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Intracranial Space Occupying Lesion with Hydatid Cyst: A Rare Infestation of Hydatid Isolate from Intracranial Cystic Lesion

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

The present case report aimed to bring into light a rare infestation of hydatid isolate from intracranial cystic lesion which was misdiagnosed as intracranial neurocysticercosis infestation at a tertiary care centre in northern India. A 16 year old female, resident of Uttar Pradesh presented to the Neurosurgery outpatient department (OPD) with a 3 months history of holocranial headache and generalised tonic clonic seizures for the past 1 year. Causes of intracranial cystic lesions other that hydatid cysts can either be abscesses or calcified neurocysticercosis or cystic and necrotic malignancies. In our case, a precoronal keyhole craniotomy and total excision of cyst followed by repeated cleaning with 3% hypertonic saline. Due to the deceiving appearance of cystic echinococcosis in the brain on CT scan, which appears as an image similar to a single cystic neurocysticercosis lesion with ring enhancement, wet mount preparation and microbiological tests are equally critical in the early diagnosis of the disease.

Keywords: Neurocysticercosis; intracranial cystic lesion; precoronal keyhole craniotomy; Echinococcous granulosus.

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1. INTRODUCTION

Echinococcous granulosus is a tapeworm causing human echinococcosis, which forms larval cysts in the human tissue. The most common definite hosts of echinococcus are carnivore animals like dogs [1]. It is a widespread zoonotic disease, where the cysts usually involve the liver but can also involve other organs like the lungs and brain [2, 3]. The hydatid cysts are isolated in the liver in 60% cases, while those in the cerebral and spinal forms are only 1–2% [4]. *E. granulosus* is frequent in southeastern region of Rajasthan in India [5]. The most common site of involvement of brain by the cyst occurs through the choroid plexus [2,6,7]. The neurohydatosis can be associated with involvement of other organs like liver or lung or it may be an isolated infection of the brain or spinal cord. In cerebral hydatid disease, supratentorial location of cystic lesions occurs in 50% to 75% of cases, while the infratentorial lesions are rare for which ultimate treatment is surgical excision [8, 9]. The cysts should be removed intact by surgical excision as spillage of contents may lead to anaphylactic shock and recurrence of hydatid disease [10]. Albendazole is administered preoperatively and postoperatively to sterilize the cyst as it decreases the tension in the cyst wall leading to reduced chances of spillage of cyst contents during surgery thus reducing the risk of anaphylaxis and recurrence [11]. In this case study we highlight a case of intracranial hydatid cyst and its diagnosis which was confused with a diagnosis of intracranial neurocysticercosis.

1.1 Aims

The aim of our case report is to bring into light a rare infestation of hydatid isolate from intracranial cystic lesion which was misdiagnosed as intracranial neurocysticercosis infestation at a tertiary care centre in northern India.

2. PRESENTATION OF CASE

A 16 year old female, resident of Uttar Pradesh presented to the Neurosurgery outpatient department (OPD) with a 3 months history of holocranial headache and generalised tonic clonic seizures for the past 1 year. On general examination, all her vitals were within normal limits, no facial asymmetry or hypoesthesia was observed and her lower cranial nerves were also within normal limits. On motor examination, tone was normal in all four limbs and power 5/5 in all the four limbs. Plantar sign showed flexion, sensory system, posterior column, and cerebellar signs were intact. Her tandem gait and lobar sign were impaired. She was advised for a Contrast enhanced Magnetic resonance imaging (CEMRI) and the results showed a large cystic space occupying lesion in left frontal region of size 4x4 cm intra-axial T1 hypointense and T2 hyperintense with contrast enhacement without any diffusion restriction (Fig 1). The lesion had a mass effect with ipsilateral effacement of lateral ventricle with subfalcine herniation. To diagnose involvement of other organs, ultrasound and computed tomography of the abdomen and high resolution computed tomography (HRCT) chest were performed on which no cystic lesions were found. The clinicians considered it as a case of Left frontal intraparenchymal single neurocysticercosis with mass effect. She was scheduled for a precoronal keyhole craniotomy and total excision of cyst on 4th July 2019. Cyst was thick translucent containing clear content. Cyst decompressed and cyst wall excised from wall of parenchyma. Postoperatively patient had improvement in headache. Rest of the post operative recovery was uneventful. Sutures were removed on 14th postoperative day and the patient was discharged on Albendazole 400 mg BD for two weeks. Patient was discharged with advice to follow up in Neurosurgery OPD.

After one month of surgery, she was suggested for a repeat Contrast enhanced computed tomography which suggested of craniotomy defect with a cystic mass lesion with contrast enhancing ring like lesion with central necrotic material and perilesional edema with mass effect (Fig 2). She was reoperated on 28th August 2019 due to repeated cystic lesion and mass effect with midline shift. Abscess cavity was decompressed and abscess was aspirated, which was thick and yellowish, there were few cysts in the cavity. Fluid was yellowish and thick. The cyst fluid and abscess pus were aspirated and sent to the Parasitology section of Department of Microbiology, of a tertiary care centre in northern India. A wet mount of the pus was prepared and observed under the 10x followed by 40x objective lens of the microscope shows few pus cells and plenty of hooklets and scolices of *Echinococcus granulosus* (Hydatid cyst) (Fig 3). No cystic lesions were found on repeat ultrasound and computed tomography of the abdomen and high resolution computed tomography (HRCT) chest, deeming it as a case of primary neurohydatosis. On histopathology of the frozen section, brain parenchyma was

observed with few scolices and hooklets, the parenchyma shows mixed inflammatory cell infiltrates comprising of sheets of foamy histiocytes, lymphocytes, eosinophils with no evidence of malignancy. No motor or sensory weakness was observed on follow up examinations. Ultrasonography findings of the whole abdomen including the pelvis and chest X-ray were within normal limits. MRI brain and spinal cord was repeated after 3 months, which showed craniotomy defect with no new lesion.

The patient underwent a left precoronal keyhole craniotomy and total excision of cyst with drainage of thick and yellow coloured pus followed by irrigation of the cavity with hypertonic saline (3%). The patient had an uneventful recovery from the surgery and was asked to continue Albendazole 400 mg BD for the next three months.

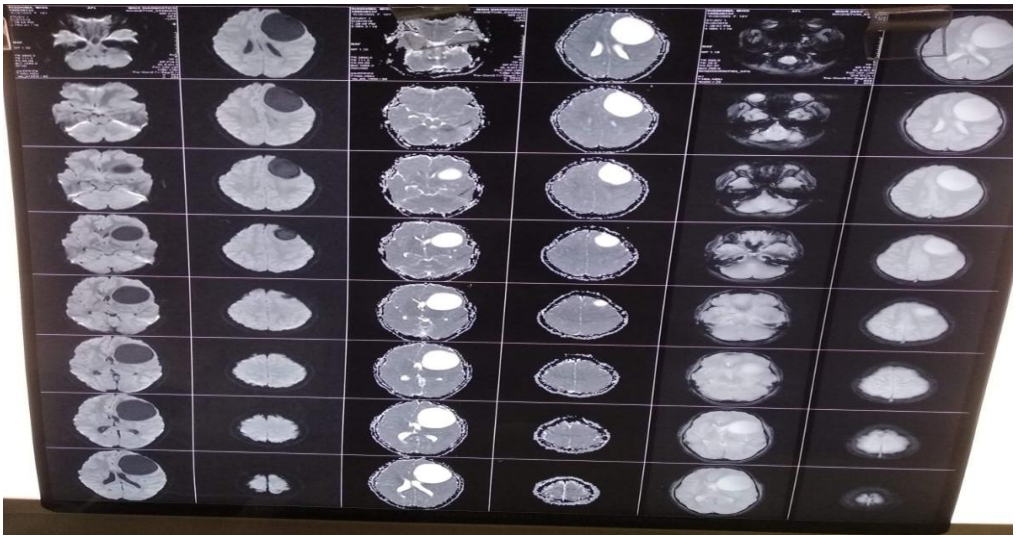

Fig. 1. Preoperative Contrast enhanced magnetic resonance imaging of the space occupying lesion in the frontal lobe

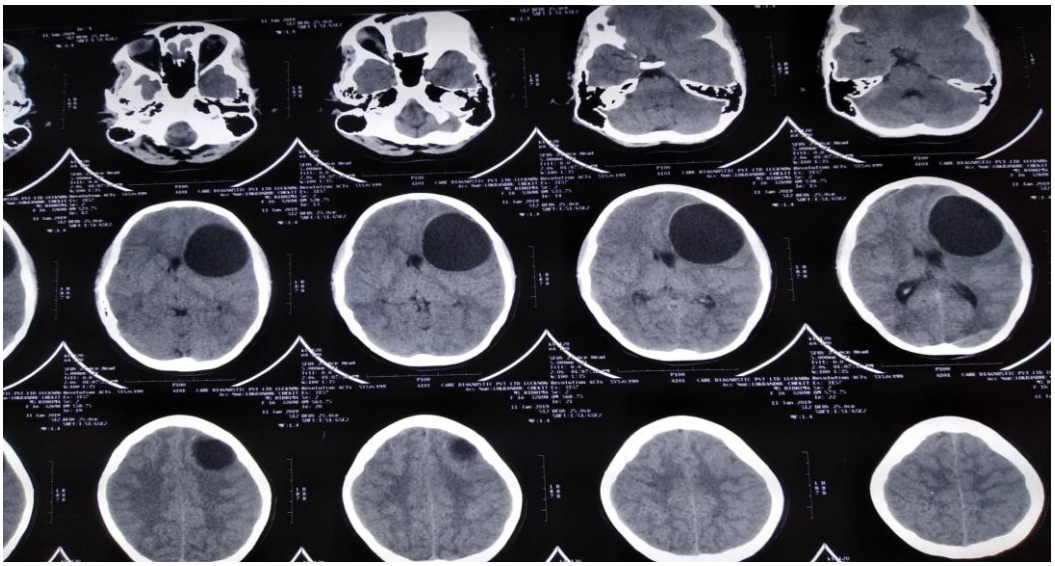

Fig. 2. Postoperative Contrast enhanced computed tomography image of space occupying lesion in the frontal lobe

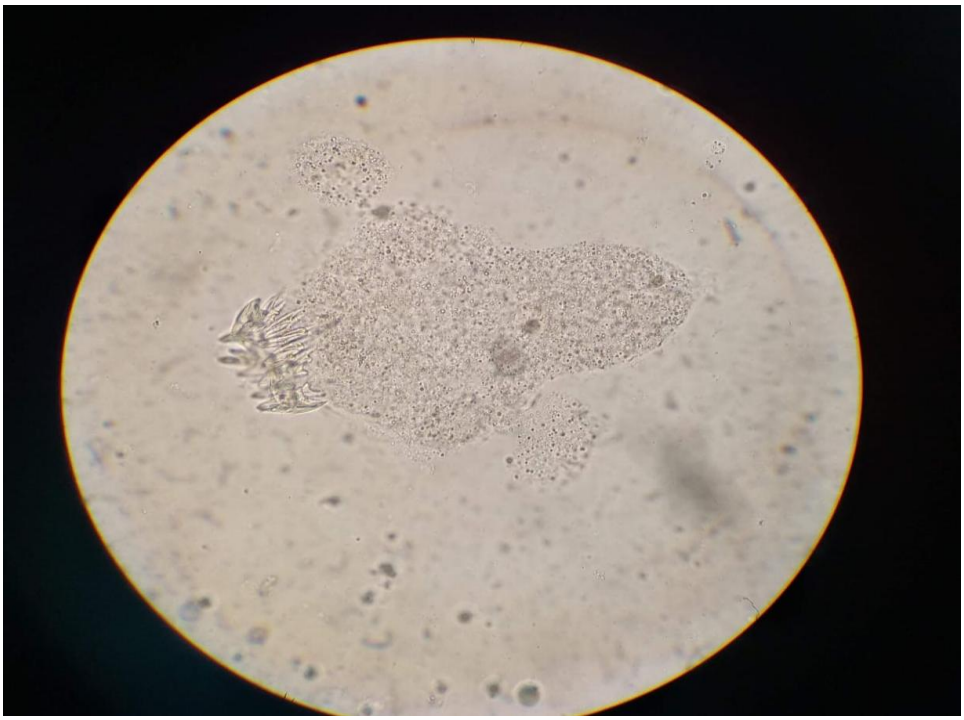

Fig. 3. Wet mount of the pus was prepared and observed under the 10x followed by 40x objective lens of the microscope shows few pus cells and plenty of hooklets and scolices of *Echinococcus granulosus*

Fig. 1 and 2 show a thick walled cystic lesion measuring 4x4 cm in the left frontal lobe which is subcalverial in location. The lesion is causing buckling of the underlying parenchyma. Mild perilesional edema is noted causing mass effect in the form of effacement of left frontal horn of lateral ventricle, left basal ganglia along with midline shift suggestive of Hydatid cyst.

3. DISCUSSION

The only pathogenic species causing human hydatid disease is *E. granulosus*. Infection in humans is caused by ingestion of food or drink contaminated with fecal material of carnivorous animals like dogs that are infected and pass stool containing tapeworm eggs [1]. India being an endemic region for hydatid disease, children in our country are thought to be infected by consuming food contaminated with eggs. Cases of intracranial hydatid disease are more common in children who form 50% to 75% of patient group. Izci *et al* [10] worked with 17 patients of intracranial hydatid disease, out of which 13 (65%) patients were children. Supratentorial hydatid cysts are often observed in case of cerebral hydatid cysts and are distributed along the terminal branches of the middle cerebral artery, usually temporo-parieto-occipitally [11]. In our case, patients also had cysts in the supratentorial location and in the left frontal lobe of the brain.

No symptoms of cerebral hydatid cysts are observed until they are considerably large. The patient usually presents with focal neurological deficits and signs of raised intracranial pressure owing to the size of the cyst and its tendency to interfere with the flow of the cerebrospinal fluid. Headache, vomiting, motor weakness and seizures are the chief presenting complains in most patients [12]. In our case the patient presented with generalised tonic clonic seizure and her tandem gait and lobar sign were impaired.

Causes of intracranial cystic lesions other that hydatid cysts can either be abscesses or calcified neurocysticercosis or cystic and necrotic malignancies. There was immense diagnostic dilemma among the clinicians on the first contrast enhanced magnetic resonance imaging film before the first keyhole craniotomy, due to a large ring enhancing space occupying lesion in the left frontal lobe. It was proved to be *E. granulosus* on wet mount prepared from the pus aspirate of the abscess cavity. Therefore microscopic examination of the cystic fluid or

abscess aspirate holds almost equal importance as the radiologic imaging techniques. Radical surgical excision is the primary treatment of intracranial hydatid cysts. Older techniques in the 1960s include injecting formalin and evacuation of the cyst [12]. The gold standard method for surgical removal of the cyst has been the Dowling technique. The operative site should be repeatedly cleaned with a solution of 3% NaCl or 10% formaldehyde in case of accidental spillage of contents. Intraoperative rupture of cysts, cases of recurrences and inoperable cases of multifocal disease are treated using albendazole [13]. In our case, a precoronal keyhole craniotomy and total excision of cyst followed by repeated cleaning with 3% hypertonic saline. Albendazole or mebendazole therapy are used for preoperative volume reduction of cysts, multiple organ involvement and brain lesions, intraoperative rupture of cysts and postoperative cases of recurrence [14,15].

4. CONCLUSION

Neurohydatosis is a rare cause of cystic lesions in the brain in non-endemic regions of India. The diagnosis of cystic echinococcosis in brain is very difficult due to misleading appearance on CT scan which presents as an image similar to the single cystic neurocysticercosis lesion with ring enhancement and thus the role of wet mount preparation and microbiological tests is equally important in early diagnosis of the disease.

CONSENT

As per international standard, parental written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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