly functioning kidney, cyst hemorrhage or infection or communicating cysts [23].

Conclusion
Genitourinary hydatid disease is an uncommon disease and the pre operative diagnosis requires a very high index of suspicion, especially in patients in endemic regions with cystic masses. Complete excision of germinal layer with daughter cysts without spillage of viable cyst contents with preoperative medical therapy is the gold standard and minimally invasive surgery is feasible in the modern era. Eradication needs breaking the continuity in the life cycle by avoiding contact of dogs to untreated fecal matter of sheep and rodents. Treating dogs per se is a reliable alternative. Adequate survey, prevention and control would involve vaccination of the lambs, de-worming of dogs and slaughter of older sheep could lead to lead to elimination of the disease in the next decade. Stringent measures enforced by the animal husbandry and food safety authorities will go a long way in bringing down the disease burden. The disease process can be brought down by measures to prevent transmission of parasite. Preventive measures include limiting the areas where dogs are allowed and preventing animals from consuming meat infected with cysts of echinococcus. The World Health Organization coordinating with the local public health authorities and services in these endemic areas should put forward a proactive and feasible method for prevention of the disease and its spread. The preventive measures in a stringent and definitive manner can lead to reduction in the mortality morbidity if not the eradication of this not so uncommon under recognized tropical disease.

References


