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An unusual case report on intracranial hydatid cyst with intraventricular extension

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Abstract

Background: We report an interesting case of a primary isolated intraventricular and intracranial hydatid cyst in 20-year-old female that mimicked bleeding cystic tumor presenting as intraventricular hemorrhage with hydrocephalus.

Case presentation: Patient presented with headache and giddiness for 1 month. On magnetic resonance imaging, there was multiloculated multi-cystic lesion in left lateral ventricle and adjacent left periventricular deep white matter. Lesion was causing compression of the body of the left lateral ventricle with temporal horn dilatation and midline shift of 17–18 mm toward right side. On investigation, it was diagnosed intracranial hydatid cyst with intraventricular extension. Patient underwent complete removal of cerebral hydatid cyst. Follow-up postoperative computed tomography scan done which revealed no residual hydatid cyst.

Conclusion: In this case, larvae of echinococcus might have passed through capillaries of liver and lungs and entered systemic circulation reaching brain. Intracerebral hydatid cysts are rare and hence, dangerous if the diagnosis and treatment is not prompt. Intracerebral hydatid cysts have slow growth rate with late-stage symptoms, morbidity and mortality are high. So, take home message is whenever intracranial cystic lesion are found on magnetic resonance imaging hydatid cyst with possible intracranial extension should be considered in differential diagnosis.

Keywords: Echinococcosis, Intraventricular hydatid cyst, Intracranial hydatid cyst

Introduction

Hydatid disease is a rare zoonotic disease caused by the larva of *Echinococcus* tapeworm. It is endemic in different parts of the world including India. Liver is the most often involved organ (75%), which is followed by lungs (15%) and spleen. It is very rare that the central nervous system is affected, in which case the cysts occupy involves the parenchyma and in extreme cases ventricles can be involved [1]. We present a case of primary isolated intraventricular and intracranial hydatid cyst in a female that mimicked a bleeding cystic tumor and presented as intraventricular hemorrhage with hydrocephalus. Very few cases of the same have been reported so far. Involvement of brain is more common in children and it has an intraparenchymal location [2]. Multiple lesions in the brain are even rarer. Majority of the intracranial hydatid cysts are solitary and are classified as primary and/or secondary. Primary intracranial hydatids are due to isolated infestation of larvae in the brain while secondary cysts are due to surgical rupture of the primary intracranial cysts or trauma. Complete surgical removal of primary hydatid cyst of the brain is often possible; however, complete removal is difficult [3].

Case presentation

A 20-year-old young female patient was presented in the emergency department of Acharaya Vinoba Bhave Rural Hospital, Sawangi (Meghe) with chief complains of headache and giddiness since 1 month which on radiological investigations turned out to be a case of

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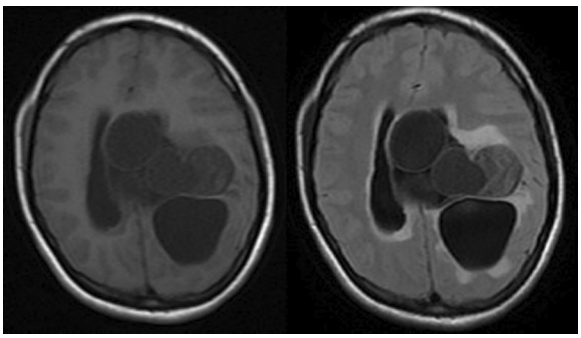


Fig. 1 Axial T1WI and FLAIR shows multiloculated multicystic lesion in left lateral ventricle and adjacent periventricular deep white matter. FLAIR shows perilesional edema

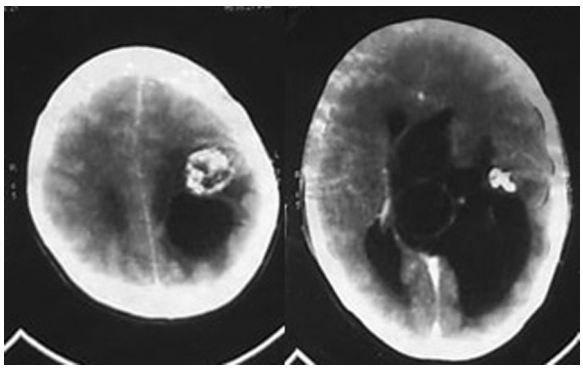


Fig. 3 Contrast-enhanced CT scan imaging showing heterogeneous cystic lesion with calcifications and multiple cystic lesions

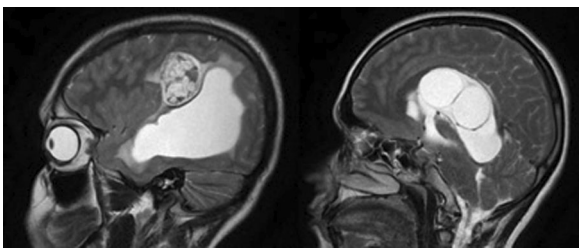


Fig. 2 T2WI Sag section shows multiple rounded cystic component which appears hyperintense. Typical crumpled appearance noted

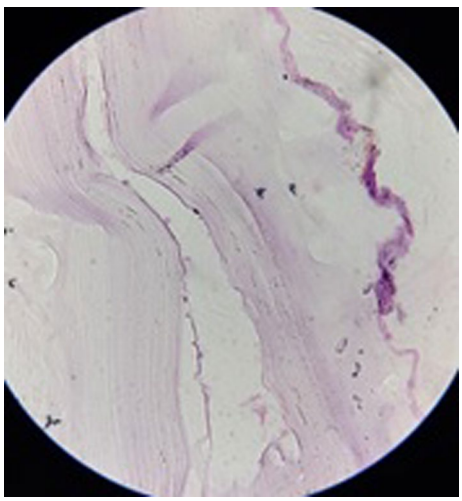
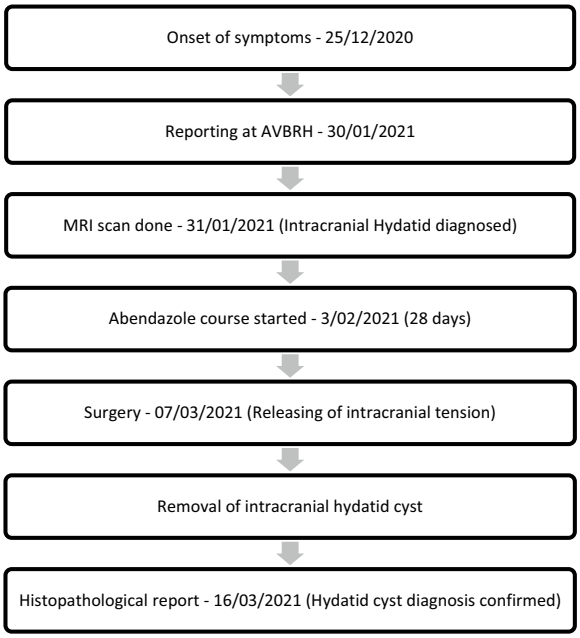


Fig. 4 Low-magnification photomicrograph showing lamellated fibrous and chitinous wall and inner germinal lining of hydatid wall

intracranial hydatid cyst with intraventricular extension. On careful history-taking from the informant (mother) and the patient herself, there was no loss of consciousness, seizure, nausea and vomiting. The vital signs were normal and on admission the patient's Glasgow Coma Score (GCS) was E4 V5 M6–15/15. Ultrasound scan of abdomen and pelvis and chest X-ray were normal. On magnetic resonance imaging (Figs. 1, 2), there were multiloculated multicystic lesions in left lateral ventricle and in the adjacent left periventricular deep white matter. The lesions had multiple rounded cystic components which were T2-hyperintense and T1-hypointense with T2-hypointense wall. On post-contrast study, the lesion showed mild septal and wall enhancement. The lesion was causing compression of body of lateral ventricle with temporal horn dilatation and associated significant rightward midline shift of 17–18 mm, perilesional edema and mild descending trans-tentorial herniation. Rest of the cerebral parenchyma showed normal gray white matter differentiation. NCCT scan revealed heterogenous cystic lesion with calcifications (Fig. 3). On physical examination, the pupils were reacting to light and cranial nerves were normal. The power in all four limbs was 5/5 with normal tone normal reflexes with no meningeal signs. There was no history of contact with pet animal and herds. On histopathology, the lesion was diagnosed as hydatid disease (Fig. 4). On magnetic resonance imaging, it was initially misdiagnosed as pilocytic astrocytoma as there were cystic lesions extending into the ventricles with few solid components. A course of albendazole was given to the patient before operation for intracranial tension reduction. After completion of the course of albendazole, the patient underwent total surgical removal of cerebral hydatid cyst. Post-operative computed tomography scan done for follow up revealed no residual hydatid cyst with no adverse or unanticipated events (Fig. 5).

A flowchart of timeline is given flowchart below.



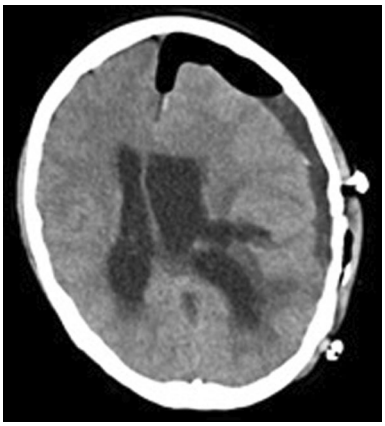


Fig. 5 Post-operative NCCT shows calvarial defect in left parietal bone with postoperative changes. No residual hydatid cyst

Discussion

Echinococcosis is a zoonotic disease caused by the larva of tapeworm Echinococcus. All four species of echinococcus documented in the literature are involved in the etiology of human infection namely—Echinococcus granulosus (cystic hydatid disease), Echinococcus multilocularis (alveolar hydatid disease), Echinococcus vogeli, and Echinococcus oligarthus [4]. In their reproductive cycle, humans are an accidental intermediate host. Liver (59–75%) is the most common site involved. This is followed by lung (27%), kidney (3%), bone (1–4%), and brain (1–2%) which is a least affected organ. Various distant unusual anatomic locations like the heart, spleen, pancreas, ovaries, parametrium, pelvis, omentum, thyroid, orbit or retroperitoneum, and muscles are also seen to be involved [3]. Definitive hosts are the carnivores, such as dogs or wolves where the adult worms inhabit in the proximal small intestine attached by the hooklets to the mucosa; whereas intermediate hosts are the herbivores such as sheep, cattle and goats housing cyst stage of the worm. Eggs of an adult tapeworm are passed in infected dog feces that is ingested by sheep while grazing. These cysts hatches into embryos in the intestine and penetrate the intestinal mucosal lining reaching the portal circulation. The embryos after reaching the host organ transform into larval echinococcal cysts in which numerous tapeworm heads (called protoscoleces) are produced via asexual reproduction. When dogs ingest the viscera containing echinococcal cysts the life cycle is completed. Humans are accidental intermediate hosts in the tapeworm life cycle [5]. Primary multiple hydatid cysts of the brain are extremely rare. Intra-cranial hydatid cysts do not show symptoms till they attain a large size. Large cerebral hydatid cysts interfere with the cerebrospinal fluid flow resulting in obstructive hydrocephalus. Headache and vomiting are the most common symptoms, whereas motor weakness and seizures can also be seen [6]. Computed tomography and magnetic resonance imaging shows well-defined smooth thin-walled spherical/oval cystic lesions with cystic component of fluid attenuation [7].

On MRI, T1-weighted and T2-weighted images show low signal intensity cyst wall. Wall calcification is rare and reported to be in < 1% of all cases. Cerebral abscesses and cystic tumors usually show surrounding oedema and peripheral rim enhancement. It is not seen in untreated or uncomplicated cases of cerebral hydatid cyst irrespective of mass effect [8]. Magnetic resonance spectroscopy and diffusion weighted imaging are used to distinguish cerebral hydatid cysts from cerebral abscesses, necrotic brain tumors including cystic lesions [9]. Generally, acetate and succinate metabolic end products are known for arising from microorganisms [10].

But, metabolic end products for hydatid cysts are non-specific and are similar as in case of abscesses and neurocysticercoses [11]. Here, in our case the magnetic resonance spectroscopy of fluid filled cystic part of lesion shows elevated choline/NAA ratio with elevated choline and pyruvate levels and the peripheral solid part shows peak of lipids.

Intra-operative cyst rupture may occur in 28% of the cases. If it ruptures into subarachnoid space of the brain, it can cause severe anaphylactic reactions.

Usually, albendazole is given in cases of recurrence, intraoperative rupture of the cysts, inoperable cases with widespread disease and is recommended before operation for intracranial tension reduction [12, 13].

Conclusion

Echinococcosis is common disease endemic in central belt of India, usually involving the liver and lungs. Intracerebral hydatid cysts being rare, it can be dangerous if not diagnosed and treated accordingly. Intra-cerebral hydatid cysts have slow growth rate and symptomatic in later stages causing high morbidity and mortality. Imaging modalities like computed tomography and magnetic resonance imaging supports in establishing diagnosis of hydatid disease, but if unusual anatomic sites with atypical imaging findings present themselves; one may have to consider differential diagnosis of cystic masses irrespective of site/location. Imaging modalities like computed tomography and magnetic resonance imaging techniques along with histopathology help the neurosurgeon to diagnose the disease correctly and further plan the treatment accordingly. A wide variety of metabolites in human brain can be examined by using non-invasive method like proton magnetic resonance spectroscopy as an additional modality to identify the nature of cerebral lesions including hydatid cysts.

Abbreviations

MRI : Magnetic resonance imaging; T1WI: T1-weighted image; T2WI: T2-weighted image; NAA: N-acetyl acetate; NCCT: Non-contrast Computed Tomography.

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Authors' contributions

RKS: designing, concept and major contribution in manuscript preparation. PAP: designing, concept, manuscript corrections, editing and reviewing. GVM: analyzed and interpreted the patient data. RPD: designing and concept. SVK: designing and concept. BSU: designing and concept. All authors read and approved the final manuscript.

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Availability of data and materials

The data is collection from the hospital system with the permission of the competent authority. The identity of the patient is not compromised at any place.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Informed consent was taken from participant(s) prior to their inclusion in this study.

Competing interests

The authors declare that they have no competing interests.

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