



## Original Article

# The Spectrum of Hydatid Disease in North Telangana: A Study of 17 Cases

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## ABSTRACT

**Introduction:** *Hydatid disease or Echinococcosis is a zoonotic disease caused by the larva of Echinococcus species and is one of the oldest known diseases to man. The disease has a worldwide distribution and is also well recognized and documented in India.*

**Aims & Objectives:** *This study was carried out with the aim of describing the epidemiology (demography, clinical presentation, imaging characteristics, and in-hospital course) of Cystic Echinococcosis (CE) in central India. The present study is done to know the incidence and various sites affected by the parasite.*

**Materials & Methods:** *A total of 17 cases were diagnosed during the period of 3 years, from December 2013 to November 2016.*

**Results:** *Females were the dominant sex affected by the disease. Hydatid of the liver was more common, especially in the right lobe. Pain in the abdomen was the most common presenting complaint. Lump in the abdomen was the most common clinical finding. Partial pericystectomy and external drainage was the most commonly performed surgery. The most common postoperative complication was wound infection.*

**Conclusion:** *This study highlights the epidemiology of Cystic Echinococcosis (CE) in the North Telanagana. The population in rural areas are more exposed to zoonotic diseases. Proper education, creating awareness, and implementing strict rules regarding the disposal of remains of slaughtered animals can help eradicate this disease.*

**Keywords:** *Echinococcosis, biceps, retroperitoneum, pericystectomy.*

## INTRODUCTION

Hydatid disease or Echinococcosis is a zoonotic disease caused by the larvae (metacestode) of the cestode species of the genus *Echinococcus* like *E. granulosus*, *E. multilocularis*, *E. vogeli* or *E. oligarthus*.<sup>[1]</sup> Classical Cystic Echinococcosis (CE) is caused by *E. granulosus* complex,

while *E. multilocularis* and *E. vogeli* are responsible for alveolar echinococcosis and polycystic echinococcosis, respectively.<sup>[2]</sup> The study of *E. granulosus* species has revealed identification of substantial phenotypic and genetic variability and several strains.<sup>[2]</sup>

Primary hydatid disease of musculoskeletal system is rare. Echinococcal cysts are usually found in liver and lungs, but can affect any part of the body. Differential diagnosis of hydatid disease should be considered for every soft cystic mass in any anatomical location, especially in areas where the disease is endemic. Hydatid disease has a worldwide distribution and causes health problems in endemic countries. The prevalence of primary muscle hydatid disease is reported to be only 0.5 – 4.7%. Hydatid disease, which is a zoonotic infection caused by larval forms (metacestodes) of tapeworms of the genus *Echinococcus* found in the small intestine of carnivores, still remains an important health problem in endemic regions. Two of the four recognized species of *Echinococcus*: *E. granulosus* and *E. multilocularis*, cause cystic echinococcosis (CE) and alveolar echinococcosis (AE) in humans, respectively. The eggs of these tapeworms excreted by carnivores may infect humans as natural intermediate host. Hydatid cysts, which generally involve the liver and the lungs, are uncommonly found in muscles; even in endemic zones. Exclusive involvement of the muscles is extremely uncommon, because implantation at this site would require passage through the filters of the liver and lung.

## MATERIALS & METHODS

The present study was conducted in Prathima Institute of Medical Sciences, Karimnagar for a period of three years. All the patients in our study came with complains of swelling in the body with associated symptoms. All routine investigations were done and special investigations related to the location of the swelling were also performed and final diagnosis of Hydatid cyst was done. All the excised mass were sent to histopathology and it was confirmed as Hydatid cyst on microscopy also.

## RESULTS

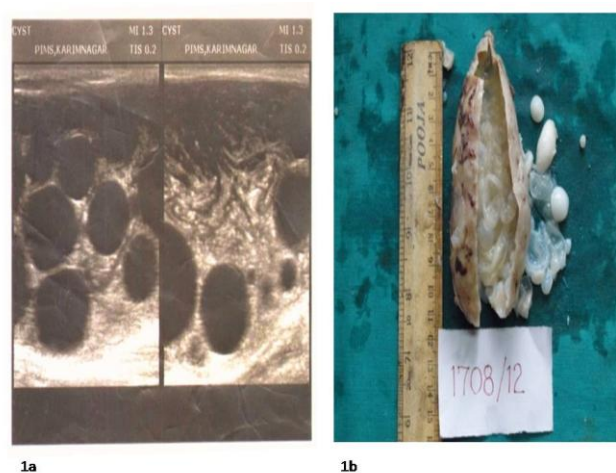
A total of 17 cases were diagnosed as Hydatid cyst in the present study. The various sites

affected were shown in Table 1. Females are more affected than males, with 12 and 5 cases.

Liver was the most common site involved in the present study, with one case each of retroperitoneum, kidney, brain and biceps muscle.

**Table 1:** Shwoing location of cases

Location	Number of cases
Liver	08
Spleen	05
Biceps brachii	01
Brain	01
Retroperitoneum	01
Kidney	01
Total	17



**Figure 1:** Ultrasonography showing multiple cystic swellings of varying sizes originating from muscle tissue (a). Gross photograph showing multiple daughter cysts (b).



**Figure 2:** CT brain showing a well defined hypodense lesion in left parieto temporal region (a). Gross picture of cyst (b).

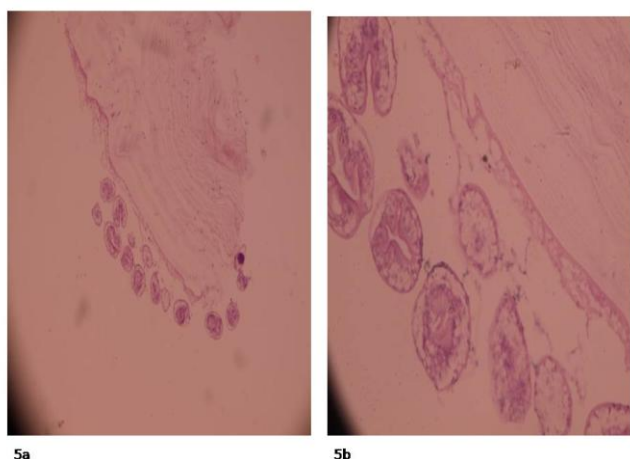




**Figure 3:** CECT showing presence of multiple large cystic lesions noted in the right lobe of liver extending into sub hepatic region largest of size 6 cm displacing the kidney anteriorly (a). Gross showing multiple daughter cysts (b).



**Figure 4:** Gross showing multiple daughter cysts from nephrectomy specimen.



**Figure 5:** Section showing cyst wall comprising of outer chitinous and inner germinative layer along with scolices and hooks. Adjacent to these there is cellular eosinophilic laminated

membranous structures seen ( a,X10 H&E) Section showing scolices and hooks of varying sizes ( b, X40 H&E).



**Figure 6:** Gross showing multiple daughter cysts excised from liver.

**DISCUSSION**

Musculoskeletal hydatid disease may either be secondary or primary. In secondary disease, there is primary location of hydatid cyst in liver, lung or spleen that has been operated or not operated. Primary hydatid disease of the skeletal muscle is rare, as the parasite has to cross pulmonary and hepatic barriers to reach the muscles. The high lactic acid level in muscle tissue is considered unfavourable for the survival of parasite.

In our study we had a case of Hydatid cyst involving biceps brachii muscle. This patient had not been operated for hydatid disease previously and investigations did not revealed any hydatid cyst in brain, liver, lung or spleen. So, this patient was diagnosed, having primary hydatid disease of musculoskeletal system [Figure 1]. Muscular hydatidosis is very rare and can cause difficulty in diagnosis, especially in the absence of typical radiological findings. The possibility of hydatid disease should always be kept in mind in the differential diagnosis of a cystic mass in the muscle, especially in endemic areas to avoid fine-needle biopsy and the consequences of spillage of cyst contents.

Cerebral Hydatid disease is a rare space occupying lesion and more common in paediatric population<sup>[3,4]</sup> The parietal lobe is the most

frequently involved region.<sup>[5]</sup> In India it is more commonly seen in kurnool district of Andhra Pradesh, madurai district of Tamil nadu and in Punjab.<sup>[5]</sup> Cerebral hydatid disease may either be secondary or primary. In secondary disease, there is primary location of hydatid cyst in liver, lung or spleen that has been operated or not operated. The primary cysts are formed as a result of direct infestation of larvae in the brain without demonstrable involvement of other organs. Primary hydatid disease of the brain is very rare, as the parasite has to cross pulmonary and hepatic barriers to reach the nervous system. The primary cysts are fertile as they contain scolices and brood capsules, hence rupture of primary cyst can result in recurrence. The secondary multiple cysts result from spontaneous, traumatic or surgical rupture of primary intracranial Hydatid cyst and lack brood capsules and scolices.

Patients with intracranial Hydatid cysts present with focal neurological deficit and features of raised intracranial pressure. The treatment of Hydatid cyst is surgical and the aim of surgery is to excise cyst in toto without rupture to prevent recurrence or anaphylactic reaction. In our study we reported one case involving central nervous system in a child who presented with headache and was diagnosed as hydatid cyst on Ultrasound and finally on biopsy [Figure 2].

Man is an accidental host in the life cycle of the *E. granulosus*. Life cycle has definitive host which is dog, intermediate host is sheep and humans are accidental intermediate hosts. When they get infected from dogs, infestation occurs when the infected cestode eggs are swallowed<sup>2</sup>. In the stomach, the outer protective coat of egg is digested and larvae are liberated. These penetrate the mucosa of proximal bowel to enter the portal system. About 85-95% of the larvae are trapped in the liver & lung & only 5-15% escape into systemic circulation to involve other organs - muscle, kidney, retroperitoneum, brain.

Retroperitoneum involvement was always thought to be secondary to rupture / spillage during surgery of liver hydatids. Primary retroperitoneal

hydatid cysts are rare. The development of cyst in liver is often responsible for severe complications including local infection, biliary fistula, rupture to peritoneum/chest. Correct pre op diagnosis is difficult to make. Diagnosis is made with good imaging techniques. We here present a case of Hydatid cyst involving retroperitoneum. Examination of abdomen revealed a lump in the right lumbar region, firm, moving up and down with respiration, lower border is well defined and is 6 fingers below the costal margin. Dullness over the swelling is continuous with the liver. It was diagnosed as Retroperitoneal Hydatid cyst [Figure 3].

Another case was diagnosed in a 32 years old male patient complaining of left sided flank pain and passage of small, pearly white balloon like grape size structures in the urine for past 15 days. USG showed a well defined cystic lesion occupying the upper and mid pole of left Kidney measuring 4x3 cm with multiple cysts of varying sizes and hyperechoic stroma. It was associated with a moderate left hydronephrosis. Radiologically it was diagnosed as Hydatid cyst. The patient underwent left nephroureterectomy through the flank extra peritoneal approach [Figure 4]. All the excised samples were sent for Histopathology and microscopy showed cyst wall comprising of outer chitinous and inner germinative layer along with scolices and hooks. Adjacent to these there is cellular eosinophilic laminated membranous structures seen [Figure 5]. Renal hydatid cysts are usually multiloculated consist of single large cyst and smaller daughter cysts of varying sizes. Hydatid cyst involving Liver and Spleen are common sites and liver was the most common site affected in our study followed by spleen [Figure 6].

Our study revealed that of all patients of CE presenting to our hospital, this disease commonly affects patients in their second and fourth decade, is seen frequently in farmers and has a female preponderance. In our study, patients with CE presented commonly with pain in abdomen followed by lump in abdomen.

In our study population, CE was commonly seen at about 20 and 40 years. Papadimitriou *et al.*<sup>6</sup> reported 25.55% patients in the age group of 21-30 years and 24.67% patients in the age group of 31-40 years. Mehta *et al.*<sup>7</sup> reported 27.08% in the second and third decade and 18.75% in the fourth decade. Irshadullah *et al.*<sup>8</sup> also reported similar distribution with the highest percentage of patients in the age group of 21-30 years (25.83%) and 20% in the fourth decade. It has been consistently seen in various studies through the years and confirmed by our study that this disease is seen in young population who are in their active years.

The risk factors identified in our study were farming and female sex, which was identical to findings of previous studies. A study done in Xinjiang province of People's Republic of China, during 1951-1990, found that maximum number of cases was detected in farm laborers.<sup>9</sup> Farming in rural areas of India involves physical labor and working with hands in the soil and contact with livestock. Numerous stray dogs are found here, and hence contamination of soil with dog feces is a common occurrence. This may probably be the reason for transmission of disease by contact with egg-contaminated plants or soil followed by direct hand-to-mouth transfer. This finding supports the hypothesis of significant environmental contamination in open areas wherein parasite eggs survive and remain infective for months under favourable conditions such as high humidity and low temperature.<sup>10</sup>

Our study established that pain and lump in the abdomen were the common presenting features of CE. According to Balik *et al.*<sup>11</sup> and Jacob *et al.*,<sup>12</sup> pain in abdomen has been the most common presenting symptom (74.01% and 85%, respectively). This was followed by the presentation of lump in abdomen, which was seen in 54.93% cases by Balik *et al.*<sup>11</sup> and 57.5% cases by Jacob *et al.*<sup>12</sup> Contrary to this, Mehta *et al.*<sup>7</sup> concluded that lump in abdomen was the most common presentation in hydatid disease (54.16%). Balik *et al.*<sup>11</sup> also found that about one-fifth (22.69%) of

their patients had fever and 5.92% patients had jaundice. Jacob *et al.*<sup>12</sup> also had eight patients (20%) with jaundice and nine patients (22.5%) had fever.

The majority of the hydatid cysts in man are seen in the liver (approximately 75%) because it is the first filtering system for all the ingested ova, which enter the portal system.<sup>13</sup> A few of these ova that have an average size of 35  $\mu\text{m}$  escape the sinusoidal system of the liver to enter the systemic circulation and pass through the lungs, which acts as a second filter for the ova, making it the second most common site for the disease (approximately 15%).<sup>14</sup> Hydatid disease of the lung occurs from the larvae, which get trapped in the arterial capillaries of the lung. Occasionally, lung may be the site of secondary metastatic hydatidosis by rupture of a liver cyst.<sup>15</sup>

Even in the liver, it has been suggested that 85% of the cysts occurred in the right lobe and 15% in the left lobe.<sup>6</sup> Most of the authors<sup>7</sup> have reported maximal involvement of the liver, with the lung being the second most common site, a trend that has been duly reflected in our study.

The most commonly performed surgery in our series was partial pericystectomy with external drainage. There are certain distinct advantages with this technique, which have also been highlighted by Al-Hureibi<sup>16</sup> who has pointed out that the external drainage would not only manage any remnants that may have been inadvertently left behind during surgery, but the drain can also be used for flushing the cavity in cases of infection and may be used for contrast study to follow the course of obliteration of the cavity. No mortality was seen in our series. Consequently, the most common complication seen was wound infection. Pre- and postoperative chemotherapy with Albendazole was given to all our patients because of its proven effects in sterilizing the cyst, decreasing the chance of anaphylaxis, decreasing the tension in the cyst wall (thus reducing the risk of spillage during surgery), and in reducing the recurrence rate postoperatively.<sup>17,18.</sup>



## CONCLUSION

This study highlights the epidemiology of CE in the rural region of Indian subcontinent. The population in rural areas are more exposed to the zoonotic diseases due to their occupation, which is mostly farming. As with other zoonotic diseases, prevention plays an important in the overall management of this disease. Environmental contamination of the soil by dog feces seems an important way of transmission of disease in rural community. Careful washing of hands and food can help tremendously in decreasing this transmission. Proper education, creating awareness, and implementing strict rules regarding disposal of remains of slaughtered animals can also help eradicate this disease.<sup>[36]</sup> Hydatid cyst should also be kept in differential diagnosis before diagnosing any cystic swelling.

## REFERNCES

1. Richter J, Orhun A, Grüner B, Müller-Stöver I, Reuter S, Romig T, et al. Autochthonous cystic echinococcosis in patients who grew up in Germany. *Eurosurveillance* 2009;14:1-7.
2. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis: A zoonosis of Increasing Concern. *Clin Microbiol Rev* 2004;17:107-35.
3. Andronikou S, Welman CJ, Kader E. Classic and unusual appearances of hydatid disease in children. *Pediatr Radiol*. 2002;32:817–28.
4. Dharker SR. Hydatid disease. In: Ramamurthi B, Tandon PN, editors. *Text Book of Neurosurgery*. 2nd ed. New Delhi: Churchill Livingstone; 1996. pp. 535–44.
5. Gupta S, Desai K, Goel A. Intracranial hydatid cyst: A report of five cases and review of literature. *Neurol India*. 1999;47:214–7.
6. Papadimitriou J. The surgical treatment of hydatid disease of the liver. *Br J Surg* 1970;57:431-3.
7. Mehta RB, Ananthkrishnan N, Gupta BK, Srivastava KK, Mehdiratta KS, Satya P. Hydatid disease in Pondicherry. *Indian J Surg* 1982;44:88-94.
8. Irshadullah M, Nizami WA, Macpherson CN. Prevalence of human hydatidosis in Uttar Pradesh. *J Commun Dis* 1989;21:114-22.
9. World Health Organization Office International des Epizooties. WHO/OIE manual on echinococcosis in humans and animals: A public health problem of global concern. World Organization for Animal Health, Paris, France: 2001.
10. Veit P, Bilger B, Schad V, Schäfer J, Frank W, Lucius R. Influence of environmental factors on the infectivity of *Echinococcus multilocularis* eggs. *Parasitology* 1995;110:79-86.
11. Balik AA, Baçođlu 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.
12. 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
13. McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *Lancet* 2003;362:1295-304.
14. Saidi F. *Surgery of Hydatid Disease*. 1 st ed. Philadelphia: W B Saunders Company Ltd; 1976.
15. Morar R, Feldman C. Pulmonary echinococcosis. *Eur Respir J* 2003;21:1069-77.
16. Al-Hureibi AA, Amert A, Al-Hureibi MA, Sharawee Z. Hepatic hydatid cysts: Presentation and surgical management in

Yemen. J R Coll Surg Edinb 1992;37:229-31.

17. Goel MC, Agarwal MR, Misra A. Percutaneous drainage of renal hydatid cyst: Early results and follow-up. Br J Urol 1995;75:724-8.
18. Kune GA, Morris DI. Hydatid disease In: Schwasyz, Ellis, editors. Maingot's Abdominal Operations. 9 th ed. Appleton and Lange; 1989. p. 1225-40.