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Cerebral Hydatid Disease Patients Admitted to Duhok City Hospitals: Management and Outcome

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Abstract

Background and objectives: The human systemic infestation with Echinococcus granulosus affects the brain in 2% of cases, cerebral hydatid cyst, which presents clinically as slowly growing intracranial mass lesion leading to variable symptomatology of raised intracranial pressure, neurological deficit, and epilepsy. The condition, when occurs, needs prompt surgical removal in order to avoid fatal consequences. The aim of this article is to report clinical data and our experience concerning cerebral hydatid cyst presentation, surgical management, complications, and outcome.

Methods: A retrospective case-series study encompassing 8 patients, four females and four males, whose ages ranged between 3.5-35 years, harboring cerebral hydatid cysts, including one cerebral hydatid abscess. The current study was conducted at Duhok Teaching Hospitals, during January 2005-May 2021. The diagnosis was established on clinical and radiological, computed tomography and magnetic resonance imaging backgrounds, and confirmed by operative findings, and histopathology (abscess cyst). Via appropriate craniotomies, all cerebral hydatid cysts, and abscess, were successfully removed, except, the recurrent hydatid cysts which needed an additional posterior fossa craniectomy to achieve total removal. There was no need for the use of advanced statistical system.

Results: Compared to their original neurological deficit at presentation, seven cases could achieve complete recovery, except, the patient with recurrence, whose recovery was delayed and suboptimal due to repeated surgeries; there was no mortality.

Conclusion: The operating surgeon should practice meticulous care avoiding the possible risk of an accidental rupture during cyst delivery. Various opinions address the pre-operative, intra-operative and post-operative use of Albendazloe, however, we are not sure whether the use of this medication could have prevented the recurrence of CHC which followed the accidental rupture.

Key words: Albendazole; Hydatid cyst abscess ; Recurrent cerebral hydatid cyst ;Single cerebral hydatid cyst.

Introduction

Hydatid cyst (HC) is regarded as one of the world's major zoonosis affecting both man and domestic animals;[1] it results from infection with the larval stage of Echinococcus granulosus.[2-3]This disease is a chronic disease and highly endemic in many parts of the world, a disease characterised by the development of slowly growing hydatid cysts which may not be detected for months or years after

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the occurrence of the initial infections.[4] Knowing its life cycle, prevalence, and geographical distribution are essential in understanding the potential public health threat it possesses to individuals who live in regions where HC is endemic, and in attempting to establish feasible control programs in these areas.[5] Cerebral hydatid cyst (CHC) infestation affects all age groups of both sexes with significant physical morbidity, and mortality if not dealt with appropriately by surgical removal. Hydatid cyst develop in various organs, the commonest site of infection is the liver, the lung, followed by other organs such as kidneys, spleen, omentum, heart, and the brain, and bone marrow cavities. Also, the CHC may become infected and present as an intracranial abscess formation. In regions with farming profile environment, cerebral hydatid cyst cases may represent occasional referral to neurosurgical services. Such presentation may include symptoms and signs of raised intracranial pressure, motor deficit, and epilepsy; this should be well thought of in current practice. In the absence of specific clinical features, the early diagnosis of the hydatid cyst depends mainly on the laboratory (immunoglobulin G, immunoglobulin M, immunoglobulin E, and immunoglobulin A) and imaging tests, including computed tomography, magnetic imaging scan, and ultrasound. The latter imaging facilities constitute the essential diagnostic armamentarium in initiating therapy and surgery.[6-7] Furthermore, the technicalities during cyst delivery need special attention and consideration while using the surgical tools, e.g, those causing unavoidable and inadvertent, tissue vibration and shaking, while using Gigli saw, and/or the bipolar diathermy; such unfortunate event(s) may lead to cyst rupture and possible recurrence, as had occurred in one of our cases in the current study.By submitting the current study, it is aiming at reporting clinical data and our experience concerning cerebral hydatid cyst presentation, surgical management, complications, and outcome.

Patients and methods

A suitable format sheet has been designed for recording relevant patient's demographic and clinical notes. All of them had both computed tomography (CT) and magnetic resonance imaging (MRI) scanning examination, in addition to routine haematological tests, chest X-ray, ECHO cardiography, and abdominal ultrasound examination to confirm or rule out the presence of HC elsewhere (e.g. hepatic or pulmonary). None had skin or serological tests. Patients were pre-operatively given anti-convulsant medication at presentation (Epitam, Levetiracetam of hikma Company), plus measures to reduce the increased intracranial pressure (ICP). Intravenous prophylactic antibiotics, Ceftriaxone, according to body weight, was given peri-operatively; however, Gentamycin-containing-normal saline solution was the priming solution which was used to wash out the tissues during surgical procedures, craniotomies. While the antibiotic therapy was continued for two weeks after hospital discharge, however, the anticonvulsant therapy was given for 6 months after surgery; none received Albendazole medical therapy. This study was performed in compliance with ethical regulations of Duhok Area Health Authority and College of Medicine, University of Duhok.

Results

These are shown in table 1. They were 4 males and 4 females; their ages ranged between 3.5 years – 35 years; they came from many other towns around Duhok City. All had single CHC; 5 on the left and 3 on the right frontoparietal regions;

they all, except case 8, had clinical and radiological symptomatology of raised ICP, focal and / or generalised epilepsy, and speech disturbance. None of the patients had eosinophilia in the peripheral blood film. Plain skull X-Ray, CT and MRI scanners films, showed supratentorial, single, cerebrospinal fluid (CSF) density/intensity, non-enhancing, cystic lesion without calcification, and the multiple CHC in the recurrent case; however, there was marked calcification, peripheral ectocyst enhancement, and the inside cyst showed water lily appearance due to in folding laminated membrane (Fig. 1-4). All cases, except case number 8, had, clinically, significant classical mass effect. All have successfully undergone CHC removal via appropriate craniotomy, including those cysts of the recurrent case; the recurrence occurred following an accidental rupture of a superficially located parietal CHC during raising the osteoplastic bone flap. Both the biochemical analysis of hydatid fluid and pathological examination of the cyst wall confirmed HC specimens (Fig. 5); only five cysts were fertile showing scolices that would give rise to daughter cysts in the intermediate host, and/or grow to a mature worm in the definite host. All patients have achieved excellent motor neurological recoveries from their deficit at presentation within few weeks post-operatively, however, the case with recurrent cysts the neurological recovery took longer time due to the need of repeated surgery; all were free of seizure and CHC during the follow-up period.

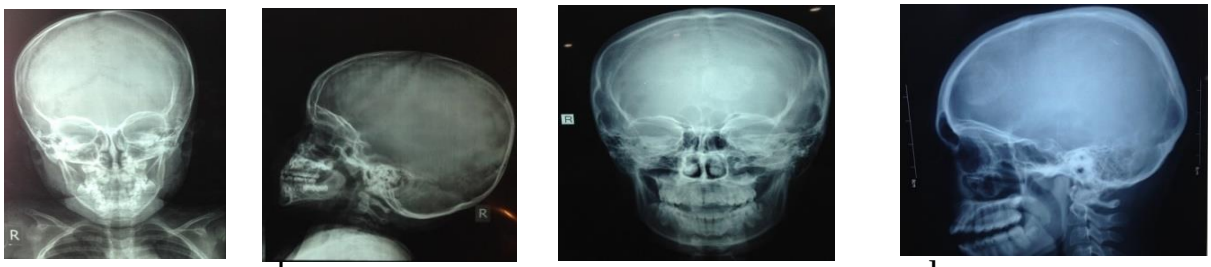
Table (1): Clinical data.

Case number	Age in years	Gender	Clinical findings	Initial CT* and MR** findings
Case 1	3.5	Female	Headache, vomiting, right sided hemiparesis, right focal epilepsy, bilateral papilloedema	Single left parietal cerebral HC***
Case 2 (Recurrent HC***)	7	Female	Headache, vomiting, focal and generalized epilepsy, left sided hemiparesis, bilateral papilloedema	Single right parietal cerebral HC***
Case 3	11	Male	Headache, vomiting, dysphasia, right sided weakness, bilateral papilloedema	Single left fronto-parietal cerebral HC***
Case 4	13	Female	Headache, vomiting, generalised epilepsy, left sided hemiparesis, bilateral papilloedema	Single right frontal cerebral HC***
Case 5	15	Male	Headache, right sided hemiparesis, bilateral papilloedema, left abducent nerve palsy	Single left fronto-parietal cerebral HC***
Case 6	15	Male	Headache, left focal epilepsy, left sided hemiparesis, bilateral papilloedema, left abducent nerve palsy	Single right fronto-parietal cerebral HC***
Case 7	35	Male	Headache, dysphasia, right sided hemiparesis, generalised epilepsy, bilateral papilloedema	Single left fronto-parietal cerebral HC***
Case 8	14	Female	Generalised epilepsy, headache.	Single left frontal infected cerebral HC***; abscess formation; marked calcification

CT*= Computed tomography. MR**= Magnetic resonance. HC***= Hydatid cyst.

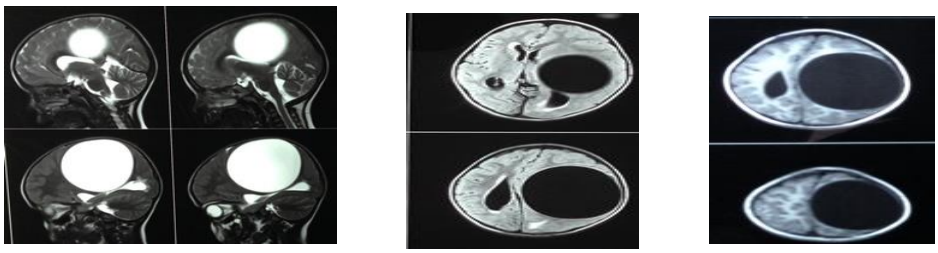
Discussion

In countries endemic with hydatidosis, regions with farming living profile and cattle raising occupation, CHC stands, although uncommon, a distinguished clinical entity and referral to Neurosurgical Units from presenting with symptoms and signs of raised ICP, motor deficit, and with or without epilepsy. Researchers have, in the setting of systemic (extracranial) HC disease, found that the age groups 11–20 and 21–30 years had higher HC infestation than other age groups, more frequent in females than in males, commoner among illiterate than literate, and more among rural than urban patients; the highest rate of infection was among housewives than other occupations[8-10]. Brain involvement with hydatid disease occurs in 2% of all *Echinococcus granulosus* infections; the growth rate of hydatid cysts in the brain has been variably reported at 1.5-10 cm per year. Lack of effective immune system in the brain, special architecture of brain tissue, patent ducts arteriosus and patent foramen ovale have been the proposed factors for isolated cerebral hydatid disease.[11] However, none of our patients had congenital heart disease.



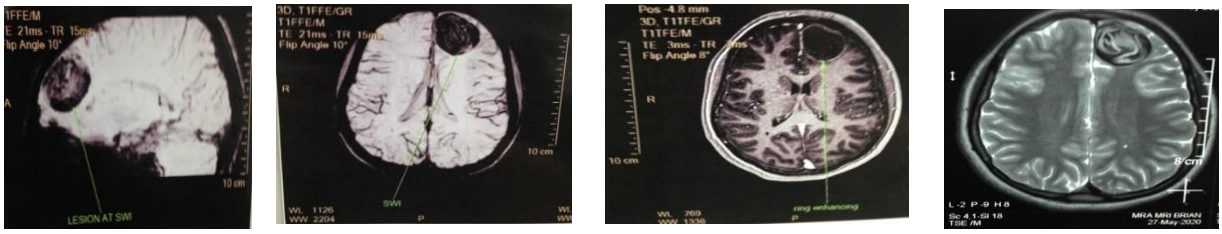
a. b. c. d.

Figure (1): Case 1. Plain skull X-ray films. a. Postero-anterior view showing left sided parietal bone bulging, a bony deformity due to long standing HC lesion growth. b. Lateral view showing coronal sutural diastasis. c. Postero-anterior view showing calcification. d. Lateral view showing calcification.



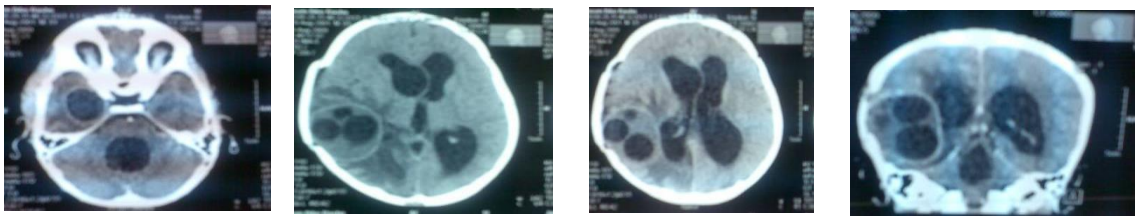
a. b. c.

Figure (2): Pre-operative MR studies. a. T2- weighted MR picture showing left sided parietal HC. b. and c. Fluid attenuated inversion recovery (FLAIR) sequence MR picture showing left sided parietal HC.



a. b. c. d.

Figure (3): Case 8. Pre-operative MR studies. a. Sagittal and b. Axial SW* images showing calcification (black inside). c. T1W image, axial view showing peripheral (ectocyst) ring-shaped enhancement. d. T2W image, axial view showing water lily appearance of HC.



a. b. c. d.

Figure (4): (case 2). CT scan pictures showing recurrent CHCs due to inadvertent rupture of the initially presenting single right parietal HC: a. Axial view showing HC in right temporal lobe and in the fourth ventricle. b and c. Axial view showing HCs in right parietal lobe. d. HCs in the coronal view.

Concerning the age, this case-series study has shown, in spite of its small size, that children, adolescent, and middle-age groups of both sexes have been affected by CHC, especially children and adolescents; this is in keeping with other studies[8-9,12].Regarding gender, this study shows that males and females are equally affected contrary to many other studies[8-9,12]; this is probably due to its small size.As far as the presenting clinical symptoms and signs, raised intracranial pressure, there are similarities between this and other studies[8-9,12]. The blood films showed normal eosinophil count, except the patient with the accidental HC rupture (and subsequent recurrence), he had marked eosinophilia; this may go with other researchers who mention that eosinophilia occurs during the larval release[13], and according to others, in 25% of cases and only when there is spillage of hydatid fluid[14].Plain skull X-ray that may show localised bony bulging in young children with CHC (Fig. 1), add some more information in the diagnostic process, however, MR and CT scans remain the golden standards in achieving the pre-operative diagnosis; the latter two, characteristically, show a spherical, well defined, non-enhancing, CSF density/intensity, cystic lesion(s) without peripheral edema[8,12]; all had significant mass effects in their imaging studies. Although other researchers found cerebral alveolar hydatidosis (caused by Echinococcus multilocularis), however, the latter was not found in the present series[15, 16]. In the present study there was no calcification in the CHC, except in the case on CHC abscess, however, others have mentioned the presence of calcified intracranial HC[17-18]. In our opinion, the CT and MR scan findings have obviated the need for serological tests that vary in their sensitivity and specificity, the latter are expensive, and need sophisticated equipment, and well trained technicians[19-21] Furthermore, quite many of them were not available at the hospital laboratory. Kohli et al had shown by MR spectroscopy (MRS), in intracranial HC, the presence of pyruvate peaks besides lactate, alanine and acetate[22]. Magnetic resonance spectroscopy has potential, albeit experimental, application in diagnosing cerebral hydatid disease. In a series, three cases of hydatid cysts have been reported to show lactate, acetate, and succinate peaks, and one case with surrounding edema revealed a choline and mannitol peak; however, none of our CHC had MRS examination.Some patients with CHC, might, also, have harbored HC in their liver, lung, and spleen; the concomitant presence of pulmonary HC dictates that it should be dealt with prior to dealing with CHC as there is the risk of pulmonary cystic rupture during endotracheal intubation with all of its possible risks, e.g. the anaphylactic shock and dissemination. However, none of the patients in the current series had extracranial HC.

a. b. c.

Figure (5): Intra-operative views: a. The HC in situ after dural reflection; b. The brain cavity following the HC delivery; c. The HC in a kidney dish with part of the ectocyst held by the toothed forceps; this HC's dimensions measured 13 cm X12 cm X 11 cm.

Surgical removal via an appropriate and adequate craniotomy is absolutely needed to deliver the CHC and save patient's life with excellent results (Fig. 5). Dowling's technique remains the preferred method, in which the cyst can be delivered by lowering the head of the operating table and instilling warm saline between the cyst and the surrounding brain parenchyma; this is possible because of minimal adhesions around the cyst wall[14-23]. However, concerning the removal of viable CHC, there is the possible risk of an accidental rupture during the use of surgical tools producing an unavoidable and inadvertent, tissue vibration and shaking, e.g. Gigli saw, as it happened in case one of this study, giving to recurrence (Fig. 4). Furthermore, during dissecting any overlying cerebral tissue from the CHC should be done carefully and patiently; we have noticed that the electrical current of the bipolar diathermy probe may puncture the cyst wall and lead to spillage of drops of hydatid fluid; in the latter event, it is managed by removing the fluid that spills out of the small hole and covering this tiny hole with layers of saline-soaked neuropatties cottonoids in order to prevent further spillage and dissemination, and, possible recurrence of the CHC. HCs were biochemically and histologically confirmed. Dealing with the CHC abscess should follow the standard management protocols similar to cerebral abscess elsewhere. In the current study, the post-operative recovery had been uneventful, including the patient with multiple recurrent CHC who was operated upon subsequently. The patients continued taking anticonvulsant therapy for about six months; all patients were free of seizure during the follow-up period. Neither recurrent CHC nor mortality has been found during the follow-up period.It may be quite interesting to mention in this context of cerebral hydatidosis that this study has dealt with patients harboring single CHC, without mortality, and without recurrence except the one that had accidentally ruptured intra-operatively; however, many researchers have referred to the rare and infrequent initial presentation of multiple CHC, excluding iatrogenic rupture, with male predominance, and with post-operative mortality of 10.7%, few recurrences between 3 months and 3 years, and an overall mortality of 17.6%; furthermore, and because of the presence of significant number of patients in their study, 34 patients, they raised the possibility of a different strain of Echinococcus granulosus[24]. Many authors recommend preoperative use of antihelmenthics to sterilise the cyst, and reduce the chances of spillage, anaphylaxis, and dissemination at surgery[25], as well as recurrence[26-27]. It is most effective in alveolar hydatid, less so for liver infection, and essentially ineffective for the diseases of bone, brain, eye, gall bladder and other sites[28-31]. However, we think that there is no need for giving Albendazole postoperatively following successful removal of the single HC intact. In the case of the recurrent CHC mentioned above, the patient fails to take Albendazole; we are not sure whether the use of this medication could have

prevented the recurrence of CHC following the accidental rupture case.

Conclusion

In regions endemic with hydatidosis, and in patients presenting with raised ICP, motor weakness, and / or epilepsy, neurosurgeons should be alert to the possibility of CHC being the offending pathological lesion, whenever the neuro-imaging has disclosed a well-defined, well-outlined, circular, single CSF-density cystic lesion. During the craniotomy procedure, every precaution should be taken not to advertently rupture the cyst in order to prevent recurrence. Successful and uneventful surgical removal of the CHC is curative and lifesaving. However, infected CHC, may, clinically, present as a cerebral abscess which should be dealt with in the standard methods.

Conflicts of interest

The authors declare that they have no conflict of interest. I would like to thank Prof. Dr. Adil Saeed, Professor of Parasitology and Dr. Bashaar Abduljabbar, Assistant Professor, Consultant Pathologist, for performing the biochemical analysis of the CHC fluid and performing the histopathological examination respectively.

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