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Cerebral hydatid cysts in children

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Abstract. Twelve children with intracranial cysts of *Echinococcus granulosus* underwent surgery during a period of 5 years and constituted 19% of all children operated on for intracranial space-occupying lesions. The more common symptoms were raised intracranial pressure (8 cases) and hemiparesis (7 cases). The total number of procedures was 14, with a standard craniotomy approach in 13. In 6 the cysts were removed without puncture or rupture, in 5 puncture and drainage were carried out before removal of capsule, and in 3 the cyst ruptured accidentally. Recurrence of multiple cysts occurred in 1 case and another patient was reoperated on twice for recurrent cysts after an operation in another center. There was no mortality. Non-bacterial meningitis occurred in 2 cases. Although drainage of the cyst contents greatly facilitates removal, it also carries a risk of contamination; thus a traditional approach via a large craniotomy seems to be the safest choice.

Key words: Cerebral hydatid cyst – *Echinococcus granulosus* infection

Hydatidosis of the central nervous system represents 2–3% of all diagnosed cases of *Echinococcus granulosus* infections. It is rare in developed countries [2, 5]. In endemic areas such as North Africa, where 27–40% of dogs and up to 80% of sheep are infected, reported prevalence of human hydatid disease reaches 13 in 100 000 [1, 3, 4]. Therefore, cases of intracranial hydatid cysts are frequently seen in local neurosurgical practices and constitute up to 10% of all intracranial space-occupying lesions [6].

Materials and methods

During the last 5 years, 15 cases of intracranial hydatid cysts were operated on in our department. Twelve were children (80%). Table 1 presents clinical data. Figures 1 and 2 demonstrate computed tomography (CT) pictures of the two largest cysts. The preferred method of treatment was craniotomy, performed in the area where the cyst was most superficial, incision and partial removal of thinned cortex, and removal of the cyst without rupture. Postoperative follow-up was irregular due to poor compliance; only one observation is available in 8 cases.

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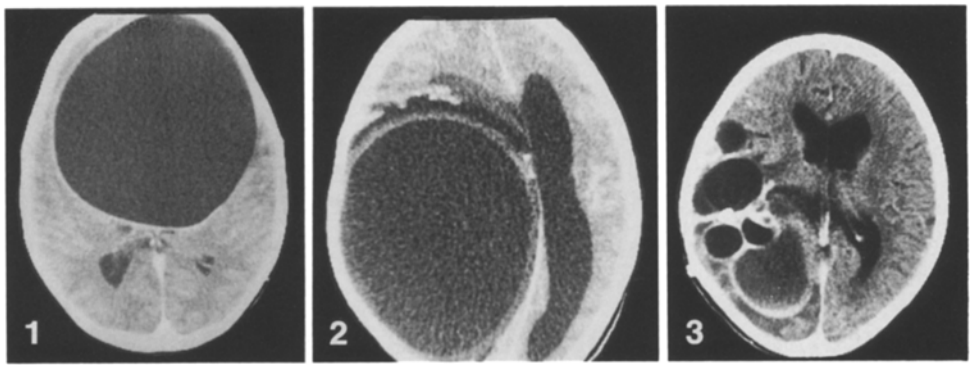
Results

Preoperative CT scan showed a single cyst in 10 cases and multiple cysts in 2. In 3 cases the cyst was surrounded by a hypodense area and an inflammatory reaction occurred around the cyst during operation. The 12 patients underwent a total of 14 operations. Although a classic approach via craniotomy was routinely used, technical details of the procedure varied according to the preferences of the different surgeons who performed the operation. In six procedures it was possible to remove the cysts without rupture and without puncture-aspiration. In the remaining eight procedures, the cysts were either electively punctured [5] or ruptured accidentally [3], thus creating the possibility of contamination of the area. There was no mortality and the only complication observed was non-bacterial meningitis in 2 cases. Two cases of recurrent cysts have been reported so far: 1 patient operated on in 1984 was readmitted 2 years later with two recurrent cysts which were successfully removed and another was admitted with multiple recurrent cysts 4 months after undergoing surgery abroad (Fig. 3). Five months after the first reoperation, control CT again showed recurrence of multiple cysts, which were subsequently removed. One year later, the patient was readmitted with meningitis, but CT did not show recurrence. In the remaining 6 cases for which follow-up information is available there were no recurrences. Three of these children, although improving, show significant retardation of mental development. Control CT scan shows a persisting cavity filled with CSF in the site of the removed cyst.

Table 1. Age, sex, and symptoms

Age (years)	4–13, av. 8
Male/female	6/6
Symptoms	
Raised ICP	8
Paresis	7
Mental retardation	2
Epilepsy	1
Meningitis	1
Duration of symptoms (weeks)	2–24, av. 10



Figs. 1, 2. Huge hydatid cysts in patients aged 4 and 11 years. Both cysts were removed without rupture

Fig. 3. Multiple recurrent cysts with an inflammatory reaction and scar formation 4 months after the first operation

Discussion

Due to extremely irregular referral patterns, no conclusions regarding incidence of intracranial hydatid disease can be drawn from this material. It was reported by Aboundaya [1] that hydatid disease was found in 0.45% of all admissions in Tripoli Central Hospital. In our practice, cases of nervous system hydatidosis constitute 5% of all and 19% of pediatric intracranial space-occupying lesions operated on during the same period. Positive serological tests for *Echinococcus* were found in 8–12% of the population in Libya [1], and we may assume that our group represents only a small fraction of cases with hydatid disease of the nervous system.

The huge size of the cysts in our patients reflects the insidious nature of the disease, as well as delays in referral. There were reports suggesting that growth rate of the cyst is about 1 cm in diameter per year [2]. The biggest cyst we operated on measured 14 cm in diameter and occurred in a 4-year-old boy (Fig. 1). In the first patient with recurrence the biggest cyst reached a size of 7 cm within 2 years. This proves that growth rate depends on individual factors and certainly can be much higher than 1 cm per year.

The cysts are surrounded by a capsule which represents glial reaction. During operation, it is most important to find a plane between this layer and the wall of the cyst which is unmistakably identified by its smooth, glistening appearance. Surgery is relatively easy when the cyst has a thick wall. Fast-growing cysts, as in our second case of recurrence, have a wall which is no thicker than arachnoid. It is extremely difficult to avoid rupture of such cysts. From a technical point of view, it is most tempting to drain the contents of the cyst and then to remove its wall (which separates spontaneously from the surrounding brain). In one procedure, done under local anesthesia, a 2-cm craniectomy was sufficient to remove a cyst measuring 10 cm in diameter, and there was no recurrence 2 years after the operation. Recently, a similar approach was advocated by Wani, using fine-bore needle puncture [6]. In our experience, each puncture invariably leads to some spilling of the contents into the operative field. We have tried to seal the puncture site with Histoacryl but it turned out that, contrary to other tissues or instruments, adhesion to the cyst wall was very weak. Therefore, we are of the opinion that the traditional approach is still the safest. Paradoxically, we find it easier to remove big cysts, which usually have a thick wall, than smaller ones, which are prone to rupture even with the most delicate handling. Development of a contamination-proof puncture and drainage technique would turn removal of hydatid cysts into a relatively minor procedure.

Incomplete follow-up in our patients and differential technical aspects of operations do not permit assessment of the risk of recurrence after puncture or rupture of the cyst. Two patients in whom the field was contaminated during operation were followed for more than 2 years and are now free of disease. Recurrent cysts in both our cases were multiple. Although reoperation was successful both times, these are extensive and very difficult procedures, carrying increased risks for the patient.

Conclusion

Intracranial hydatid cysts are frequently seen as a cause of increased intracranial pressure in patients from endemic areas. Pure cystic lesions in these patients, particularly in children, should always be approached as suspected hydatid even if serological tests are negative. Large operative fields should be prepared for possible craniotomy if initial bur hole exposure and inspection of the cyst wall reveals the presence of *Echinococcus*. Until a safe method of puncture and drainge of the cyst is established, the classic craniotomy approach is the safest.

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