Child's Nerv Syst (1995) 11: 679–686

ORIGINAL PAPER

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Cerebral hydatid cyst in children

Experience of 27 cases

Received: 20 November 1994

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Abstract The authors present 27 cases of cerebral hydatid cyst (CHCy) treated between 1980 and 1992. These cases of CHCy represent 2.8% of all cases of expansive nontraumatic lesions in children. Most of the patients were between 6 and 10 years of age. There was a substantial prevalence of male patients – 18 cases (66.6%). Most of the children with CHCy were from rural areas. The cysts were all located in the cerebral hemispheres (none in the posterior fossa). Usually two or three lobes were affected and the cysts were most often retrorolandic. Only 8 patients (29.6%) also had pulmonary or hepatic infestation. All patients were operated on immediately the diagnosis was established. Operative mortality was very low (1 patient died immediately after surgery). There have been 11 cases of recurrence; all these patients have been reoperated on. Neurological sequelae were mainly partial and general seizures. Epilepsy developed postoperatively in five patients. Paresis, which usually disappears in time, was the most common motor disturbance. Two other important postoperative complications were subdural effusion and ventricular dilatation (six cases). There have been five cases of multiple recurrences (four of these patients have since died). Treatment with albendazole was used in three cases without significant results.

Key words Computed tomography
Cerebral hydatid cyst
Echinococcosis · Albendazole

Introduction

Hydatid disease (echinococcosis) of the nervous system is by definition a neurological disorder caused by damage to nervous tissues by the development of vesicles or cysts, especially in the cerebrum, by the larval stage of the tapeworm *Echinococcus granulosus*. Hydatidosis of the skull or vertebrae is also possible, although rare. In the spinal cord a subarachnoid localization is exceptional. The cerebral hydatid cyst (CHCy) develops in soft tissue and grows without resistance, thus forming a single spherical vesicle which may reach a considerable size before declaring its presence clinically. When this occurs, the symptoms are usually those of raised intracranial pressure, particularly in children. The lesion can be accurately shown by computed tomography (CT) and definitive surgical cure, with complete extirpation of the disease, is possible. CHCy represents the most familiar neurological manifestation of CNS hydatid disease.

In this study we reviewed 27 pediatric patients with CHCy operated on in the period 1980–1992, when CT was available. The 52 patients with CHCy operated on before CT was available (give a total of 79 cases), were not included in the study because some diagnostic errors were made and there was no accurate postoperative follow-up.

Patients

Twenty-seven children underwent surgery for CHCy between 1980 and 1992. The youngest patient was 3 years and 4 months old. The

largest age group was the 6- to 10-year-old group (Table 1), and the mean age was 8.2 years. There was a 2:1 preponderance of boys (18 boys, 9 girls).

Results

Clinical findings

Headache and vomiting were the predominant symptoms (Table 2), followed in order of frequency by hemiparesis, visual disturbance, and seizures. Papilledema was present in 24 patients (88.8%). Secondary optic atrophy was detected in two patients. Only one patient had a normal funduscopic examination. Visual field changes (homonymous hemianopia and quadrantanopia) were frequent findings. Mental changes occurred with disturbances of consciousness, but generally these pediatric patients tended to tolerate the presence of the cyst well, remaining lucid.

Imaging studies

Plain skull radiographs showed highly significant changes: suture diastases and thinning of the vault (especially in children under 7 years of age). Cerebral angiography was done in only five cases and showed an avascular space-occupying lesion. CT is the best method for diagnosis. On CT CHCy is highly characteristic. The compelling abnormality is a large, well-circumscribed lesion, round and several centimeters in diameter. A unilocular cyst is usually in one or two lobes (Fig. 1). We performed CT before and after surgery in all cases.

All patients underwent a chest X-ray and abdominal ultrasound to reveal any hydatid disease in the lung and liver. In eight children presenting with CHCy, other areas of dissemination of hydatid disease were found on admission.

Cyst sites

The sites of the lesions were as follows: one lobe affected, 7 cases (group 1); 2 lobes affected, 11 cases (group 2); 3 lobes affected, 8 cases (group 3); multiple CHCy, 1 case (group 4; Table 3). In the 8 cases in group 3 the cysts were of giant dimensions. This element had a strong influence on the possibility of total removal and also on the rates of recurrences and postoperative complications.

Operative findings

All patients were operated on. In the majority, a large craniotomy and wide cortical incision were performed and warm saline solution was injected through one or two cath-

Table 1 Distribution by age group of 27 children with cerebral hydatid cysts

Age	No. of cases	%
0– 3 years	0	–
3– 5 years	8	29.6
6–10 years	11	40.8
11–16 years	8	29.6
Total	27	100

Table 2 Neurological findings in 27 children with cerebral hydatid cysts

Neurological signs	No. of cases	%
Increased ICP (headache and vomiting)	26	96.2
Hemiparesis	19	70.3
Visual field modification	12	44.4
Papilledema	24	88.8
Secondary optic atrophy	2	7.4
Seizures	6	22.2
Ataxia	3	11.1
Involuntary movements	2	7.4
Somnolence	1	3.7

Table 3 Site of cerebral hydatid cysts

Group	Site	No. of cases	%
1	Parietal	3	
	Frontal	2	
	Temporal	2	
	Occipital	0	
		7	25.9
2	Frontotemporal	4	
	Frontoparietal	2	
	Parietotemporal	5	
		11	40.7
3	Frontoparietotemporal	4	
	Temporoparietooccipital	3	
	Hemisphere	1	
		8	29.6
4	Multiple	1	3.7
Total		27	100

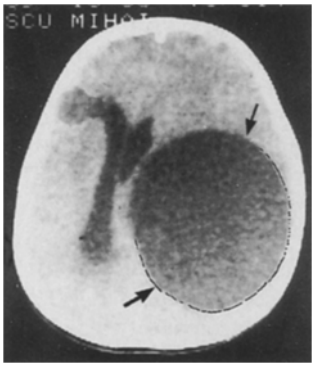

Fig. 1 Computed tomographic (CT) scan: unilocular right temporoparietal giant cerebral hydatid cyst (CHCy)

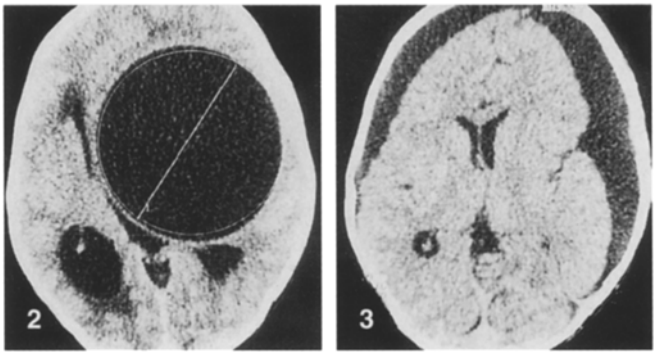

Fig. 2 CT scan: unilocular left temporoparietooccipital giant CHCy

Fig. 3 CT scan: same patient as in Fig. 2 after placement of a subdural shunt

eters to remove the cysts without rupture. We achieved total removal of the cyst without rupture in 17 patients (63 %). In 10 patients (37 %), the cysts ruptured during surgery but were immediately suctioned and total removal was achieved. Usually the rupture of the cyst occurred in the cases in which the lesion was very large. In all cases the operative area was irrigated with hypersaline solution when the cyst ruptured at surgery.

Complications

Motor disturbances is the form of preoperative hemiparesis were found with a relatively high frequency (70.3 %), but this usually disappeared in time, while reappearance represented a sure sign of cyst recurrence. Postoperatively epilepsy developed in five children who had not had convulsions preoperatively; the convulsions were controlled by antiepileptic therapy. Postoperative seizures occurred mostly in the patients who suffered recurrences. Among ocular complications were visual field alterations that have not disappeared postoperatively (six cases).

Subdural effusion and ventricular dilatation were observed on the postoperative CT scans of six patients. Subdural effusion (three cases) was treated by a subdural-peritoneal shunt (Figs. 2, 3). For hydrocephalus appearing after the total resolution of subdural effusion, a ventriculoperitoneal shunt was inserted (two cases).

After the removal of a large CHCy, a porencephalic cyst developed in five children. A cyst-peritoneal shunt was placed in three cases because the porencephalic cysts induced the symptoms of intracranial hypertension.

Superinfection of the operated zone occurred in one patient operated on for multiple CHCy. One patient died immediately after surgery, because the cyst was adherent to the superior longitudinal sinus, and when sudden decompression occurred, massive venous bleeding began.

Recurrences

There were 11 recurrences (40.7 %). These include all cases in which total removal of the CHCy could not be achieved. Insemination of cerebral tissue with daughter cysts and hexacanth embryos is extremely dangerous and produced recurrences situated either in the bed of the primary cyst or in the cerebral ventricles by way of CSF-pathway dissemination. All patients with recurrences were reoperated on. Five of them underwent multiple operations, and four of these five died despite all efforts with local application of hypersaline solutions.

Medical therapy

Three patients in whom the CHCy ruptured at surgery or who had systemic hydatid disease received albendazole treatment. Albendazole prolonged the interval between operations in all patients with recurrences.

Discussion

Epidemiology and geographic distribution

Hydatid disease is endemic in sheep-raising regions of the world, notably South America, New Zealand, Australia, the Middle East, and the Mediterranean area. In all series, including ours (22 cases, 81.5 %), most of the children with CHCy were from rural areas.

Pathophysiology and hydatid infestation

The adult tapeworm, *Echinococcus granulosus*, is to be found in the gut of the dog. Dogs are infected by eating uncooked sheep meat containing hydatid cysts. Water, plants, and the skin of the dog may become contaminated with the eggs of tapeworms; children who often play with them are most exposed to infestation. The alimentary tract is the usual path of infection in man. The eggs lose their enveloping layer in the stomach, after which the hexacanth embryos can pass through the wall of the gut, principally the duodenum and the small intestine, thence passing into the veins of the portal system. Most of them are trapped in the liver, but the embryos can pass through any vessel admitting a red corpuscle, and may thus reach the right side of the heart and thence the lungs. They may remain in the pulmonary capillary network, but some pass even this barrier to reach the left side of the heart and the systemic circulation. Once the hexacanth embryo has arrived in a tissue it will form the hydatid cyst. The brain offers ideal conditions for the growth of the cyst, and daughter cysts arise more frequently there.

Brain involvement

Involvement of the brain occurs in 2% of all *Echinococcus granulosus* infections [2]. The majority of cases (approx. 50–75%) of CHCy are seen in children [10]. The statistics of the Neurosurgical Clinic of Bucharest (1936–1992) show a total of 131 cases of CHCy. The distribution was: 79 children, 52 adults (60% children, aged 0–16 years).

Comparing the incidence of CHCy with that of all brain tumors, we found only two cases of CHCy among the 2000 cases in Cushing's classic series [7]. The percentages found in other series included: Obrador and Ortiz Gonzales [17]: 2%; Paillas et al. [19]: 3% (12% in children). In the two series previously reported from Turkey [13, 18], CHCy comprised 2.3% and 3.4% respectively of all intracranial space-occupying lesions. In our series CHCy represented 2.8% of all intracranial space-occupying nontraumatic lesions (962 cases operated in 1980–1992).

Clinical diagnosis

CHCy is the prototype of a benign, slow growing tumor. Headache and vomiting were the most common presenting symptoms in this series, as in other series reported in the literature [9]. Other symptoms, such as hemiparesis, seizures, visual field alteration, and gait disorders, may vary with the location of the cyst. Macrocrania or local cranial bulging can be seen in young children [14]. Papilledema is usually present in patients with intracranial hydatid cyst at the time of diagnosis [5]. The majority of our patients (26 cases, 96.2%) had either papilledema or secondary optic atrophy. Epileptic seizures do not represent an important sign in the clinical diagnosis of CHCy (17% in one series [2] and 22.2% in ours). A bilateral tremor, more marked on one side, appeared in CHCy involving the basal ganglia. For clinical diagnosis the tetrad of Schroeder [21] is very suggestive: (a) a country-dwelling child; (b) good general condition; (c) raised intracranial pressure; (d) no marked focal findings.

Other sites of CHCy

CHCy may also be found in other sides. In recent studies [24] hepatic, pulmonary, and other locations were found in only 10%. In our series 8 patients (29.6%) presented hydatid infestation in multiple organs. Primary hydatid infestation of the myocardial muscle and afterwards of the brain has also been described [15].

Laboratory investigations

Biological tests were mentioned in the section on pathophysiology. Without disturbance and consequent dissemination, the hyaline CHCy, does not cause immunological reactions.

Electroencephalography

It is clear that CHCy is not an important cause of epilepsy. The delta wave activity always disappears directly after extirpation of the cyst, and its reappearance is a significant indication of a recurrence.

Imaging techniques

Plain skull radiographs may show highly significant changes: suture diastases and thinning of the vault.

On cerebral angiography, Arana-Iniguez and San Julian [3] have described four features typical of CHCy: (a) marked arterial displacement (large mass lesion); (b) total absence of vessels within the mass; (c) lack of pathological changes in the vessels themselves; (d) pericystic circumferential displacement of the vessels. Cerebral angiography was useful until CT examination became routine.

On CT CHCy appear as a cystic mass, spherical in shape, and with an absorption value similar to that of cerebrospinal fluid. CHCy generally cause significant ventricular distortion and midline shift. They can be differentiated from brain abscess and cystic astrocytoma by the absence of significant rim enhancement, perifocal edema, and mural nodule. Calcification of CHCy is very rare, less than 1%.

Cerebral edema around the cyst is not a common finding, but it can occur. Contrast enhancement of the lesion is not typical, since the lesion is usually quiescent and incites very little inflammatory response. However, if an inflammatory response is excited, a thin rim of contrast enhancement may be detected [1], limited to the capsule region (usually only a segment). The rim of enhancement can be quite prominent around a multivesicular, secondary cyst-type lesion [18].

An all-body CT scan is extremely useful for detecting other hydatid infestation sites.

Magnetic resonance imaging (MRI) will come to be more widely used for diagnosis and surgical planning in cerebral hydatid disease. MRI scans show details that cannot be seen on CT [6]. The cysts are of the same density as water. The hydatid sand is not visualized on CT, but MRI has the potential to detect these cyst contents. The mass effect caused by these large cysts is less than expected, given the size of the lesion. Both the brain parenchyma and calvarium slowly adapt to the cyst.

Sites

CHCy are usually distributed in the territory of the middle cerebral artery. Most of the cysts are located in the supra-

tentorial region [5]. A hydatid cyst of the posterior fossa is very rare; no case was met in our series. In some cases CHCy have reached impressive volumes, occupying two or even three cerebral lobes (40.7% and 29.6% respectively of the cases in our series). We can therefore affirm that more than 75% of CHCy in our series were very large or even giant in volume (19 cases, 70.3%) – a fact that surely had an effect on the technical possibilities of total removal and also on the rates of recurrence and postoperative complications.

Multiple CHCy

In our series we had only one case (3.7%) of CHCy secondary to previous rupture of a primary CHCy (Fig. 4). After complete removal of the CHCy a very serious superinfection occurred that was very difficult to stop. Multiple primary CHCy have also been described [4, 20]; in such cases the myocardial muscle must be carefully investigated as the source of the multiple intracranial CHCy.

Growth rate of CHCy

On the basis of serial CT scans, Vaquero et al. [26], and Sierra et al. [23] have respectively suggested average growth rates of hydatid cysts in the brain of 1 and 5 cm per year.

Alveolar echinococcosis

Our records contain no case of cerebral alveolar echinococcosis due to the tapeworm *Echinococcus multilocularis*. These cerebral lesions are extremely rare; Jacquet et al. [12] found only 18 cases in the literature in addition to one personal case.

Treatment

The treatment of CHCy is surgical. The aim of surgery must be to remove the cyst without damaging its walls, thus preventing the hydatid liquid from coming into contact with the tissue (Fig. 5). The procedure may be summarized as follows: (1) a wide osteoplastic flap; (2) exposure of the cyst by a wide incision in the nervous tissues; (3) an inclined position of the head such that gravity will assist expulsion of the cyst; (4) gentle saline irrigation between the surface of the cyst and the surrounding cerebral substance by one or two rubber catheters. With this technique, we found that the saline irrigation alone, without the use of any metallic instrument, was able to remove the cyst totally.

No intracystic irrigation with 10% formalin was done in any case, because it is too dangerous for brain. Neither did we use any intracystic injections of 0.5% silver nitrate, hydrogen peroxide, 1% aqueous iodine. Hypertonic saline solutions were used to destroy the residual larvae, especially in those cysts that ruptured intraoperatively.

Postoperative course

Operative mortality

Advances in surgery have greatly improved the operative prognosis of CHCy. Today we no longer see cases of death as a result of decompensation of intracranial hypertension before operation. In our series we had only one operative death.

Neurological sequelae

Motor disturbances such as hemiparesis are frequent in patients in whom the motor tract is directly compressed by

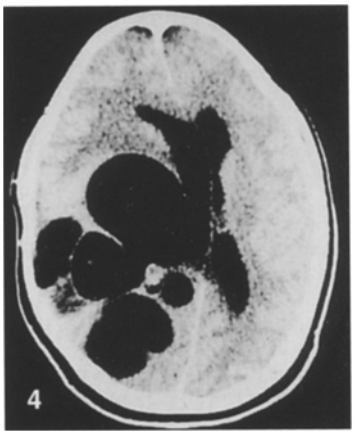

Fig. 4 CT scan: multiple CHCy

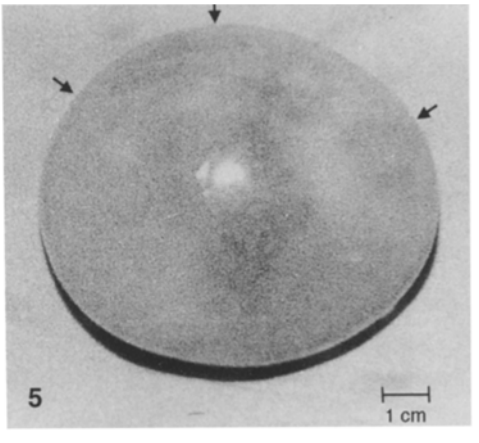

Fig. 5 Gross appearance of a unilocular CHCy after total removal

the CHCy. In these cases there is spectacular disappearance of the motor disturbance postoperatively. Postoperative seizures are generally controlled by antiepileptic drugs. Recurrence and refractoriness of seizures to treatment are a sure sign of CHCy recurrence.

Ocular sequelae are significant. In our series the most important ocular sequelae were visual field alterations that remained postoperatively (six cases). Today there are no more cases of severe visual loss and blindness due to secondary optic atrophy. We have not had any case of blindness after rapid removal of the CHCy – a sequel reported by other authors [10].

Postoperative complications

These can be multiple, and include subdural effusion, ventricular dilatation, and porencephalic cysts. All these are treated surgically by insertion of a subduro-, ventriculo- or cystoperitoneal shunt. In such cases CT follow-up is mandatory for all patients.

Chemotherapy with albendazole

Complete surgical removal of CHCy is often impossible, and treatment can be difficult, especially when the cysts involve vital structures or are widely disseminated. Recent developments suggest that chemotherapy with albendazole is a useful adjunct and in certain circumstances can replace surgery. We used albendazole in three cases of CHCy recurrence.

Albendazole is a broad-spectrum oral antihelmintic drug. It blocks glucose uptake by larval and adult stages of susceptible parasites, depleting their glycogen stores and decreasing the formation of adenosine triphosphate. As a result, the parasite is immobilized and dies. Successful treatment with albendazole for hydatid cysts in other parts of the body concomitantly with the brain has been reported [16, 23]. The albendazole treatment regimen is: 10 mg/kg three times daily for 4 months. In the successful cases, no CHCy were found after this treatment. Twelve months later, calcification was observed at the side where the CHCy had previously been located [25]. Golematis et al. [11] analyzed 44 patients who had been treated with albendazole and found that large cysts were reduced considerably in size, while small ones disappeared. A World Health Organization multicenter study clearly showed the efficacy of albendazole in 112 patients and confirmed that the follow-up for an objective evaluation for the efficacy of treatment should be at least 12 months [8]. On the basis of albendazole experience, Singounas et al. [24] suggest that patients with central nervous system hydatid disease should be treated for at least 3 months and monitored for up to 12 months before they are considered cured.

Recurrences

Cyst recurrences are extremely dangerous because they are difficult to remove, many being situated very close to ventricular walls, so that CSF-pathway dissemination is possible at any time. In our series we recorded 11 recurrences (40.7%), all patients being reoperated on. Five of our patients with recurrences had to undergo multiple operative interventions, and four of the children finally died.

Conclusions

Cerebral hydatid cyst (CHCy) is a neurosurgical lesion which can appear in children from countries where hydatid disease is endemic. CT scanning gives a maximum of noninvasive, efficient diagnostic information. Total surgical removal of the CHCy must be performed in all cases. It is important to avoid cyst tear or rupture. If recurrence occurs, surgical reintervention is necessary and everything must be done to ensure that total removal of the cysts is achieved. We believe that the most frequent and important postoperative sequelae are the ocular disturbances, mainly disturbance of the visual field.

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EDITORIAL COMMENT

This article by Ciurea et al. presents a large series of cases of pediatric cerebral hydatid cyst with a considerable amount of information concerning the findings, neurodiagnostic procedures, surgical technique, and complications. Although the article as a whole contains neither new nor striking data that are not present in the textbooks, it is interesting in terms of emphasizing certain facts in central nervous system hydatidosis. The disease is thought to be confined to certain geographical areas and is essentially unknown in developed countries. As travel and migration between countries is rapidly increasing, however, the prevalence of this type of infestation should be expected to rise, especially in developed countries. Therefore, physicians in countries other than the endemic areas should also keep in mind the possibility of a hydatid cyst when confronted with cystic lesions in the central nervous system. Besides their usual hemispheric localization, cerebral hydatid cysts are reported to be found virtually in any part of the cranium, including the sella, parasellar area, ventricular system, cerebellum, and brain stem [1–4].

As mentioned in the article, the aim of surgical intervention has to be removal of the cysts *intact*, which necessitates bearing in mind the possibility of hydatid cyst when dealing with cystic masses preoperatively. Intact removal especially of primary cysts in cerebral tissue is curative, whereas spilling of the cyst contents is responsible for recurrences and most of the morbidity and mortality. The cyst wall along with the cyst contents should always be forwarded for histopathological examination to differentiate a fertile, primary cyst from a silent secondary cyst, especially in cases where the cyst cannot be removed totally.

CT scanning has enormously improved our diagnosis and surgical planning for these lesions. Moreover, multiple occurrence of the lesions is much better appreciated with CT than with any other imaging technique. In planning the cortical incision for evacuating the cyst, the closest and most superficial part of the cyst to the bone flap has to be determined, and CT is invaluable for this.

Recently, reports have begun to be published on the MRI findings of central nervous system hydatidosis. In our experience, MR images provide additional information on the exact localization and extent of the cyst(s), especially in spinal and paraspinal forms. Much more interestingly than that, however, is that by using different sig-