

A Retrospective Descriptive Study of Hydatid Disease in Unusual Location Carried Out in Tertiary Care Hospital

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Abstract

Background: Hydatid disease or echinococcosis is an important zoonosis, and a serious public health problem both globally, and especially in endemic country like India. This disease is usually found in liver and lungs, but unusual sites in the body can pose a diagnostic challenge. The aim of this study was to evaluate the epidemiology, clinical picture and different localisations of hydatid cyst and to suspect this disease in any cystic lesion affecting any organ in the body, especially in endemic areas of the world. *Material and Methods:* Total of 72 cases were analysed and a retrospective descriptive study was carried out in our institute over a period of 6 years from 2010 to 2015. We screened all the histopathology records diagnosed with hydatidosis and details are collected from their case sheet. *Results:* Out of 72 patients, the dominant sex affected by the disease were females 45(62.5%) and 27(37.5%) males. Hydatidosis more commonly affect in the second decade of life followed by third decade. Overall pain in abdomen was the most common presenting complaint. Among the unusual sites most common presentation was solitary, cystic swelling. Liver was the most commonly affected organ. We reported 11 cases of hydatidosis in unusual localisations. *Conclusion:* Hydatid disease is still a problem and burden in rural regions of India. A high index of suspicion, radiological investigations as well as histopathological examination is necessary in establishing the diagnosis of hydatid disease at unusual sites in the body.

Keywords : Hydatid Disease; Echinococcosis; Unusual.

Introduction

Hydatid disease or Echinococcosis is a zoonotic infection, caused by larval forms of *Echinococcus granulosus*. It has a world wide distribution and causes most important health problems in endemic countries like India. In our country this disease is highly prevalent in Kashmir, Andhra Pradesh, Tamil nadu, and central India [1,2]. It is estimated that world wide incidence of hydatidosis is about 100,000-300,000 cases annually and is known to occur in at least 100 countries [3,4].

Hydatid cyst disease can occur in any site of the body most commonly involving the liver (55-70%) and

the lung (18-35%). The two organ can be affected simultaneously in about 5-13% of cases. The other rare sites involved by hydatid cyst are kidney, muscle, peritoneal cavity, spleen, pancreas, gall bladder, thyroid, breast, brain, supraclavicular region, pericardium, diaphragm, pleural cavity and thigh [5]. The exact percentage of site involvement varies and the exact incidence of unusual locations is difficult to ascertain as they are only reported as case reports.

Echinococcus cyst following a primary infection may inhabit any anatomic site from head to toe [5]. Primary definitive host is carnivorous animal, intermediate host is herbivorous animal. Human act as a accidental intermediate host after ingesting viable oncosphere containing eggs, which are shed in the faeces of the definitive host. The ingested ova penetrate the intestinal wall, reach the portal system and from there to liver where most of them lodged in the hepatic sinusoids [6]. A few ova may pass through the

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liver(first filter) and reach the lungs(second filter) and then into the systemic circulation, causing hydatid disease in other organs. A possible dissemination through lymphatic channels accounts for cases with hydatid cysts at uncommon sites [7].

Clinical features was varying, majority of patients presented with pain abdomen with lump in right upper quadrant in patient with hydatid cyst of liver. Pulmonary hydatid disease presented with fever, chest pain and cough. Parotid hydatid cyst presented as parotid swelling mimicking pleomorphic adenoma. Hydatid cyst in Paravertebral muscle was presented as painless swelling over right side of back with no history of trauma, fever, weight loss. Muscle hydatidosis presented as soft tissue lump. Omental and pelvic hydatid cyst disease presented as cystic disease, diagnosed with help of CT scan and MRI [8].

It may present in an unusual manner in this usual and uncommon sites. In the literature, it is very rare to see the disease in the sites reported in this article. Localization at unusual sites in body can have an atypical presentation, which pose a diagnostic challenge and mislead the treatment including inadvertent use of interventional investigations that may complicate the outcome and prognosis.

In this study we have defined various epidemiological, clinical factors and anatomical location of hydatid cyst. Due to its presentation in unusual manner and in unusual sites, it is necessary to suspect this disease in any cystic lesion affecting any organ in the body, especially in endemic areas of the world, hygienic measures is to be educated. So, that the prevalence and burden of disease can be minimized

Material and Methods

A retrospective, descriptive study was carried out in a tertiary care hospital attached to a medical college situated in a rural area, which has a high zoonotic exposure. This study was conducted on approval from ethical committee during a period of 6 years from 2010-2015. We included all the patients admitted in our hospital, with cases confirmed histopathologically as cystic hydatidosis. The samples included the specimens from those patients diagnosed as hydatidosis received in histopathology department. Tissue was already processed by routine paraffin techniques and sectioned stained by haematoxylin and eosin stain.

The patients were evaluated regarding their age, sex, occupation, site of cyst, unusual presentations,

unusual localizations. Radiological details including ultrasonography(USG- site of CE, size of the cyst), computed tomography (CT Scan) records, routine lab investigation, serology, histopathological diagnosis and operative details were collected from their case sheets. In all the cases enucleation or evisceration of cyst was done by open surgery or laproscopic surgery. One patient with brain involvement had left against medical advice. We excluded all patients whose hospital records were not available. The data obtained were compared with other national and international research studies to compare the statistical equation.

Criteria for diagnosis of cystic hydatid disease are presence of lamellated structure of ectocyst, germinal layer, scolices and protoscolices on Hematoxylin and Eosin (H&E) stained paraffin- embedded sections

Results

A total number of 72 patients with hydatid disease were identified. Majority of the patients were females (62.5%) followed by males (37.5%). The male to female ratio is 1:1.67. Age of the patient were ranged from 7 years to 70 years. Most of the patient involved were in the 2nd decade(21 cases, 29.16%) followed by 3rd decade(16 cases, 22.22%). Age and sex distribution are shown in Table 1.

Housewives had the highest rate of infection(26 cases, 36.1%) followed by farmers (19 cases,26.38%), daily wage workers (15 cases, 20.8%), and others (12 cases,16.67%).

Most common sites involved according to incidence are : liver (54.16%), lung (30.55%). 11 patients had unusual localisations. Kidney (4.16%), Muscle (2.77%) and pelvic cavity (2.77%) are the most frequently sites involved after liver and lung. The organ distribution of the hydatid cyst is shown in Table 2 and details of site of unusual cases are shown in Table 3. In this series, out of the total study population of 72 patients, 61(84.7%) had single organ involvement and 11 patient (15.27%) had more than one organ involvement

The main presenting symptom of the study population with liver involvement was pain in abdomen followed by lump, anorexia, vomiting and hepatomegaly. Pulmonary hydatid disease (Figure 1) presented with chest pain, cough, hemoptysis and fever. Renal involvement is asymptomatic. Muscle involvement usually revealed as painless enlarging soft tissue masses. Brain involvement present with focal neurological deficit, features of raised intracranial pressure and a solitary cystic lesion. Hydatid cyst of parotid region (figure 2) present as

gradually increasing swelling, freely mobile cystic lesion. Pelvic hydatid cyst present with lump in lower abdomen, constipation on and off (Figure 3) .

Diagnosis in all cases of unusually location require radiological support either by USG, CT – alone or in conjunction. Histopathological findings confirmed the diagnosis(Figure 4). Gross appearance of the cyst is

like pearly white cyst wall. Microscopic examination of cyst wall of hydatid cyst shows outer fibrous layer, middle lamellated cutical layer, and inner germinal layer with brood capsules. Daughter cysts having invaginated morphology alongwith scolex and germinal lining.

Table 1: Age and sex distribution of hydatid cyst disease

Age in years	Number of Male	Number of Female	Number	Total Cases	Percentage
0-10	2	-	2		2.77%
11-20	3	5	8		11.11%
21-30	6	15	21		29.16%
31-40	4	12	16		22.22%
41-50	4	7	11		15.27%
51-60	5	4	9		12.5%
61-70	3	2	5		6.94%
Total	27(37.5%)	45(62.5%)	72		100.0%

Table 2: Distribution of hydatid cyst disease based on localisations

Organs	Number of cases	Percentage
Liver	39	54.16%
Lungs	22	30.55%
Kidney	03	4.16%
Muscle	02	2.77%
Pelvic cavity	02	2.77%
Peritoneal cavity	01	1.38%
Mesentry/omentum	01	1.38%
Brain	01	1.38%
Parotid	01	1.38%

Table 3: Details of age, sex and unusual site of hydatid cyst

S. No.	Age, Gender	Site
1.	22 ,Female	Perineal region
2.	65, Female	Peritoneum
3.	15, Female	Left Kidney
4.	44, Female	Upper left arm
5.	45, Female	Right kidney
6.	60, Male	Intracranial
7.	20, Male	Right Paravertebral Muscle
8.	40, Male	Right upper arm
9.	28, Female	Right kidney
10.	29, Female	Pelvic cavity
11.	35, Male	Left parotid gland



Fig. 1: Photograph of Hydatid cyst lung



Fig. 2: [A]Photograph of Hydatid cyst parotid gland. [B] Pearly white cyst wall



Fig. 3: (A,B) Photograph showing excised pelvic hydatid cyst

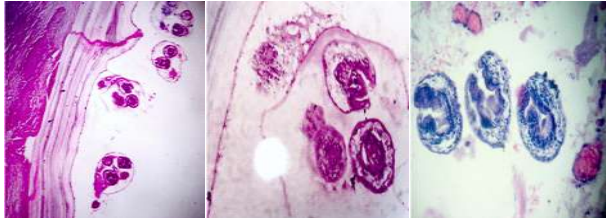


Fig. 4: Photomicrograph showing [A] Cyst wall of hydatid cyst having outer fibrous layer, middle lamellated cuticular layer, and inner germinal layer with brood capsules(H and E,10X) [B,C] Photomicrograph showing daughter cysts having invaginated morphology alongwith scolex and germinal lining(40X)

Discussion

Hydatid disease is one of the world's most well known geographically widespread parasitic zoonosis and is a serious concern in the endemic region due to severe disease, increase in morbidity and considerable economic loss. Most human infection are due to *E. granulosus* transmitted between domestic dogs and livestock. The annual incidence is varying, 1 to 200 per 100000 population [9].

Our study revealed that, this disease commonly affect patients in their second decade followed by third, this is in accordance with other studies [10-14]. It has been consistently seen in various studies and confirmed by our study that this disease is seen in young patients who are in their active years.

The risk factor identified in our study were females, mainly housewives. These results are consistent with reports from other studies [15,16]. It can be postulated that since in the rural community, females are more involved in activities related to farming and herding livestock. And probably this may be the reason for transmission of disease by contact with eggs-contaminated plants or soil followed by direct hand-to-mouth transfer [17].

There are no specific local or general symptoms and signs of hydatid disease. Most common presenting symptom in liver hydatidosis, in our study was pain in abdomen, followed by lump, which is in accordance with other studies [18,19]. But it is in contrary to study

in which lump was the presenting symptom [14]. The presenting symptom in unusual localisations are mainly solitary, cystic lesion associated with or without pain and localized symptom of neurological deficit, pain and hemoptysis.

The disease usually involves the liver and lungs, but any organ or tissue can be involved by hydatid disease. So apart from liver and lung, other localization where this disease is considerably seen are peritoneal cavity(10-16%), muscle(0.5-4%), kidney(1.5-4%), pelvic cavity(1-3%), brain(3%), retroperitoneum(1%), mesentery/omentum and parotid gland [20]. In the present study, the involvement of kidney(4.16%) were predominant among unusually located hydatid cyst. Such results were reported by previous study [18,22]. Hydatid disease of kidney account for 2 to 3 % of all hydatid disease, this is in accordance with our study with a slight higher rate [20,23]. Primary muscular hydatid cysts are also rare accounting for 3% of all patient in previous study. Our study reveals similar results account for (2.77%) with hydatidosis [20,24]. Incidence of hydatidosis in pelvic region in our study is (2.77%), which is also similar to previous study [25]. Hydatid disease localization in other sites are also rare, but with a slight higher rate in comparison to previous study [11]. This may be explained because of small sample size.

In our study, Renal involvement may be alone or in combination with liver involvement. Primary muscle hydatidosis is also rare, with only selected reports in the literature [26]. Muscle hydatidosis has low incidence, possibly because of muscle lactic acid content and muscle contraction, two factors that likely prevent cyst growth in striated muscle.

Diagnosis of this disease is based on clinical suspicion and epidemiological data, but is often made difficult by the variable signs and symptoms in case of unusual location. Laboratory tests include the haematological tests, biochemical liver function tests have low sensitivity. The casoni skin test has been abandoned as a diagnostic tool since 1980. Other serological tests like antibody titer to hydatid antigen by ELISA, latex agglutination and indirect haemagglutination tests may be helpful [27]. USG and CT are very useful in revealing well-defined cysts with thick or thin wall, containing daughter cysts, septa, calcification and hydatid sand can be visualized [28,29]. FNAC is also useful diagnostic method, as it doesnot cause any complication and can help in diagnosis and useful when serology is negative as by some authors [28]. While other discourages FNAC for the diagnosis. So clinical suspicion, radiological investigation and histopathological examination are necessary in establishing diagnosis.

Conclusions

Hydatid disease is a common health problem in India, prevalent in rural areas, where population are unaware of this parasite. Higher infection rate is seen in housewives and farmers, who are mainly involved in rearing cattles and herding. In spite of its usual location, unusual localization of this disease should be considered in the differential diagnosis of any cystic mass in all anatomic locations as the presentation is with non specific symptoms. A high index of suspicion, radiological investigations as well as histopathological examination is necessary in establishing the early diagnosis of hydatid disease at unusual sites in the body. Improvement in personal hygiene and awareness regarding disease is required and will result in control and eradication of this disease.

References

1. Reddy CR, Narasiah IL, Parvathi G, Rao MS. Epidemiology of hydatid disease in Kurnool. *Indian J Med Res.* 1968; 55:499-507.
2. Amir-Jahed AK, Fardin R, Farzad A, Bakshandeh K. Clinical echinococcosis. *Ann Surg.* 1975; 182(5):541-6.
3. Richter J, Orhun A, Gruner B, Muller-Stover I, Reuter S, Romig T, et al. Autochthonous cystic echinococcosis in patients who grew up in Germany. *Eurosurveillance* 2009; 14:1-7.
4. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis: A zoonosis of Increasing Concern. *Clin Microbiol Rev* 2004; 17: 107-35.
5. G. Grosso, S. Gruttadauria, A. Biondi, et al. Worldwide epidemiology of liver hydatidosis including the Mediterranean area. *World Journal of Gastroenterology* 2012; 18:1425-1437.
6. Prousalidis J, Tzardioglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. *World J Surg* 1998; 22:17-22.
7. Saidi F.: Nyhus LM, Beker JR, Fsicler JE. Treatment of echinococcal cyst. In. *Mastery of Surgery.* 3rd Edn. Little, Brown and Company 1998; 1035-2
8. Wani RA, Wani I, Malik AA, Parray FQ, Wani AA, Majid A .Hydatid disease at unusual sites. *International Journal of Case Reports and Images* 2012; 3(6):1-6.
9. Parija SC. A textbook of medical parasitology: 2nd edition. Madras: All India publishers and distributors; 2004; 220-9.
10. Rao SS, Mehra B, Narang R. The spectrum of hydatid disease in rural central India: An 11-year experience *2012; 5(3):225-230.*
11. Mathur PN, Parihar S, Joshi CP, Kumawat JL. Hydatid disease-still endemic in the southern region of state of Rajasthan, India: a clinical study carried out in tertiary care hospital *Int Surg J.* 2016 Nov; 3(4): 1802-1805.
12. Papadimitriou J. The surgical treatment of hydatid disease of the liver. *Br J Surg* 1970; 57:431-3.
13. Irshadullah M, Nizami WA, Macpherson CN. Prevalence of human hydatidosis in Uttar Pradesh. *J Commun Dis* 1989; 21:114-22.
14. Mehta RB, Ananthkrishnan N, Gupta BK, Srivastava KK, Mehdiratta KS, Satya P. Hydatid disease in Pondicherry. *Indian J Surg* 1982; 44:88-94.
15. Baharsefat M, Massoud J, Mobedi I, Farahnak A, Rokni M B Seroepidemiology of human hydatidosis in Golestan province, Iran. *Iranian J. Parasitol.* 2007; 2: 20-24.
16. Pezeshki A, Kia EB, Gholizadeh A, Koohzare A. An analysis of hydatid cyst surgeries in Tehran Milad hospital, Iran, during 2001-2004. *Pak. J. Med. Sci.* 2007; 23:138-140.
17. Al Barwari SE, Saeed IS, Khalid W, AlHarmni KI. Human hydatidosis in Arbil, N Iraq *Jour Islamic Aca Sci.* 1991; 4:330-5.
18. Balik AA, Basoglu M, Celebi F, Oren D, Polat KY, Atamanalp SS, et al. Surgical treatment of hydatid disease of the liver: Review of 304 cases. *Arch Surg* 1999; 134:166-9.
19. Langer JC, Rose DB, Keystone JS, Taylor BR, Langer B. Diagnosis and management of hydatid disease of the liver: A 15 year North American experience. *Ann Surg* 1984; 119:412-7.
20. Wilkinson A, Mahore S, Chikhlikar K, Gohad G, Bothale K Hydatid cysts presenting in uncommon sites- an uncommon case series *SEAJCRR* 2014; 3 (1):645-656.
21. Balik A, Celebi F, Basoglu M, Oren D, Yildirgan I, Atamanalp S S. Intra-abdominal extrahepatic echinococcosis. *Surg. Today* 2001; 31:881-884.
22. Col C, Col M, Lafci H. Unusual localizations of hydatid disease. *Acta Med Austriaca* 2003; 30:61-64.
23. Gogus C, Safak M, Baltaci S, Turkolmez K. Isolated renal hydatidosis: experience with 20 cases. *J Urol* 2003; 169(1):186-9.
24. Verasaci A, Scuderi G, Rosato A, et al. Rare localizations of echinococcosis; personal experience. *ANZ J Surg* 2005; 75:986-1.
25. Tarcoveanu E, Dimofte G , Bradea C , Crumpei F , Anton R , Moldovanu R. Multiple peritoneal hydatid disease after rupture of a multivesicular hepatic hydatid cyst : case report . *J Gastrointestin Liver Dis* 2006; 15:301-5.
26. Khanna AK, Prasanna GV, Khanna A. Unusual sites

- of hydatid cysts in India. Trop Doct 2005; 5:233-235
27. Abu-eshy SA: Some rare presentations of hydatid cyst (Echinococcus granulosus). R. Coll, Surg. Edinb: 1998; 43:347-52.
28. Safioleas MC, Misiakos EP, Kouvaraki M, Stamatakos MK, Manti CP, Felekouras ES. Hydatid Disease of the Liver A Continuing Surgical Problem. Arch Surg 2006; 141:1101-8.
29. Battyany I, Herbert Z, Rostas T, Vincze A, Fulop A, Harmat Z et al. Successful percutaneous drainage of a giant hydatid cyst in the liver. World J Gastroenterol; 2006; 12(5):812-14.
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