

Giant Cerebral Hydatid Cyst in a Five-Year Old Child: A Case-Report

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Abstract

Background

Cerebral hydatid cyst is a rare condition even in endemic parts of the world, which usually happens in children. Huge cysts are usually presented with neurological deficits and symptoms of increased intracranial pressure. Here, we present the case of a five-year-old child with solitary huge cerebral cyst.

Case Presentation

A five-year old girl presented with symptoms of headache, nausea, and vomiting in the emergency department of Ninth of Dey Hospital, Torbat Heydariyeh, Iran. She had referred to several doctors and received symptomatic treatments. In the initial impression, the patient had no neurological deficit but a suture splitting in physical examination and laboratory findings showed no eosinophilia. The brain computed tomography (CT) scan showed a huge cystic lesion suggestive for hydatid cyst. The cyst was removed intact and postoperative treatment with albendazole was initiated. The patient was discharged in good condition with no recurrences in a six-month follow-up.

Conclusion

Huge cerebral hydatid cyst is a very rare condition, usually occurring in solitary form and in children. Surgical removal and postoperative medical treatment should be considered in these cases.

Key Words: Child, Cerebral, Echinococcus, Hydatid Cyst, Granulosus.

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1- INTRODUCTION

Hydatid cyst is a parasitic larval disease mainly caused by *Echinococcus granulosus* and sometimes by *Echinococcus multilocularis*. The development cycle of larva usually involves one intermediate host from herbivores (such as sheep and cattle), and one definitive host from carnivores (e.g. dogs, foxes, jackals, and wolves) (1). Humans usually become infected through oral-fecal routes by consuming vegetables, milk, and other foods that are contaminated with canine fecal material or through direct touch with carnivores (2).

The process of infection occurs through the ingestion of parasite eggs and the subsequent hatching of the eggs in the gastrointestinal tract (3). The eggs then enter the portal blood flow and reach the liver. Most of the eggs settle in the liver and do not cross the hepatic barrier. However, some conquer the barrier and enter the lung. It is estimated that around 70% of all hydatid cysts are formed in the liver and about 25% are located in the lungs (4). The larva rarely enters the systemic circulation and spread into other organs, forming hydatid cysts in other solid organs. Central nervous system is a rare involvements site (5). Even in regions that are endemic for hydatidosis, the brain involvement accounts for less than 1 percent of involvements (6). Cerebral hydatid cysts frequently present as solitary supratentorial cysts and usually involve parietal lobe through middle cerebral artery, which is most commonly affected due to the embolic nature of the infestation (7). This involvement may be primary without any other organ involvements or secondary to cysts originated from other organs e.g. cardiac cyst (8). Herein, we present a case of primary giant hydatid cerebral cyst in a 5-year-old girl.

2- CASE REPORTS

A five-year-old girl from rural areas around Mashhad, Iran, presented to the pediatric emergency department of Ninth of Dey Hospital, Torbat Heydarieh, Iran, with intermittent episodes of headache, nausea, and vomiting over the previous 2.5 years. During this period, the patient had referred to several physicians and only received some symptom-oriented treatments without any imaging study.

In physical examination, the patient was completely conscious and alert without any neurological deficit. She also had no signs of papilledema and her proximal and distal muscle forces were normal. The only notable finding in her examination was a left coronal suture splitting. Computed tomography (CT) scan revealed a huge round frontoparietal hypodense cystic lesion that was suggestive of hydatid cyst. The huge mass caused significant mass effect, shifted the midline structures, and collapsed left cerebral ventricle causing some degrees of hydrocephalus (**Figure.1**).

Laboratory assessment of the patient was relatively normal and showed no prominent finding. Laboratory findings are illustrated in **Table-1**. The patient's abdominal sonography was not suggestive for liver involvement. Moreover, chest X-ray and lung CT scan was done to assess lung involvement and showed nothing indicative for hydatidosis. The patient underwent frontoparietal craniotomy, gross removal of the cyst was performed using Dowling's technique, and the cyst was removed intact without any rupture in its wall or any opening and spillage into the ventricular system. Splitting of the left coronal suture was evident during the surgery (**Figure.2**).

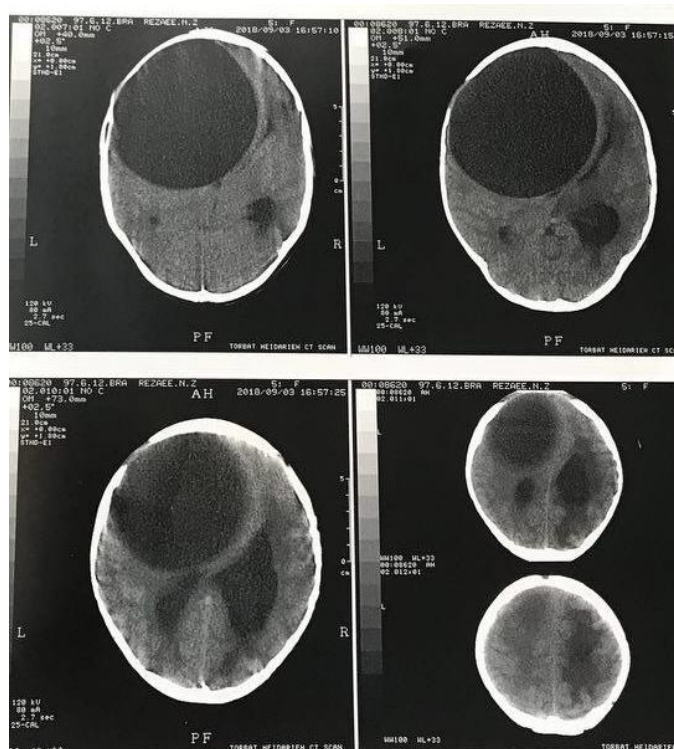


Fig.1: The first head CT scan of the patient in axial view.

Table-1: Laboratory findings of the patient.

Parameter	Result
White Blood Cell count (per 10 ³ ml)	12
Polymorphonuclear cells (%)	70
Lymphocytes (%)	25
Monocytes (%)	3
Eosinophils (%)	2
Red Blood Cell count (per 10 ⁶ ml)	5
Hemoglobin (g/dl)	12.5
Hematocrit (%)	38.6
Platelet count (per 10 ³ ml)	336
Blood Culture	Negative
Lactate Dehydrogenase (U/L)	428
Creatine Kinase (IU/L)	22
Erythrocyte Sedimentation Rate (mm/h)	11
C-reactive protein (mg/L)	1
Sodium (mEq/L)	132
Potassium (mEq/L)	4.4
Arterial Blood Gas	
pH (mmHg)	7.4
PCO2 (mmHg)	34.1
PO2 (mmHg)	58.4
HCO3 (mEq/L)	21.3
Base Excess (mEq/L)	-3.3
O2 saturation (%)	90.4

PCO2: Partial pressure of carbon dioxide; PO2: Partial Pressure of Oxygen; Bicarbonate.



Fig.2: Suture splitting due to compressive effect of the cyst is evident during surgery.

The removed cyst was over 10 cm in diameter and had a volume of about 400 ml as shown in **Figure.3**. Postoperative treatment was commenced with Albendazole (200 mg daily) under supervision of infectious disease specialist. The patient stayed in the intensive care unit (ICU) five days after the operation and she was then transferred to the neurosurgery ward for

another seven days of stay. She was discharged in good condition, with no fever, seizure, cerebrospinal fluid leak, or neurological deficit, on the 13th post-operative day. In the six-month follow-up, our patient showed no complication. The follow-up CT image of the patient is shown in **Figure.4**.

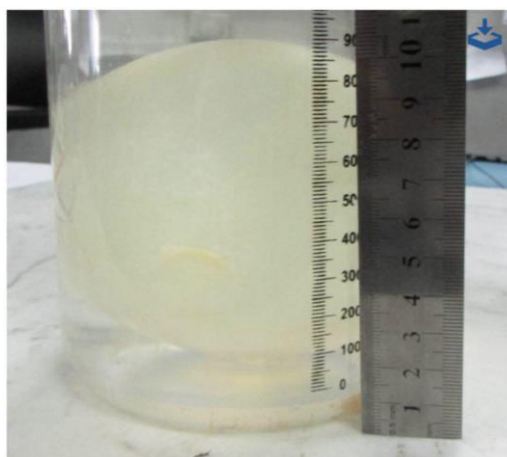


Fig.3: The macroscopic view of the resected cerebral cyst showing its volume and diameter.

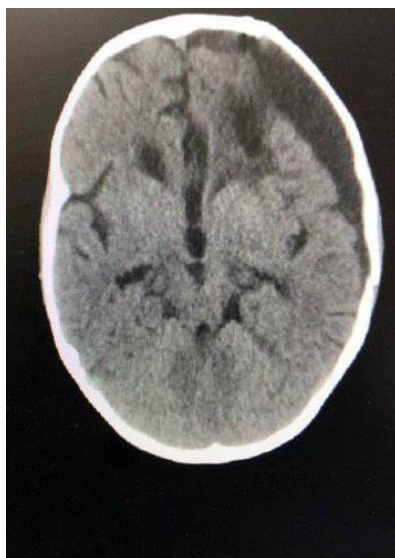


Fig.4: Follow- up CT- scan of the patient.

3- DISCUSSION

Hydatid cyst is a parasitic disease that occurs frequently in Asian, African, Australian, and Middle Eastern countries (9). This disease is endemic in different parts of Iran. It mainly involves liver and lungs, but is rarely found in the brain (10). This condition is more frequent in children and is hypothesized that it might be due to the presence of patent ductus arteriosus (11). Most of the cerebral hydatid cysts are solitary; however, multiple cysts have also been reported. The dissemination from liver and lungs to the brain could be due to anatomical variations that form a shunt for the parasites to escape (12). Primary cysts result from direct infestation of the brain without any other solid organ involvement. Secondary cysts usually are a result of spontaneous, traumatic, or perioperative rupture of primary cysts. We presented a solitary cyst in the brain of a 5-year-old girl who presented with sole symptoms of nausea, vomiting, and headache. The cyst was primary and without any involvement in other organs (13). Humans can become infected through ingestion of *Echinococcus* eggs, which might be found in contaminated foods or water. In some

cases, however, the direct contact with the carnivores may cause infection (12). Therefore, living in rustic areas can be considered a risk factor for the disease. In the case of our study, the residence of the patient in rural areas and her contact with dogs, sheep, and other animals and subsequently contaminated food and water, might have been the source of infection. Giant cerebral hydatid cysts are reportedly symptomatic and accompanied by progressive neurological deficits (3). Furthermore, symptoms of increased intracranial pressure are evident in most of the patients. Further neurological symptoms are also reported in the literature such as seizures (13, 14). Surprisingly, in the case of our study, the child only had unspecific symptoms such as headache, nausea, and vomiting without any neurological deficit. The insidious growth of the cyst and its subacute condition makes the diagnosis difficult. It is believed that the growth rate of this cyst is about 1 cm in year (13). This might be the reason for misdiagnosis and neglecting of imaging studies in the experience our patient had with several previous physicians. A part of the approach in

patients with suspicion of cerebral hydatid cyst is radiologic imaging. The patients usually undergo CT or magnetic resonance imaging (MRI), which often reveals a huge, round well-margined cystic lesion without marginal enhancement, suggestive of hydatid cyst (15). Further assessment with MR spectroscopy and diffusion weighted MR imaging may also be helpful to detect intracranial hydatid cysts. However, it is not indicated in most cases (16). Important differential diagnoses for a cerebral mass, such as cystic tumors, arachnoid cysts, abscess, and porencephalic cysts should always be considered when managing these cases (17). Our radiologic findings in this report showed a solitary round, well-margined cyst with over 10 cm diameter, indicative of a huge hydatid cyst. Laboratory findings are not always helpful in diagnosis of hydatid cyst, but eosinophilia may be present in 25% of the patients (18).

Surgical approach should be used for removal of cerebral cysts. It is paramount to perform the surgical removal of the cyst with great care, so that the cyst does not rupture, in order to prevent subsequent complications such as spread of parasites and anaphylactic reactions (8). The most commonly used technique is Dowling's-Orlando that involves continuous warm saline irrigation and insertion of a rubber catheter between the cyst and the brain. The rupture of the cysts usually accompanies a great risk for the spread of scolexes (7). In case of cyst rupture, cyst aspiration must be performed along with placement of 10% formalin soaked cottonoid and continuous saline irrigation for about 5 minutes after removing the capsule (19). Albendazole at a dose of 400 mg per day should be initiated immediately after the surgery and continued over an eight-week period for adult patients (20). In our study, since the case was a 5-year-old child, we used a 200-mg dose of albendazole.

4- CONCLUSION

Cerebral hydatid cyst is a rare condition even in the endemic regions such as Iran. Huge cerebral cysts may present with neurological signs and deficits, or they may only show symptoms of increased intracranial pressure such as headache, nausea, or vomiting. Imaging studies such as CT or MRI should be considered in these cases for accurate diagnosis. Treatment involves surgical removal of the cyst as well as postoperative albendazole.

5- CONFLICT OF INTEREST: None.

6- REFERENCES

1. Vennarecci G, Manfredelli S, Guglielmo N, Laurenzi A, Goletti D, Ettorre GM. Major liver resection for recurrent hydatid cyst of the liver after suboptimal treatment. *Updates in surgery*. 2016;68(2):179-84.
2. Hamzavi Y, Nazari N, Mikaeili A, Parandin F, Faizei F, Sardari M. Prevalence of Hydatid Cyst in slaughtered livestock in Asadabad Slaughterhouse during 2014-2015. *Pajouhan Scientific Journal*. 2016;14(3):58-66.
3. Yaya-Loo HJ, Montalvo GR, Vega G. Uncommon Localizations of Echinococcosis: Primary Cerebral and Cardiac Hydatid Cyst. *Ann Clin Pathol*. 2017;5(2):1106.
4. Mandal S, Mandal MD. Human cystic echinococcosis: epidemiologic, zoonotic, clinical, diagnostic and therapeutic aspects. *Asian Pacific journal of tropical medicine*. 2012;5(4):253-60.
5. Guzel A, Tatli M, Maciaczyk J, Altinors N. Primary cerebral intraventricular hydatid cyst: a case report and review of the literature. *Journal of child neurology*. 2008;23(5):585-8.
6. Fdaladdin YAJ, Alsaggaf AI, Wakid MH. Comparative epidemiological studies on Echinococcosis of local and imported livestock in Al-madina Al-munawwarah in Saudi Arabia. *The Egyptian Journal of Hospital Medicine*. 2018;50(1):108-26.

7. Polat G, Ogul H, Sengul G. Hydatidosis Following Giant Cerebral Hydatid Cyst Operation. *World neurosurgery*. 2018;118:14-5.
8. Shih RY, Koeller KK. Bacterial, fungal, and parasitic infections of the central nervous system: radiologic-pathologic correlation and historical perspectives: from the radiologic pathology archives. *Radiographics*. 2015;35(4):1141-69.
9. Davoudi JD, Safari M, Shirazi S, Bahman Shabestari A, Dolatkhah A. A Survey of hydatidosis surgical cases in Kermanshah province of Iran during 2012-2013. *Journal of Zoonotic Diseases*. 2016;1(1):40-6.
10. Richter J, Profis E, Holtfreter MC, Orhun A, Müller-Stöver I, Dedelen H, et al. Anaphylactic shock ensuing therapeutic puncture of an echinococcal cyst. *Parasitology research*. 2015;114(2):763-6.
11. Pezeshki A, Akhlaghi L, Sharbatkhori M, Razmjou E, Oormazdi H, Mohebali M, et al. Genotyping of *Echinococcus granulosus* from domestic animals and humans from Ardabil Province, northwest Iran. *Journal of helminthology*. 2013;87(4):387-91.
12. Luo K, Luo D-H, Zhang T-R, Wen H. Primary intracranial and spinal hydatidosis: a retrospective study of 21 cases. *Pathogens and global health*. 2013;107(2):47-51.
13. Beyhan Y, Yılmaz H, Taş ZC, Yıldızhan R, Kotan Ç, Batur A, et al. A Rare Case of Secondary Hydatid Cyst: Uterus and Colon Locations in the Same Patient. *Turkiye parazitolojii dergisi*. 2019;43(3):149-51.
14. Brizuela M, Sarkis C, Gonzalez R, Paulin P, Lubieniecki F, Berberian G. Cerebral hydatid disease: report of six pediatric cases. *Revista chilena de infectologia: organo oficial de la Sociedad Chilena de Infectologia*. 2017;34(3):270-5.
15. Aydin MD, Karaavci NC, Akyuz ME, Sahin MH, Zeynal M, Kanat A, et al. A New Technique in Surgical Management of the Giant Cerebral Hydatid Cysts. *Journal of Craniofacial Surgery*. 2018;29(3):778-82.
16. Borkar SA, Verma N, Joseph SL, Kale SS, Mahapatra AK. Magnetic resonance imaging of pediatric primary cerebral hydatidosis. *Journal of pediatric neurosciences*. 2017;12(3):298.
17. Karaaslan A, Borekci A. Emergency surgical treatment in primary cerebral hydatid cyst: A case report and review of the literature. *South Clin Ist Euras*. 2017;28(2):143-6.
18. Kandemirli SG, Cingoz M, Olmaz B, Akdogan E, Cengiz M. Cerebral Hydatid Cyst with Intraventricular Extension: A Case Report. *Journal of tropical pediatrics*. 2019. DOI: 10.1093/tropej/fmy080
19. Farajirad E, Farajirad M, Khajavi M, Shojaie SR. Central Nervous System Hydatid Disease: Clinical Analysis of 99 Cases in Qaem Hospital of Mashad University of Medical Sciences, Iran. *Neurosurgery Quarterly*. 2016;26(1):1-4.
20. Alomari MS, Almutairi MK, Alali HM, Elwir JS, Alola SA, Alfattoh NI, et al. Primary giant cerebral hydatid cyst in an 8-year-old girl. *Asian journal of neurosurgery*. 2018;13(3):800.