

Young girl with multiple intracranial hydatid cyst

Nani Sen¹, Debal Laha², P K Gangopadhyay², B.C. Mohanty¹

¹Department of Neurosurgery, ²Dept. of Neuro-Medicine,
Calcutta National Medical College, Kolkata – 700014, INDIA

Corresponding Author:

Nani Sen, MS

Tel: +91 94322 63664

E-mail: dr.nanisen@gmail.com

ABSTRACT

8 years young girl presented with uncontrolled seizures in obtunded and bedridden state. MRI brain showed multiple extra-axial coalescent cystic lesions in bilateral frontotemporo parietal regions – diagnosed as multiple hydatid cyst. We used irrigation saline during surgery and a total 35 cysts were removed intact, one by one separately and histologically confirmed as hydatid cyst. Postoperatively, patient showed marked neurological improvement and was seizure free.

KEY WORDS: Hydatid cyst, Multiple, Intracranial

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Introduction

Hydatid disease or echinococcosis is caused by infestation with larval stage of tapeworm echinococcus. The main pathogenic species for human are echinococcus granulosus which produce cystic lesions. ¹⁻⁴ All organs may be involved, with brain being involved in only 1-2% of all infestation. ³⁻⁶ Cerebral hydatid cyst are 2-3 times more common in children than adults. ⁶⁻⁸ 90% of cases have solitary lesion,⁹ most frequently supratentorial, and in the middle cerebral artery territory. ³⁻¹¹

The diagnostic tools including CT and MRI lead to early and correct preoperative diagnosis. The usual appearance is large, intra parenchymal non enhancing hypodense lesion with a well circumscribed border and no perilesional oedema. ^{2,10}

Case Report

8 years female child presented with complaint of urinary incontinence for 2 months, weakness of all 4 limbs for 1 month and seizure for 3 weeks. The patient was apparently asymptomatic 2 months back. She developed urgency and frequency of micturition with occasional incontinence of urine. She initially developed weakness of left lower limb followed by right upper limb and gradually all four limbs. Patient developed generalized tonic clonic seizures with rolling of eyeballs, right side more than the left with frothing from mouth, associated with occasional involuntary passage of urine and faeces for last 3 weeks. Patient also had history of vomiting, diminished speech for 1 month. For last 10 days there was impairment of consciousness and she was bedridden for last 7 day.

Patient had no history of fever or headache. She had no visual complaint, with no history of cranial or spinal trauma. She had past history of contact with a street cat. She was from poor socio economic status with hygienic habits receiving antiepileptic drug (sodium valproate) before admission without control of seizure.

Examination

General and systemic examinations were within normal limits. There was spontaneous eye opening without any response to verbal command even though, though the patient responded

to painful stimuli. Speech was severely impaired and cranial nerve examination showed horizontal nystagmus on looking towards left side. Motor examinations revealed hypertonia with dystonic tremor of right hand. The power could not be properly assessed. Deep tendon jerks were exaggerated, plantar reflex was bilaterally extensor. Other systems were normal. Informed consent was obtained from the patient.

Investigation

Routine hemogram, serum electrolytes and liver function tests were within normal limits, chest x-ray revealed no abnormality, interictal EEG showed paroxysmal generalized dysrhythmia. USG of abdomen showed small cystic lesions in the spleen but there was no lesion in the liver.

MRI of brain showed multiple extra-axial coalescent rounded cystic lesions involving bilateral frontotemporoparietal regions measuring 8 cm. × 8.7 cm, without contrast enhancement but with significant mass effect. Diameter of each cyst ranged from 1-3 cm.

Pre-surgical - patient was given sodium valproate and levetiracetam in standard dose. After MRI of brain, she was given albendazole.

Surgical - Bifrontal Craniotomy was performed. Dura was tense, on opening the dura from right side, there were multiple rounded cysts extraaxially and the cyst wall was transparent, with clear plane between the cysts and the arachnoid. After splitting the arachnoid membrane, cysts appeared to come out of the brain. A6Fr infant feeding tube was passed between the cysts and gentle irrigation with normal saline done. All 35 cysts were removed one by one separately without causing rupture inside the brain. Immediate postoperative stage was uneventful. Histological section of the cyst showed evidence of hydatid cyst.

Postoperatively, the patient responded to verbal command on day 1 and spoke meaningful words on day 2 after operation. However, seizures continued for about 5 days and abnormal behavior along with pathological laughter (which started on day 1) persisted for about two weeks thereafter. Gradually, there was significant improvement of cognition along with speech

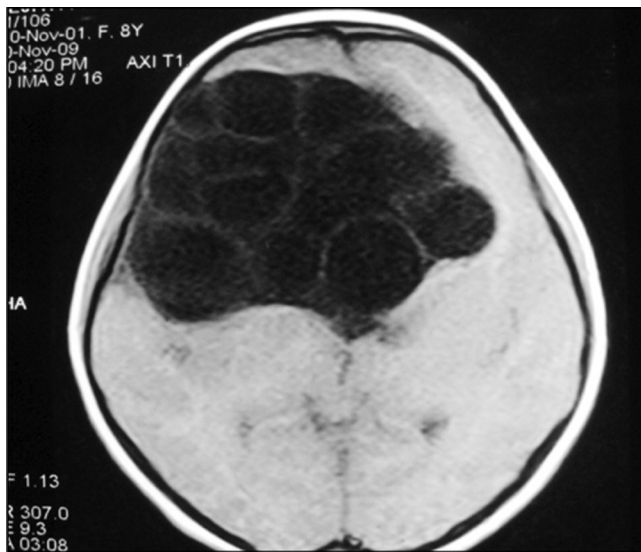


Fig. 1: T₁ MRI sequence (axial) shows multiple coalescing thin walled cysts of varying sizes in both fronto-parietal areas.

and remarkable improvement of power of upper and lower limbs, became ambulatory (with support) within 2 weeks.

Repeat USG abdomen (at 2 months) revealed no lesion in the spleen

Follow up CT scan at 2 months after surgery showed one cystic lesion in right frontal area. Albendazole was given for six weeks in standard doses but the anti-epileptic drugs were continued.

Discussion

Intracranial hydatid cyst is rare, with a reported incidence of 1-2%.⁶ Intracranial hydatid cyst is more frequently localized in supratentorial compartment. The most common site for intracranial hydatid cyst is parietal lobe. All the four cases reported by Dharker et al¹² and 3 out of 5 cases of intracranial hydatid cyst reported by Balasubramaniam et al¹³ both exhibited involvement of parietal lobe. The other less common sites reported are skull,¹³ cavernous sinus,¹⁴ eyeball,¹⁵ pons,¹⁶ extradural,¹⁷ cerebellum and ventricles.¹⁸ It was reported that the growth rate of hydatid cyst in brain was 1.5 to 10 cm/year.^{18,19} In our case, the total size of the hydatid cysts were 8cm × 8.7cm. The cyst wall is whitish and transparent, it is involved bilaterally with the fronto-temporo-parietal region. Solitary hydatid cyst is common, multiple cyst are rare. In our case we had 35 multiple cysts.

The patient with intracranial hydatid cyst showed focal neuro-surgical deficit and features of raised intracranial pressure as a result of interference with pathway of CSF flow. Index case patient presented with focal neurological deficit and features of raised ICP. With the help of MRI and CT scan it was characteristically shown that hydatid cyst is a spherical, well defined, non enhancing cystic lesion without peripheral oedema.

Surgery is the treatment of hydatid cysts with an aim to excise the cyst without rupture in order to prevent recurrence and anaphylactic reaction. Various surgical options were summarized by Aranalniguez.²⁰ However, the most popular technique is to use irrigation saline with mild force between the cyst wall and bed interface in order to deliver this cyst intact. We used

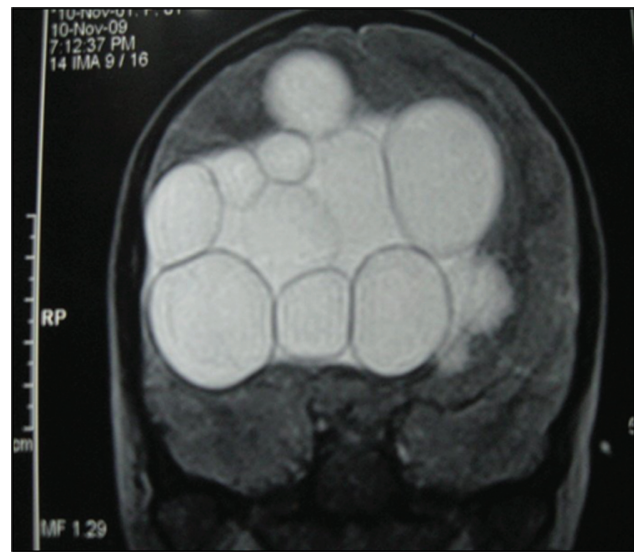


Fig. 2: T₂ MRI sequence (coronal) shows bilateral fronto-temporal multiple cysts.

this method for delivery of the cysts. On review we failed to find as many cysts being removed from a single patient successfully. The patient had clinically uncontrolled seizures and obtunded sensorium and she was bedridden before operation.

After surgery, sensorium improved dramatically and seizure was controlled. The patient began to speak from day two and walked with support after two weeks of surgery. At 'two months' follow-up, the patient was seizure free and was fully ambulatory with normal sensorium and enjoying normal life.

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