



The scenario of Hydatid cyst disease in epidemic areas of Andhra Pradesh – evaluation and analysis

Md Khader Faheem N*, N Nusrath, B Syama sundara rao, G Raja Ram, Sushma C, Y Subramanyam, K Ramesh

ABSTRACT

Background: Hydatid cyst is due to Echinococcus species a Cestode parasite. Commonly found species affecting humans are Echinococcus granulosus and Echinococcus multilocularis which cause Cystic Echinococcosis (Hydatid disease) and Alveolar Echinococcosis respectively. It is characterized by slow growing cyst in visceral organs. Dog is definite host and man is incidental host. It is commonly found in sheep and cattle rearing areas of the world like Eastern Europe, Australia, South America and South Asia.

Aim: To study the epidemiology and clinicopathological picture of Hydatid disease.

Objectives: To estimate the burden of Hydatid disease over the society.

Materials and methods: A review of 118 cases of Hydatid disease was carried out in various hospitals in central and southern epidemic zones of Andhra Pradesh, India. A broad descriptive prospective study was done for a period of 3 years i.e., from Jan 2009 - Dec 2011.

Results: During this three year study period (Jan 2009 to Dec 2011) 118 Hydatid disease cases were reported, from 3 different districts of Andhra Pradesh. Male predominance was observed 85(72.03%) and females accounted for 33(27.96%) cases. M:F – 2.5:1, majority of cases were distributed in 3rd decade, 20(25.42%). Mean age of presentation was 36.34(±12.29). About 33(27.96%) cases were farmers by occupation. Liver was found to be the most common site – 63(53.38%). Solitary cyst was seen in maximum number of cases – 86(72.88%). The mean size of cyst was 4.11(±1.38). The most common clinical presentation was asymptomatic – 26(22.03%).

Conclusion: Hydatid disease still is an emerging problem and is a course of challenge to all the medical practitioners. It is waging a war, with its roots spread deeply in society. Due to its non-specific clinical presentation and lack of awareness regarding the parasite in society it is being overlooked very commonly. Thus, it is necessary to formulate an accurate pathway of approach for the diagnosis, management and prevention of the disease.

Key words: *Echinococcus granulosus, Hydatidosis, small tapeworm of carnivores*

Introduction

Hydatid cyst is due to Echinococcus species – Cestode parasite commonly known as small tape worms of carnivorous animals. There are predominantly two species affecting the human population; Echinococcus granulosus and Echinococcus multilocularis causing Cystic Echinococcosis (Hydatid disease) and Alveolar Echinococcosis respectively. The other two species found very rarely in humans are Echinococcus vogeli and Echinococcus oligarthrus. Hydatid diseases are characterized by a slow growing cyst, commonly seen in visceral organs likely – Liver, Spleen, Lung, etc., It is a

common disease in sheep mostly in areas of Eastern Europe, Australia, South America and South Asia [1]. In India highest prevalence is reported from Andhra Pradesh and Tamilnadu [2,3]. This cyclozoonotic disease continues to be the most vexing socioeconomic problem in many parts of the world [4]. Although the disease is eradicated in most of the parts of the world, it still remains a serious endemic health problem in certain developing countries [5,6]. As Dog is definitive host, the research is still under progress over the aspects of the common occurrence of adult Echinococcus granulosus in the stray dogs and on the

Hydatid disease caused by its metacestode in man and his livestock. In spite of the clear epidemiological picture, the accurate prevalence is still not established due to the lack of awareness in the public regarding the rampant and fatal disease. The study was undertaken to assess the magnitude of the problem and to estimate the present prevalence and burden of disease with clinicopathological correlation in southern and central districts of Andhra Pradesh for a period of 3 years i.e., from 2009-2011, which is an epidemic zone for Hydatid disease.

Aims and Objectives

To estimate the burden of Hydatid disease over the society and assess - present prevalence, distribution and establish clinicopathological correlation - in epidemic areas of Andhra Pradesh

Materials and Methods

The records of all patients admitted to the various hospitals and health centers of southern and central parts of Andhra Pradesh with surgically and histopathologically proven Hydatid cysts over a period of three years from Jan 2009-Dec 2011 were carefully examined. The data of all the patients included in the study was extracted regarding the age, sex, place (address), occupation, habitat, site of involvement of cyst, number of cysts, size of cyst(s), clinical features, radiological evaluation, routine lab investigations and histopathological diagnosis. The data retrieved was considered to be the criteria of diagnosis. In all the cases observed, enucleation of cysts or evisceration of the organ was done; cyst fluid was examined microscopically under wet-mount preparation. The cysts were fixed in 10% formalin and subjected to histopathological examination. Thus, a final criterion of diagnosis was established. Inclusion of the subjects under the study was done only after histopathological evaluation of the cyst. The data obtained was compared with other national and international research studies to derive the statistical equations.

Results:

A total of 118 patients with Hydatid disease were identified. Among the above – 85 (72.03%) were male and 33 (27.96%) were female. The M:F ratio was 2.5:1. The majority of patients were in 3rd decade - 20 (25.42%). More number of males were seen to be afflicted with the disease in 5th decade – 20 (16.94%) and comparatively more number of females were of 3rd decade – 12 (10.16%). The distribution of Hydatid disease in occupational groups

in accordance with sex is depicted in Figure: 1. The Mean age of presentation was 36.34 (± 12.29); Median – 35.5; Mode – 28; Range of observation (12 to 68)56; Variance Standard Deviation – 166.22; Population Standard Deviation – 12.83; Variance Population Standard Deviation – 164.81. Among the three epidemic areas included in the study Nandyala (Kurnool District) shared maximum number of cases – 74 (62.71%) followed by Kadapa 22 (18.64%) and Chittoor – 22 (18.64%). All the three areas show male predominance in distribution.

The study included a population from various socio-economic strata of society basing on their occupation. About 33 (27.96%) of patients were farmers; the observations of occupation based data tabulated in Table: 1. All classes show major distribution in male sex from Nandyala (Kurnool district) in different age groups (house wives are an exception)

Majority of cysts were found in liver – 63 (53.38%) (Figure: 2); followed by spleen – 30 (25.42%) (Figure: 3); lung – 13 (11.08%); liver + spleen – 5 (4.23%); omentum – 3 (2.54%); kidneys – 2 (1.69%); diffuse abdominal – 1 (0.84%) (Figure: 4) and spine – 1 (0.84%) (Figure: 5). The graphical representation depicted in Figure: 6. The age group varied for different anatomical location of cysts, tabulated in Table: 2.

Maximum number of cases presented with a single cyst – 86 (72.88%) and the majority were seen in the liver. Two cysts were seen in 17 (14.40%); 3 cysts – in 3 (2.54%); 4 cysts – in 3 (2.54%); 5 cysts – in 3 (2.54%) and >5 cysts – in 6 (5.08%) cases. Highest numbers of cysts reported were 12 cysts in a 47 year old male from Kadapa district; who was an employee (clerk). The largest cyst in this case measured 7.2 cms. There was a significant variation in size of the cysts. Majority of cysts were <3cms – 81 (68.64%). The Mean size was 4.11 (± 1.38); Median - 4; Mode – 3.5; the Range of distribution was (2 to 10) 8; Variance Standard Deviation – 1.91; Population Standard Deviation – 1.37; Variance Population Standard Deviation – 1.89. [Z score = -0.4420 (Standardized Random Variable X)]. The two-tailed P value equals 0.6585. By conventional criteria, this difference is considered to be not statistically significant.

The clinical data of all the patients were extracted from the records. There was a varied clinical presentation of cases depending upon the site and visceral organ involved. The most common presentation of disease was incidental diagnosis, and patients were asymptomatic – 26 (22.03%).

Most of the patients with Hepatic Hydatid disease presented with pain abdomen (right upper quadrant) – 19(16.10%). Hepatomegaly was observed in 17 (14.40%) patients.

TABLE: 1. DISTRIBUTION OF HYDATID DISEASE CASES BASED ON CCUPATION AND SEX

Age in years	FARMERS		LABOUR		HOUSEWIVES		STUDENTS		EMPLOYEES		RETIRED		UNEMPLOYED	
	M	F	M	F	M	F	M	F	M	F	M	F	M	F
11-20	0	0	1	1	0	0	7	5	0	0	0	0	0	0
21-30	5	1	9	1	0	6	2	0	2	4	0	0	0	0
31-40	10	0	7	2	0	9	0	0	1	0	0	0	0	0
41-50	11	0	7	1	0	4	0	0	2	0	0	0	0	0
51-60	6	0	3	0	0	0	0	0	0	0	0	0	5	0
>60	0	0	0	0	0	0	0	0	0	0	3	0	4	0
TOTAL	32	1	27	4	0	19	9	5	5	4	3	0	9	0
GRAND TOTAL (%)	33(27.96%)		31(26.27%)		19(16.10%)		14(11.86%)		9(7.62%)		3(2.54%)		9(7.62%)	

M- Male; F- Female

TABLE: 2. DISTRIBUTION OF HYDATID DISEASE CASES BASED ON ANATOMICAL SITE AND SEX

Age in years	LIVER		SPLEEN		LUNGS		LIVER + SPLEEN		OMENTUM		KIDNEY		DIFFUSE ABDOMINAL		SPINE		TOTAL(%)		GRAND TOTAL(%)	
	M	F	M	F	M	F	M	F	M	F	M	F	M	F	M	F	M	F		
11-20	4	2	2	1	2	1	0	0	0	0	0	1	0	0	0	0	0	8(6.77)	5(4.23)	13(11.01)
21-30	8	7	4	3	3	2	1	0	1	0	1	0	0	0	0	0	0	18(15.25)	12(10.16)	30(25.42)
31-40	9	8	3	2	4	1	0	1	1	0	0	0	0	0	0	0	0	18(15.25)	11(9.32)	29(24.57)
41-50	11	2	6	2	0	0	2	0	1	0	0	0	1	0	0	0	0	20(16.94)	5(4.23)	25(21.86)
51-60	7	0	6	0	0	0	0	0	0	0	0	0	0	0	1	0	0	14(11.86)	0	14(11.86)
>60	5	0	1	0	0	0	1	0	0	0	0	0	0	0	0	0	0	7(5.93)	0	7(5.93)
TOTAL	44	19	22	8	9	4	4	1	3	0	1	1	1	0	1	0	0	85(72.03)	33(27.96)	118(100)
GRAND TOTAL (%)	63(53.38)		30(25.42)		13(11.08)		5(4.23)		3(2.54)		2(1.69)		1(0.84)		1(0.84)					

M- Male; F- Female

Fig: 1. OCCUPATION AND SEX BASED DISTRIBUTION OF HYDATID CYSTS

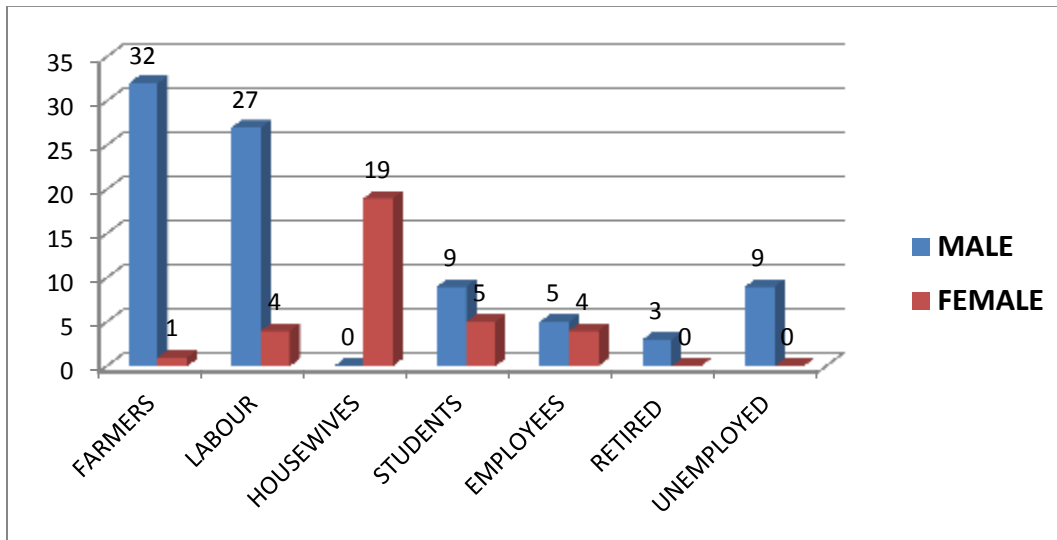


Figure 2. Tender coconut peel appearance of Hydatid cyst wall enucleated from liver



Figure 3. Multiple Hydatid cysts in Spleen



Figure 4. Enucleated Multiple Hydatid cysts from Omentum



Figure 5. MRI showing Hydatid cyst at T8 region of Spine

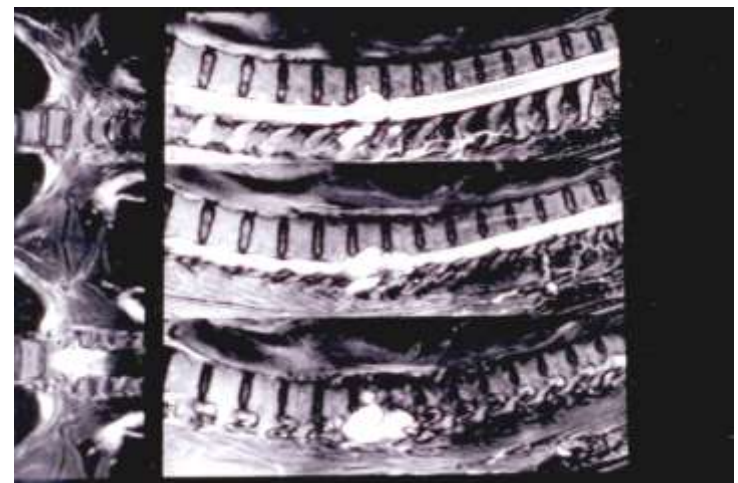
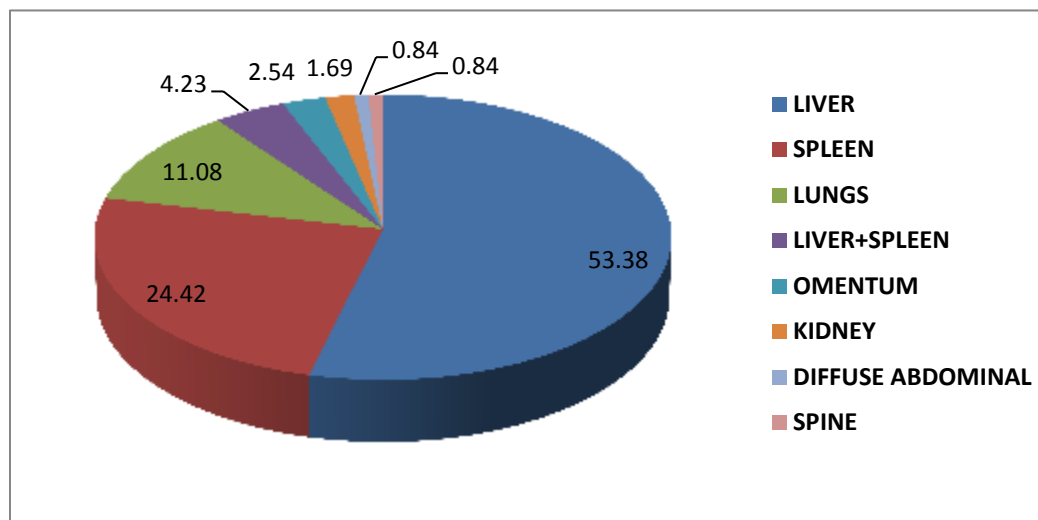
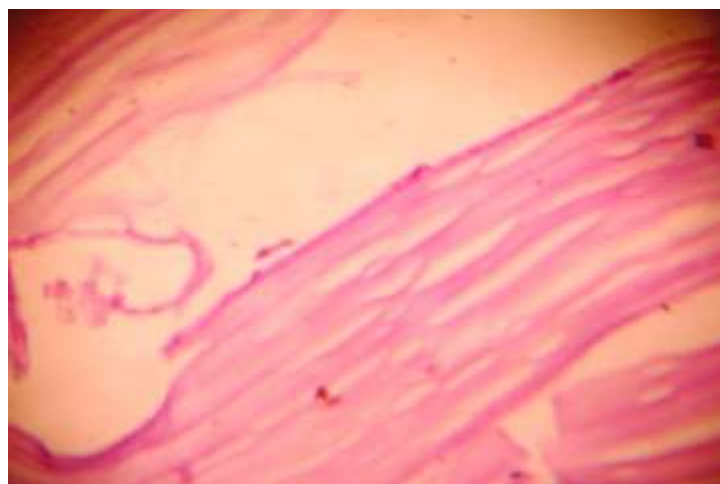


Fig: 6. ANATOMICAL DISTRIBUTION OF HYDATID CYSTS**Figure: 7. Microphotograph - Lactophenol blue wet mount preparation of cyst fluid showing hooklets and daughter cysts (100x)****Figure: 8. Microphotograph – Laminated membrane of Hydatid cyst wall (100x)**

Majority of Splenic Hydatid disease cases showed asymptomatic presentation – 12 (10.16%); Splenomegaly was seen in 15 (12.71%) of patients. Pulmonary Hydatid disease presented with chest pain, cough, fever and hemoptysis. But, the majority of cases were asymptomatic – 5 (4.23%). Cases showing combined Liver and Splenic Hydatid disease showed hepatosplenomegaly, with diffuse pain abdomen, fever, jaundice and abdominal distension. Omental Hydatid disease showed fever with pain abdomen. Diffuse abdominal Hydatid disease showed the distribution of cysts in liver, spleen, omentum etc., Clinically high grade fever with vomitings and diarrhea was presenting complaint. The Renal Hydatid disease was absolutely asymptomatic.

Hydatid cyst of spine presented with paraplegia. The (Magnetic Resonance Image) MRI imaging showed cystic lesion at T8 with cord compression. (Figure: 5).

The cysts were enucleated and some viscera were eviscerated surgically. The cyst fluid was aspirated and observed under LCP (Lactophenol cotton blue preparation) wet mount preparation (Figure: 7) and then were subjected to histopathological examination. Cytological evaluation showed hooklets of the parasite and histological examination showed inner nucleated germinal layer, an outer anucleated chitinous layer and the contents of the cysts showed brood capsules and scolices – together known as Hydatid Sand. (Figure: 8).

Discussion

There still exists a dilemma of obtaining accurate figures on the prevalence of Hydatid disease, as in the majority of cases the disease manifests with a very few specific signs and symptoms [4]. A considerable number of cases present to clinician in an asymptomatic state and the diagnosis will be made incidentally or accidentally. As such, Hydatid disease is an endemic in India. The annual incidence of Hydatid disease per 1,00,000 persons varies from 1 to 200 [7]. A descriptive study conducted over a period of 3 years i.e, from 2009-2011 at various hospitals and health care centers in 3 districts of Andhra Pradesh [Nandyala (Kurnool dist); Kadapa; Chittoor] revealed 118 cases of Hydatid disease in a strange and peculiar pattern of distribution in accordance with age, sex, occupation, with a varied and unstable clinical picture and anatomical distribution.

In the present study, the maximum number of cases was seen in 3rd decade – 30 (25.42%) and affliction is seen from 11 years to 68 years. This finding was observed in all the other similar studies done by various research workers [8-15]. There were some contrast results in other studies, which stated that, it may be anywhere between 2nd to 6th decades [16-18]. This might be attributed to the chronicity and non-specific (asymptomatic) presentation of the disease in majority of cases.

In relation to the sex, the majority of cases were males – 85 (72.03%), M:F ratio was 2.5:1, which was a similar observation in the majority of studies performed in relation to Hydatid disease [12,14,17,19-21]. Some studies showed a minimal confliction regarding sex distribution, where female preponderance was observed [4,10,15,16,22-26]. The distribution of sex was compared with various national and international studies from different parts of the world. Thus, a varied and diverse picture was obtained. The differences in the reports were due to difference in socioeconomic, traditional, cultural variations in different regions in India as well as in other parts of the world. In southern part of India considerable proportion of men are actively involved in livelihood activities of farming, routine labour and animal breeding and agriculture; compared to women, thus are more prone and exposed to infections and diseases. Various animal experiments were performed to relate the distribution of sex [27]. An interesting finding was male Mice were more susceptible to contract the Hydatid disease than the female species. The basis propounded regarding this result was that, the female goadotrophins (estrogens) have an inhibitory action on level of

parasitization, while male hormone (testosterone) had a little of any such effect or else, might even increase the susceptibility of the host infection [4].

The present study had focused on burden over the society due to the illness of Hydatid disease. The livelihood and occupation of the individuals were taken into consideration. Maximum number of cases were notified in farmers – 33 (27.96%) with majority being men 32 (27.11%). Among females the maximum numbers of cases were housewives – 19 (16.10%). In a similar researches done by Al Barwari et al., [4] and Jawed Akther et al., [15] maximum cases were females, housewives accounting around 37.90% and 39.32% respectively. The majority of males according to Al Barwari et al., [4] were students (23.90%). Present study correlated with the above one regarding female population, whereas male students accounted for about 7.62% only. Farmers and housewives are more prone to the disease as, they are more involved in household activities related to animal breeding and agriculture in South Indian rural areas.

A diverse organ involvement is a common feature of any representative sample of Hydatid patients. The analysis of the present sample of patients revealed that liver being the most common site followed by the spleen. The comparison of the distribution of Hydatid cysts in various organs was done and study correlated with the majority of other similar research works. Still there were some minor differences with some other research workers which are tabulated in Table: 3.

The higher rate of Hepatic infection may be attributed to the fact that liver acts as a primary filter in the human body and lung is often thought to be the second filter [4]. There was a predominance of single organ involvement (93.37%) over the multiple organ involvement (7.63%), which was a similar finding in most of the research works done on Hydatid cysts [4,6,9,15,26]. It is widely accepted that primary cysts are mostly solitary in nature.

The sizes of the cysts were quite variable 99 (83.89%) patients showed cysts <5cms; 19 (16.10%) patients show cysts >5cms size. In contrast to study done by Alghoury et al., [26] where 94% showed cysts >5cms and only 6% showed cysts <5cms.

The majority of cases showed a single cyst – 86 (72.88%); Multiple cysts were seen in 32 (27.11%) cases. An equivalent observation was notified in a study done by Alghoury et al., [26] where 61% of single cysts and 39% of cases showed multiple cysts.

TABLE: 3. COMPARITIVE ANALYSIS OF ANATOMICAL DISTRIBUTION OF HYDATID CYSTS

(n)	LIVER (%)			SPLEEN (%)			LUNG (%)			LIVER + SPLEEN (%)	KIDNEYS (%)			OMENTUM (%)	DIFFUSE (%)	OTHERS (%)		
	TOT	RL	LL	BL	TOT	R.Lu	L.Lu	B.Lu	TOT		RK	LK	BK					
Present study (2012)	118	53.38	58.73	38.09	3.17	25.42	11.08	53.84	30.76	15.38	4.23	1.69	50	-	50	2.54	Abd – 0.84	Spinal – 0.84
Beckett et al., (1945) [28]	-	75	-	-	-	2	15	-	-	-	-	-	-	-	-	2	-	Muscle; Brain; Breast; Heart; Orbit - 5
Al Barwari et al., (1991) [4]	153	41.20	-	-	-	4.60	24.2	56.75	24.32	8.10	2	7.18	45.45	36.36	18.18	3.90	-	Pancreas; Gall Bladder; Breast; Thyroid – 16.3
Alghoury et al., (2010) [26]	66	57.58	65.78	15.79	18.43	-	25.75	58.82	41.18	-	-	3.03	-	-	-	-	-	Ovarian; Uterine; Brain; Iliac fossa – 9.09
Rukmangadha et al., (2010) [14]	34	35.5	-	-	-	8.8	26.5	-	-	-	-	2.9	-	-	-	-	-	Subcutaneous tissue; Breast; Retrouterine; Synovial; Brain – 26.3
Jawed Akther et al., (2011) [15]	117	69.32	62.5	26.13	11.36	3.38	14.53	-	-	-	-	1.71	-	-	-	1.71	Abd – 0.85	Muscle – 3.38

TOT – Total; RL – Right Lobe; LL – Left Lobe; BL – Both Lobes; R.Lu – Right Lung; L.Lu – Left Lung; B.Lu – Both Lungs; RK – Right Kidney; LK – Left Kidney; BK – Both Kidneys; Abd – Abdominal

Clinical picture gives important information in parasitic diseases, in deriving the site of infestation. Thus, in the present study an attempt was made to study the clinical scenario of Hydatid disease in various organs of the body. Overall – commonest presentation of Hydatid disease was as such, asymptomatic – 22.03% of cases. Pain abdomen was the primary complaint in Hepatic and Splenic Hydatid disease. Pulmonary Hydatidosis was asymptomatic in the majority of patients. A Hydatid cyst of spine was observed which presented with paraplegia. The present study correlated with study done by Rukmangadha et al., [14] to some extent regarding clinical presentation of Hydatid disease in common sites. Some other general signs were

examined in accordance with the site of involvement and general condition of the patient. Hepatomegaly was observed in 16.94% of cases; Splenomegaly in 15.25% and Anemia with pallor in 28.81%. A similar observation was done by Jawed Akther et al., [15]

Conclusion

Hydatid disease is still an important health problem in India, with its expansion in various parts of South India. It is still raging a war over the communities from low socioeconomic status who are unaware about this demonic parasite. Thus, there is a quite an impact of morbidity due to Hydatid

disease over the society due to its higher infliction rate in farmers and housewives. Thus, the present study attempts to throw some light regarding the emphasis needed to improve personal hygiene and promotion of awareness regarding Hydatid disease in the community. In this study we have also stated that, the disease is not an exception to any organ of the body. We have come across some rare sites, including- Spleen, Omentum; Diffuse abdominal and Spinal Hydatidosis. The care of self-hygiene and creation of awareness are two small things which can generate a great difference in controlling this dreadful disease.

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AUTHOR(S):

1. Dr.Md Khader Faheem N., Assistant Professor, Dept. of Pathology, S.V.Medical College, Tirupati
2. Dr.N Nusrath, Ex-Senior Resident, Dept. of General Medicine, S.V.Medical College, Tirupati
3. Dr.B Syama sundara rao, Assistant Professor, Dept. of Pathology, Shantiram Medical College, Nandyala
4. Dr.G Raja Ram, Assistant Profesor, Dept. of Microiology, S.V.Medical College, Tirupati
5. Dr.Sushma C, Ex-Senior Resident, Dept. of Pathology, NRI Medical College, Guntur
6. Dr.Y Subramanyam , Professor & Head (i/c), Dept. of Pathology, RIMS, Kadapa
7. Dr.K Ramesh, Consultant Surgeon & Director, KR Hospitals, (Ex- Assistant Professor, Dept. of General Surgery, S.V.Medical College) Tirupati

CORRESPONDING AUTHOR:

*Dr.Md Khader Faheem N,
Assistant Professor,
Dept. of Pathology,
S.V.Medical College,
Tirupati, Andhra Pradesh,
India
Email: faheemkhader@gmail.com
Ph: +919989610786*

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