



SPECTRUM OF CYSTIC ECHINOCOCCOSIS –A MEDICAL COLLEGE EXPERIENCE IN MADHYA PRADESH

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ABSTRACT

Objectives: To study the epidemiology of cystic echinococcosis in Madhya Pradesh; to study the various common and uncommon site of presentation and the risk factors associated with cystic echinococcosis. **Methodology:** This was a descriptive retrospective study conducted for 5yrs from March 2011 to February 2016 in L.N Medical College & Research Centre, Bhopal. Clinically and radiologically suspected 58 patients of cystic echinococcosis (CE) which were serologically positive and histologically proved were enrolled in this study. **Results:** Female predominance was noted with 38(65.5%) females and 20 (34.48%) males with a male: female ratio of 1:1.52. The most common age group was 30-40 years .75.86% (n-44) of the patients resided in rural areas and only 24.13%(n-14) were urban residents Abdominal CE presented mostly with abdominal pain. Hepatomegaly was observed in 15 patients and jaundice was present in one patient of hepatic CE. Splenic CE cases showed splenomegaly in 3 patients. Pulmonary CE presented with mainly with chest pain and cough. **Conclusion:** The risk factors are female, rural, housewife and farmers, unsafe drinking water with exposure to animals such as dog and sheep. Treatment options for cystic hydatid disease are surgery, drug therapy and percutaneous drainage .The most common surgical procedure performed was a hydatid cystectomy. CE is a neglected disease causing immense morbidity and mortality and hence of public health concern there is an urgent need to create awareness among public and common people who are more prone to the condition.

KEYWORDS: cystic echinococcosis, hydatidcyst;, neglected disease.

INTRODUCTION

Cystic Echinococcosis (CE) is the most widespread zoonotic disease caused by larvae of dog tapeworm known as Echinococcus granulosus (EG) which causes cystic disease mainly in liver and lung and also in other organs, known as hydatid cyst disease.^[1] CE is a neglected disease causing immense morbidity and mortality and hence of public health concern, especially in poor countries where livestock is raised in association with dogs in a low hygienic environment and where sheep husbandry is an important agricultural industry.^[2] More than 1 million people are affected with echinococcosis at any one time.^[2] In India CE is endemic in different regions including Central India but the highest prevalence is found in Andhra Pradesh, Saurashtra and Tamil Nadu.^[2]

CE is of special importance due to its wide geographic distribution and socioeconomic impact, not only with respect to the medical costs associated with it but also with regard to the loss of livestock because of they are the intermediate host.^[3] According to statistics given out by WHO, the annual societal cost of CE amounts to US

\$150 million in the subcontinent of India alone.^[4] Hence as CE lead to considerable morbidity and socio-economic losses in many regions of the world, recently it was included in the WHO strategic plan (2008-2015) for the control of NTDs and was put under the neglected zoonosis subgroup “neglected zoonotic diseases” (NZD).^[3] According to WHO, these diseases are called “neglected” as these have not been given enough importance at national or international levels by researchers and health policy makers alike even in endemic areas.^[5]

Diagnosis, prevention and control of CE are still at the juvenile stage, despite the pandemic occurrence of the disease. Usually the condition goes unnoticed until the cyst triggers some physical disabilities. Hence, there is an urgent need to create awareness among public and common people who are more prone to the condition. Also there is a paucity of data from Indian studies in terms of clinical presentation, risk factors, investigations and hospital course. Hence, this study was carried out with the aim to characterize the epidemiological, clinical, radiological, pathological features and risk factors of

patients with CE in our region i.e Madhya Pradesh with reference to the current context.

METHODOLOGY

The clinical study of CE was conducted in LN Medical College and Research Centre, Bhopal, Madhya Pradesh. It is a 750-bedded tertiary care hospital situated in a zoonotic endemic area.

TYPE OF STUDY- Descriptive retrospective study.

DURATION OF STUDY- The study was conducted for 5yrs from March 2011 to February 2016.

PLACE OF STUDY- L.N Medical College & Research Centre, Bhopal.

The study was approved by institutional ethics committee.

Inclusion criteria: all patients with histopathologically confirmed CE were included in the study.

Exclusion criteria: Clinically and radiologically suspected patients of CE who did not undergo surgical cystectomy and histopathological examination were not included in the study.

Information of all patients included in the study were reviewed from the hospital records. Data collected were documented under the following headings:

- (a) Demographic data (age, gender, occupation, rural or urban residence and history of pet ownership);
- (b) Clinical features and physical examination of abdomen and chest.
- (d) Investigations such as complete hemogram with total leucocyte count, eosinophil count, blood sugar and serum bilirubin were noted. Serological test such as indirect haemagglutination test (IHT) and the enzyme-linked immunosorbent assay (ELISA) were done for immunodiagnosis.
- (e) Radiological study-(1)Posterior-anterior chest X-ray in the erect position (2) Ultrasound study (site of CE, number of cyst, size of the cyst in centimetre) (3)CECT abdomen and MDCT chest for confirmation whenever required and afforded by patient due to cost.(f) Treatment and follow up- Surgery with adjuvant antihelminthic.

The primary modality of surgery utilized was the hydatid cystectomy (open or laproscopically) in which the cyst cavity was opened after scoliceal deactivation. The scoliceal agent used was hypertonic saline. Cyst cavity was packed with omentum or was closed with 2-0 chromic catgut suture or drained externally. All extra hepatic hydatids including that of the spleen were entirely excised with the pericyst intact. A similar cystectomy procedure was followed for lung hydatids and renal hydatid, with the difference that the drain was left within the cyst cavity and connected to an external

underwater seal bottle in lung hydatid. In cases dealt laproscopically hydatid trocar cannula spill proof technique was used. Complications were noted. A recent technique PAIR (ultrasound guided percutaneous aspiration with infusion of scoliceal agents and re-aspiration) was done in 4 patients.

The first follow up ultrasound was usually performed at one month. Subsequent follow visits ranged from a period of three months to one year.

Histopathological examination. Diagnostic criteria-visualisation of a lamellated structure of a cyst wall, germinal layer, scolices and protoscolices on Hematoxylin and Eosin (H&E)-stained paraffin embedded sections. The cysts were staged according to the World Health Organization International Working Group on Echinococcosis (WHO-IWGE) standardized classification for ultrasound (US).^[6]



FIG.1 CECT ABDOMEN classical hydatid cyst of liver

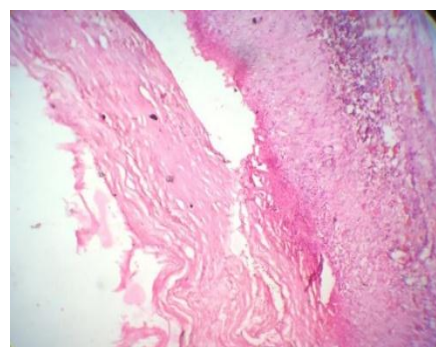


FIG -2 slide of hydatid cyst wall under H & Estain



FIG-3 ultrasound picture of hydatid cyst

OBSERVATION TABLES

TABLE –1. DEMOGRAPHIC STATUS

AGE DISTRIBUTION			
	AGE IN YEARS	NUMBER	PERCENTAGE
	0-10	0	0
	11-20	5	8.6
	21-30	15	25.8
	31-40	17	29.3
	41-50	12	20.68
	51-60	6	10.34
	61-70	3	5.17
	TOTAL	58	100
SEX DISTRIBUTION			
	MALES	20	34.48
	FEMALES	38	65.5
RESIDENCE			
	URBAN	14	24.13
	RURAL	44	75.86
OCCUPATION			
	HOUSEWIFE	31	53.44
	STUDENT	5	8.6
	FARMER	15	25.86
	EMPLOYEE	7	12.06
HISTORY OF PET ANIMAL			
	PRESENT	38	65.66
	ABSENT	12	20.68
	NOT KNOWN	8	13.79
SOURCE OF DRINKING WATER			
	SAFE	21	36.20
	UNSAFE	36	62.06

TABLE-2.CLINICAL FEATURES

CLINICAL FEATURES			
	VAGUE SYMPTOMS	8	13.79
	ABDOMINAL PAIN	15	25.86
	LUMP(MASS)	10	17.24
	PAIN + MASS	9	15.51
	PAIN + FULLNESS	6	10.34
	HEPATOMEGALY	15	25.86
	SPLEENOMEGALY	3	5.17
	FEVER	2	3.44
	COUGH + FEVER	2	3.44
	COUGH + CHEST PAIN	5	8.62
	PALLOR	5	8.62
ORGAN INVOLVED			
HEPATIC	LIVER	39	67.24
PULMONARY	LUNG	8	13.79
ABDOMINAL	KIDNEY	1	1.72
	SPLEEN	3	5.17
	ABDOMINAL CAVITY	1	1.72
	MESENTERY/OMENTUM	1	1.72
	OVARIES	1	1.72
OTHERS	MUSCLE	2	3.44
	BREAST	1	1.72

TABLE-3 DIAGNOSTIC INVESTIGATIONS

ULTRASONOGRAPHY			NO	%
	NO.OF CYST	ONE	39	6.72
		TWO	8	13.79
		MULTIPLE	11	18.9
	SIZE OF CYST	RANGE		3-15CM
		<5CM	23	39.65
		5-10 CM	30	51.75
		>10 CM	5	8.62
		MEAN		6.94CM
PATHOLOGICAL CONSIDERATIONS				
	ESR	Normal	38	65.5
		Elevated	20	34.48
	Eosinophilia	Present	15	25.86
		Absent	43	74.13
	Serum Bilirubin	>3mg%	1	1.72
	IHT	Negative/Doubtful (<1/320)	5	8.6
		Positive (P1/320)	53	91.37
	ELISA		56	96.55
	HPE		58	100

TABLE-4 CLASSIFICATION (WHO-IWGE standardized classification)[6]

STAGE-I	CE-1	24	41.37
STAGE-II	CE-2	14	24.13
STAGE-III	CE-3A	12	20.68
STAGE-IV	CE-3B	5	8.62
STAGE-V	CE-4	3	5.17

TABLE -5 TREATMENT MODALITIES (SURGICAL)

TREATMENT MODALITIES			
SURGERYPAIR	Partial Pericystectomy (open/lap)	28	48.27
	Partial Pericystectomy with omentoplasty/suture	8	13.79
	Thoracotomy with enucleation and ICD	7	12.08
	Splenectomy	3	5.17
	Excision of cyst	3	5.17
	Explorative laprotomy with excision of cyst	4	6.89
	Excision of renal cyst with external drainage	1	1.72
2)MEDICAL- anti-infective drug treatment;	Albendazole(400mg BD)for 4 mths	58	100
3)PAIR percutaneous treatment of the hydatid cysts	Hepatic	3	5.17
	Pulmonary	1	1.72

RESULT

Clinically and radiologically suspected 58 patients of cystic echinococcosis (CE) which were serologically positive and histologically proved were enrolled in this study. The demographic data of 58 patients who were treated surgically in LN Medical College and Hospital from March 2011 to February 2016 (5yrs) is shown in Table I.

Most of the patients presented with varied clinical presentations depending upon the site and visceral organ involved. (TABLE-II) The spectrum of presentation is listed in Table II As regards the laboratory investigations of cases in TABLE III.

Classification and staging of cystic echinococcosis based on US findings as per WHO-IWGE standardized classification was done in all cases (Table 4). The majority of cysts were unilocular cysts, CE1 (n-24, 51.37%) Stage I, followed by Stage II CE2 (n-14, 24.13%), Stage III CE3a (n-12, 20.68%), Stage IV CE3b (n-5, 8.62%) and Stage V CE4 (n-3, 5.17%).^[6]

STATISTICAL ANALYSIS

The collected data were tabulated and statistically analysed using SPSS software (statistical package for social science) version. Mean [Standard Deviation (SD)] as a measure of central tendency for continuous variables, percentages and number (proportions) for discrete variables were used. We analysed normally

distributed continuous variables by Student's test, proportions by Chisquare test.

DISCUSSION

The worldwide incidence of cystic echinococcosis is estimated to be about 100,000-300,000 cases annually.^[2] It is endemic in many regions of the world including India where the annual incidence of CE persons varies from 1 to 200 per 1,00,000 (105) person years.^[7]

EG inhabits the gastrointestinal tracts of dogs and other wild carnivores as its primary host. Domestic animals, such as sheep, cattle, goats and camels, serve as intermediate hosts as a part of the natural life cycle. By contrast, humans represent an aberrant host, who are infected accidentally where the larval parasite develops into a hydatid cyst.^[3] Humans acquire primary CE by faecal-oral route by consumption of food, water or soil contaminated with echinococcus eggs in carnivore faeces or through direct contact with animal hosts as feeding the dogs, from viscera of slaughtered livestock etc; hence CE is considered as a rural & occupational disease.^[7]

In our study, female predominance was noted with 38(65.5%) females and 20 (34.48%) males with a male: female ratio of 1:1.52. While the most common age group was 30-40 years (29.3%) and the least age group affected was >60 years (5.17%), 75.86% (n=44) of the patients resided in rural areas and only 24.13% (n=14) were urban residents. Farmers and housewives were the most commonly affected occupational groups. 65.66% of patients had history of pet animals, hence exposure to dog sheep etc. Safe drinking water was not available frequently so 62% were exposed to eggs of echinococcosis. Hence the risk factors are female, rural, housewife and farmers, unsafe drinking water with exposure to animals such as dog and sheep.

Hydatid cysts have been reported from every part of the human body like liver, lungs, peritoneum, bones, ovaries, breast, brain, etc, but have special predilection for liver and lungs. Clinical features of CE varies widely depending upon the site of localization, size and the presence or absence of localized and systemic complications.^[7] Predominantly CE presented as hepatic CE in 39 (67.24%) patients, of which 30(51.72%) occurred in the right lobe (67%) and 9 in the left (15.51%). Rest 19(32.75%) were extrahepatic, out of which 7(12.06%) were abdominal CE. Eight (13.79%) presented as pulmonary hydatids, of which four were in the left lung. Rare presentations included one (1.72%) in breast and 2(3.44%) in muscles of legs. Similar findings were seen by MERKEL ET AL.^[8]

Abdominal CE presented mostly with abdominal pain (n=15, 25.86%), 9(15.51%) patients had tender mass and 6 (10.34%) had abdominal fullness. Out of 10 (17.24%) patients presenting with painless, palpable mass, 7(12.06%) had abdominal mass, rest one (1.72%) was in breast and 2(3.44%) in muscles of leg. Hepatomegaly

was observed in 15 patients (25.86%) and jaundice was present in one patient (1.72%) of hepatic CE. Splenic CE cases showed splenomegaly in 3 (5.17%) patients. Pulmonary CE presented with mainly with chest pain and cough (n=5, 62%) rest had cough (n=2, 3.44%), fever (n=2, 3.44%) cough with fever in 2(3.44%) patients and one patient had dyspnoea.

Pallor was present in 5(8.62%) patients. ESR was elevated in 20 cases (34.48%) and eosinophilia was noted in only 15 cases (25.86%). Serum bilirubin was increased only in one patient. Serological diagnosis was conducted using an indirect hemagglutination assay (IHA) which was found positive in 53(91.37%) and ELISA for echinococcosis was found positive in 56(96.55%) cases. The cysts were staged according to the World Health Organization International Working Group on Echinococcosis (WHO-IWGE) standardized classification for ultrasound (US).^[6] Cyst wall of all patients histopathologically showed inner nucleated germinal layer, an outer anucleated chitinous layer and the contents of the cysts having brood capsules and scolices in some. Recent advances in imaging modalities play an important role in diagnosis, classification and evaluation of response to treatment of CE. These findings are in consensus with Haliloglu M, Saatci I, Akhan O, Ozmen MN, Besim A who studied a spectrum of imaging findings in pediatric hydatid disease.^[9,10]

In addition, the immunological tests complement the clinical and radiological diagnostic work-up of CE.^[11,12,13] Definitive diagnosis is done by histopathological documentation of lamellated membrane and scolices. CE are often expensive and complicated to treat, requiring extensive surgery and/or prolonged drug therapy. Ultrasonography was done in all patients. Solitary cysts (n=39, 67.2%) were more common than double (n=8, 13.79%) or multiple cysts (n=11, 18.9%). Cyst size ranged mostly 5–10 cm (51.75%) with a mean size of 6.94 cm. Also, more cysts (51.72%) were found in the right lobe of the liver.

Treatment options for cystic hydatid disease are surgery, drug therapy and percutaneous drainage.^[14,15] The most common surgical procedure performed was a hydatidcystectomy, total or partial in all 58 cases. Partial Pericystectomy, open or laproscopically was done in 28(48.27%), partial pericystectomy with omentoplasty was done in 8 patients of hepatic hydatid (13.79%) In pulmonary hydatidosis, thoracotomy with enucleation and intercostal drainage was done in 7 (12.06%) patients. Splenectomy was done in 3 cases (5.17%). Explorative laprotomy with excision of cyst was done in 4(6.89%) cases and excision of renal cyst with drainage was done in 1(1.72%) patients. In extra abdominal cases, excision of cyst was done in 3 cases (5.17%) None of the patients died due to complications of the disease. There has been no re-representation as recurrence as yet in these patients.

Recent techniques as PAIR technique –ultrasound guided percutaneous aspiration with infusion of scolicalid agents and re-aspiration was done in 4 (6.89%) patients one of pulmonary and 3 of hepatic CE, Also adjuvant medical treatment is given to all patients with albendazole, 400mg twice a day, starting at least 4 days prior to removal and staying for more than 4 weeks.^[16] CE lead to considerable morbidity and socio-economic losses in many regions of the world, recently it was included in the WHO strategic plan (2008-2015) for the control of NTDs and was put under the neglected zoonosis subgroup “neglected zoonotic diseases” (NZD).^[17] According to WHO, these diseases are called “neglected” as these have not been given enough importance at national or international levels by researchers and health policy makers alike even in endemic areas.^[18]

Diagnosis, prevention and control of CE are still at the juvenile stage, despite the pandemic occurrence of the disease. Usually the condition goes unnoticed until the cyst triggers some physical disabilities. Hence, there is an urgent need to create awareness among public and common people who are more prone to the condition. Also there is a paucity of data from Indian studies in terms of clinical presentation, risk factors, investigations and hospital course.

CONCLUSION

The risk factors are female, rural, housewife and farmers, unsafe drinking water with exposure to animals such as dog and sheep. Treatment options for cystic hydatid disease are surgery, drug therapy and percutaneous drainage. The most common surgical procedure performed was a hydatid cystectomy, total or partial. Diagnosis, prevention and control of CE are still at the juvenile stage, despite the pandemic occurrence of the disease. Usually the condition goes unnoticed until the cyst triggers some physical disabilities. Hence, there is an urgent need to create awareness among public and common people who are more prone to the condition.

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