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Case Report

Primary Cerebral HydatidCyst: A rare case of parasitic brain disease from non-endemic zone

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Abstract: Primary Cerebral hydatid cyst (CHD) is absolutely rare phenomenon in *Echinococcus* infection. Even in the endemic zone CHD is consist of 2% of all intra-cranial space occupying lesions (SOL). We are describing a 7-y-boy from non-endemic area with Primary CHD having no extra cranial echinococcosis presenting right sided hemiparesis, partial seizure and bilateral papilloedema. Computed tomography (CT) of brain showed a large cyst in left hemisphere, characteristics of the lesion suggested hydatid cyst. Neurosurgical intervention was taken to deliver the cyst intact and antihelminthic medication advocated. Symptoms were improved shortly and after two years follow -up no recurrence was found. In conclusion, neurological deficit with typical cystic SOL of brain should have considered a hydatid cyst even in non-endemic zone because neurosurgical management and pharmacotherapy may have been life-saving and cure for the SOL.
Keywords: Primary Cerebral Hydatid cyst, non-endemic zone, CT of Brain, Albendazole.

INTRODUCTION

Hydatid disease (HD) is caused by the larval stage of *Echinococcus*tapeworm, endemic in sheep rearing countries [1]. The literature describe the epidemiology is complex [1] in farming rural areas, sheep- dog (intermediate host and definitive host respectively) domestic animals milieu increase the chances of human infection by the eggs of infected feces. As India is not a sheep raising country, HD is not common except south India [2].

The major public health concerns in the world is caused by infestation of *Echinococcusgranulosus* [1] that the most commonly affect liver (77%) and lung (43%) as a cystic lesion [3]. CHD is rare, 1- 2% of all intracranial SOL [4].The mean age for HD is 40 years[2] but CHD is common in children [4].

Imaging (USG, CT) and ELISA (detecting specific antibody) are recommended for diagnosis. Albendazole (ovicidal, larvicidal, and vermicidal) and surgery is the mainstay of treatment [5, 6].

CASE REPORT

A 7-y-boy from rural West Bengal was presented in our institution with partial seizure involving right sided limbs and right sided hemiparesis insidious onset with progressive over two months. The boy used to play with pet dogs. On examination, vital signs were normal and his Glasgow Coma Scale (GCS) was 15/15. Power was 3/5 in right side. Deep reflexes are exaggerated in right side with extensor plantar. Direct Ophthalmoscope revealed bilateral papilloedema.

Routine blood examination was normal. CSF examination could not be done due to raised intracranial tension. CT scan of brain revealed a single large(8×9 cm) spherical, well-defined, thin-walled ,non enhancing homogeneous, CSF fluid density cystic lesion in left pareito-temporal area with mid-line shift.(Figure 1) ELISA (IgG and IgM) for *Echinococcus*was negative. The provisional diagnosis is hydatid cyst in brain. USG abdomen and Chest X ray was done to exclude hydatid cyst in liver and lungs, but no other focus were detected. Echocardiography was normal.

Patient was referred to neurosurgical department for definitive management. The large cyst was excised out meticulously to avoid rapture or spillage. Biopsy of the lesion confirmed hydatid cyst. A post-operative CT scan showed total excision of CHD and an empty space (Figure 2). Post-operative, Albendazole 15 mg/kg was

started and continued for four months and anticonvulsant for two years.

The patient showed marked improvement in his neurological deficit and was discharged in stable condition after two weeks with close follow-up for two years.

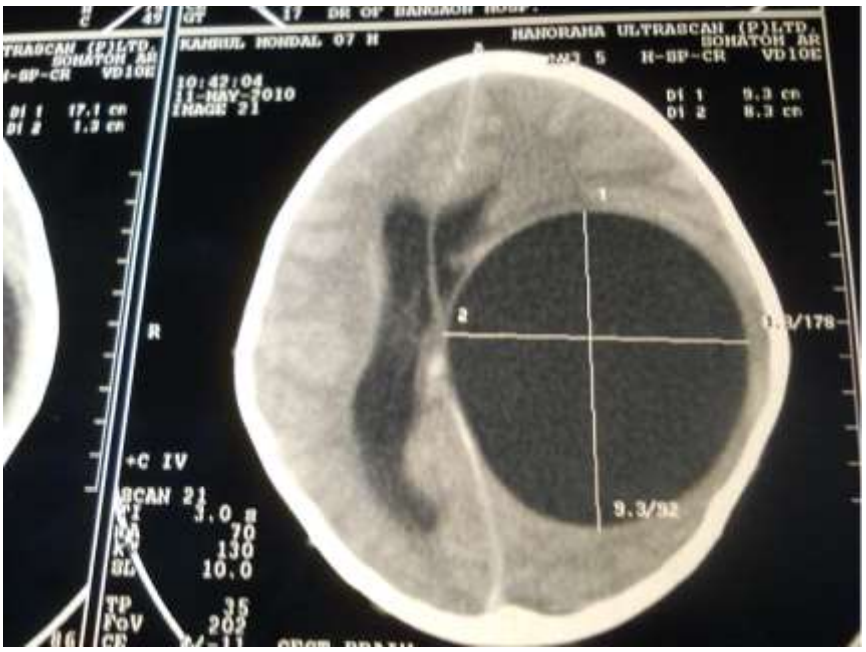

Fig 1: CT scan of brain revealed a single large (8×9 cm) spherical, well-defined, thin-walled, non enhancing homogeneous, CSF fluid density cystic lesion in left pareito-temporal area with mid-line shift

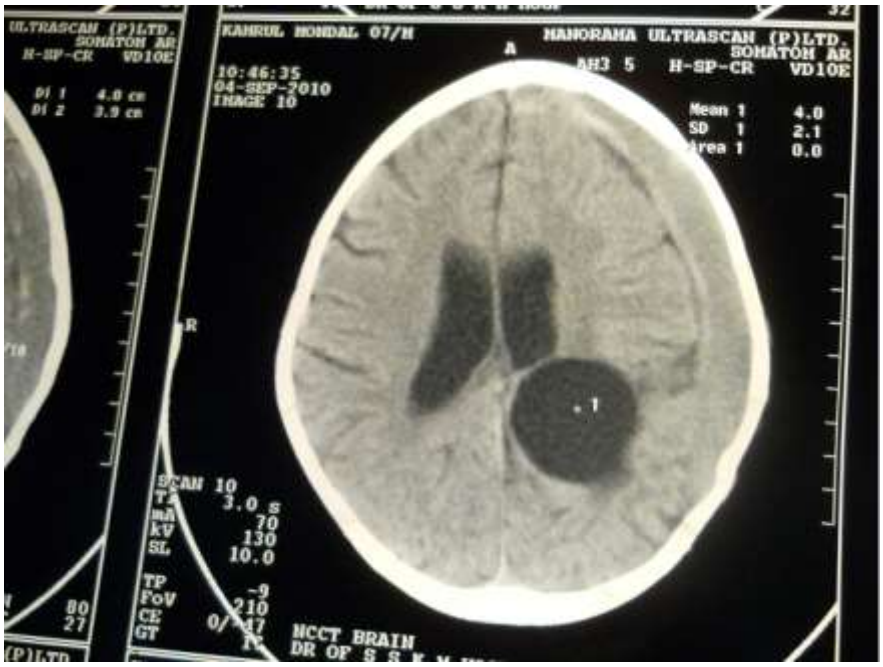

Fig 2:A post-operative CT scan showed total excision of CHD and a empty space

DISCUSSION

Hydatid disease is a re-emerging zoonotic disease in the world [1]. Although cases had been reported from every state of India, HD is not common in the country[2] except Andhra Pradesh,Tamil Nadu and Punjab where HD highly prevalent [7]. Grosso G *et al.;*depicted in geographical distribution that West Bengal is non- endemic for *Echinococcus*granulosus[1].

CHD is consisting of 2% of all echinococcosis [2, 5, 8, 9]. CHD is rare intracranial SOL[5, 9] and incidence rate in India is 0.2% **[**7]CHD is more

prevalent in children[7] than adult, our patient one of the youngest patient till published in literature. In addition, 25-30% patients are suffering from patent of ductusarteriosus [10], but our patient had normal echocardiogram. Sometimes contact- history with pet dog like our case was found previously in India [11].

The commonest site of CHD is middle cerebral territory (MCT) [4]commonly affects parietal lobe [12, 13]. CHD is classified by primary and secondary [4, 5, 8]. The larvae directly infest the brain and develop primary CHD without affecting the other organ. It has scolices and brood capsules, so primary CHD fertile. Secondary CHD arise from either rapture of primary CHD or multiple scolices come out from left heart after rapture of cyst in the heart. Apart from MCT, intracranial hydatid cyst also found in intraventricular, posterior fossa, orbit [14, 15].

Clinical symptoms develop insidiously, most commonly headache, vomiting at early period [4, 16]. In addition, hemiparesis, seizure, ataxia, diplopia and coma have been reported [4, 15, 16]. Headache, vomiting, bilateral papilloedemaare signs of increased intracranial pressure are very common in CHD [4, 16] that is as a consequence of huge size and/or obstruction of CSF flow [4].

CT scan shows distinctive features [17] well defined, spherical, smooth, CSF density cyst and no perilesional edema or enhancement, if present those are indicative of brain abscess or cystic malignancy.

There is some debate about primary CHD. Even though clinically no extracranial cyst or infection has been concomitantly found [18], but parasitic infestation may have been found in other organ almost 90% of CHD in postmortem [19]. It is hypothesized that parasite infest at early of life, this grows into cyst in brain but not in other organ, as brain has softer tissue and limited responses of immune mechanism. Similarly, ELISA test may have been false negative in CHD due to poor immunological response in brain [3, 5, 20].

CHD is primarily managed by Antihelminthic and surgical removal of cyst. Albendazole alone or combined with praziquintal is the best pharmacotherapy. Albendazole is primary mode of treatment for: infant, small, deep or inoperable CHD and also highly effective as adjuvant with surgery. After Albendazole therapy, cyst may have disappeared (48%) or decreased in the size (28%) [20]. The standard dosage of Albenazole is 10-15 mg/kg/day (max 800 mg) orally, two divided doses,for 2 months [21] but may extend up to 6 months [22]. Surgical excision of cyst is the gold standard treatment for CHD [5, 6]. Meticulous care should have taken to excised en-bloc to prevent anaphylactic shock and recurrence .Recurrence has been seen in 25% of cases so long duration follow up is advised [23].

Surgical removal, albendazole for 4months and antiepileptics had been advocated in our patient and followed up to two years.

CONCLUSION:

In conclusion, a child presents with focal neurological deficit, seizure and bilateral papilloedema for short duration and CT of brain is showing large, non-enhancing cystic lesion that can be CHD even in non-endemic zone. Prompt diagnosis and surgical intervention and Albendazole therapy can have been lifesaving and cure of the disease.

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Conflicts of interest:

There are no conflicts of interest

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