Giant intracranial hydatid cyst: A report of two cases and literature review

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A B S T R A C T

Hydatid disease is a zoonosis caused by *Taenia echinococcus*. The three main varieties *Echinococcus granulosus, E. multilocularis* and *E. vogeli* are primarily found in dogs and are transmitted to man by fecal–oral route. Commonly affected organs are liver, lungs and spleen. Brain is involved only in 2-5% cases. The authors herein present two cases of giant intracranial hydatid cysts managed at department of neurosurgery, Rajendra Institute of Medical Sciences, Ranchi, India.

Key words: Echinococcus, hydatid cyst, taenia, zoonosis

INTRODUCTION

Human echinococcosis, also known as hydatid disease, results from infestation with the tapeworm *Echinococcus* granulosus which lives in the intestinal tract of dogs. This zoonotic disease occurs worldwide and is most common in agricultural regions. The definitive hosts are various carnivores, commonly dogs. Mammals (sheep and cattle) are intermediate hosts.

Man is infected by the fecal-oral route with the ingestion of egg which after digestion releases the hexacanth embryo. This embryo penetrates the intestinal mucosa and reaches the portal circulation and then to the liver where a hydatid cyst develops. The CNS is affected only when the embryo passes through the capillary filters of liver and lungs to enter the systemic circulation. Liver is affected in 65% cases, lungs in 15-20% and brain in 2-5% cases.^[1] In India, hydatid disease is seen more commonly in Kurnool district of Andhra Pradesh, Madurai district of Tamil Nadu and in Punjab.^[1,2] Cerebral hydatid disease is a rare entity in the state of Jharkhand and few cases have been observed.

We present here two cases of giant cerebral hydatid cyst. The relevant literature has been reviewed.

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	www.ijns.in DOI: 10.4103/2277-9167.94378

CASE REPORTS

Case 1

A 10-year-old boy presented to us with complaints of headache for 4 months, aphasia for 1 month, right hemiparesis for 15 days and recurrent vomiting for 10 days. There was no history of convulsions, fever or trauma.

On examination, the patient's GCS was E2V1M5. Right hemiparesis was apparent along with hypertonia, plantar up going and exaggerated deep tendon reflexes on the same side. Bilateral papilloedema was observed.

MRI of brain was done which showed a symmetrically rounded, well-defined cyst in the left temporoparietal region [Figures 1a-b]. The cyst was hypointense on T1W, hyperintense on T2W and showed little perilesional edema. There was no enhancement of cyst wall. A diagnosis of cerebral hydatid disease was made. Serological tests for hydatid disease were positive.

The patient was operated with a standard left temporoparietal free bone flap (bone was thinned out). Cortical incision was made and cyst wall reached. While the cyst was being delivered, there was an accidental rupture of cyst. The cyst was solitary and $10 \times 11 \times 10$ cm in dimensions [Figure 1c]. It was delivered in toto followed by irrigation with 3% saline and systemic administration of corticosteroids. Duroplasty was done and bone flap was replaced. The patient was given broad spectrum antibiotics, anticonvulsants, corticosteroids and albendazole.

The boy started vocalizing from the second day and at the time of discharge after 10 days, he had grade 3

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power on the right side with improved speech. Post op CT revealed subdural hypodense collection on both sides [Figure 1d]. The patient, being asymptomatic, did not require a V-P shunt.

At 2-months follow up, the child came walking to the OPD with fluent speech. He had returned back to school. Albendazole was given for 4 months.

Case 2

A 16-year-old male presented to us with complaints of rightsided weakness for 2 years, headache for 1 year, vomiting for 3 months and diminution of vision for 4 months.

On examination, the patient was confused. Vision was limited to finger counting at 1 m. Right-sided hemiparesis was apparent. There was generalized hypertonia, reflexes were exaggerated on both sides and plantar response was up going bilaterally. CECT of brain was done as he could not afford an MRI. CT revealed a large frontotemporo-parietal cyst on the left side with septations and intracystic hemorrhage [Figures 2a and b]. The cyst wall did not enhance and there was little perilesional edema. The serological tests were positive.

The patient was operated with a large fronto-temporoparietal free bone flap and cyst reached after cortical incisions. As the cyst wall was being delivered, the cyst wall ruptured with spillage of daughter cysts [Figure 2c]. The cyst was excised, cavity irrigated with hypertonic saline and systemic corticosteroids were administered. Post op CT showed a large hypodense collection at the lesion site [Figure 2d]. A cysto-peritoneal shunt was done. The patient was discharged in full consciousness, with improved vision and power on right side. Albendazole was advised

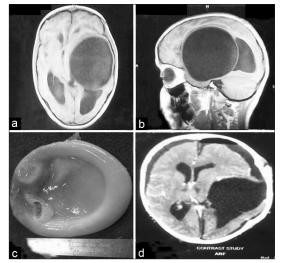


Figure 1: MRI images (a) axial and (b) sagittal of case 1 showing hydatid cyst in left hemisphere; (c) Hydatid cyst specimen removed after surgery; (d) Post operative NCCT head following complete excision of the cyst

for 4 months. The boy was brought to our emergency after one and a half months in unconscious state. He had convulsions due to poor compliance with drugs. He was appropriately managed and subsequent follow up showed that the boy had regained normal function.

DISCUSSION

Hydatid disease has been known to mankind since the days of Hippocrates. The first reports of cerebral hydatid disease and vertebral echinococcosis were made in 1807 by Guesnard and Chaussier.^[3] The causative tapeworm, Echinococcus, is found primarily in dogs, but also in wolves, foxes, sheep, goats and camels. Mammals act as intermediate hosts.

Clinical Presentation and Diagnosis

Cerebral hydatid disease is more common in pediatric population.^[1,4] Headache and vomiting are the most common presenting features.^[5-7] Papilloedema, ataxia, diplopia, hemiparesis, VI nerve palsy, Gerstman's syndrome^[8] and focal nerve deficits may also be seen.

MRI is the investigation of choice.^[6,9,10] It shows a welldefined cyst with high signal intensity on T2W image and low signal intensity on T1W image. The cyst wall shows little contrast enhancement and there is little perilesional edema. CT scan, Casoni and Wenberg test, indirect hemagglutination and ELISA are also used for diagnosis.^[11]

Most cases are located in the supratentorial compartment,^[1] mostly in the parietal lobe.^[12] Other sites are skull, cavernous sinus, eyeball, pons, cerebellum and ventricles.^[13] Cyst may be solitary or may include multiple daughter cysts.

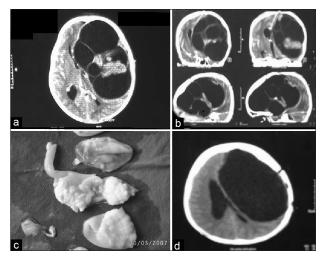


Figure 2: MRI images (a and b) showing multiloculated hydatid cyst in left hemisphere, sagittal of case 1 showing hydatid cyst in left hemisphere; (c) Hydatid cyst specimen removed after surgery; (d) Post operative NCCT head following complete excision of the cyst

Treatment

Treatment is essentially surgical with total cyst excision using Dowling episiotomies.^[1] Irrigation with hypertonic saline may be used in case of accidental cyst rupture. Small or inoperable cysts may be treated medically with albendazole 10-15 mg/kg/day in 3 divided doses for 4 months.^[13] Praziquantel is an alternative drug.

In the present report, both cases being giant cysts saw accidental rupture and were irrigated with hypertonic saline with subsequent administration of albendazole. In our view, there are high chances of spillage in case of giant cysts and hence local irrigation with hypertonic saline and post operative administration of albendazole should be routinely done.

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How to cite this article: Mallik J, Kumar A, Sahay CB, Minj TJ. Giant intracranial hydatid cyst: A report of two cases and literature review. Indian J Neurosurg 2012;1:80-2. Source of Support: Nil, Conflict of Interest: None declared.

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