Cerebral hydatid cysts: technique and pitfalls of surgical management

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Object. Hydatid cysts are rare, but most often they occur in the liver and lungs. Cerebral manifestation is very rare, and surgery is the main treatment. The goal of surgery is to remove the cysts in toto without rupture. The aim of this study was to investigate the surgical technique of removing cerebral hydatid cysts and to show the possible pitfalls of surgery.

Methods. This study included 17 patients who underwent surgery for an intracranial hydatid cyst. The Dowling technique was used in all patients. This technique is based on the large cranial opening, careful handling, meticulous cortical dissection, and removal of the cyst by hydrostatic assistance.

Results. The hydatid cysts were removed unruptured in 88% of the cases. The cysts ruptured intraoperatively in 2 patients, and the ventricular system was opened in 1 of them. Anaphylactic reaction or chemical meningitis did not occur, but recurrence and spinal seeding were observed in the follow-up of these patients. The thin cyst wall, periventricular locations, and microadhesions to the surrounding brain tissue were the main surgical problems. None of the patients died after the surgery.

Conclusions. Although this technique seems safe and easy, there are some pitfalls concerning the cyst location and surgical approach. Successful management requires a flexible therapeutic strategy and meticulous dissection by an experienced surgeon. (DOI: 10.3171/FOC/2008/24/6/E15)

Key Words • brain • hydatid cyst • pitfall • surgery

Human echinococcus is caused by a tapeworm, Echinococcus granulosus, which forms larval cysts in human tissue. Humans acquire the infection by ingesting the eggs. Once swallowed, the eggs hatch embryos that invade the bloodstream and pass via the portal vein to the liver where most larvae are entrapped and encysted. Some may reach the lungs and develop into pulmonary hydatid cysts. This is an endemic manifestation that is more frequently found in Mediterranean countries, the Middle East, South America, and Australia, and this entity affects particularly those involved with sheep and cattle raising. Brain involvement is rare and occurs in 1–2% of all cases of hydatidosis. Fifty percent to 75% of intracranial hydatid cysts are seen in children. Because of this 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. Preoperative and postoperative albendazole may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery), and reduce the recurrence rate.

We retrospectively reviewed our experience with cerebral hydatid cysts and attempted to show the pitfalls of surgical treatment. Our goal was to ascertain whether this technique is the optimal method for the treatment of cerebral hydatid cysts.

Clinical Materials and Methods

Patient Population

Between 1993 and 2007, 17 patients underwent surgical management for cerebral hydatid cysts at the Departments of Neurosurgery of Gulhane Military Medical Academy and Atatürk University. Informed consent was obtained from all patients or from the parents of the patients. The patient population consisted of 13 children (65%) and 4 adults (24%) with cerebral hydatid cysts. Eleven patients (65%) were male and 6 (35%) were female, with a mean age of 13.6 years (range 3–21 years).

Preoperative Assessment

After recording of the initial complaints, all patients were examined by a team of neurosurgeons and pediatricians (for children). They had previously undergone a com-
Complete investigative workup including complete blood analysis and plain x-ray studies.

All patients underwent neurological and neuroimaging evaluation during the preoperative period. For neuroimaging assessment, CT scans and MR images were obtained. Patients were assessed before and just after the operation by using CT scanning and in the final follow-up by using MR imaging. Preoperative MR images were obtained in patients who had cystic lesions observed on the initial CT scans (Fig. 1). Once the cyst was detected on neuroimages, serological tests such as enzyme-linked immunosorbent assay or indirect hemagglutination were applied for the diagnosis. Abdominal ultrasonography and chest CT scanning were performed after the diagnosis of hydatid disease during either the preoperative or the postoperative period. Medical treatment with albendazole commenced preoperatively to inactivate protoscolices and continued during the postoperative period.

**Surgical Technique**

Surgery was performed in all patients. The timing of surgery was based on the neurological and neuroimaging findings in patients. Early cyst extirpation (within 24 hours) was reserved for giant hydatid cysts that caused severe neurological deficits, and late extirpation (> 24 hours) was performed in patients who had relatively small cysts and needed to undergo further evaluation for the differential diagnosis.

After induction of general endotracheal anesthesia, exposure was gained through a curvilinear skin incision appropriate for the cyst location. After cutting the periosteum, a large craniotomy was performed with an air-powered drill and a craniotome. The bone was then carefully separated from the underlying dura mater, which can be very fragile. Cyst fluid was observed in some patients on illumination when the cranium was opened. 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. If the cyst was deeply located, the dura was more easily opened. Under microscopic visualization, meticulous cortical dissection was performed to reach the cyst wall following the dural opening. Microtechniques and microinstruments were used during the dissection. Because the cyst wall was fragile, moist cottonoid strips were gently inserted around the overlying cyst wall. Every attempt was made to avoid retraction on the cyst wall. Adhesions around the cyst wall were gently cut. Once the entire cyst wall had been exposed, fine plastic tubes were inserted around the wall. Saline solution (0.9%) was used to dissect the wall from the brain parenchyma and to facilitate the hydatid removal (Fig. 2). The operation table was tilted toward the lesion site, and the head of the patient was lowered to make the cyst removal easy with the help of gravity. Valsalva maneuver and counterpressure to the surrounding brain were also used to aid the delivery of the cyst. A small bowl was used to collect the cysts (Fig. 3). Aspiration or irrigation of the cyst before extirpation was not performed, but the area was irrigated with 20% saline in case of an unintentional cyst rupture. After the removal of the hydatid cyst, the dura was closed in a watertight fashion.
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Postoperative Management

Computed tomography scanning was performed in all patients routinely on the 1st postoperative day. Postoperatively, all patients received antihelminthic agents such as albendazole 10 mg/kg twice daily for 3 months.

All patients underwent follow-up for ≥ 1 year after the operation with the aid of serial CT scans and MR imaging. Patients who had hydatid cysts in other organs such as liver, kidney, or spleen were referred to the Departments of Surgery and Pediatric Surgery for further treatment.

Results

Seventeen consecutive patients with cerebral hydatid cysts underwent surgical treatment during a 14-year period, and they had a mean follow-up period of 14 months (range 12–24 months). Headache was the most presenting symptom (12 patients [70.5%]), followed by weakness in the extremities in 7 (41%). The mean duration of the symptoms was 7.5 weeks (range 1–24 weeks). The hydatid cyst was located in the left cerebral hemisphere in 10 patients (58.8%) and the right hemisphere in 7. The hydatid cyst was solitary in 16 patients (94%) and multiple in 1.

Early operation was performed in 5 patients (29%) who had giant hydatid cysts that produced worsening neurological symptoms, and late operation was reserved for 12 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 15 cases (88%). Intraoperative cyst rupture occurred in 2 patients, and the cyst site was irrigated with 20% saline. The cyst fluid was drained into the ventricular system in 1 patient because the ventricular wall was accidentally opened. Anaphylactic reaction or chemical meningitis did not occur. Whole-body CT scanning showed that 7 patients (41%) had cysts elsewhere in the body including the kidney, liver, and lung. Recurrence of cerebral hydatid cyst and spinal seeding were observed in only 2 patients (11.7%) who experienced intraoperative cyst rupture, and they underwent reoperation for cyst extirpation. None of the patients died after surgery, and all of them underwent follow-up with the aid of serial CT scanning and MR imaging. The summary of all cases is reported in Table 1.

Discussion

We have presented our surgical experience with 17 cases of cerebral hydatid cysts. We particularly focused on the surgical technique and tried to show the difficulties and pitfalls of surgery.

The preoperative diagnosis is important for surgical planning and successful outcome. The diagnosis can be made using cranial CT scanning or MR imaging, which may provide signs of a hydatid cyst specific enough to achieve a definitive diagnosis. A large intraparenchymal cystic lesion with a well-defined border is the usual appearance on CT scans of the cranial hydatid cyst. The cyst fluid is generally isodense or slightly hyperdense compared with cerebrospinal fluid. Rim enhancement and pericystic edema are less common than in other cystic lesions. Cyst calcification may seldom occur. On T2-weighted MR images, the hydatid cyst appears as a cystic lesion with a hypointense halo around the cyst capsule.

The treatment of cerebral hydatid cysts is principally surgical. The primary goal of the operation is total cyst extirpation without rupture.12 Many different techniques of cyst removal have been proposed and all of them emphasize atraumatic techniques to avoid cyst rupture. The Dowling technique,3 later improved by Arana-Iniguez and San Julian,2 has been widely used for the surgical treatment of hydatid cysts of the central nervous system. The essential steps of this technique are the following: creation of a large flap; careful handing during all operative steps to avoid monopolar coagulation; opening the atrophic cortex overlying the cyst over an area with a diameter no less than three quarters of the diameter of the cyst; and allowing the cyst to fall out by just lowering the head of the operating table and flushing warm saline between the cyst and surrounding brain.25

Despite the advancements in microsurgical operative techniques and instrumentation, cerebral hydatid cysts pose

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Clinical Presentation</th>
<th>Cyst Location &amp; Diameter (in cm)</th>
<th>Other Location</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>16, F</td>
<td>rt weakness</td>
<td>lt occipital &amp; parietal, 2 &amp; 1.5</td>
<td>spleen, kidney</td>
<td>rupture</td>
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<td>2</td>
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<td>headache</td>
<td>lt parietal, 3</td>
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<td>good</td>
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<tr>
<td>3</td>
<td>9, F</td>
<td>headache, nausea, vomiting</td>
<td>rt occipital, 2.5</td>
<td>liver</td>
<td>good</td>
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<tr>
<td>4</td>
<td>28, M</td>
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<td>rt posterior parietal, 3</td>
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<tr>
<td>6</td>
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<td>lt parietocerebral, 3.5</td>
<td>kidney, liver</td>
<td>subdural effusion</td>
</tr>
<tr>
<td>7</td>
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<td>rt weakness</td>
<td>lt parietocerebral, 3</td>
<td>liver</td>
<td>good</td>
</tr>
<tr>
<td>8</td>
<td>21, M</td>
<td>headache, visual blurring</td>
<td>lt occipital, 6</td>
<td>none</td>
<td>good</td>
</tr>
<tr>
<td>9</td>
<td>16, F</td>
<td>headache, nausea, vomiting</td>
<td>rt parietocerebral, 2.5</td>
<td>none</td>
<td>good</td>
</tr>
<tr>
<td>10</td>
<td>11, F</td>
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<td>lt temporalparietal, 3</td>
<td>none</td>
<td>subdural effusion</td>
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<tr>
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<td>rt frontal, 3</td>
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<td>subdural effusion</td>
</tr>
<tr>
<td>13</td>
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<td>lt parietal, 2.5</td>
<td>none</td>
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<td>lt parietal, 3</td>
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<tr>
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<td>20, M</td>
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<td>rt parietal, 3.5</td>
<td>liver, lung</td>
<td>good</td>
</tr>
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<td>18, M</td>
<td>headache, rt weakness</td>
<td>lt frontoparietal, 4</td>
<td>liver</td>
<td>good</td>
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</table>
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.

There are many reports suggesting that the Dowling technique is the most effective surgical procedure for the removal of cerebral hydatid cysts. However, some pitfalls exist with this technique concerning surgical methods, instruments, and cyst location. The best operative approach to a cystic lesion in the brain should be based on the site and the size of the cyst, and the relationship of the lesion with the other neural and vascular structures.

It is impossible to reach a cerebral hydatid cyst without opening some neural structures if the cyst is not immediately below the dura. The surgical approach to the hydatid cyst may require cortical incisions, and the brain may be retracted to expose an external wall of the cyst. The incision and retraction of the cerebral cortex to reach the cyst wall may produce variable results; in some cases there has been no deficit, and in others the deficit has been transient or permanent. Sometimes the cortex and cyst wall were not clearly separated. At this point, a cottonoid is placed on the cyst wall to provide protection and to maintain the identification of this cleavage plane during cyst removal.

Adhesions around the cyst wall can be troublesome. During the release of these adhesions, venous bleeding is not often encountered and only requires gentle tamponade or meticulous use of bipolar cautery for control. It is imperative to properly address each bleeding source because one cannot safely separate the cyst wall under a pool of blood. Cutting of these adhesions with microsurgical tools is essential.

The hydatid cyst may be located close to the lateral ventricle wall, which is termed periventricular hydatid cyst. The cyst wall may be attached to the ventricle wall. This is a very dangerous location, and particular attention should be paid during the removal of these cysts. In those cases, a safe plane of cleavage must be developed and the ventricle wall should be preserved. If a tear occurs in the cyst wall, it may prematurely rupture. This is quite a catastrophic event if the cyst fluid mixes with cerebrospinal fluid that is released through the ventricular opening. In these cases, the portion of the cyst wall, which is attached to the ventricular wall, should not be handled with force, and saline solution should not be used at this point to kill the larvae because it could cause an anaphylactic reaction, chemical meningitis, subsequent death, or serious neurological deficits. Although aspiration may facilitate the removal of these cysts, it carries the risk of contamination if contents are spilled.

The hydatid cyst may be giant in size and there may be no cortical tissue between the cyst wall and dura. This situation may be observed on preoperative neuroimages or on illumination after the craniotomy. Meticulous dural opening with fine microscissors is required in such cases to avoid any damage to the wall of cysts that lie just below the dura. The technique for treating multiple or deep-seated hydatid cysts is not different, but some modifications are needed. In the case of multiple cerebral hydatid cysts, the largest one must be targeted first, and then the other cysts must be removed during the same session. Staged surgery is an alternative for multiple hydatid cysts. The position of the head may be changed during the removal of each cyst. Deep-seated hydatid cysts must be removed via a cortical approach to prevent rupture. The risk of rupture in multiple or deep-seated hydatid cysts is high when compared with single and superficial cysts.

Safety is enhanced during the removal of the cyst if adequate exposure is obtained. The access corridor to the cyst must be maintained and protected throughout the removal process, but not at the expense of trauma due to further dissection or manipulation once the corridor has been established. Because of the depth of the operative field, the operating microscope is uniformly used for magnification and illumination.

Although the principal treatment is surgery, pre- and postoperative administration of albendazole may be considered to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall, and reduce the recurrence rate. Corticosteroids may be used to reduce pericystic edema preoperatively. We placed all patients on a regimen of albendazole postoperatively (10 mg/kg twice daily for 3 months), but we did not prescribe corticosteroids.

**Conclusions**

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

**References**

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