



INTRA-ABDOMINAL DISSEMINATED HYDATID CYST: A SINGLE TERTIARY CENTRE BASED OBSERVATIONAL STUDY

General Surgery

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ABSTRACT

Background: Abdominal disseminated hydatid cyst disease or Abdominal Hydatidosis is a zoonotic disease caused by a tapeworm *Echinococcus granulosus*. Most common site affected in abdomen is liver, but it can affect any part of the body. It can be primary or secondary; primary disseminated intra-abdominal Hydatidosis is rare, accounting 2% of all intraabdominal cysts. **Objective:** Determine the demographic variations, presentation patterns, radiological staging, types of surgical management and outcomes of patients operated for intra-abdominal hydatid cyst (IAHC). **Methodology:** A prospective descriptive study of patients admitted and operated for abdominal Hydatidosis of 11 cases. **Results:** Eleven patients whose mean age is 36.9 years were operated for IAHC. Females (72.7%) were mostly affected. Commonest presenting symptom was abdominal pain (90.9%). History of contact with dog was in nine patients (81.8%). Ultrasonography of whole abdomen and Contrast CT scan of whole abdomen were main imaging studies done on 11(100%) and 8 (72.7%) patients, respectively. Cysts measuring more than 10cm were most common findings in both imaging. Liver is primary site of involvement, 9 (81.8%) cases. Seven patients (63.6%) underwent deroofting, evacuation, marsupialization and omentoplasty (DEMO). There was no recurrence, but 4 (36.4%) patients faced post-operative complications. **Conclusion:** Early diagnosis and avoidance from pets should be done as it may occur anywhere of body. USG and CT remain preferred imaging. DEMO was most common surgery.

KEYWORDS

“Hydatid Cyst”, “Disseminated Hydatidosis”, “*Echinococcus granulosus*”

INTRODUCTION:

Hydatid disease is caused by infestation by the tapeworm *Echinococcus spp.*; mainly two of all known species *E. granulosus* causes hydatid cyst (HC) and *E. multilocularis* causes alveolar hydatid disease [1,2]. Canids and ungulates are the definitive and intermediate hosts in the life cycle of *E. granulosus*. Human is an accidental intermediate host. HC is endemic in Africa, Eastern Europe, Middle East, New Zealand and Mediterranean region [1-3]. HC affects both sexes equally, at all age groups, but mostly affects middle- age group. HC may affect any part of the body, most commonly liver, lung and brain [4-8]. Intra-abdominal hydatid cysts (IAHCs) in addition to liver can be seen in peritoneum, spleen, kidney, pancreas, omentum and biliary tree [6,7,9]. Hydatidosis can be primary or secondary [10]; Primary occurs due to accidental ingestion of *E. granulosus* eggs [11], while secondary occurs due to rupture of cysts [12]. A number of studies have revealed that peritoneal hydatidosis develop by daughter cysts (secondary cysts) [13,14].

Primary peritoneal hydatidosis is rare, accounts only for 2% of cases of abdominal hydatidosis [15].

Symptoms of IAHCs are variable and depends on site, size and stage of development of cysts [16]. As cysts grow slowly in early stage, IAHCs are asymptomatic. Most commonly presents with abdominal pain, nausea, loss of appetite and weight and low-grade fever [6, 16].

Diagnosis of IAHC depends on ultrasonography of abdomen and/ or CECT scan [17,18]. Radiological classification of stages of cyst development done by Gharbi Classification and WHO classification. Laboratory findings include eosinophilia, positive serology like enzyme-linked immunosorbent assay (ELISA), radioallergosorbent test (RAST) and the “arc 5” antibody test [17]. Ultimate confirmation of diagnosis done by histopathology and demonstration of parasitic elements in surgical specimen [2]. The typical hydatid cyst has a three-layer wall surrounding a fluid cavity; outer layer is called Pericyst, mother layer is called Ectocyst and the germinal layer is called Endocyst [2].

Management of IAHC can be medical, surgical or both. Percutaneous approaches are also options [19]. Options and approaches of surgical intervention depends on size, number and site of cysts and mostly done deroofting, evacuation, marsupialization and omentoplasty (DEMO). Resection of part of or the entire affected organ may be necessary. Pre- and post-operative albendazole reduces the rate of recurrence [6,20]. The aim of our study to determine sociodemographic characteristics, presentation patterns, surgical intervention options and outcomes of the patients operated at our hospital.

METHODS AND MATERIALS:

A prospective descriptive study of 11 patients admitted and operated through emergency admission or OPD admission and operation done either elective way or emergency way in our Tertiary Hospital. IAHC is defined by any HC developed in abdominal viscera, peritoneal cavity, omentum, retroperitoneal space or organ. Data is collected in a pretested data collection format by our junior surgical resident. Data was checked for completeness, cleaned, coded, entered and analysed with SPSS version 20. The associations of different variables when applicable were tested for significance in chi-square analysis. Considering confidence level of 95%, a P value of < 0.05 was considered significant in all statistical comparisons. Ethical clearance was obtained from institutional review committee.

RESULTS:

Our study included 11 patients; age ranged from 18 years to 58 years with a mean of 36.9 years. The most affected age group is 31-40 years (Table. 1). Females are predominant, accounting 8 (72.7%) of the patients, making female to male ratio 2.7: 1. Majority of the patients came from urban settings (63.6%).

Most of the patients were admitted on elective (90.9%) basis. Emergency admissions contributed only one (9.1%) of the cases.

Table. 1: Sociodemographic characteristics of patients of IAHC

Features	Number	Percentage (%)
Age group (years):		
1 - 10	0	0%
11 - 20	1	9.1%
21 - 30	2	18.2%
31 - 40	4	36.4%
41 - 50	3	27.3%
51-60	1	9.1%
>60	0	0%
Gender Distribution:		
Male	3	27.3%
Female	8	72.7%
Address:		
Urban	7	63.6%
Rural	4	36.4%
Pattern of Admission:		
Elective	10	90.9%
Emergency	1	9.1%

Most common complaint at presentation was Abdominal pain (10, 90.9%), followed by fever, abdominal lump and loss of appetite/ weight 81.8%, 72.7% and 72.7% respectively (Table.2). Nausea/

vomiting was complained by 5 (45.5%) patients. Jaundice was found in 2 (18.2%) patients. An abdominal mass was documented in 9 (81.8%) cases while hepatomegaly and splenomegaly were present in 5 (45.5%) and 4 (36.4%) patients, respectively. Ascites and abdominal guarding were present in one case (9.1%) each. Nine patients (81.8%) had history of close contact with dog. History of smoking and alcohol intake were present in 4 (36.4%) and 5 (45.5%) of cases, respectively.

Table.2: Clinical Findings of the patients with IAHC

Complaints at presentation:	Number	Percentage (%)
Abdominal Pain	10	90.9%
Nausea/ Vomiting	5	45.5%
Abdominal Lump	8	72.7%
Appetite / Weight loss	8	72.7%
Fever	9	81.8%
Haematuria	1	9.1%
Jaundice	2	18.2%
Asymptomatic	1	9.1%
Physical Examination:		
Abdominal mass	9	81.8%
Hepatomegaly	5	45.5%
Abdominal Tenderness	3	27.3%
Jaundice	2	18.2%
Chest Findings	0	0%
Fever	8	72.7%
Splenomegaly	4	36.4%
Ascites	1	9.1%
Guarding / Rigidity	1	9.1%
Personal History:		
H/O contact with Dog/ Pets	9	81.8%
H/O Smoking	4	36.4%
H/O Alcohol intake	5	45.5%

Complete blood count was done for every patient and leucocytosis was found in 7 (63.6%) patients. Liver enzymes were normal, but only in two (18.2%) patients had elevated direct and total bilirubin. Abdominal ultrasonography and CECT abdomen were the main imaging studies done on 11 (100%) and 8 (72.7%) patients, respectively (Table.3). In both intracavitary daughter cysts were seen in 45.5% (USG) and 50% (CT scan) of the patients. A diagnosis of HC was ascertained by intraoperative findings of different parasitic material and layers and daughter cysts in 95% of cases and confirmed by histopathology.

Table.3: Imaging Results of the patients with IAHC

Ultrasonography of Whole Abdomen:			
Characteristics	Description	Number	Percentage (%)
Cyst Size	>10 cm	7	63.6%
	5 – 10 cm	3	27.3%
	<5 cm	1	9.1%
Cyst Nature	Clear fluid, unilocular (I)	1	9.1%
	Clear fluid, multiseptated (III)	3	27.3%
	Total/ partial separation of membranes (II)	2	18.2%
	Intracavitary native cysts (daughter cysts) (III)	5	45.5%
	Slid like mass (IV)	0	0%
	Calcified walls (V)	0	0%
CT scan Whole Abdomen Findings:			
Characteristics	Description	Number	Percentage (%)
Cyst Size	>10 cm	5	62.5%
	5 – 10 cm	2	25%
	<5 cm	1	12.5%
Cyst Nature	Clear fluid, unilocular (I)	1	12.5%
	Clear fluid, multiseptated (III)	2	25%
	Total/ partial separation of membranes (II)	1	12.5%

Intracavitary native cysts (daughter cysts) (III)	4	50%
Slid like mass (IV)	0	0%
Calcified walls (V)	0	0%

Pre-operative Albendazole was given to all 10 (90.9%) patients for at least three days as per patient's body weight (not exceeding 800 mg/ day). Most common incision was midline incision followed by right subcostal incision. Liver was most involved organ 9 (81.8%) followed by spleen and peritoneum 7 (63.6%) each (Table.4). Mesentery and small intestine were involved in 5 (45.5%) and 4 (36.4%) cases respectively. Biliary tree was involved in two (18.2%) of the patients.

Table.4: Intra-operative findings of Hydatid cysts involving different viscera

Organs involved	Number	Percentage (%)
Liver	9	81.8%
Spleen	7	63.6%
Small Intestine	4	36.4%
Mesentery	5	45.5%
Peritoneum	7	63.6%
Bladder Wall	1	9.1%
Large Intestine	1	9.1%
Omentum	3	27.3%
Urinary Bladder	1	9.1%
Biliary tree	2	18.2%

Seven (63.6%) patients underwent deroofing (of the cyst wall), evacuation (of the content), marsupialization (of the edge) and omentoplasty (obliteration of the cavity by placing omentum) (DEMO) (Table.5). Splenectomy was done in two (18.2%) cases. Pericystectomy, Excision of cyst and En bloc bowel resection were done in one (9.1%) case each. In all cases there were moderate to severe adhesions between cyst and surrounding structures.

Table.5: Types of surgery in patients with IAHC

Types of Surgery	Number	Percentage (%)
DEMO	7	63.6%
Peri cystectomy	1	9.1%
Splenectomy	2	18.2%
Excision	1	9.1%
En bloc bowel resection	0	0%
Laparoscopic drainage	0	0%

Intra-operatively hollow viscous perforation occurred in one (9.1%) case, contamination with cyst fluid/ rupture occurred in one (9.1%) case (Table.6).

Table.6: Intra-operative Complications

Complications	Number	Percentage (%)
Hollow viscous injury	1	9.1%
Bleeding	2	18.2%
Contamination with Scolices	1	9.1%
Anaphylaxis	0	0%

Four (36.4%) of the patients developed post-operative complications including three (27.3%) cases with surgical site infection, one (9.1%) case with bile leak. Bile leaks stopped with follow up with drainage tube. No patient recurred with same during 6 months follow up (Table.7).

Table.7: Post-operative Complications

Description	Number	Percentage (%)
Surgical site Infection	3	27.3%
Bile Leakage	1	9.1%
Sinus tract formation	0	0%
Recurrence	0	0%

DISCUSSION:

IAHC is very rare condition. Though this disease is supposed to be common in rural settings, our findings showed a relatively higher occurrence in patients of urban area. Bilutse et. al. also showed 60% of their patients with HC of liver to be from urban settings [6]. This may be due to immigration of patients from rural area to urban for economic reasons or in urban area patients were bound to be in close contact with pets due to space crisis.

The finding of young adults affected more commonly, shown in

literatures from Ethiopia and other developing countries [3,6]. Studies from Bahir Dar, Northern Ethiopia and Addis Ababa showed HC were common in third decade of life (21 – 30 years) with a mean age of 37 and 33.5 years, respectively [3,6]. Studies by Ahmadi et. al. and Mouaqit et. al. showed mean age of 40 years and 39.5 years, respectively [16,21].

Our study showed a female preponderance for IAHC like other studies of Kebede et. al. and Ahmadi et. al. where females made up 83% and 58% of the patients [3,16]. Similar findings revealed from studies by Ghartimagar et. al., Alghoury et. al. and Jastaniah et. al. [22-24]. Females have the disease more than males due to their more intimate and frequent contact time with domestic pets.

Like other literatures abdominal pain was the most common presenting symptoms in our study [6,16]. Malik et. al. and Akhter et. al. also showed abdominal pain as most common presenting symptoms, 76.8% and 89.5%, respectively [25,26]. Bilutse et. al. reported abdominal pain as main presenting complaint in 84% cases [6]. This is easily explainable as liver is the most common affected organ and due to stretch of liver capsule and irritation of parietal peritoneum patients complains of pain.

Pre-operatively diagnosis of HC was made with imaging studies and confirmed by intraoperatively and followed by histopathology. Abdominal USG and CT scan is very good investigation defining size, site and stages of development of cysts. The sensitivity and specificity of USG in detecting HC reported in literature were very good as per study by Wuestenberg et. al. and Marrone et. al. [17,18]. The rate of CT utilization was less than that of USG [18]. The reason for this could be that USG was the first screening and diagnostic imaging which revealed the diagnosis.

The finding of liver in general was the most common site involved in IAHC in our study is in agreement of studies by Kebede et. al., Bilutse et. al. and Ahmadi et. al. [3,6,16]. Liver is most commonly affected organ as it is the first organ which takes parasite infested portal venous system by Pedrosa et. al. [2].

Major physical finding was abdominal mass mostly due to hepatomegaly and followed by splenomegaly. This is most likely due to huge size of cyst >10 cm in more than 60% cases. This is understandable as small cysts are asymptomatic and patients do not seek medical intervention [2,8,16].

Management options for HC ranges from simple cyst drainage to resection of whole or part of involved organ as per studies by Bilutse et. al. and Gourgiotis et. al. [6,27]. Major from surgery as per our study was DEMO. This was mainly due to nature of cysts in which most are intraparenchymal liver cysts or peritoneal HC with extensive surrounding structure adhesions. The outcome of DEMO was also comparable to literatures [6,27]. Though pre-operative scolicalid agent administration rate was low, use of pre-operative and long term post-operative albendazole administration was good as per WHO guidelines [28].

CONCLUSION:

Abdominal pain and mass are most common complaint and findings of patients with IAHC. Patients should be made aware of avoidance of close contact from domestic pets. Adult age group is most commonly affected, so we should think of some deworming programme for those who are in frequent contact time of domestic animals. We should conduct a multicentre large scale prospective study to determine all clinical findings and management options to establish a guideline about its management.

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