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Original Article

Successful surgical management of intracerebral hydatid cyst in children: timing, procedure, and adjuvant treatment

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Abstract

Background
Cerebral hydatid cysts are very rare, and surgery, with its inherent risk of rupture and spillage of cyst contents, has been the traditional treatment. The goal of surgery is to remove the cysts without rupture.

Aim
The aim of this review was to investigate the surgical technique of removing cerebral hydatid cysts in children, to show the possible pitfalls of surgery and to assess the effect of adjuvant preoperative and postoperative medical treatment.

Patients and methods
This retrospective analysis included four patients who underwent surgery for an intracranial hydatid cyst at the Department of Neurosurgery, Matarya Teaching Hospital. The Dowling technique was used in all patients. Preoperative and postoperative treatment with albendazole was added in all cases.

Results
One patient had multiple hydatid cysts within a very thin membrane and all were removed intact, and the other three patients had solitary cyst and all were removed intact by Dowling technique. Anaphylactic reaction or chemical meningitis did not occur. The thin cyst wall, periventricular locations were the main surgical problems in two cases, and microadhesions to the surrounding brain tissue were other obstacles during the removal of the other two cysts.

Conclusion

Most intracranial hydatid cysts are seen in the pediatric population. Dowling's technique is the most effective method of surgical removal of the cysts, and recovery depends on successful cyst extraction without rupture.

Keywords: Albendazole, hydatid cyst, multiple intracranial cyst, primary, surgery

Introduction

Hydatid disease is a common parasitic disease in the highly endemic countries of North Africa, Middle East, and Central Asia. The pathogenic agent is hexacanth embryo of Echinococcus granulosus. It is accidentally transmitted to humans either through the ingestion of infected food, or following direct contact with a definitive host, usually a dog [1–5].

Brain involvement is rare and occurs in 1–2% of all cases of hydatidosis. Overall, 50% to 75% of intracranial hydatid cysts are seen in children [6].

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Hydatid cysts of the brain are usually single, spherical, unilocular, and may be large; in rare instances, they can be multiple and embolic. Although an intracranial single lesion is nearly always primary, multiple lesions are frequently secondary. Multiple hydatid cysts resulting from the rupture of

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a primary cyst are acephaloceles; they are infertile and have no broad capsule. However, very rarely, a multiple larval intake may cause primary multiple cerebral hydatid cysts [2].

Because of its rarity, experience with intracranial hydatid disease at a single institution has been very limited, and the Dowling technique is widely used as a surgical treatment [7].

Preoperative and postoperative albendazole may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall, and hence reduce the risk of spillage during surgery and the recurrence rate [8].

Patients and methods

Patient population

This retrospective analysis includes four patients with cerebral hydatid cysts operated between 2012 and 2019 in the Department of Neurosurgery, Matarya Teaching Hospital. Three children of 9, 10, and 12 years old were males, and a young female of 14 years old. The symptoms and clinical signs have been reviewed.

Preoperative evaluation

The first child of 9 years old presented with manifestations of increased intracranial pressure (e.g. drowsiness, repeated vomiting unrelated to meals, headache, blurring of vision, and papilledema on fundus examination) starting 2 weeks before seeking for medical advice and exacerbating 3 days before admission. The female child presented also with manifestations of increased Intracranial pressure (ICP) hydatic cyst (HC) started 5 weeks before admission. The second (10 years old) and third (12 years old) male children presented with epileptic seizures several months before admission, which were controlled by antiepileptic after being completely investigated in the neuroscience ICU.

After clinical evaluation a computed tomography (CT) brain and MRI were obtained and the diagnosis of cerebral hydatid cyst was confirmed. Multiple cysts were confirmed in one case (the boy of 9 years old), mostly included within the same membranous coverage (Fig. 1), and solitary cysts were present in the other cases. Chest radiographies and abdominal and cardiac ultrasounds were performed to find any other visceral localization.

Adjuvant medical therapy (albendazole) was prescribed to all cases before surgery according to the body weight. The suggested dose is 10 mg/kg/day in four 1-month courses, separated by 15-day intervals.

Surgical timing and procedure

The timing of surgery was based on the neurological and neuroimaging findings in the patients. Early cyst extirpation was reserved for giant hydatid cysts that caused severe neurological deficits, and late extirpation performed in patients who had relatively small cysts and needed to undergo further evaluation for the differential diagnosis. The first child (9 years old) was taken for surgery after 48 h because

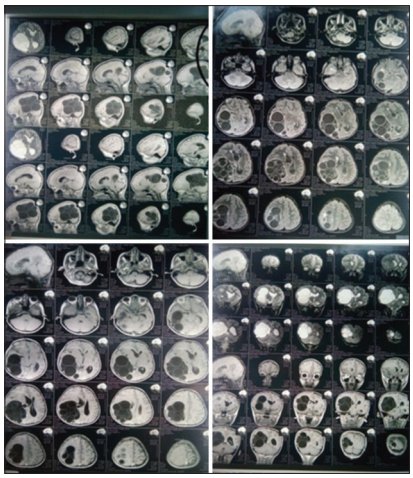

Figure 1: Preoperative MRI of a 9-year-old male child presented with drowsiness and repeated vomiting unrelated to meals and blurring of vision living in direct contact with dogs.

of the patient clinical status of increased ICP, and the female child was operated upon after the first week of the medical treatment, whereas the two other cases were operated upon after completing the first cycle of treatment. All cases provided written and spoken informed consent before surgery.

Surgical procedure depended mainly on accurate studying of the preoperative radiological investigations and accurate localization of the lesion. A wide craniotomy was performed in all cases depending on the site of the lesion which was right parietooccipital in the 9-year-old child and the female child, left frontoparietal (parasagittal) in the 12-year-old child, and right frontal in the last one.

Moist cottonoid strips were inserted around edges of the craniotomy before dural opening, and others were inserted on brain surface after dural opening around the suspected cyst site. Microinstruments were used, and great care was taken during the dural opening of these cysts, because in many cases, there is no cortical tissue between the cyst wall and the dura.

Meticulous cortical dissection was performed to reach the cyst wall following the dural opening. Microtechniques and microinstruments were used during the dissection.

The Dowling technique has been widely used for the excision. A soft rubber catheter was inserted between the hydatid cysts and surrounding brain tissue, and warm saline was injected through this catheter. The position of the operative table was modulated to facilitate cyst extirpation. This technique was successful in excising all cysts without any intraoperative complications, for example, rupture except in excising the cyst of left frontoparietal (parasagittal location), whereas microadhesions make it difficult to remove the cyst by this technique only, and meticulous microdissection was needed to remove the cyst after meticulously inserting moist cottonoid strips between the cyst wall and the surrounding brain tissue.

Postoperative management

CT scanning was performed in all patients routinely on the

early postoperative days during the first week. Postoperatively, all patients received albendazole 10 mg/kg twice daily for 3 months. Two patients underwent follow-up for more than 1 year after the operation with the aid of serial CT scans and MRI with no evidence of recurrence, and two patients are still in early follow-up.

Results

Four patients with cerebral hydatid cysts underwent surgical treatment during a 7-year period, and they had a mean follow-up period of 12 months (range: 6–20 months). Two cases presented with manifestations of increased intracranial pressure, mainly headache, and two cases presented with seizures. The mean duration of the symptoms was 8 weeks (range: 1–15 weeks).

The hydatid cyst was located in the left cerebral hemisphere in one patient and the right hemisphere in three patients. The hydatid cyst was solitary in three patients and multiple in one patient.

Early operation was performed in two patients who had giant hydatid cysts that produced worsening neurological symptoms, and late operation was reserved for two patients whose conditions were relatively good. The Dowling technique was used as the surgical method in all cases. The hydatid cysts were removed unruptured in all cases. Whole body scanning showed no cysts elsewhere in the body. No recurrence of cerebral hydatid cyst noted in any of the cases taking in mind early follow-up in two of them. None of the patients died after surgery (Figs. 2–5).

Discussion

Hydatidosis is caused by *Echinococcus granulosis*. It occurs commonly in dogs, primary host. Intermediate hosts like man get infected accidentally by ingestion of feces. The eggs hatch in the intestine and burrow through to the portal circulation. The embryos that survive are able to survive in many tissues most commonly liver and lungs. Even after the pulmonary filter, a few still make it to the systemic circulation and reach any tissue such as brain, heart, and bones [9].

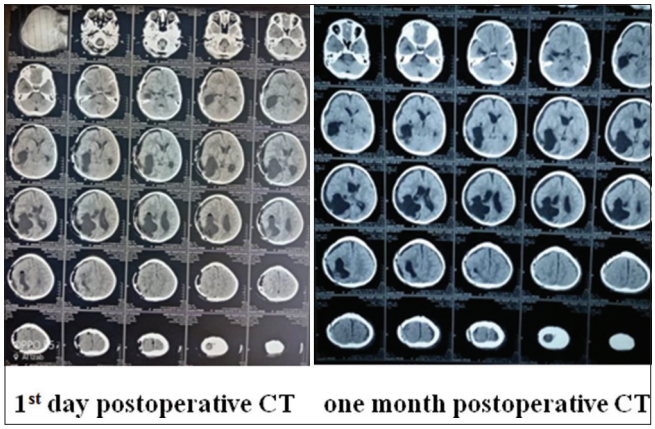

Figure 2: Postoperative computed tomography brain in the first day and after 1 month.

Recently, a WHO strategic plan (2008–2015) added the disease to the list of neglected tropical diseases, which is a collective term used for diseases sharing a few common characteristics such as being more common in tropical countries and that have not received enough importance at national or international levels [10].

Patients with cerebral HCs may also have cysts in other organs. Less than 20% of the patients with intracranial hydatidosis demonstrate other organ involvement.[11] Tuzun *et al.*[12] stated that other organ involvement with brain HC reaches up to 80–90% in postmortem examinations, whereas in clinical practice, concomitant extracranial cysts were not often demonstrated.

Cerebral hydatid cysts are more common in the pediatric population; 50–75% of cerebral hydatid cysts occur in children, and they are more frequently located in the supratentorial region. The parietal lobe is the most popular site for such cysts, because the parasites are usually distributed in the watershed of the middle cerebral artery [13]. Other reported sites included the subarachnoid space, the ventricles, the pons, the cerebellum, the aqueduct of Sylvius, the extradural space, and the diploic space of skull bones [14–16].

The development of symptoms is slow, neurological deficits appear late and are often preceded by signs of increased intracranial pressure. Other symptoms, such as weakness in the limbs and gait disorders, may vary with the location of the cyst [17].

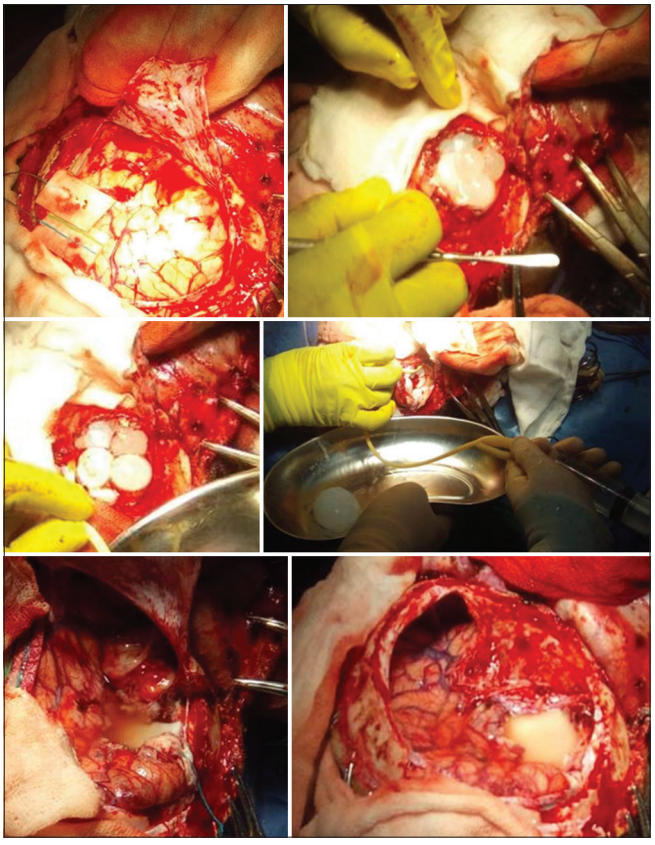

Figure 3: Surgical steps for the removal of the multiple hydatid cysts.

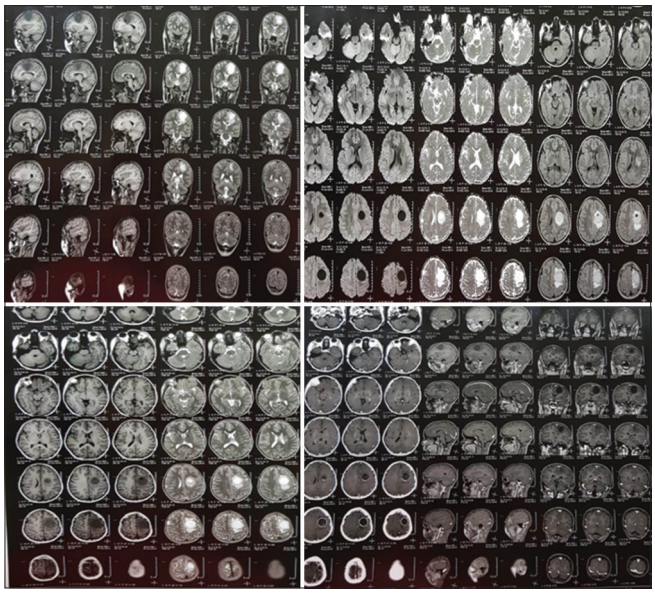

Figure 4: Preoperative MRI of a 12-year-old male child presented with epileptic seizures.

In children and adolescents, the clinical picture usually includes the cardinal symptoms of increased intracranial pressure, whereas focal findings such as hemiparesis, speech disorders, and hemianopsia, sometimes associated with epileptic seizures, are more prevalent in the older age group [18].

Cerebral HC should be considered in children with cystic brain lesions even in nonendemic areas. As serological tests of brain HC frequently yield false-negative results due to an inadequate immune response, the most reliable methods for reaching a diagnosis are radiological and histopathological examinations [19,20].

The preoperative diagnosis is important for surgical planning and successful outcome. The diagnosis can be made using cranial CT scanning or MRI, which may provide signs of a hydatid cyst specific enough to achieve a definitive diagnosis [7]. MRI is superior to CT for demonstrating neural involvement, multiple cysts, cyst capsule detection, and in delineating the relationship of the cyst with adjacent structures [1].

Recently, a MRI classification was proposed for cerebral CHD on the lines of the WHO categorization of hepatic E. granulosus cysts. This categorized the cysts on the basis of their fertility, activity, and imaging morphology. Type 1 cystic echinococcosis (CE1) includes fertile active cysts that appear unilocular and spherical with a clear visible wall. Type 2 cysts (CE2), which are also active, are unilocular mother cysts containing multiple vesicles arranged peripherally along the cyst wall. Type 3 (CE3) are transitional forms containing scolices and are seen as maternal cysts entirely filled by multiple daughter cysts. Type 4 (CE4) cysts show detached membranes giving the water-lily sign, and type 5 (CE5) are calcified lesions. Both CE4 and CE5 are inactive cysts that have lost their fertility [21].

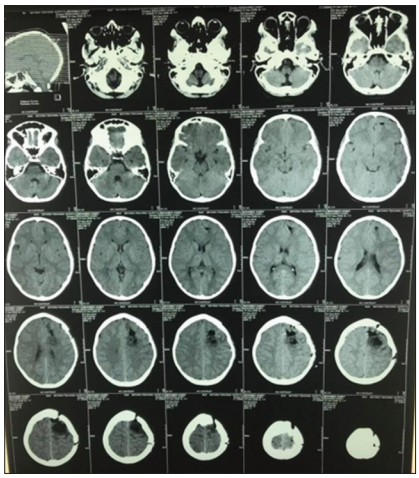

Figure 5: One-week postoperative computed tomography brain.

The treatment of cerebral hydatid cysts is principally surgical. The primary goal of the operation is total cyst extirpation without rupture [6].

Many different techniques of cyst removal have been proposed, and all of them emphasize atraumatic techniques to avoid cyst rupture. The Dowling technique has been widely used for the surgical treatment of hydatid cysts of the central nervous system [7].

Compared with mebendazole, albendazole is a better treatment option for hydatid disease, because of its better resorption [22]. Albendazole can be beneficial for inoperable patients, with multiple cysts, and in the lungs and liver hydatid disease [23,24]. It can be administered orally at a dosage of 10–15 mg/kg/day, and administration should be continuous without interruptions. However, the optimal dosage of albendazole and optimal duration of treatment are still unknown [25].

In this review, we wanted to share our limited experience on surgical management of these of four cases of hydatid cyst and the role of adjuvant medical treatment with Albendazole.

We started medical treatment in all cases preoperatively once diagnosis was evident. The duration of preoperative treatment depended on the clinical presentation and the need to rapidly decompress the brain. The suggested dose is 10 mg/kg/day in four 1-month courses, separated by 15-day intervals. Postoperative adjuvant treatment continued for three months in all patients without any drug-related complications.

The Dowling technique with cautious insertion of soft rubber catheter between the cyst and surrounding brain tissue with slow injection of normal saline to facilitate cyst delivery from surrounding brain tissue was our surgical technique in all cases, and it was successful. Moreover, cyst removal was assisted by changing the position of the head.

Adhesions around the cyst wall can be troublesome. During the management of our cases, we faced that during the surgical excision of the frontoparietal parasagittal hydatid cyst in the 12 years old male child. Venous bleeding was encountered

during microdissection and only required gentle tamponade by cottonoid.

The hydatid cyst may be giant in size, and there may be no cortical tissue between the cyst wall and dura. Meticulous dural opening with the meticulous microdissection is required in such cases to avoid any damage to the wall of cysts that lie just below the dura.

The most important factor in determining prognosis is intact removal of the cysts.[26] The intraoperative rupture rates for intracranial hydatid cysts were 16.9% and 25.6% in the cooperative study and in the literature survey conducted by Altinors *et al.* [27]. The accepted procedure in cases of intraoperative rupture is aspiration of the cyst contents, extirpation of the cyst wall, and as a precaution, extensive irrigation of the surgical field with hypertonic solution to prevent recurrence [27,28].

The reported recurrence rate in the brain due to cyst rupture at surgery was 40.7% [29]. It was recommended that patients should be followed up regularly to recognize possible recurrence, dissemination, or complications of the disease [19].

Successful surgical excision was achieved in all cases without any complications apart from mild right-sided hemiparesis (mainly upper limb) in the parasagittal one which resolved after a short course of physiotherapy. No patient developed anaphylaxis. No recurrences were noted during the follow-up taking in mind the short follow-up period of the last two cases.

Owing to the vast empty space remaining after removal of large cysts, stretching of the bridging veins may lead to subdural hematoma or subdural effusion. Spontaneous recovery may occur in most cases, but decompression surgery will be required in some cases. A large cavity that remains after removal of large cysts can lead to severe complications such as cortical collapse, hyperpyrexia, cerebral edema, and cardiopulmonary failure.

Following the removal of the cyst and the rapid decline of intracranial pressure, fatal complications such as pontine hemorrhage have been reported.

Conclusion

Despite the advancements in microsurgical operative techniques and instrumentation, cerebral hydatid cysts pose a challenge for the surgeon because of the following characteristics: they are usually diagnosed when they are large in size; they have a very thin cyst wall; the neurological deficits are often minimal in their presentation despite the location and the large size of the cyst; and they are sometimes located deep or near the ventricular wall and require retraction of vital structures or meticulous cortical dissection and good outcome is desired.

Successful treatment of cerebral hydatid cysts requires thorough knowledge of the potential pitfalls and keys to avoid intraoperative mistakes and cyst rupture. Meticulous dissection and delicate separation of the cyst wall from the adjacent cortex provide excellent results.

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Conflicts of interest

There are no conflicts of interest.

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