CASE REPORT

Recurrent multiple primary intracranial hydatid cysts: a case report

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Abstract

Brain hydatid cysts are rare and mostly occur as solitary supratentorial cysts in children. Multiple primary intracranial hydatid cysts are even rarer. A 26-year-old cattle farmer, diagnosed as a case of brain hydatidosis, had undergone multiple surgeries. He presented with headache and right hemiparesis, and was re-diagnosed with recurrent hydatid disease. He underwent surgery using Dowling technique and was discharged on tablet Albendazole. Brain hydatidosis should be ruled out in patients who present with raised intracranial pressure symptoms in endemic regions. Recurrent disease may require multiple surgeries and long-term treatment and follow-up.

Keywords: Hydatid cyst, Primary, Multiple, Echinococcus, Intracranial.

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Introduction

Hydatid disease in humans is a zoonotic, parasitic infection caused by cestode species of the genus Echinococcus.[1] It is endemic in regions where cattle farming is extensively practised, such as Mediterranean countries, Central Asia, the Middle East, South America, and Australia.[2]

The definitive hosts are dogs, wolves, and foxes, while intermediate hosts are sheep, cattle, rodents, and humans.[1,3] Cerebral hydatid cysts are rare (2%) and occur mostly in children as single cysts supratentorially; rarely, they may occur in infratentorial distribution.[3,4] Surgery using Dowling technique is the treatment of choice.[2] The use of pre- and post-operative Albendazole decreases the recurrence of the disease.[2] We present a unique case of a patient with recurrent primary intracranial brain hydatidosis, treated by surgery and Albendazole therapy.

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Case Report

A 26-year-old male from a rural area presented to the outpatient department of Naimat Begum Hamdard University Hospital, Karachi, on September 7, 2023, with complaints of generalised headache for five months and progressive right hemiparesis for three months. Nine years ago, he was diagnosed with hydatid brain disease, as he had developed paraparesis, urinary incontinence, vision blurring, and deafness over two years. From 2016 to 2017, he underwent four craniotomies for the removal of recurring hydatid cysts causing pressure symptoms. After the last surgery, he remained symptom-free for seven years on Albendazole. In social history, the patient was a farmer by profession with frequent exposure to cattle and dogs. He lived in suboptimal sanitary conditions.

Examination showed previous craniotomy scars in the right frontal, right occipital, and left frontal regions of the head. The patient was oriented to time, place, and person. The tone of the right arm and leg was increased, power was 3/5 on the right upper and lower limbs, reflexes were diminished and he could not stand without assistance. Fundoscopy revealed bilateral papilledoema. Cranial nerves and left limb motor examination was unremarkable. CT of the abdomen with contrast revealed no intra-abdominal hydatid cysts.

MRI showed multiple intra-axial cysts with internal daughter cysts and a 3×3.3×4 cm lesion in the right parieto-temporal lobe causing mass effect, vasogenic oedema, midline shift, and ventricular effacement (Figure 1). Removal of the right parieto-occipital cyst and surrounding cysts was performed using Dowling's technique and hydrodissection, avoiding rupture. Post-operatively, he was discharged on Albendazole 400mg, twice daily. Histopathology confirmed hydatid cyst. On one month follow-up, it was observed that the right-sided weakness had improved, but the patient had developed aphasia and left-sided hemiparesis. The patient was subsequently lost to follow-up.

Discussion

Two clinically important species cause hydatid disease, Echinococcus granulosus and Echinococcus multilocularis. The former has a more worldwide distribution and causes solitary cysts, whereas the latter

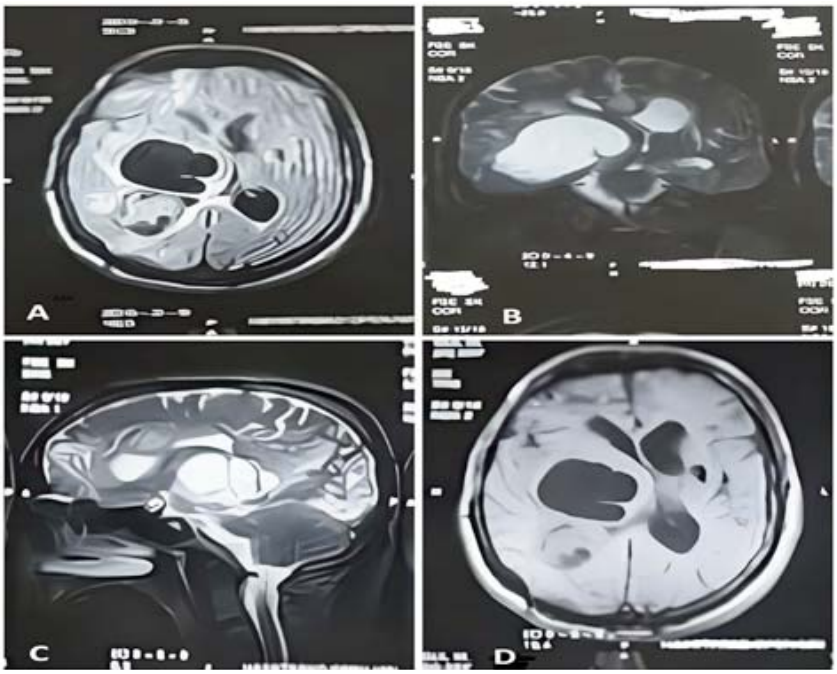

Figure-1: Preoperative magnetic resonance imaging showing multiple intracranial cystic lesions; (A) axial, flair MRI sequence; (B&C) T2-weighted, coronal and sagittal sections and (D) T1-weighted, contrast enhanced axial sequence.

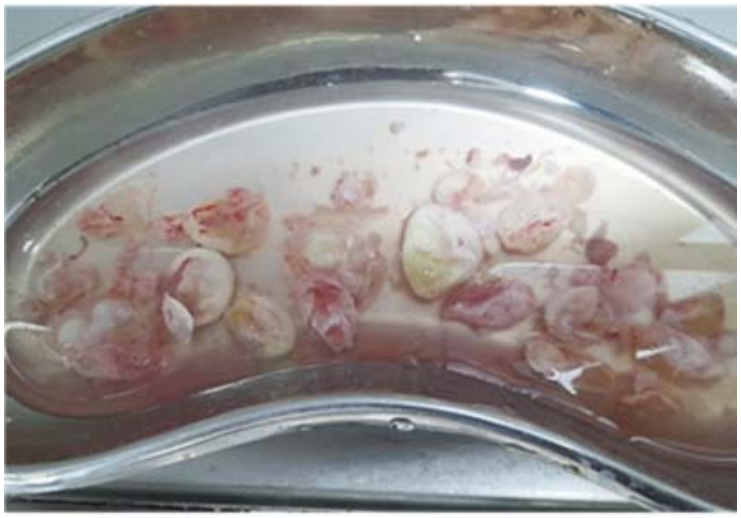

Figure-2: Macroscopic findings of multiple cysts extracted after surgery.

which is mostly seen in North America, Northern and Central Eurasia, causes multiple cysts, has a tendency to disseminate to distant organs, and can be fatal.[1] Only 42% of the patients infected with E. multilocularis have a good long-term prognosis.[4]

Brain cysts are rare (2%) and usually seen in a supratentorial distribution, mostly in hemispheres.[3,4] Rarely, they may present in infratentorial regions such as the brainstem, basal cisterns, and ventricles.[3,4] In the present case, multiple cysts were dispersed bilaterally, as well as infratentorially. Garg et al. discussed the possible aetiology for multiple cysts, including ingestion of multiple parasitic eggs causing multiple primary cysts, rupture of primary cysts, dissemination from other organs resulting in capillary invasion, and pre-existing left ventricular pathology.[5]

The patient usually presents with raised ICP symptoms like focal neurological deficit, headache, nausea, vomiting, papilloedema, and impaired level of consciousness, while seizures are rarely seen.[3] Gautam et al. reported three cases of hydatid cysts of the brain, which showed hemiparesis common among all.[6] Izci et al. conducted a study on 17 patients with brain hydatid cysts, showing headache as the most common symptom followed by weakness in the extremities.[2] This was relevant to this patient as well.

Brain hydatid cysts grow slowly, about 1cm/year, so symptoms occur late in the course of the disease.3 Akrim et al. reported patients with multiple cerebral hydatid cysts experiencing recurrence of symptoms seven years after the excision of the primary cyst.[7] The current patient had a similar presentation.

Garg et al. reported a case of multiple bilateral primary hydatid cysts in which the patient had eosinophilia, and enzyme-linked immunosorbent assay for granulosus antibody was positive.[5] CBC commonly shows eosinophilia.[3] The current patient had no eosinophilia, and serological testing could not be performed as it was unavailable in this institute. He did not demonstrate extra-cerebral cysts, and histopathology showed cysts with a germinal layer and protoscoleces, indicating that they were primary. Secondary cysts are infertile, lack a germinal layer, and are formed by primary cysts when they rupture through surgery, trauma, or spontaneously.[7] Multiple primary cerebral hydatid cysts are uncommon.[5,7]

The diagnostic investigations include CT and MRI, the latter being superior.[3,6] MRI of the brain with contrast in the present patient showed multiple thin-walled cysts, containing daughter cysts within, in various brain regions. Large ones caused compression and extensive oedema.

The most effective procedure for the removal of brain hydatid cyst is Dowling technique, improved by Arana-Iniguez and San Julian.[2] Izci et al. demonstrated that among patients undergoing Dowling's technique, 15 out of 17 had no cyst rupture, recurrence was seen in two patients, and only three had subdural effusions with one haemorrhage[2] The current patient underwent Dowling technique without any gross cyst rupture or anaphylaxis.

Literature shows that in cases of multiple deep-seated hydatid cysts, larger cysts should be excised first to decrease chances of rupture, and staged surgery could be an alternative.[2]

Albendazole is indicated in inoperable multifocal disease or cysts in vital brain structures.[8] Its pre- and post-operative use reduces the chance of anaphylaxis and content spillage by decreasing cyst wall tension and recurrence of the disease.[2] It leads to cyst disappearance in 48% cases and substantial reduction in another 28%.[4] Kalaitzoglou et al. reported a case of inoperable multiple brain hydatid cysts with complete cyst resolution on Albendazole without recurrence.[8] Praziquantel increases Albendazole serum concentration fourfold and can be used concurrently.[4] Despite being young, immunocompetent, and adherent to Albendazole therapy for four years, the current patient experienced disease recurrence in the absence of any extra-cerebral cysts. This raises the possibility of treatment-resistant cysts, subtherapeutic drug penetration into deep-seated lesions, or the presence of dormant intracerebral cysts not detected on initial imaging. Concomitant Praziquantel may be considered to enhance antiparasitic efficacy.

Conclusion

Multiple primary intracranial hydatidosis is rare but should be ruled out in endemic areas. The mainstay of treatment is surgery; however, in multiple recurrences with multiple cysts, Albendazole along with Praziquantel should be considered with or without surgery.

Consent for publication: Verbal consent was taken from patient for publishing this case report and to protect the identity we have not included any personal details of the patient.

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Conflict of Interest: The head of the department of the hospital where the patient was treated is also the co-author of this article..

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AUTHOR'S CONTRIBUTION:
SH: Data collection, writing, reviewing, drafting and final approval.
HR: Data collection, writing, editing and final approval.
MB: Writing, reviewing, editing and final approval.
SAQ: Supervision, writing, validation, editing and final approval.
RR: Primary neurosurgeon managing the case, concept, reviewing and final approval.