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Intracranial Hydatid Cyst: A Report of Three Cases in North-West India

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Abstract

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Human echinococcus is caused by tapeworm, *Echinococcus granulosus*, which forms larval cysts in the human tissue. Incidence in the cerebral form is only 1–2%. This localization can be associated with the involvement of other organs such as liver or lung or may be an isolated infestation of the brain or spinal column. Surgical removal of the intact and unruptured cyst is advised to prevent local recurrence that may require further surgery and long-term treatment with parasitocidal agents. We report three cases who presented with headache, vomiting, hemiparesis with decreased visual acuity, and convulsions. MRI showed a giant hydatid cyst in all three cases which was removed surgically and the patient was successfully discharged. Successful treatment of hydatid cyst requires preoperative diagnosis and meticulous surgical technique for complete excision of cyst without rupture under perioperative coverage of albendazole to avoid recurrence and anaphylaxis.

Keywords: *Albendazole, Echinococcus, hydatid cyst*

INTRODUCTION

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Human echinococcus is caused by a tapeworm, *Echinococcus granulosus*, which forms larval cysts in the human tissue. The definite hosts of echinococcus are various carnivores, the common one being the dog. All mammals (more often sheep and cattle) are intermittent hosts. Humans get infected through the faeco-oral route by ingestion of food or milk contaminated by dog feces containing ova of the parasite or by direct contact with dogs. The incidence of hydatid disease varies greatly in different geographical areas. It is much more frequently found in South America, Australia, Middle East, and parts of North Africa than in Europe and North America.[1] In our country, *E. granulosus* is relatively common in southeast Rajasthan.

The most common location of hydatid cyst is the liver (60%). Cerebral and spinal hydatid cysts are rare. Incidence of the cerebral form is only 1–2%.[2] This localization can be associated with the involvement of other organs such as liver or lung or may be an isolated infestation of the brain or spinal column. Fifty to seventy-five percent of cerebral cases appear in childhood with supratentorial location whereas infratentorial lesions are quite rare.[3]

Because of this rarity, experience with intracranial hydatid disease at a single institution has been very limited, and the Dowling technique is widely used as a surgical treatment.[4] Preoperative and postoperative albendazole may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall, and hence reduce the risk of spillage during surgery and the recurrence rate.[5]

CASE REPORT

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Three cases of intracranial hydatid cysts were treated during the period of 2013–2016 in the neurosurgical department at the Government Medical College, Kota, representing an incidence of 1% of all space-occupying lesions operated during this period. The mean age of presentation was 12.5 years. Two patients (66.6%) were in the age group of 10–15 years and the remaining one case was less than 10 years old. The male to female ratio was 2:1. In one case, there was a history of contact with pet dogs. All three patients were from rural areas of southeast Rajasthan. The duration of symptoms varied from 1 month to 2 years. The clinical features varied from headache associated with occasional vomiting, decreased visual acuity, hemiparesis, and convulsions depending on the location of cyst in the brain. Hemiparesis was the commonest finding in all the cases. Papilledema and seizures were seen in two cases. In all three patients, radiological investigations included CT scan and MRI, which revealed a solitary cyst in the temporoparietal region in two patients and one was found to have had multiple cysts in the parietal lobe [Figures 1 and 2]. X-ray of the chest and ultrasonogram of the abdomen were done in all three cases. These investigations failed to reveal any associated hydatid cyst in the lungs and abdomen.

The hydatid cysts were totally excised in all the three cases and necessary precautions to prevent rupture and dissemination of hydatid were taken during the surgery; despite these precautions, rupture of cysts occurred in one case (33.3%). Following rupture, no evidence of anaphylactic reaction was noted. One patient with solitary cyst in the temporoparietal lobe had recurrence of hydatid cysts and presented with multiple cysts after 6 months of the first surgery. The patient was reoperated at another institute and all the cysts were removed during the second surgery. However, recurrence led to reoperation at our institute. After surgery, albendazole was given for 6 months. All three patients have been followed up from 6 months to 1 year with no recurrence in any case [Figures 1 and 2]. All patients have shown good recovery from neurological dysfunction.

DISCUSSION

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In hydatid disease, the only pathogenic species for humans is *E. granulosus*. Humans become infected by ingesting tapeworm eggs passed from an infected carnivore, especially dogs, which most frequently happens when individuals handle or have contact with infected carnivores or inadvertently ingest food or drink contaminated with fecal material containing tapeworm eggs.[1] However, because India is an endemic region, all the children in our series may have been infected by eating ingested food. Intracranial hydatid disease is considered a childhood disease. Fifty to seventy-five percent of intracranial hydatid cysts are seen in children. Izci *et al.*[6] reported a series of 17 patients with intracranial hydatid cysts and 13 (65%) of these patients were children. Cerebral hydatid cysts are often supratentorially localized in the distribution of the terminal branches of the middle cerebral artery, usually temporo-parieto-occipitally.[7] In our case, patients also had cysts in the supratentorial location and in the distribution of the terminal branches of the middle cerebral artery.

Brain hydatid cysts are relatively rare and only account for up to 2% of total cases. The actual percentage may be higher than what we have in the literature, due to underreporting. Brain hydatid cysts can be primary (single) or secondary (multiple). The primary cysts are fertile as they contain scolices and brood capsules, hence rupture of primary cyst can result in recurrence.[8] The secondary multiple cysts result from spontaneous, traumatic, or surgical rupture of the primary intracranial hydatid cysts and they lack brood capsule and scolices. The secondary intracranial hydatid cysts are therefore infertile and the resultant risk of recurrence after their rupture is negligible. Primary multiple cysts are uncommon and isolated case reports of primary multiple hydatid cysts can be found in the literature.[8,9]

The wall of the cyst consists of an inner endocyst (germinal layer) and outer ectocyst (laminated layer). The host reacts to the cyst forming a pericyst (fibrous capsule), which provides nutrients to the parasite. In the brain, due to minimal reaction, the pericyst is very thin. The endocyst produce scolices that bud into the cyst cavity and may sediment within the hydatid cavity, commonly known as hydatid sand.[10]

The commonest presentation is that of a child or young adult with signs and symptoms of raised intracranial pressure. In adults, focal neurological signs like hemiparesis, hemianopia, speech disorders, or seizures were usually first to appear, whereas in children the clinical picture was dominated by symptoms of raised intracranial pressure.[11] In our case, headache was the earliest symptom in all cases. Vomiting was often associated with headache. Variations of symptomatology such as hemiparesis, somnolence, and convulsive fits with lowering of visual acuity were noted. Seizure occurred in two of three patients with

cerebral hydatid cysts. Examination may reveal impairment of general health and somnolence. Macewen sign, papilledema optic atrophy, visual field defects, hemiparesis, and signs of cerebellar dysfunction and stiffness of the neck may occur.

On CT scan, a solitary cyst appears as well-defined, spherical, smooth, thin-walled, and homogeneous, with an inner density similar to CSF, and nonenhancing walls. The wall may appear isodense to hyperdense on CT scan and, rarely, may become calcified. There is usually no surrounding brain parenchymal edema, which if exists along with ring enhancement, indicates inflammation and infection. Ring enhancement and perilesional edema differentiates brain abscesses and cystic neoplasms from uncomplicated hydatid cysts. These findings can, in fact, sometimes cause dilemma and misdiagnosis and lead to catastrophic events. The cyst shows low-signal intensity on T1-weighted, and high-signal intensity on T2-weighted MRI. MRI may also show perilesional edema not seen on regular CT scan imaging.[12] MRI may prove superior in determining the exact cyst location, presence of super-added infections and cystic contents, and also in surgical planning and ruling out other diagnostic possibilities. We strongly recommend MRI for better evaluation of cystic brain lesions.

The treatment of cerebral hydatid cysts is principally surgical. The primary goal of the operation is total cyst extirpation without rupture. Many different techniques of cyst removal have been proposed and all of them emphasize atraumatic techniques to avoid cyst rupture. The Dowling technique, later improved by Arana-Iniguez and San Julian, has been widely used for the surgical treatment of hydatid cysts of the central nervous system. The essential steps of this technique are the following: creation of a large flap; careful handling during all operative steps to avoid monopolar coagulation; opening the atrophic cortex overlying the cyst over an area with a diameter no less than three quarters of the diameter of the cyst; and allowing the cyst to fall out by just lowering the head of the operating table and flushing warm saline between the cyst and surrounding brain.[13]

Despite the advancements in microsurgical operative techniques and instrumentation, cerebral hydatid cysts pose a challenge for the surgeon because of the following characteristics: they are usually diagnosed when they are large in size; they have a very thin cyst wall; the neurological deficits are often minimal in their presentation despite the location and the large size of the cyst; and they are sometimes located deep or near the ventricular wall and require retraction of vital structures or meticulous cortical dissection. Many reports suggest that the Dowling technique is the most effective surgical procedure for the removal of cerebral hydatid cysts.[14] However, some pitfalls exist with this technique concerning surgical methods, instruments, and cyst location. The best operative approach to a cystic lesion in the brain should be based on the site and the size of the cyst, and the relationship of the lesion with the other neural and vascular structures.

Definitive treatment is complete removal of the cystic lesion by surgery followed by medical treatment with albendazole to avoid recurrence. Isolated case reports showed complete disappearance of multiple intracranial hydatid cysts with albendazole therapy with a daily dose of 10 mg/kg, taken three times a day for 4 months. Preoperative and postoperative albendazole treatment may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the spillage during surgery), and reduce the recurrence rate.[5] All our patients were operated upon and total surgical removal was possible. Albendazole treatment was started in one case before the second operation for recurrence and continued for 6 months postoperatively.

CONCLUSION

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Successful treatment of hydatid cyst requires preoperative diagnosis and meticulous surgical technique for complete excision of cyst without rupture under perioperative coverage of albendazole to avoid recurrence and anaphylaxis.

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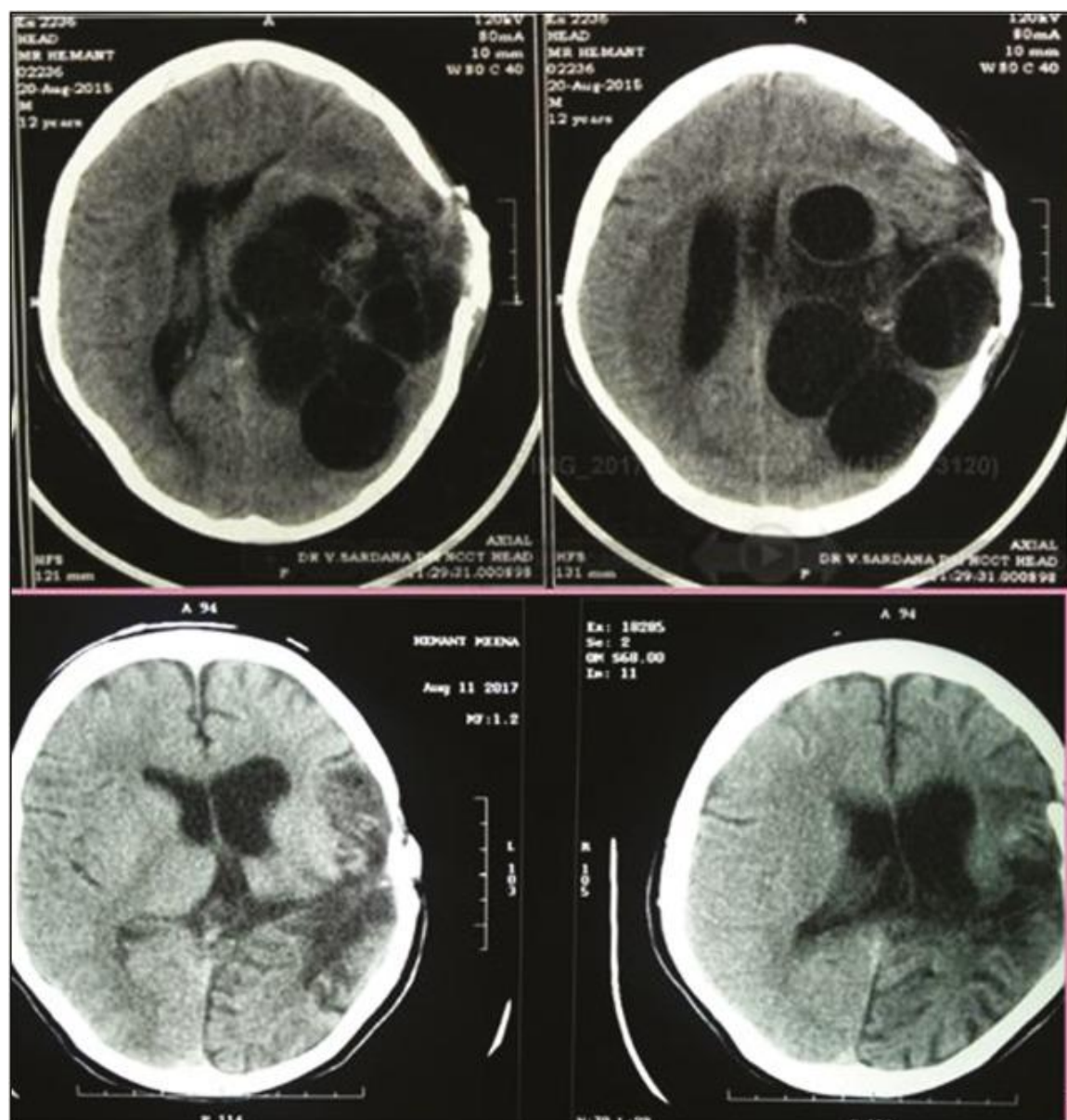
Conflicts of interest

There are no conflicts of interest.

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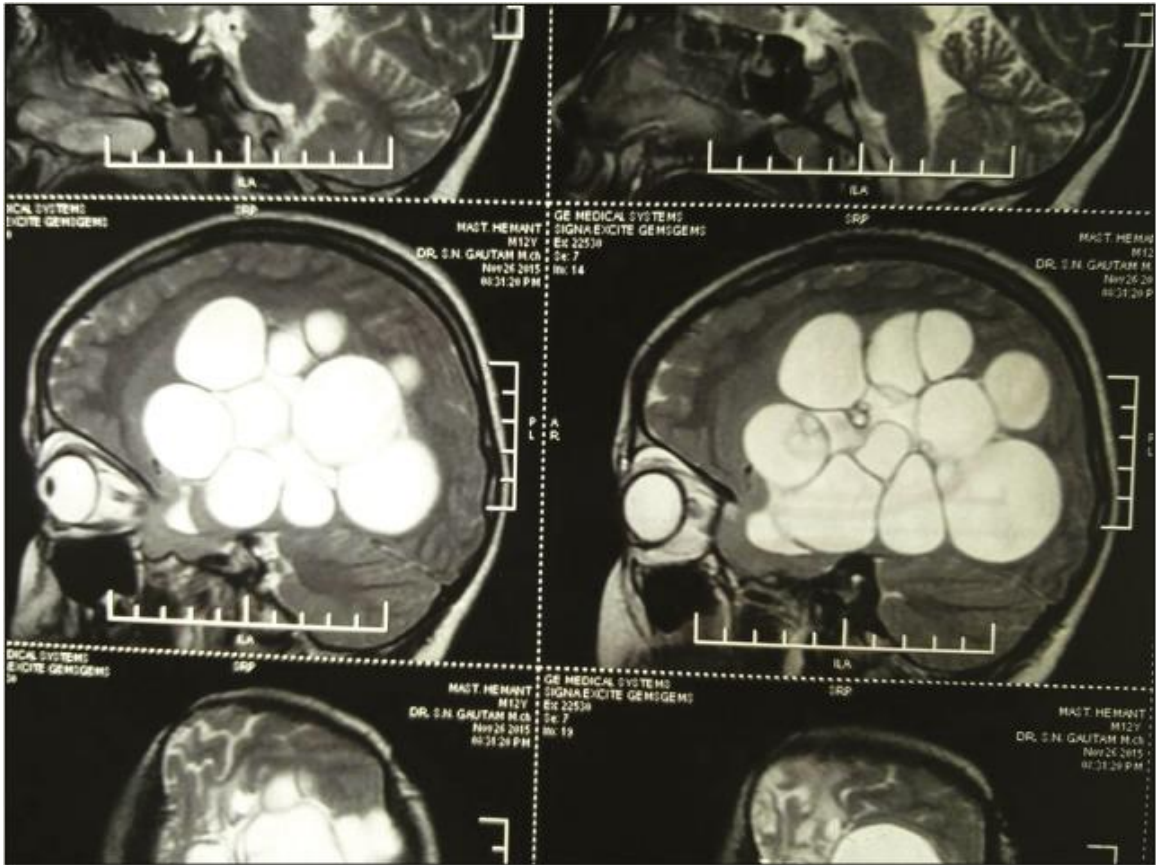
Figures and Tables[Go to:](#)

Figure 1

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Pre operative NCCT head showing multiple hydatid cysts and post operative image showing no residual

Figure 2



Pre operative scan

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