

Intracranial hydatid cyst. Clinical features and outcomes of surgical treatment of a series of 8 Iraqi cases

Zaki N. Hasan, MBCHB, FICMS,
Wissam J. Sagban, MBCHB, FICMS,
Aqeel K. Hatim, MBCHB, FICMS,
Mohammed A. Assad, MBCHB, FICMS.

Hydatid cyst is a parasitic infestation caused by the ingestion of the encysted larvae of the parasite *Echinococcus granulosus*. It is endemic in Australia, the Middle East, central Europe, South America, and Mediterranean countries. The cysts are most frequently distributed in the liver in 75%, and the lung in 15%. Brain involvement accounts for only 1-2% of all hydatid cysts in humans, and 2% of all intracranial cystic lesions. Humans are accidental intermediate hosts and get infected by ingestion of food contaminated by dog feces, or by direct contact with dogs. The eggs hatch into larvae in the stomach and small intestine, which then penetrates the gut wall into the vessels of the portal system and then become trapped in the liver where they form the hydatid cysts. Rarely, the larvae bypass the liver sinusoids into the systemic circulation reaching the lungs and occasionally the brain.¹ The most common location of the hydatid cyst in the brain is along the tributaries of the middle cerebral artery and subarachnoid spaces. Intracranial hydatid cysts are slow growing, spherical, solitary and uni-locular; they contain translucent fluid and daughter cysts resembling small white grapes. They could be as large as 5-10 cm. Protoscolices within the cyst form a granular deposit known as hydatid sand.^{2,3} Clinically, the patients usually present with focal neurological deficit and features of raised intracranial pressure. A diagnosis is made on CT scanning depicting thin-walled, spherical, non-enhancing CSF-attenuation cyst. The serological diagnosis is complementary as hydatid cysts often induce no or low antibody responses. In routine laboratory practice, enzyme linked immunosorbent assay (ELISA) using hydatid cyst fluid antigen is often used.^{3,4} The treatment of hydatid cyst is medical by Albendazole and surgical excision of the cyst in toto. Various surgical techniques were commonly used for cyst removal; the first widely accepted surgical method was the Dowling-Orlando technique,^{3,4} which was later improved by Arana-Lniquez,^{3,4} and which included direct puncture and aspiration of the cyst fluid through a small hole in the cyst wall, and expulsion of the cyst through a small cortical incision over the cyst using insufflations of air in the contra lateral ventricle. However, the most popular technique was

using peri-cystic hydraulic irrigation of saline with mild force between the cyst wall and brain interface in order to deliver the cyst intact without rupture, which was the technique used in our cases.^{3,4} The objective of the current study was to assess the clinical presentations, radiological features, and outcome after combined surgical and medical treatment protocol of intracranial hydatid cyst cases in Iraq.

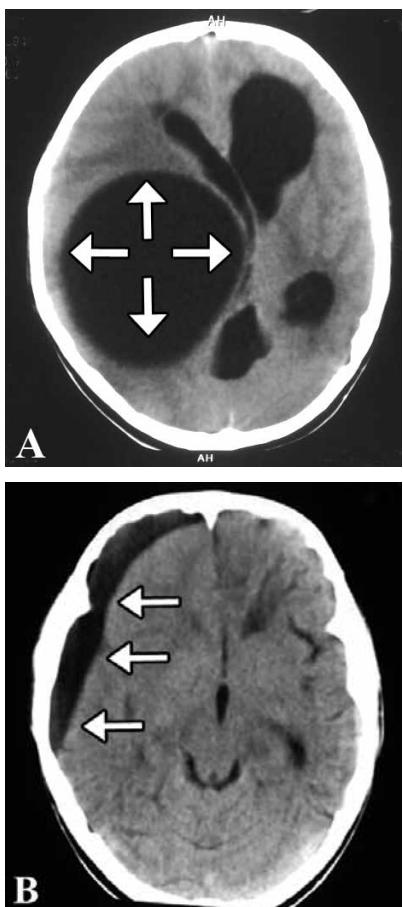
This is a case series report of 8 patients with intracranial hydatid cysts. All of the patients were treated at the Neurosciences Hospital in Baghdad, Iraq and followed up for 30 months after treatment. The duration of study was from January 2006 to April 2012. All patients consented to participate in the study, which was conducted according to the Principles of the Helsinki Declaration, and approved by the Scientific Committee of the Neurosciences Hospital. Patient history was taken according to a detailed form, and a consultant neurologist performed clinical examination including neurological assessment. All patients had full blood count, and triple radiological assessment in the form of chest x-ray, abdominal ultrasound, CT of the brain before and every 3 months after surgical excision of the hydatid cyst. The diagnosis was made mainly by CT scan, and confirmed by postoperative histopathology. Trans esophageal echocardiography assessment was carried out postoperatively to exclude the presence of patent ductus arteriosus. All cases received Albendazole (10 mg/kg/day) for 3 months postoperatively, and underwent the surgical procedure using peri-cystic hydraulic irrigation of saline with mild force between the cyst wall and brain interface to deliver the cyst intact without rupture. The results of the study were tabulated using Minitab 16 statistical software (Minitab Inc., State College, PA, USA).

There were 6 males (75%), and 2 females (25%) with a male to female ratio of 3:1. The age range was from 6-35 years. Five patients (63%) were aged between 6-15 years; the other 3 were 19, 30, and 35-years-old. The mean age was 16.5 ± 11 . All patients had lived in rural areas. The duration of symptoms varied from one month to 3 years, with a mean duration of 17.5 months. Table 1 summarizes the presenting complaints and neurological deficits found on clinical examination. Imaging of the

Disclosure. The authors declare no affiliation or financial involvement with organizations or entities with a direct financial interest in the subject matter or materials discussed in the manuscript. No funding was received for this work from any organization.

Table 1 - Presenting complaints and neurological deficits on clinical examination of Iraqi intracranial hydatid cyst patients.

Symptom	Number	(%)
Headache	8	(100)
Hemiparesis	7	(87.5)
Vomiting	6	(75)
Seizures	5	(62.5)
Papilledema	6	(75)

**Figure 1** - Brain CT scan of a 13-year-old female: A) preoperatively showing well-defined large cystic lesion with a thin wall and smooth outline measuring 10-12 cm in the right parietal lobe associated with severe mass effect on the right lateral ventricle and midline shift to the left side with clear content and absence of contrast enhancement. B) Postoperatively showing complete removal of the cyst with residual subdural effusion.

brain by CT scan characteristically showed spherical well-defined non-enhancing cystic lesions, thin-walled without surrounding cerebral edema, and the fluid

density was that of the CSF (Figure 1a). The brain CT scan findings are summarized as follows: 7 patients had a solitary cyst in the brain (87%), and one patient had multiple cysts. The cyst was found in the parietal lobe in 7 patients (87%), 5 on the right side, and 2 on the left, and one in the left occipital lobe (13%). The sizes of the cysts ranged from 6-11 cm, with a mean cyst size of 8.1 ± 1.30 cm. Six patients (75%) had a cyst size of 10 cm and above, and 2 patients (25%) had a cyst size below 10 cm. One patient was found to have a hydatid cyst in the liver on ultrasound and was referred to general surgeon who removed the hepatic cyst 2 months after the cranial procedure. None of the patients had evidence of pulmonary hydatid cyst on chest x-ray.

The brain is a rare site of involvement by hydatid cysts; it most commonly affects the liver as the larvae get trapped in the hepatic sinusoidal plexus. The second common place for hydatid cyst is the lung, where the second filter of alveolar capillaries traps the larvae that have escaped from the liver. Once they bypass the lungs (one possible explanation is patent ductus arteriosus), the larvae can travel and implant anywhere in the body. The brain is considered the fourth common site after liver, lungs, and peritoneum.³ There are no previous statistical figures on the infestation incidence in Iraq, but an incidence of 1:2000 was recorded in Turkey, the northern neighboring country.³ The present study showed 63% predominance in the pediatric age group; similar to the results of Per et al.⁵ The above pediatric age predominance is in contrast to the findings of Ahmadi et al.,⁶ who found 42.8% of his series were between 21-40 years. The higher pediatric incidence was attributed to higher patent ductus arteriosus in the childhood group. None of our patients had patent ductus arteriosus. Also, close contact and association of children with sheep, dogs, and pets, as well as lack of health education and carelessness all lead to a higher pediatric incidence. All of our patients had lived in rural areas and all had direct contact with animals such as dogs, cattle, sheep, and donkeys. All of our patients had drunk raw non-pasteurized milk. The incidence rate of patients from rural areas was 40% in Indian study.⁷

The supratentorial compartment location was the only site found in the present study. The parietal lobe is the most common site, and was seen in 87% of the present series. These results are in agreement with Turgut,³ and Gupta et al.⁷ Our study did not report any hydatid cyst of the posterior fossa, which is a very uncommon site for the disease, and was recorded in one patient by Ahmadi et al.⁶ The more common supratentorial involvement was related to the straight continuation of the internal carotid artery into the

middle cerebral artery, and the higher blood flow in the predominant middle cerebral artery. Ventricular hydatid cyst was not reported in our study, and was reported in one case out of 5 in the Indian series reported by Gupta et al.⁷

Multiple intracranial hydatidosis is a rare disease with serious neurological manifestations, high recurrence, and mortality rates comparable sometimes to malignant disease is exceedingly rare. The causes of multiple infestations and their mechanisms are not clearly understood. Multiple intracranial cysts raise a suspicion that they arise as an embolization from hydatid cysts located within the heart.²⁻⁴ In our series, we reported one patient out of 8 with multiple hydatid cysts, similar to the results of Gupta et al⁷ who reported 40% (2 cases out of 5) with multiple cysts, 40% of Per et al's study,⁵ and in 53 out of 276 of Turgut's study.³ Our study showed one patient out of 8 [25%] had both intracranial and liver hydatid cyst, this figure is compatible with other studies.^{2,7} The duration of the disease in the present study is also comparable with other studies, and may represent the cyst growth rate that varies between 1-10 years.^{2,7} Symptoms of raised intracranial pressure, headache, weakness, vomiting, and seizures were the most common clinical features, and this is compatible with other studies.^{2,7} We had successful complete removal of the cyst using the peri-cystic hydraulic method with only 3 patients developing transient subdural effusion (Figure 1b). All our patients had full clinical recovery, and this rate of success was better than reported in Ahmadi et al's series,⁶ and by Turgut et al.³

In conclusion, hydatid cyst was more common in the childhood age group, male gender, and in rural people. In our series, the most common site was in the supratentorial location, and mostly in the parietal lobe. We had only one case with multiple cysts. There was

no record of any other unusual sites. Finally, surgical treatment by craniotomy and total cyst removal with hydraulic pressure combined with more than 3 months Albendazole oral therapy should be adopted as the optimum therapy for brain hydatid cysts.

Received 9th October 2012. Accepted 6th January 2013.

From the Department of Neurology (Hasan) and the Department of Community Medicine (Assad), Alkindy College of Medicine, Baghdad University, and the Department of Neurosurgery (Sagban), and the Department of Neurology (Hatim), Neurosciences Hospital, Baghdad, Iraq. Address correspondence and reprint requests to: Dr. Zaki N. Hasan, Professor of Neurology, Department of Neurology, Alkindy College of Medicine, Baghdad University, Al-Nahdha Square, PO Box 47188, Jadriya, Baghdad, Iraq. Tel. +964 7706067660. E-mail: zaki_nooh@yahoo.com

References

1. Bekçi TT. Diagnosis and Treatment of Human Hydatid disease. *Eur J Gen Med* 2012; 9 (Suppl 1): 15-20.
2. Vishal K, Vibhuti. Neuroimage--hydatid cyst of brain. *J Assoc Physicians India* 2010; 58: 173.
3. Turgut M. Intracranial hydatidosis in Turkey: its clinical presentation, diagnostic studies, surgical management, and outcome. A review of 276 cases. *Neurosurg Rev* 2001; 24: 200-208.
4. Nourbakhsh A, Vannemreddy P, Minagar A, Toledo EG, Palacios E, Nanda A. Hydatid disease of the central nervous system: a review of literature with an emphasis on Latin American countries. *Neurol Res* 2010; 32: 245-251.
5. Per H, Kumandas S, Gümüs H, Kurtsoy A. Primary soliter and multiple intracranial cyst hydatid disease: report of five cases. *Brain Dev* 2009; 31: 228-233.
6. Ahmadi NA, Badi F. Human hydatidosis in Tehran, Iran: a retrospective epidemiological study of surgical cases between 1999 and 2009 at two university medical centers. *Trop Biomed* 2011; 28: 450-456.
7. Gupta S, Desai K, Goel A. Intracranial hydatid cyst: a report of five cases and review of literature. *Neurol India* 1999; 47: 214-217.

COPYRIGHT

Whenever a manuscript contains material (tables, figures, etc.) which is protected by copyright (previously published), it is the obligation of the author to obtain written permission from the holder of the copyright (usually the publisher) to reproduce the material in Neurosciences. This also applies if the material is the authors own work. Please submit copies of the material from the source in which it was first published.