

Clinical presentation and Management of Patients with Hydatid disease in a tertiary care teaching hospital in India

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Abstract

Background: Hydatid cyst disease or cystic echinococcosis (CE) is a zoonosis affecting lungs and liver causing a major health care problem with significant morbidity in endemic areas.

Objective: To assess various risk factors, clinical presentation and treatment modalities employed in treatment of Hydatid cyst disease patients.

Methodology: A prospective observational study involving 25 patients of hospitalized patients was conducted in a tertiary care teaching hospital from August 2016 to October 2018. Clinical examination, radiological diagnosis and pathological evaluation of all the patients were performed. Chemotherapy with albendazole (10 mg/kg) and surgical removal of cyst were main stay of therapy employed for patients with follow up for 6 months to 2 years.

Results: 28% of patients in study belong to 51-60 years of age group and from agricultural rural areas. Out of total 25 patients, 16(64%) and 9(36%) were women and men respectively. Total 7 patients had contact with dog/sheep and 60% patients had eosinophilia. In 68% of patients had single cyst in the right lobe of liver. 40% patients were treated conservatively with albendazole and 60% patients treated surgically. Most commonly performed procedure was decompression of cyst with deroofing and cyst excision followed by either external tube drainage (36%) in majority of cases or omentoplasty (16%). Post-operative complications were seen in 4 cases which includes development of biliary fistula (2, 13.33%), infection of remaining cavity and wound infection occur in one patient (6.66%) each.

Conclusion: Decompression of cyst along with albendazole chemotherapy proves to be an effective intervention for better clinical management of hydatid cyst.

Key words: hydatid disease, cystic echinococcosis, surgical management of CE, albendazole

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I. Introduction

Human echinococcosis is a parasitic disease caused by tapeworms of the genus *Echinococcus*. The two most important forms in humans are cystic echinococcosis (hydatidosis) and alveolar echinococcosis. (1) Humans are infected through ingestion of parasite eggs in contaminated food, water or soil, or after direct contact with animal hosts. Echinococcosis is often expensive and complicated to treat and may require extensive surgery and/or prolonged drug therapy. More than 1 million people are affected with echinococcosis at any one time. (2)

CE is found worldwide, especially where livestock breeding and farming are widespread and in areas where human, definitive and natural intermediate hosts are found in close proximity. (3) In the human host, a hydatid cyst can lead to life-threatening complications, such as cyst rupture, with possible anaphylactic shock, the spread of new cysts, and bacterial infection. CE is often under-diagnosed because it is frequently a silent condition that develops over several years and whose symptoms are only apparent when compression of internal organs occurs. (4) Human infection with *E. granulosus* leads to the development of one or more hydatid cysts located most often in the liver and lungs, and less frequently in the bones, kidneys, spleen, muscles and central nervous system. The asymptomatic incubation period of the disease can last many years until hydatid cysts grow

to an extent that triggers clinical signs, however approximately half of all patients that receive medical treatment for infection do so within a few years of their initial infection with the parasite. (2, 5)

Abdominal pain, nausea and vomiting are commonly seen when hydatids occur in the liver. If the lung is affected, clinical signs include chronic cough, chest pain and shortness of breath. (2,3) Other signs depend on the location of the hydatid cysts and the pressure exerted on the surrounding tissues. Non-specific signs include anorexia, weight loss and weakness. Ultrasound, CT scan and MRI are frequently used for the diagnosis of the diseases. (2)

The estimated prevalence of CE in endemic areas ranges from 1%–10%; however, the incidence rate in hospitals is about 1000 times less than the prevalence as only a small proportion of patients who develop severe symptoms seek medical care. (6, 7) In India, CE is present in most parts of the country. A previous study from our center in North India has shown an overall seroprevalence of 15.1% over a period of 20 years in clinically suspected cases of CE. (8) CE may be responsible for a loss up to 3.6 million disability-adjusted life year worldwide. (9) Echinococcal cysts continues to be a common surgical condition in many rural parts of India, carrying a significant risk of morbidity and mortality. (6) Most of the patients presenting with serious complications of hydatid disease require a surgical procedure. Late manifestation of symptoms creates a challenging situation in diagnosis of CE however serological analysis conjugated with radiological diagnosis and selection of appropriate treatment modality is a key to reduce the rate of morbidity and mortality. The treatment options for the disease vary from medical line of management to various surgical procedures which also poses significant challenge to surgeons and sometimes complicated by life threatening outcomes.

Therefore, the present study was conducted to know the clinical features, epidemiology, risk factors and management of the CE in western India.

II. Methodology:

This was a prospective observational study involving diagnosed patients of Echinococcal cysts disease hospitalized in a surgical ward of a tertiary care teaching hospital from August 2016 to October 2018. The study protocol was approved by the institutional ethics committee and written informed consent was obtained from all patients before enrolling them for the study.

All the patients admitted in the surgical ward with diagnosis of Hydatid disease were interviewed and their demographic data, exposure to risk factors, clinical features were asked. Their management including investigations and treatment were recorded from their case files. A total of 25 patients suffering from hydatid liver disease were included in this study based on a meticulous clinical examination, radiological diagnosis using USG abdomen, CT scan, X-ray chest and pathological findings (detection of Echinococcus granulosus in biopsy specimen).

All patients underwent surgery after required pre-operative preparation. They were prescribed with albendazole (10 mg/kg) for one month before surgery, and this medication was continued postoperatively for 21 days. Cystotomy of pericyst (fibrous capsule) was performed on all patients and complete excision of ectocyst and endocyst (both parts of the actual parasite) was done. Capitonnage was done wherever feasible. All cavities were irrigated with scolicidal agent. 5% betadine was used for all lung cysts. The case with both liver and lung involvement was dealt with a double thoracotomy instead of a laparotomy as liver involvement was in the 7th lobe and access would have been very difficult. Liver was approached transdiaphragmatically. After meticulous removal of each and every daughter cyst, the cyst cavity was irrigated with 3% hypertonic saline. The diaphragm was meticulously repaired after placing a drainage tube in the liver cyst cavity, also through the phrenotomy opening. Tube All patients received amoxicillin and potassium clavulanate combination (50-100 mg/kg/day in 3 divided doses), and amikacin (15 mg/kg/day in 2 divided doses) for 7 days. Amoxycylav was continued till chest drain was in situ. All cysts were larger than 5 cm in diameter; hence scolicidal agent (like hypertonic saline or 5% betadine) was injected in cyst cavity after aspiration of cyst component and then completely resected except in the liver cyst case in which excision and post-excision irrigation was done. In all cases, cavity was also irrigated by the same solution after excision. All resected cysts were sent for histopathological examination.

All 25 cases were studied and a follow up for a minimum of 6 months to 2 years after the treatment during the study period for recurrence of cyst. Data was analyzed in terms of age, gender, clinical features, diagnostic investigations, operative technique, post-operative complications, mortality and duration of hospital stay.

Statistical analysis:

Data was analysed using Microsoft excel 2013. All the data represented as actual frequencies, percentage, mean, standard deviation as appropriate. For association analysis chi square test was used and p value less than 0.05 was considered significant.

III. Results:

Total 25 patients included in the study of Hytid disease. Total 28% pf patients belonged to an age range of 51-60 years, followed by 31-40 years (24%). Out of a total of 25 patients, 9 were males and 16 were females. As shown in Table 1, history of contact with dog or sheep was there in 28% of patients. Agriculture was the most common occupation of patients (40%) followed by labourers (24%).

In the present study, the commonest symptom of hydatid cyst of liver was mass per abdomen, 16 patients (64%) followed by pain abdomen in 14 patients (56%) as shown in table 2. Raised eosinophil count, i.e. >5% of leukocytes was present in 60% cases.

In this study the right lobe harboured the cyst in 68% cases and the left in 32% cases. Single cyst was present in 17 cases (68%) while multiple cysts was present in 8 cases (32%). Univesicular cyst was present in 14 cases (56%) and multivesicular cyst was present in 11 cases (44%). (Table 3)

In this study, 40% of patients with hydatid cysts in liver were treated conservatively by medical treatment with albendazole. In surgical treatment most commonly performed procedure was decompression of cyst with deroofting and cyst excision followed by either external tube drainage (36%) in majority of cases or omentoplasty (16%). Partial peri-cystectomy was done in two patient (8%). Post-operative complications were seen in 4 cases. Two patients (13.33%) had developed biliary fistula. Infection of remaining cavity and wound infection occur in one patient (6.66%) each. (Table 4)

Table-1 Demographic details of study patients (n=25)

| Sr. no. | Parameter | No. of Cases (%) |
|----------|--|------------------|
| 1 | Age in years | |
| | 11-20 | 3 (12) |
| | 21-30 | 2 (8) |
| | 31-40 | 6 (24) |
| | 41-50 | 4 (16) |
| | 51-60 | 7 (28) |
| | >60 | 3 (12) |
| 2 | Gender | |
| | Male | 9 (36) |
| | Female | 16 (64) |
| 3 | History of Contact with dog/sheep | |
| | Present | 7 (28) |
| | Absent | 18 (72) |
| 4 | Occupation | |
| | Labourers | 6 (24) |
| | house wives | 5 (20) |
| | Students | 2 (8) |
| | Agriculturist | 10 (40) |
| | Others | 2 (8) |

Table-2 Clinical features and laboratory investigations of the study patients (n=25)

| Parameter | No. of Cases (%) |
|----------------------------|------------------|
| Presenting Symptoms | |
| Abdominal Mass | 16 (64) |
| Pain in abdomen | 14 (56) |
| Fever | 4 (16) |
| Pruritus | 1 (4) |
| Jaundice | 2 (8) |
| Eosinophil count | |
| 0-5 % | 10 (40) |
| > 5 % | 15 (60) |

Table-3 Site, numbers and types of hydatid cyst in study patients (n=25)

| Parameter | No. of Cases (%) |
|-------------------------|------------------|
| Location of Cyst | |
| Right Lobe | 17 (68) |
| Left Lobe | 8 (32) |
| Number of Cyst | |
| Single | 17 (68) |
| Multiple | 8 (32) |
| Type of Cyst | |
| Univesicular | 14 (56) |
| Multivesicular | 11 (44) |

Table-4 Treatment and postoperative complication of hydatid cyst in study patients (n=25)

| Treatment Modality | No. of Cases (%) |
|---|-------------------------|
| Medical | 10 (40) |
| Operative | 15 (60) |
| 1. Decompression and excision with | |
| a) External drainage and tube insertion | 9 (36) |
| b) Omentoplasty | 4 (16) |
| 2. Partial peri cystectomy | 2 (8) |
| Post-operative complication | No. of Cases (%) |
| Biliary fistula | 2 (13.33) |
| Infection of remaining cavity | 1 (6.66) |
| Wound infection | 1 (6.66) |

IV. Discussion

Recent disease control programs have significantly reduced worldwide incidence and prevalence of helminth disease. However Echinococcosis is still remains a common health problem encountered by various developing countries due to potential serious complications. (1, 3) Documented evidences have been reported from various countries regarding increase in the incidences of cystic echinococcosis with an annual incidence of 1-200/100000 population in India. (4, 8) This study has evaluated the pattern of presentation, epidemiology and management of patients of hydatid cyst disease in western India.

In current study the overall CE prevalence was observed to be higher in females as compared to males which concur with findings of previous studies. (10) This result can be explained by gender roles in endemic regions where females play a major role in domestic activities including food preparation, caring for daily routines of pet dogs and tending gardens. (10) Patients with elderly age in 51-60 years of age group were associated with higher incidence of CE which was matched with the results of other studies. (11) In our study 7 cases were associated with history of contact with dogs which is an important risk factor for development of CE disease. Similar association was established in other reported studies wherein development of CE disease was related to contact with dogs which act as definitive hosts, harboring the adult form of the tapeworm in their intestines contaminating the soil and water through their faeces. (12) Even direct or indirect contact with dog faeces while cleaning through contamination of hands with *E. granulosus* eggs excreted by dogs appears to be the most important pathways of transmission for human CE. (13) People with agriculture or livestock farming were potentially more vulnerable population to development of human CE. Similar observations were been made and reported in previous studies also. This was contributed to poor awareness regarding the role of dogs in CE transmission. Majority of farmers leave dogs faeces unattended on the farm ground leading to accidental contamination of soil, water, vegetables and other crops ultimately leading to human CE infection. (14)

Liver (64-75%) and lungs (15-21%) are most commonly associated organs affected by CE. (15) A similar observation was made in our study with palpable mass per abdomen with pain as most frequent clinical symptom. Right lobe of the liver is more affected compared to the left lobe of the liver while other organs like brain, kidney, heart, skin, bones, spleen and muscles are rarely associated with CE. (15) Most of primary CE infections are associated with development of single cyst while 20-30% patients have multiple cyst. Similar findings were obtained in our study as well with majority patients suffering from single cyst. (15, 16) Various

radiological investigations including Ultrasound Sonography (USG), computed tomography (CT), Magnetic resonance imaging (MRI), are used for precise and differential diagnosis of hydatid cyst associated with deep seated lesions. Non-invasive immunodiagnostic techniques and radiologic imaging are widely used in differentiation of echinococcal cysts from benign cysts, mycoses and abscesses. (15) Conventionally surgery and chemotherapy with benzimidazole compounds were the only approaches for the treatment of echinococcal cysts. In cases of small cyst with < 4 cm diameter with thin wall, chemotherapy is most effective. (17) In our study, treatment with albendazole resulted in betterment of patients with echinococcal cysts which was in agreement with several previous studies. Preoperative chemotherapy is associated with sterilization of cyst and reduces the risk of recurrence. Similar successful observations pertaining to results of chemotherapy of hydatid disease were also been made and reported in previous studies wherein albendazolesulfoxide, an active metabolite of albendazole shows activity against protoscolecocytes of echinococcusgranulosus in in vitro cultures and penetrates in hydatid cyst. (17-19) Even the risk of recurrence in patients treated with albendazole therapy was found to be minimum. Various new treatment modalities like cyst puncture, aspiration, reaspiration have been introduced. However the accuracy and efficiency of these treatment processes needs further validation. (20, 21) For immediate and appropriate cure where medications fail to improve the condition, surgery of intact hydatid cyst still remains the first choice with best potential as it involves total removal of cyst without any adverse effects of spilling the contents. External drainage, pericystectomy or omentoplasty are most commonly performed surgical procedure. Peripheral cysts and pedunculated cyst are excised entirely through obliteration of the cyst cavity after evacuation of the cystic content. Decompression of cyst accompanied by external tube drainage which retain its value due to simple and safe process which is consistent with results of earlier studies. (21-23) Omentoplasty is considered to be the best possible surgical alternative for treatment of uncomplicated hepatic hydatid cysts due to complete elimination of the parasite, lowest surgical site infection rate and lack of intraoperative spillage. Omentoplasty also supports healing of the raw surface with resorption of serosal fluid with less rate of recurrence and residual infection. (22) Secondary infections like rupture into biliary tree, peritoneum or adjacent structures are most common post-operative complications observed in 10-15% patient population which was found to be consistent with other studies. Early diagnosis and treatment of cystobiliary communications reduces postoperative morbidity. (22-24)

This study has highlighted the clinical presentation and epidemiology of the CE in western India along with its management modalities used. Few limitation of the study include single centre and limited number of cases. Further larger studies involving multiple centres can be useful for understanding present trends of hydatid disease in India.

In conclusion, Cystic echinococcosis should be diagnosed and treated at an early stage before the growth and spread of cyst which can cause complications in later stages. Medical chemotherapy in adjuvant to surgery is considered to be the ideal choice of treatment as they ensure complete removal of parasite with lower incidence of recurrence, fewer postoperative complications and better outcome. Medical treatment alone should be advisable to comorbid patients small cysts (<5 cm) in the liver and lungs. Management of the residual cavity through external drainage or omentoplasty is an important segment which should be focused carefully to minimize the rate of morbidity and recurrence. Omentoplasty and external tube drainage can be considered as most promising modalities but should be selected carefully based on the patient's condition and physician's expertise. Patient education and awareness can also play very important role in prevention of such diseases.

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