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Case Report

Giant cerebral hydatid cyst with intraventricular extension: A rare case report with imaging findings ☆,☆☆

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ABSTRACT

Hydatid cyst is the condition caused by larvae of the parasite Echinococcus granulosus, usually involving the liver, lung, and spleen. Involvement of the cerebrum with a hydatid cyst is a rare entity, comprising 2%-3% of all cases of hydatidosis. Intraventricular extension of cerebral hydatid cysts occurs in only a limited percentage of cases. Cerebral hydatid cysts can present with nonspecific symptoms, usually with features of raised intracranial pressure. Therefore, it is crucial to consider cerebral hydatid cysts as a potential diagnosis, especially in pediatric patients with nonspecific neurological symptoms, particularly those from areas endemic to the parasite. We report a case of a 9-year-old girl who presented with progressive headache and vomiting, with imaging findings suggestive of a giant hydatid cyst in the brain. She was managed surgically with a good outcome.

Introduction

Hydatid cysts are the cystic lesions seen in human echinococcosis, also known as hydatidosis or hydatid disease. This is endemic in Nepal and caused by the ingestion of larval forms of cestode *Echinococcus spp.* especially *E. granulosus*. This disease most commonly involves liver followed by lung and spleen [1,2]. Intracranial hydatid cyst comprises 2-3% of all hydatid cysts. The disease remains asymptomatic for long duration,

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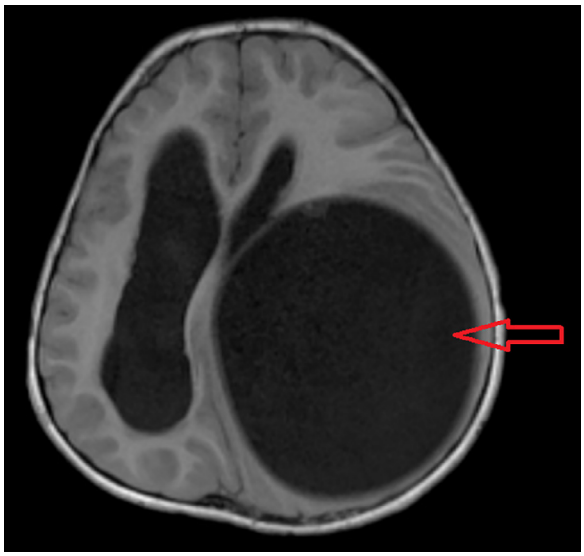

Fig. 1 – T1-weighted MRI Brain axial plane image showing hypo intense cystic lesion in left occipital parietal lobe.

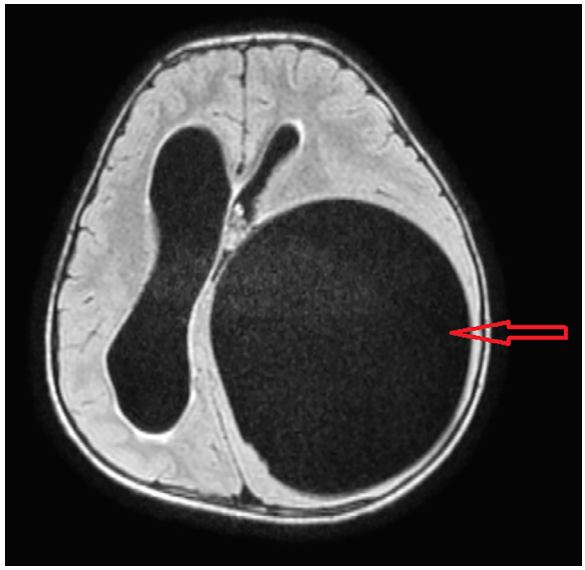

Fig. 2 – FLAIR MRI Brain axial plane image showing cystic lesion with mass effect without surrounding edema.

until the cyst grows large enough to become symptomatic [1]. Intracranial hydatid cyst may grow up 1-10cm per year [3]. Patient presents with nonspecific symptoms, usually with features of high intracranial pressure like headache, vomiting, altered mental status, rarely seizures and hydrocephalus [4]. Most common complications of brain hydatid cysts are rupture of the cyst and anaphylactic reaction [3]. Imaging technique like computed tomography and/or magnetic resonance imaging of brain are reliable methods for diagnosis. For definitive diagnosis histopathological examination can be done [5].

Due to paucity of literatures and rarity of the case, we are describing our experience with giant hydatid cyst with intraventricular extension in a nine-year-female, who was managed surgically without intra or postoperative complications.

Case presentation

A 9-year-old girl was referred to our center for evaluation of headache. She had been complaining of headaches for the last 15 months. The headache occurs more in the occipital area, without radiation, is of the on-and-off type, progressively increasing in severity, more pronounced during the morning, and relieved by taking analgesics, without specific aggravating factors. The headache is associated with multiple episodes of nonprojectile vomiting without bile and blood mixed. Her headache increased in intensity over the last one and a half months. There was no history of loss of consciousness, abnormal body movements, fever, weakness of limbs, or weight loss. She has a normal bowel habit and bladder function. She represents a lower-income family from a rural area of Nepal.

On examination, she appeared well and was oriented to time, place, and person. Her Glasgow Coma Scale rating was 15/15. Her vitals were as follows: temperature 98.7 degrees Fahrenheit, blood pressure 90/60 mmHg, pulse rate 96 beats per minute, and respiratory rate 16 breaths per minute. Her general physical examination and systemic examination revealed no abnormalities. Motor and sensory examinations were normal.

On investigations, her hemoglobin was 13.3 g/dl (Ref. 11.2-14.5 g/dl), WBC count reported 7,930/mm^3 ([Ref. 3, 800-10,400/mm^3), and platelet counts were 258,000/mm^3 ([Ref. 1,53,000-3,61,000/mm^3). Her renal function tests, liver function tests, serological tests for HIV and Hepatitis B, chest x-ray, ultrasonography of the abdomen and pelvis, and urine analysis were normal. Magnetic resonance imaging was performed to rule out intracranial causes, which showed a large, well-circumscribed, round, thin-walled, nonenhancing cystic lesion of cerebrospinal fluid attenuation in all sequences, measuring 11.2×9.6×10.8 cm with no surrounding edema in the left occipital-parietal lobe. This cystic lesion was causing mass effect as evident by compression of the frontal horn, body, and occipital horn of the left lateral ventricle and dilatation of the right lateral ventricle and temporal horn of the left lateral ventricle with contralateral midline shift of 14mm (Figs. 1 and 2). Additionally, there were multiple T2 hypo intense linear membranes within the cyst with crenated margins of the cyst wall at places (Fig. 3) Similar membranes and multiple daughter cysts were also noted in bilateral lateral ventricles (Fig. 4). There were no diffusion restrictions and no evidence of calcification or enhancing components or hemorrhage on imaging.

Based on these imaging findings, a diagnosis of cerebral hydatid cyst with intraventricular extension was made, and she was admitted to the neurosurgical ward for surgical removal of the cyst. Parietal-occipital craniotomy with dissection of the dura mater was performed, and using the Dowling-Orlando technique, a cyst measuring 15×15×7 cm in size was delivered. (Fig. 5) After cyst removal, the ventricles were irrigated, and a ventricular drain was inserted. A postoperative CT scan showed a large space without residual matter. Postoperatively, ant seizure medication (levetiracetam 500 mg twice a day) was

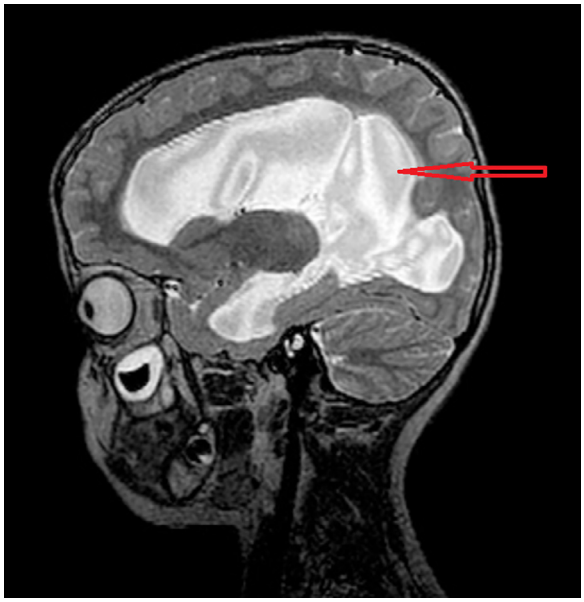

Fig. 3 – T2-weighted MRI Brain sagittal plane image showing hypo intense linear membranes within the cyst.

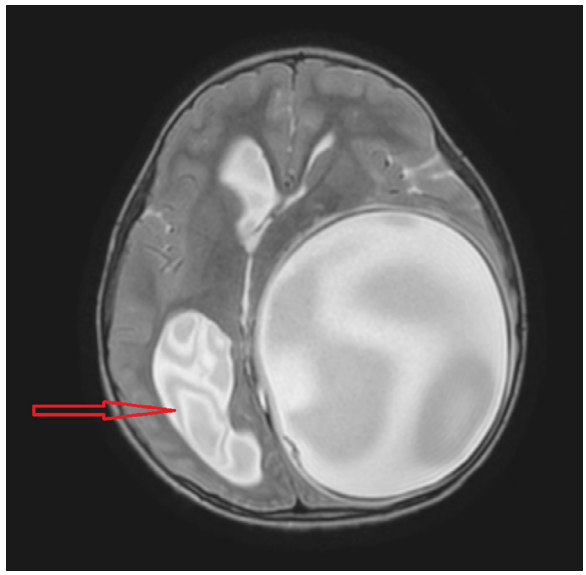

Fig. 4 – T2-weighted MRI Brain axial plane image showing daughter cysts in lateral ventricles.

prescribed for the prevention of seizures. Additionally, treatment with albendazole (15 mg/kg) was started and continued for 1 month for the prevention of recurrence of the hydatid cyst with routine monitoring of blood counts and liver function tests (in view of the potential hepatic and hematological adverse effects of albendazole). The patient showed marked improvement in headaches and was discharged after 15 days of hospital stay with close follow-up.

Discussion

Hydatid cyst is an emerging zoonotic parasitic disease worldwide, caused by the larval stage of *Echinococcus granulosus* [6]. There are four species of Echinococcus infecting humans: *Echinococcus granulosus, Echinococcus multilocularis, Echinococcus vogeli, and Echinococcus oligarthrus*. The definitive hosts of this tapeworm are carnivores, commonly dogs, and intermediate

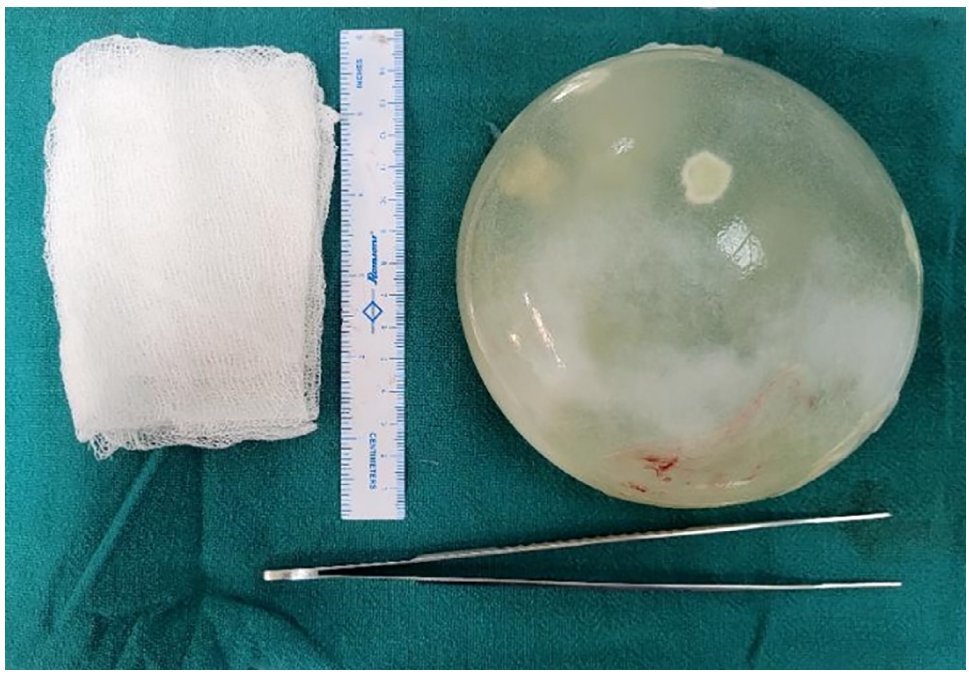

Fig. 5 – Figure showing gross hydatid cyst delivered after craniotomy.

hosts like cattle, goats, pigs, and humans become infected by ingesting definitive host feces [7]. After entering the intestine, eggs hatch and penetrate the wall. From there, they enter the circulation and reach the liver where they may form cysts or move towards the lungs. Some cysts are able to reach systemic circulation even after filtering through the pulmonary circulation and lodge into different organs such as the brain, heart, kidneys, orbit, spinal cord, spleen, spermatic cord, and soft tissues [6].

Hydatid disease is globally distributed and more prevalent in regions such as the Mediterranean, Russia, China, North and East Africa, Australia, Central Asia, and South America [7]. The highest prevalence occurs in rural areas [8]. Our patients come from rural areas of Asia, where contact with dogs is a risk factor for the disease. Therefore, in patients from endemic areas where contact with dogs is common, hydatid disease should be considered as a differential diagnosis in the evaluation of headaches and cystic lesions on the brain in imaging.

Hydatid disease commonly involves the liver, followed by the lungs, abdominal cavity, and brain [9]. Intracranial hydatid cysts comprise 2-3% of all hydatid cysts, with 1-2% of all intracranial lesions being hydatid cysts [10]. Mostly, cysts are supra tentorial in the territory of the middle cerebral artery [4]. Intraventricular extension of cerebral hydatid cysts occurs in a limited percentage of cases, with a limited number of reported cases to date [11]. Additionally, hydatid cysts are more common in children and young adults [6]. The presented case involved the supra tentorial region with ventricular extension in a child.

Hydatid cysts in the brain can be classified as primary and secondary. Primary cysts are solitary and due to direct infestation of larvae in the brain, whereas secondary cysts are multiple and due to the rupture of primary cysts during surgery, trauma, or spontaneously [12]. Secondary cysts are thought to arise from the scoleces released from the left side of the heart due to cyst rupture in the heart [6]. Primary cysts are fertile, whereas secondary cysts are infertile. In our case, the cysts were identified only in the brain without involvement of other organs, so they are of the primary type.

Usually, patients with hydatid cysts are asymptomatic as cysts take a longer time to grow. Symptoms depend on the area of the brain compressed by the cyst. Typically, patients present with headache, vomiting, papilledema, neurological deficits, and seizures [13]. Our patient presented with features of increased intracranial pressure such as headache and vomiting. Therefore, a space-occupying lesion should be considered in the differentials in pediatric patients with features of increased intracranial pressure.

Imaging techniques are essential for the proper diagnosis and management of hydatid cysts. CT and MRI imaging show well-defined, smooth, thin-walled, spherical, homogeneous cystic lesions with no contrast enhancement, no calcification, and no perilesional edema. CT scans are superior in detecting calcification, whereas MRI is better in detecting cyst capsule, multiplicity, and anatomic relationship of the lesion with adjacent structures [14]. . Perilesional edema and calcification are usually seen in differentials like cerebral abscess and cystic neoplastic lesions, which are rarely reported in hydatid disease [15]. In our case, which showed a large, well-circumscribed, round, thin-walled, nonenhancing cystic lesion of cerebrospinal fluid attenuation in all sequences measuring 11.2×9.6×10.8 cm with no surrounding edema in the left occipital-parietal lobe, helped us to make the diagnosis and rule out differentials. Multiple daughter cysts were also noted in bilateral lateral ventricles. The presence of daughter cysts in MRI is one of the rare but pathognomonic features [5].

Surgical removal of the cyst without rupture is the gold standard treatment method, although medical treatment with two benzimidazoles (albendazole or mebendazole) is promising in conditions with small and inoperable patients and patients with cystic lesions in multiple organs. For surgical removal, the Dowling-Orlando technique is an effective method. In this method, the head is positioned lower than the operation table, and a large craniotomy flap can be made depending on the size and site of the lesions. The surgical area is covered with cotton soaked in normal saline to prevent spillage in case of rupture. Then, the cyst is removed using hydro dissection. Prior to and after craniotomy, the surgical field must be cleaned with a scolicidal agent to prevent recurrence as minimal spillage can lead to new cyst formation (1 ml of cyst fluid contains 4,000,000 scoleces). Ventricular irrigation with a scolicidal agent and placement of a drain were done for intraventricular daughter cyst removal. Medical treatment with albendazole (10-15 mg/kg) given monthly separated by 14 days for at least three courses is given to prevent recurrence. Chemotherapy is more effective among younger patients. In our case, surgical removal using the Dowling-Orlando method was done without rupture, and albendazole was prescribed for prevention of recurrence

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Conclusion

Intracranial hydatid cyst with intraventricular extensions is rare disease and usually affects pediatric age group of endemic areas. Cerebral hydatid cyst should be kept in mind

as differentials in case of cystic lesion in imaging findings of brain. Patient can be treated successfully with surgical method without rupture.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Author contributions

BP was involved in the conception of the report. SS, SY, PKK, BG, AG and RKS were literature review, initial manuscript preparation, manuscript critique and review, and the final manuscript preparation. All authors have read and approved the final manuscript.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

REFERENCES

[1] CDC - DPDx - Echinococcosis.2019. Accessed January 13, 2024. https://www.cdc.gov/dpdx/echinococcosis/index.html.

[2] Akrim Y, Barkate K, Arrad Y, et al. Multiple cerebral hydatid cysts: a case report. Cureus 2022;14(5):1–2. doi:10.7759/cureus.25529.1-2.

[3] BABOLI S, BABOLI S, SOLEIMAN MEIGOONI S. Brain hydatid cyst with atypical symptoms in an adult: a case report. Iran J Parasitol 2016;11(3):422–5.

[4] Ebouda FM. Cerebral hydatid disease | Radiology Reference Article | Radiopaedia.org. Radiopaedia. doi:10.53347/rID-31026. 1-2 Revision 13

[5] Thakur SH, Joshi PC, Kelkar A, Seth N. Unusual presentation of hydatid cyst – ruptured intraventricular hydatid. Indian J Radiol Imaging 2017;27(3):282–5. doi:10.4103/ijri.IJRI_70_16.

[6] Giant Cerebral Hydatid Cyst in a Child- A Case Report and Review of Literature | British Journal of Medical Practitioners. Accessed July 12, 2023. https://www.bjmp.org/content/giant-cerebral-hydatid-cyst-child-case-report-and-review-literature.

[7] Almulhim AM, John S. Echinococcus Granulosus. In: *StatPearls*. StatPearls Publishing; 2023. Accessed July 12, 2023. http://www.ncbi.nlm.nih.gov/books/NBK539751/.

[8] Echinococcosis. Accessed July 12, 2023. https://www.who.int/news-room/fact-sheets/detail/echinococcosis.

[9] Santosh T, Patro MK, Bal AK, Behera B. Hydatid cyst at unusual locations: report of two cases. Hum Pathol Case Rep 2017;8:59–61. doi:10.1016/j.ehpc.2017.03.004.

[10] Cerebral Hydatid Cyst with Intraventricular Extension: A Case Report | Journal of Tropical Pediatrics | Oxford Academic | My Library | Zotero. Accessed July 10, 2023. https://www.zotero.org/paudelbinod/items/6UZ2MVB6/library.

[11] Kandemirli SG MD, Cingoz M MD, Olmaz B MD, Akdogan E MD, Cengiz M MD. Cerebral hydatid cyst with intraventricular extension: a case report. J Trop Pediatr 2019;65(5):514–19. doi:10.1093/tropej/fmy080.

[12] Singh RK, Patwa PA, Mishra GV, Dhande RP, Kashikar SV, Unadkat BS. An unusual case report on intracranial hydatid cyst with intraventricular extension. Egypt J Radiol Nucl Med 2022;53(1):52. doi:10.1186/s43055-022-00727-x.

[13] Cureus | Multiple Cerebral Hydatid Cysts: A Case Report | Article. Accessed July 13, 2023. https://www.cureus.com/articles/98245-multiple-cerebral-hydatid-cysts-a-case-report#!/.

[14] Bükte Y, Kemaloglu S, Nazaroglu H, Ozkan U, Ceviz A, Simsek M. Cerebral hydatid disease: CT and MR imaging findings. Swiss Med Wkly 2004;134(31-32):459–67. doi:10.57187/smw.2004.10711.

[15] Taslakian B, Darwish H. Intracranial hydatid cyst: imaging findings of a rare disease. BMJ Case Rep 2016;2016:bcr2016216570. doi:10.1136/bcr-2016-216570.

[16] Ganjeifar B, Ghafouri M, Shokri A, Rahbarian Yazdi F, Hashemi SA. Giant cerebral hydatid cyst: a rare case report. Clin Case Rep 2021;9(3):1774–8. doi:10.1002/ccr3.3908.