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Review Article

Cerebral intraventricular echinococcosis in an adult

Sharad Pandey, Deepa Pandey[1], Neeraj Shende, Anurag Sahu, Vivek Sharma

Departments of Neurosurgery, Sir Sunderlal Hospital, IMS, BHU, [1]Clinical Microbiology, DLW, Varanasi, Uttar Pradesh, India

E-mail: *Sharad Pandey - drsharad23@yahoo.com; Deepa Pandey - deepalohani02lhmc@gmail.com; Neeraj Shende - neerajshende11@gmail.com; Anurag Sahu - anuragsahu25@rediffmail.com; Vivek Sharma - neurosurgeryimsbhu@gmail.com
*Corresponding author

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Abstract

Background: Echinococcosis in humans occurs as a result of infection by the larval stages of taeniid cestodes of the genus echinococcus. Intracranial hydatid cysts usually develop at an intraparenchymal site. Hydatid cyst within the cerebral ventricle is quite unusual.

Methods: We reviewed the literature on adult intraventricular hydatid cyst and found case reports mainly in children with an only handful of cases in adults. We reported a rare case of cerebral intraventricular (left lateral ventricle) hydatid cyst in a 21-year-old adult female.

Results: Although cerebral hydatid cysts are most commonly seen in children and young adults cerebral intraventricular hydatid cyst are comparatively rarer in adults.

Conclusion: The possibility of infection with *Echinococcus granulosus* should be included in the differential diagnosis of raised intracranial hypertension in patients from endemic areas.

Key Words: Adult, cerebral intraventricular hydatid cyst, children, *Echinococcosis*

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INTRODUCTION

Echinococcosis or hydatid disease, an endemic zoonotic disease, occurs as a result of infection by the larval stages of taeniid cestodes of the genus echinococcus. The two most commonly associated with human disease are *Echinococcus granulosus* whose cyst has limiting membrane and *Echinococcus multilocularis* (alveolar) which are less common but more serious and is almost always fatal. Several studies have shown that these diseases are an increasing public health concern and that they can be regarded as emerging or re-emerging diseases.[31] The incidence of intracranial hydatid in India is 0.2%.[9,39,43] Hydatid cysts constitute 3–4% of all intracranial space occupying lesions.[6,12,22,26,28,44]

CASE REPORT

Twenty-one-year-old adult female with rural background presented in the OPD with chief complaints of headache since 3 years and difficulty in walking since 6 months. Physical examination was unremarkable. A neurologic examination revealed bilateral papilledema. The hematological investigation was within normal limits. No abnormality was found in X-ray chest, echocardiography, ultrasonography (USG) abdomen, and pelvis. Computed tomography (CT) scan of brain revealed mild asymmetrical dilatation of left ventricle with normal size right lateral, third and fourth ventricles without periventricular edematous changes suggesting possibility of outlet obstruction of lateral ventricle [Figure 1a]. Magnetic

resonance imaging (MRI) of the brain showed evidence of altered signal intensity complex cystic lesion measuring, approximately 4 cm × 3 cm × 3 cm, noted in left lateral ventricle near foramen of Munro leading to widening of left lateral ventricle with mild midline shift toward right side. Few small daughter cysts noted within the cyst. No significant postcontrast enhancement noted within the lesion [Figure 1b]. Magnetic resonance spectroscopy (MRS) showed acetate peak. No evidence of any other lesions in ventricles and brain parenchyma noted. There was no family history of hydatid infection. The patient underwent left frontal craniotomy. Intraoperative USG of the cystic mass showed a hypoechoic cystic lesion in the left lateral ventricle with another small cystic lesion within this hypoechoic lesion [Figure 2a]. The left lateral ventricular wall including the choroid plexus was exposed. The cystic mass was removed successfully by gentle irrigation of the cleavage plane between the cyst wall and the brain interface with saline [Figure 2b]. After cyst removal, the ventricle was irrigated several times with 3% hypertonic saline. The histopathologic findings were consistent with hydatid cyst [Figure 3a and b]. The postoperative course was uneventful, and she was discharged 10 days after surgery. The patient was continued on anthelmintic albendazole 10 mg/kg 3 times daily for 3 months.

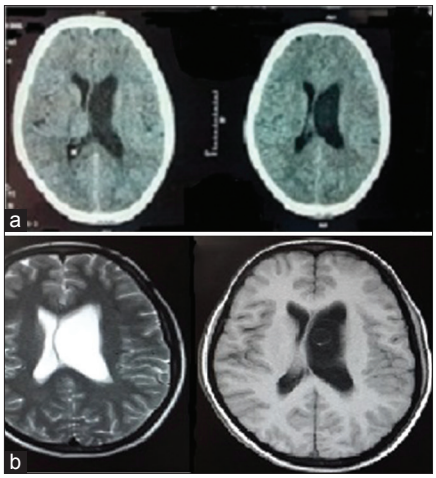

Figure 1: (a) Noncontrast computed tomography brain showing asymmetrical dilatation of left lateral ventricle. (b) Magnetic resonance imaging of the brain showing complex cystic lesion in left lateral ventricle and cyst within cyst appearance of hydatid

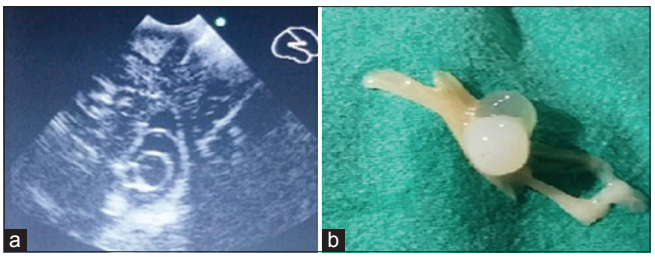

Figure 2: (a) Intraoperative ultrasonography is showing a hypoechoic cystic lesion in left lateral ventricle with another small cystic lesion within this lesion. (b) Hydatid cyst after excision

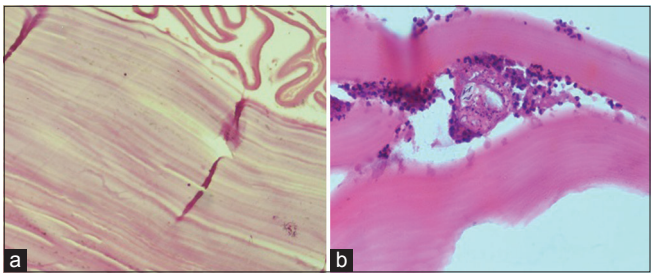

Figure 3: (a) Microphotograph (H and E, ×40) showing ectocyst (laminated wall) of hydatid cyst. (b) Microphotograph (H and E, ×40) showing ectocyst (laminated wall), scolex containing multiple hooklets with mixed inflammatory infiltrate

DISCUSSION AND REVIEW OF LITERATURE

Echinococcosis is also known as hydatidosis or hydatid disease. Six species have been recognized, but four are of public health concern: *E. granulosus* (causes cystic echinococcosis), *E. multilocularis* (causes alveolar echinococcosis), and *Echinococcus vogeli* and *Echinococcus oligarthrus* (which cause polycystic echinococcosis).[32] The two most commonly associated with human disease are *E. granulosus* whose cyst has limiting membrane and *E. multilocularis* (alveolar) which is less common but more serious and is almost always fatal.[2,5] The latter lacks limiting membrane thus can grow aggressively.[31] Two new species have recently been identified: Echinococcus shiquicus in small mammals from the Tibetan plateau and *Echinococcus felidis* in African lions, but their zoonotic transmission potential is unknown.[1] Though the mortality directly due to echinococcosis is low (4–5%) but it can be a serious problem.[16,25]

Echinococcosis is a zoonosis in which the definitive host is a carnivore that harbors the adult tapeworm in the small intestine.[31] The carnivore becomes infected by ingesting the larval form in the tissue of the intermediate host.[31] The intermediate host, chiefly herbivorous animals but also humans, become infected by ingestion of tapeworm eggs, passed in carnivore feces.[31]

The greatest prevalence of cystic echinococcosis in human and animal hosts is found in countries of the temperate zones including southern South America, the entire Mediterranean littoral, southern and central parts of the former Soviet Union, Central Asia, China, Australia, and parts of Africa.[33,48] In India, the hydatid disease is more commonly seen in the Kurnool district of Andhra Pradesh, Madurai district of Tamil Nadu, and Punjab.[10]

Hydatid is a Greek word meaning "a drop of water." Hydatid cyst commonly occurs in the liver (55–70%), lung (18–30%), followed by the kidneys muscles, spleen, soft tissues, brain, and bone.[14] On the contrary,

Geramizadeh reported that the most common locations of the hydatid cyst, after the lung and liver, to be the central nervous system (CNS), orbit, musculoskeletal system, cardiovascular system, kidney, and urinary tract.[15]

Hydatid cysts constitute 3–4% of all intracranial space occupying lesions.[6,12,22,26,28,44] We reviewed the literature and found a handful of cases on cerebral intraventricular hydatid cyst [Table 1].[3,7,8,11,13,17,18,20,23,29,38] Brain involvement in hydatid disease, especially with respect to intraventricular cyst location, is rare and occurs primarily in children.[42] Only three cases reports were available in adults pertaining to cerebral intraventricular hydatid cyst.

The most common location in CNS infection is the hemispheric parenchyma, in the perfusion territory of the middle cerebral artery, especially in the parietal lobe.[7] Cerebral hydatid cysts are well encapsulated and vascularized and rarely degenerate.[19] The other less common reported sites are pons, cerebellum, basal ganglia, extradural, skull, cavernous sinus, ventricles, and eyeball.[11,41]

The cerebral hydatid cysts are usually slow growing; the growth rate has been variably reported between 1.5 and 10 cm/year.[35,45] They may reach a considerable size before the patient becomes symptomatic. As a rule, the hydatid cyst of the brain tends to be solitary and spherical.[1]

Clinically patients present with nonspecific signs and symptoms, most common ones being a headache, papilledema, vomiting, and symptoms of raised intracranial pressure can be seen.[5,7,25] Focal symptoms are usually depend on the site and size of the lesion.

Cerebral hydatid cysts are most commonly seen in children and young adults.[7] Children are more commonly affected than adults.[12,21,42] The review of the literature on cerebral intraventricular hydatid cyst till date showed that children were more commonly affected than adults.[3,7,8,11,13,17,18,20,23,29,38]

Among the laboratory investigations the immunoblot test, where available is the test of choice (98% specific and 91% sensitive). The "arc 5" antigen is present in *E. granulosus* hydatid fluid and can be identified easily as it shows a well-defined immunoelectrophoretic pattern. Arc 5 test is also diagnostic except for cross reactions with *Taenia solium* cysticercosis infection. Several other tests enzyme-linked-immunosorbent-assay, indirect hemagglutination, immunofluorescence are useful, but both false negative and false positive results are common.[31] The Weinberg and Casoni serologic tests are of little practical value in confirming the diagnosis of cerebral echinococcal disease.[25,30]

Imaging studies such as CT scan and MRI are necessary for preoperative diagnosis.[1] These play a

Table 1: Cases of cerebral intraventricular hydatid cyst reported till date (*cases in adult)

Year reported	Reported by	Age /gender	Clinical features	Duration	Intraventricular location in CNS	No. of CNS cysts
1992	Copley IB *et al* [8]	NA (02 children)	Headache, dizziness		Lateral ventricle	01
1993	Diren HB *et al* [11]	10/F	Headache, right proptosis		Frontal horn of the right lateral ventricle	02 (another in Left frontal region- 01)
1999	Gupta S *et al* [17]	Not Available	Focal neurological deficits and features of raised intracranial pressure		Lateral ventricle	01
2002*	Aydin MD *et al* [3]	18/M	Headache, blurred vision, vertigo, vomiting		Right lateral ventricle	Quadruplet (fourcystic masses, 2-8 mm in diameter)
2004	Iyigun O *et al* [20]	2/M	Focal neurological deficit		Right lateral ventricle	01
2004	Bukte *et al* [7]	7/F	Cerebellar deficit, ataxia		Posterior fossa,4th ventricle	01
2005	Evliyaoglu C *et al* [13]	7/F	Headache, nausea, vomiting		Lateral ventricle	01
2007*	Maurya P *et al* [29]	25/F	Complex partial seizures for 3 months. bifrontal headache for the last 2 months, and bilateral papilledema		Right lateral ventricle	01
2008	Guzel A *et al* [18]	10/F	Headache	10 months	Right lateral ventricle	01
2009	Kamath SM *et al* [23]	6/F	Headache, vomiting since 10 days, left hemiparesis since 1.5mths		Right lateral ventricle	01
2013*	Prasad RS *et al* [38]	20/M	Headache	3months	Third ventricle	01
2014*	Present study	21/F	Headache	3 years	Left lateral ventricle	01
			Difficulty in walking	6 months		

CNS: Central nervous system

major role in patient management as symptoms are quite often nonspecific. The differential diagnosis of intracerebral hydatid cysts [Table 2] includes cystic lesions such as a porencephalic cyst, arachnoid cyst,

Table 2: Differential diagnosis of cerebral hydatid cyst

Cystic cerebral lesion	Typical features	Radiological findings
Neurocysticercosis	Usually more numerous lesions, common CNS locations are the gray matter-white matter junction and deep sulci, less common in subarachnoid spaces and ventricles (especially IVth ventricle)	Multiple lesions in different stages of development. 1) Vesicular stage- cyst fluid isodense to CSF on CT and isointense to CSF on MRI studies, with a small dot inside. E−/CE-.2) Colloidal vesicular stage- larva begins disintegrating, an intense inflammatory response seen around cyst, resulting in a fibrous capsule, identified by MRI. Cyst wall CE+ on CT or MRI with PE+. Cyst fluid has increased density on CT scans and increased intensity on MRI. 3) Granular nodular stage- cyst becomes granulomatous nodule with peripheral gliosis, calcifications +/−, following contrast administration on CT scan surrounding E+, CE+. Isointense on T1and iso to hypointense on T2-weighted images, showing nodular or ring enhancement.4) Nodular calcified stage- small calcified nodules may be seen, E−, CE−
Cerebral toxoplasma abscesses	Focal brain lesions mostly localized to the basal ganglia, but also in other brain regions	Surrounded by E+++, NCCT -usually multiple low density areas, on CECT, CE−,NE−,RE−.MRI -hypointensity on T1−& variable intensity on T2-weighted images, due to presence of hemorrhage and/or calcification. Post contrast T1-weighted images may exhibit a highly suggestive abscess aspect: "the target sign" with rim-enhancement and central hypointensity with a little eccentric nodule of contrast inside the mass. Usually characterized by surrounding moderate oedema in the periphery, with hypointensity on T1 weighted and hyperintensity on T2-weighted images. Typical radiological findings comprise multiple, ringenhancing lesions in both cerebral hemispheres
Arachnoid cyst	Benign, probably congenital lesion, localized in the intra-arachnoidal space, usually supratentorial. May have large sizes but generally do not communicate with the ventricles, not spherical in shape, not surrounded entirely by brain substance, are extra-axial masses that may deform adjacent brain, have an irregular inner border	Are well circumscribed, having the same signal intensity as CSF at CT scans and all MRI sequences, with no contrast enhancement
Porenephalic cyst	Usually not spherical in shape, not surrounded entirely by brain substance result from insults to normal brain tissue	Lined by gliotic white matter that could easily be demonstrated with MRI
Epidermoid cyst	Benign, congenital most common at the CP angle (≈50%), sellar & parasellar regions, diploe, rhomboid fossa, 4thventricle/ brainstem, corpus callosum, pineal gland. May develop within the frontal, parietal, or petrous bone & may destroy the inner and outer table of the cranial bone to cause soft-tissue swelling under the scalp well-demarcated, encapsuled lesions, with a whitish capsule of a mother-of-pearl-sheen (pearly tumor) lined by stratified squamous epithelium & are filled with debris, keratin, water,& cholesterol crystals. Cyst rupture may produce intense chemical meningitis. Epidermoids can be differentiated usually by their lobulated, vessel engulfing, self moulding behaviour	On CT scans, appear as well demarcated hypodense lesions that resemble CSF and do not enhance with contrast agents. Most of them show low signal on T1-weighted and high signal on T2-weighted MRI sequences
Hydatid cyst	Most common location in CNS is the hemispheric parenchyma. Rare sites- subarachnoid spaces, lateral ventricle, and cerebellum. Most cysts contain clear fluid, usually associated with small daughter cysts and a granular deposit of scolices.	On CT or MRI examination, the hydatid lesions appear as large, spherical, cystic masses well demarcated from the surrounding brain parenchyma,with cyst fluid isodense with CSF on CT scans and isointense with CSF on MRI studies with no surrounding oedema. CE- partial or complete involving the cystic wall. The peripheral capsule of the cyst can usually be seen on MRI imaging, and calcification of the wall is better identified on CT imaging

E: Edema, CE: Contrast Enhancement, PE: Perilesional edema, NE: Nodular enhancement, RE: Ring-like enhancement, +: Present, −: Absent, CNS: Central nervous system, CFS: Cerebrospinal Fluid, MRI: Magnetic resonance imaging

epidermoid cyst, pyogenic abscess, parasitic diseases involving CNS such as neurocysticercosis, cerebral toxoplasma abscesses.[7,34]

Both CT and MRI demonstrate a spherical, well-defined, smooth, thin-walled, homogeneous cystic lesion with a fluid density similar to the cerebrospinal fluid (CSF), with or without septations or calcification.[7] On unenhanced CT, the cyst wall is isodense or hyperdense to brain tissue.[7,11,19] The cyst wall usually shows a rim of low signal intensity on both T1- and T2-weighted images. Calcification of the wall is rare, being <1%.[7,12,19] The presence of daughter cysts is considered pathognomonic but has been rarely reported.[19,36] In this case, radiologist reported the presence of daughter cysts on MRI, one of the rare but pathognomic findings. Compression of the midline structures and ventricles are seen in most of the cases, however, surrounding edema, and rim enhancement are usually absent in untreated or uncomplicated cases.[36,47] Edema is not a feature of intracranial hydatid. When present, edema and postcontrast enhancement indicate ongoing inflammation.[24] The presence of significant edema may indicates rupture of the cyst and may be present in postoperative cases. Such cases are difficult to differentiate from other cystic lesions with enhancement and peripheral edema such as abscesses, large granulomas or cystic gliomas.[37]

In cerebral cystic echinococcosis, CT and MRI findings are more typical than in cerebral alveolar echinococcosis. On CT and MRI, cerebral alveolar echinococcosis lesion appears as a solid, semisolid, or multilocular cystic mass with definite margins. Calcification and surrounding edema are common. Contrast enhancement occurs within the region of inflammatory reaction around the cysts.[4,40,46] The advantages of CT and MRI are enumerated in Table 3.

Another, one of the recent modalities is MRS. It uses 1H signals to determine the relative concentrations of target brain metabolites. Few MRS studies are available to give comprehensive information on cerebral echinococcosis. Kohli *et al*. performed *in vivo* and *in vitro* studies in a patient of intracranial hydatid cyst. Besides lactate, alanine and acetate, a large resonance for pyruvate was observed. MRS pattern appeared different from the other cystic lesions of the brain, and they suggested MRS as an adjunct to imaging in the differential diagnosis of intracranial hydatid. The role of MRS in monitoring drug therapy was also highlighted.[27] In our patient, *in vivo* MRS study showed acetate and succinate peak.

Table 3: Advantages of CT and MRI

CT scan	MRI
The exact size and number of hydatid cysts in the brain can be determined	MRI provides additional information in the exact localisation of the cyst. It is better in detecting multiplicity, defining the anatomic relationship of the lesion with the adjacent structures and helps in surgical planning
CT is superior in detecting calcification of the cyst wall or septa	In complicated or recurrent disease, surrounding oedema can better be demonstrated owing to the inherent capability of the imaging modality in revealing subtle differences in the tissue content

CNS: Central nervous system, MRI: Magnetic resonance imaging

Other investigations such as USG whole abdomen and pelvis, echocardiography, X-ray chest were unremarkable. These investigations are necessary to differentiate primary and secondary hydatid cysts. Primary intracranial hydatid infection is caused by embryos passing hepatic and pulmonary barriers. These are the most common types and are solitary. Secondary cysts are usually multiple which may follow embolization of ruptured cardiac cyst or spontaneous, traumatic and surgical rupture of a primary cyst in other organs.[18]

Histopathology helps in confirming the diagnosis. Hydatid cyst (cystic echinococcosis) consists of three layers:

- Endocyst – in which is the inner germinal layer. Scolices, brood capsules, and daughter cysts originate from this layer by endo-proliferation or internal budding. To the contrary, a cyst of alveolar echinococcosis proliferates by external budding of the germinal membrane and progressively infiltrates the surrounding tissue
- Ectocyst - is the outer laminated layer
- Pericyst - signifies host reaction to the cyst. It is a fibrous capsule, which contains blood vessels that provide nutrients for the parasite.

Rapid decompression caused by evacuation of a large cyst may result in disturbances in autoregulatory mechanisms such as sudden decrease in pressure followed by cerebral edema, hyperpyrexia, cardiorespiratory failure, subdural collection, and the development of a porencephalic cyst which needs to be watched in postoperative period.[18]

Medical management involves the use of protoscolicidal agents such as praziquantel, albendazole. Praziquantel does not pass through the hydatid cyst wall, but high levels are seen in the CSF. As it has a powerful, lethal action on free scolices, it may be used in the event of spillage of the cyst fluids. Although protoscolicidal agents do not penetrate large hydatid cysts in sufficient quantity, highly soluble albendazole can used over a prolonged period for the treatment of small multiple cysts in inaccessible sites. A combination of praziquantel and albendazole is more effective than either drug used alone.[10]

CONCLUSION

Cerebral hydatid cysts although rare, can prove to be dangerous if not diagnosed and treated earlier. Slow growth rate and appearance of symptoms in later stage

of the disease adds to the morbidity and mortality. Therefore, clinical suspicion in cystic lesions of CNS is important for a correct diagnosis. Imaging techniques and histopathology aid neurosurgeon to reach a correct diagnosis and plan treatment accordingly. Preventing infection in humans depends on the education to improve hygiene and sanitation.

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