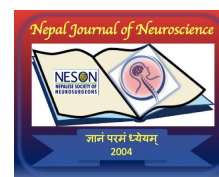


Retrospective Analysis of Hydatid Cyst Patients and Incidence of CNS Involvement in Southern Rajasthan

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Abstract

Introduction: Echinococcosis also known as hydatid disease is an endemic zoonotic disease with an estimate of 2,00,000 new cases per year worldwide. Its primary definitive host is dog and man being an accidental intermediate host. The most common organ affected is liver, brain, and spine. Surgery is the mainstay of treatment and medical management is reserved for selected cases. Our objective was to retrospectively evaluate the prevalence of craniospinal hydatid cyst disease in southern Rajasthan.

Materials and method: A retrospective analysis of all cases of craniospinal hydatid disease managed at our department was done from 2013 to 2021 and data was analysed.

Results: A total number of eight cases were reported with an incidence of 1.8% of all craniospinal space-occupying lesions during the study period with a male to female ratio of 3:1, mean age of presentation 18.5 years, four out of eight patients were in paediatrics age group. All cases were managed surgically with preoperative rupture in one case and recurrence in two cases. Albendazole was given to cases only with rupture or recurrence.

Conclusions: Craniospinal hydatid disease should be suspected in all non-enhancing cystic brain lesions especially in endemic regions. In all patients, surgical excision should be done preferably with medical management reserved for inaccessible lesions, unfit patients, rupture and recurrent cases only. The study concludes that primary surgery with gross total excision with no or minimal spillage of cyst content intraoperatively has good results with recurrence rate of only 12.5%.

Keywords: Craniospinal hydatid, Echinococcosis, Albendazole, Craniotomy

Introduction

Human infection with the larval form of two species of Echinococcus parasite, i.e., *E. granulosus* and *E. multilocularis* is known as 'Hydatidosis'. Hydatid - Greek word for 'watery cyst' disease is a rare but significant parasitic disease in endemic areas. It is most frequently seen in the Middle East, South America, New Zealand, and Mediterranean countries¹. In India it is known to be endemic in Andhra Pradesh and Tamil Nadu, provinces in southern India⁵ and high cases have been

reported in south eastern parts of Rajasthan in last few years⁷. The lack of accurate case reporting and genotyping currently prevents any precise mapping of the true epidemiologic picture.

Cystic echinococcosis (CE) disease results from being infected with the larval stage of Echinococcus granulosus, a tiny tapeworm (two to seven millimetres in length) found in dogs (definitive host), sheep, cattle, goats, foxes, and pigs, amongst other intermediate hosts. The adult Echinococcus multilocularis (1.2–4.5 mm long) resides in small intestine of definitive host. Humans are aberrant intermediate host and become infected by ingesting eggs. Oncospheres are released in the intestine and cysts develop within the liver. Metastasis or dissemination to other organs (e.g., lungs, brain, heart, bone) may occur if protoscolices are released from cysts, sometimes called "secondary echinococcosis." 60% to 70% of hydatid cysts are located in the liver and 10% to 15% are located in the lungs. Brain involvement occurs in 1-2% of all Echinococcus granulosus infection. Cerebral hydatid cysts are usually supratentorial, whereas infratentorial lesions are quite rare. Cerebral hydatid disease is more common in the paediatric population and young adults with approximate 50-70% incidence rate. Spinal hydatid cysts account for 1% of all cases of hydatid disease^{19,14,11}.

An osseous location is seen 0.5% to 2% and approximately half of them are located in vertebrae. While 50% of the vertebral involvement is seen in the thoracic area and 20% is in the lumbar area. Sacral and pelvic involvement is rare^{6,10,2}. The disease usually spread to spine by direct extension from a pulmonary,

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abdominal, or pelvic infestation and most commonly affect the dorsal region of spine ^{2,12,16}. Usually, the cerebral cyst is solitary and often seen in the parietal lobe ⁴ while spinal cysts are mostly multiple and epidural in location. In any endemic area of HD, disease should be considered in any patient presenting with cysts or cystic masses in any organ of the body with unusual site or presentation. The aim of this study was to report cases of hydatid cyst that has been confirmed by histopathology in the past eight years (2013-2021) concerning the rare occurrence of cerebral, spinal hydatid cysts with their clinicopathologic findings.

Materials and Methods

A retrospective study was done and data was collected from Government College Kota. From 2013 to 2021, a total of 62,892 histopathological examinations were performed, out of which 498 were total histopathological confirmed cases of hydatid cyst and a total of eight CNS cases were found, seven from the brain and one in the spinal cord. The total incidence was 1.96%.

Table 1: Comparison of number of histopathological examination (HPE)

| Total no of HPE | No. of hydatid cyst | No. of Hydatid cases in CNS |
|-----------------|---------------------|-----------------------------|
| 62,892 | 498 | 8 |

Table 4: Case details of patients with craniospinal hydatid cyst.

| AGE & SEX | PRESENTATION & DURATION OF SYMPTOMS | LOCATION AND NUMBER | MANAGEMENT | RECURRENT | INTRAOPERATIVE RUPTURE | ALBENDAZOLE | RISK FACTORS | FOLLOW UP |
|------------|--|--|------------|-----------|------------------------|------------------------|------------------------|--|
| 12 year/ F | Headache, seizures, right hemiparesis, duration six months | Left temporo-parietal, single | Surgery | No | No | No | H/o contact to pets | Lost in follow up |
| 14 year/ M | Headache, vomiting, left hemiparesis, three months | Right temporo-parietal, single | Surgery | No | No | No | No | Four years |
| 5 year/ M | Headache, vomiting, left hemiparesis, three months | Right temporo-parietal, single | Surgery | No | No | No | H/o contacts with pets | Lost in follow up and presents with recurrence after three years |
| 30 year/ M | Headache, vertigo, tinnitus two months | Right CP angle, single | Surgery | No | No | Yes | No | Two months |
| 8 year/ M | Headache, vertigo, tinnitus two months | Right CP angle, single | Surgery | Yes | No | Yes (After recurrence) | No | Eight years |
| 60 year/ M | Headache, right hemiparesis five months | Left parietal, single | Surgery | No | No | No | No | One year |
| 23 year/ F | Lower backache, paraparesis, urinary incontinence 3 months | Right paraspinal and intraspinal, multiple | Surgery | No | No | Yes | No | Two month |
| 33 year/ M | Headache, vomiting, reduced vision in right eye 3 months | Right mastoid region, single | Surgery | NO | No | No | No | Lost in follow up and presents with recurrence after 3 years |

Observation and Results:

A total number of cases were studied in terms of different demographic properties. When we compared the number of cases in males and females in different age groups, following inference was drawn.

Table 2: For total number of cases hydatid cyst.

| AGE GROUP | MALE | FEMALE |
|-------------|------|--------|
| < 20 years | 74 | 27 |
| 21-40 years | 114 | 72 |
| 41-60 years | 50 | 38 |
| >60 years | 25 | 8 |
| TOTAL CASES | 263 | 145 |

It was observed that most of the patients were young adults (45.58%) in the age group of 21-40 years and Male: Female ratio was found to be 1.8:1.

Table 3: For Craniospinal cases.

| AGE GROUPS | MALE | FEMALE |
|-------------|------|--------|
| < 20 years | 3 | 1 |
| 21-40 years | 2 | 1 |
| 41-60 years | 1 | 0 |
| >60 years | 0 | 0 |
| TOTAL CASES | 6 | 2 |

Whereas the patients with CNS disease was found to be more commonly affecting young males (50% of cases) and with Male: Female ratio of 3:1.

The sign and symptoms of the disease varied according to site of presentation. Following results were found when we observed the frequency of symptoms.

Table 5: frequency of sign and symptoms of brain and spinal cases.

| Sign & Symptoms of brain | Percentage | Signs & Symptoms of spine | Percentage |
|--------------------------|------------|---------------------------|------------|
| Headache | 100% | Urinary in-continence | 100% |
| Vomiting & hemiparesis | 57.1% | Backache | 100% |
| Seizures | 28.6% | | |
| Vertigo & tinnitus | 14.3% | | |
| Reduced visual acuity | 14.3% | | |

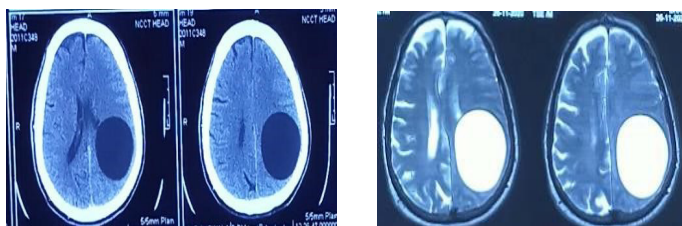


Figure 1[a]: Preoperative scans showing the location of intracranial hydatid cyst

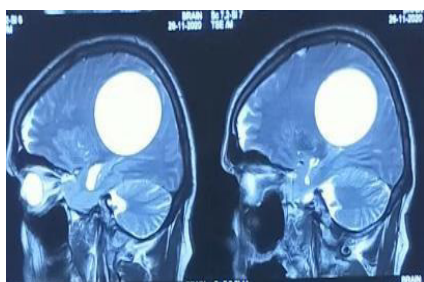


Figure 1[b]: Preoperative scans showing the location of intracranial hydatid cyst

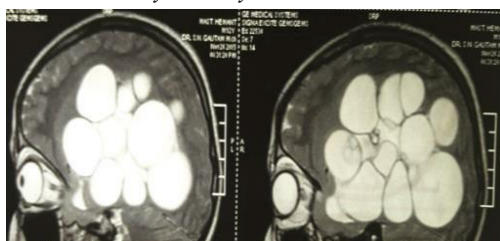


Figure 1[c]: Preoperative scans showing multiple intracranial hydatid cysts

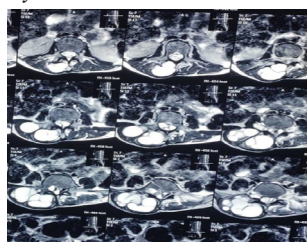


Figure 1[d]: Preoperative scans showing multiple intraspinal hydatid cysts

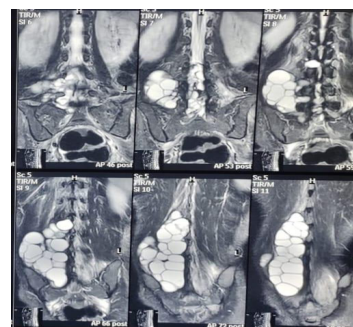


Figure 1[e]: Preoperative scans showing multiple paraspinal hydatid cysts

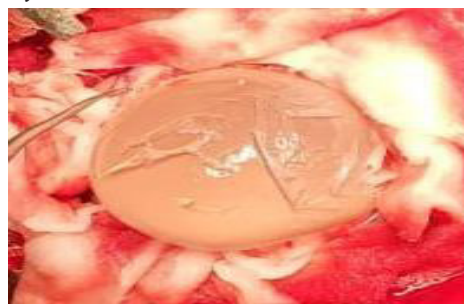


Figure 2: Delivery of intracranial hydatid cyst using Dowling's technique

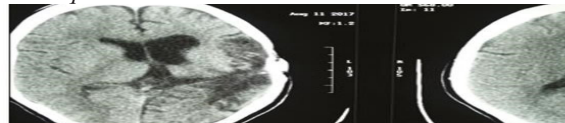


Figure 3: Post operated scan showing complete excision of cyst

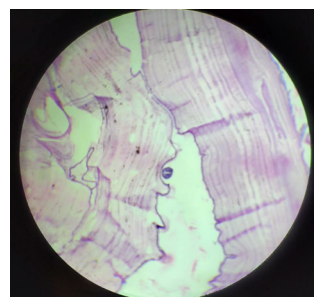


Figure 4[a]: Histopathological microphotograph [40X] showing laminated ectocyst, scolex and inflammatory infiltrate



Figure 4[b] Histopathological microphotograph [40X] showing laminated ectocyst, scolex and inflammatory infiltrate

Discussion and review of literature:

In Indian set up we have found three case series^{9,21,22} with number of different cases that have been compared below.

Table 6: Comparison of different studies.

| | Tanki et.al | Gupta et. al | S.N Gautam et. al | S. Shukla et.al | Current series |
|-------------------------------|--|---|--|--|--|
| Number of cases | 9 | 5 | 6 | 3 | 8 |
| Mean age of presentation | 11.5 years | 13.4 years | 21.2 years | 9 years | 18.5 years |
| Paediatric patients | 9/9 [100%] | 4/5 (80%) | 4/6 (66.66%) | 2/3 (66.66%) | 4/8 (50%) |
| Duration of symptoms | One month to two years | One month to two years | One month to six months | Fifteen days to four months | Two to six months |
| Male: female | 5:4 | 3:2 | 5:1 | 2:1 | 3:1 |
| RISK FACTORS | 7/9 | 2/5 | 2/6 | NA | 2/8 |
| 1.Contacts to pets | 8/9 | 3/5 | 6/6 | NA | 8/8 |
| Rural location | | | | | |
| Symptoms | Seizures (m/c), hemiparesis, Vomiting, headache | H e m i p a r e s i s [Most common], Seizures | H e a d a c h e (Most common), Hemi-paresis, Vomiting, Seizures | Headache, vomiting, seizures, decreased vision, backache, urine retention | Headache, vomiting, seizures, decreased vision, backache, urinary incontinence |
| Location of cysts | Four parietal solitary cysts, Two frontal solitary, One parietooccipital solitary, | One patient each with frontal, lateral ventricle, | Two patients each with temporoparietal solitary cyst, | One temporoparietooccipital | Two patients each with Temporo-Parietal solitary cyst |
| | Two parietal multiple cysts | parietal. Two patients with multiple cysts in parietal and temporoparietal region | one with lateral ventricle, one parietal, one mastoid, one with multiple temporoparietal cysts | solitary cyst, one with C6, C7 spinal canal multiple, one cauda equine below conus solitary cyst | one with lateral ventricle, one parietal, one mastoid, one with multiple temporoparietal cysts, one with paraspinal & intraspinal multiple |
| Intraoperative rupture | Three cases | Two cases | One case | Nil | One case |
| Anaphylaxis following rupture | Nil | nil | Nil | Nil | Nil |
| Recurrence | Two cases, one year of surgery | Two cases; six months to one year after primary surgery | One case, six months following surgery | NA | Two cases, three years following surgery, one with six months of surgery |
| Albendazole | Everyone, 10mg/kg for two months | To two patients following recurrence, 10 mg/kg for one month | Two patients, one with rupture another with recurrence, 10mg/kg up to four cycles | Everyone, 10mg/kg for three months | Three patients, 10mg/kg up to four cycles |
| Follow up period | Six months to five years | Six months to eight years | Six months to seven years | NA | one month to eight years |

Hydatidosis of the spine was first described by Churrier in 1807¹⁸ Guesnar reported first case of cerebral hydatid⁵. Intracranial hydatid forms about 1-2% of all intracranial space-occupying lesions¹⁴.

Spinal involvement in hydatid disease is the result of the portovertebral shunt and the centre of the vertebral body is first¹⁵ site to be involved. Multiple vertebral involvement is unusual, owing to the relative resistance for invasion of the disc space. It usually starts in the vertebral body and grows slowly because of the resistant nature of bone. When the bone is breached by cysts and extradural space is violated, the neurologic deficit with unremitting pain ensues. A study has also concluded that a possible mechanism of spreading the cysts to lumbosacral spinal intradural space is through cerebrospinal fluid flow, after entering the thoracic spinal level from the primary pulmonary hydatid disease via transforaminal due to local invasion⁹.

On CT head, there is a hypodense non-contrast enhancing oval homogenous cystic mass lesion with thin walls and smooth margins with pressure effect on surrounding brain parenchyma as per the size of the lesion with no surrounding oedema. The MRI with its superior soft-tissue resolution is the most sensitive diagnostic method as well as the method of choice for the determination of the extent of the disease. A study by Berk et al stated that MRI of the lesions has a unique appearance, a sausage-like appearance with two dome-shaped ends, thin and regular walls and with no septation or debris in the lumen. The lesions are occasionally spherical³. In T1W images of the cyst, wall appears iso- or slightly more hypo-intense than the cyst content and enhance slightly after contrast injection. T2W images demonstrate a low-intensity rim which correlates to the pathological findings of reactive fibrosis and degeneration surrounding the parasitic membrane. The spinal hydatid can be classified according to Dew/Braithwaite and Lees classification.

Table 7: Dew/Braithwaite and Lees classification.

| | |
|--------|----------------------------|
| Type 1 | Intramedullary |
| Type 2 | Intradural, extramedullary |
| Type 3 | Extradural, intraspinal |
| Type 4 | Vertebral |
| Type 5 | Paravertebral |

In our spinal case, a 23-year female who had multiple extradural intraspinal as well as multiple paravertebral cysts, presented to us with symptoms of lower backache, paraparesis, urinary incontinence for three months. It is a mixed presentation of both type three and type five of this classification. Surgical removal is the first and most effective option for treating spinal canal hydatid cysts. The preferred technique includes the Dowling technique later modified by Arana Iniguez and San Julian. But radical excision is almost impossible in hydatid disease of the spine because of absence of distinct anatomic planes and the existence of neural structures, hence has high local reoccurrence rate. A course of Albendazole (800 mg daily in two divided doses) is continued for one to six months (usually three months). Albendazole is preferred over Mebendazole. Lam et al reported an inoperable case of recurrent spinal hydatidosis treated with a combination of Albendazole plus Praziquantel survived for 34 years¹³.

Conclusions:

With this study, we concluded that total incidence of CNS hydatid disease in south-eastern Rajasthan is found 1.8% that is more than the previously calculated incidence of 1%^{20,11,17}. This study had the advantage of long term follow up that has been conducted so far for the CNS cases. The study also concluded that a good primary surgery with gross total excision of cyst with no or minimal spillage of cyst contents intraoperatively has good results with recurrence in only 12.5% of cases.

Consent: Proper consent has been taken from legal guardians of patients to use their data for academic publication with assurance of non-disclosure of identity to the best of the extent.

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