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MULTIPLE PRIMARY HYDATID CYSTS OF THE BRAIN*

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All reported cases of multiple hydatid cysts of the brain have resulted from spontaneous, traumatic or surgical rupture of a primary solitary cyst[1-6]. As far as we know, only one case of multiple primary cerebral hydatid cysts had been previously described[7]. We also present such a case. An extremely rapid growth of cerebral hydatid cysts in children is suggested.

Case Report

A 15-year-old boy was admitted to Erciyes University Hospital with a two-month history of severe headache and intellectual deterioration. Ten days prior to admission he developed progressive dysphasia.

Neurological examination revealed bilateral papilledema, expressive dysphasia, and obtundation. Skull films demonstrated evidence of raised intracranial pressure. Computerized tomography (CT) showed four spherical, well-defined cystic lesions occupying the left supratentorial hemisphere. Of these, three were located in the frontoparietal region, and one measuring one cm in diameter, in the occipital region. The cerebral hemisphere was markedly compressed and the ventricular system was displaced to the opposite side. The lesions contained a fluid with a Hounsfield unit value similar to that of cerebrospinal fluid, and enhancement of a peripheral rim of the compressed brain was noted (Fig. 1). The CT appearance was diagnosed as multiple hydatid cysts. Laboratory studies were normal and there was no evidence of hydatid disease elsewhere.

During the operative procedure a large, frontoparietal bone flap was turned, the dura and arachnoid were opened, and a cortical incision was made over the most surperficial part of the cysts. The cysts were encountered at a depth of two mm.

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Saline was injected into the cyst-brain interface and a cyst measuring six cm across was delivered intact. After the removal of this superficial and largest cyst, four other cysts were exposed underneath. Of these, two were four cm, and two were two and half cm in diameter. All of the four cysts were removed intact using saline irrigation assisted by changing the position of the patient's head. A deep-seated cyst measuring one cm was tightly surrounded and could not be delivered by irrigation. The entire contents of this cyst were aspirated, and the collapsed cyst was lifted away. Large cotton tampons soaked in 3% NaCl were applied to the large cavity and left in place a couple of minutes. Then, the cavity was thoroughly and repeatedly washed with saline. No attempt was made to remove the small single occipital cyst. It was thought that deferring the surgical intervention and waiting for the evolution of the cyst would be the best choice. Microscopic examination of the pearly white cysts showed the characteristic ectocyst and scolices.

The postoperative course was uneventful. The patient's neurological condition markedly improved, and he was free of symptoms when discharged on the fifteenth day after admission. Seven months later, a new CT performed because

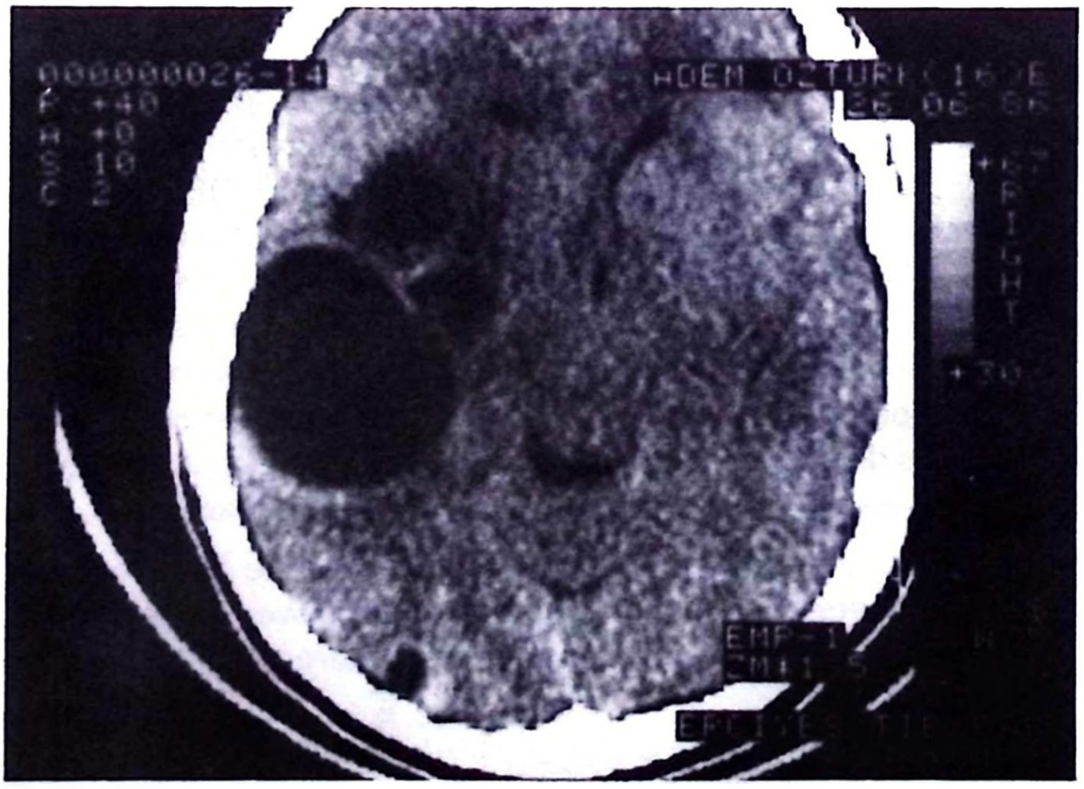

Fig. 1: CT scan showing multiple hydatid cysts occupying the left supratentorial region. Note the peripheral rim enhancement following intravenous contrast injection, and the small occipital hydatid cyst.

of dysphasia, revealed a large recurrent cyst in the sylvius. The small occipital cyst detected in the first CT was found to be seven cm in diameter (Fig. 2). These two cysts were removed intact using the same operative technique. The patient recovered rapidly after the second operation. He has had no symptoms since then and no cyst recurrence during a year follow-up period.

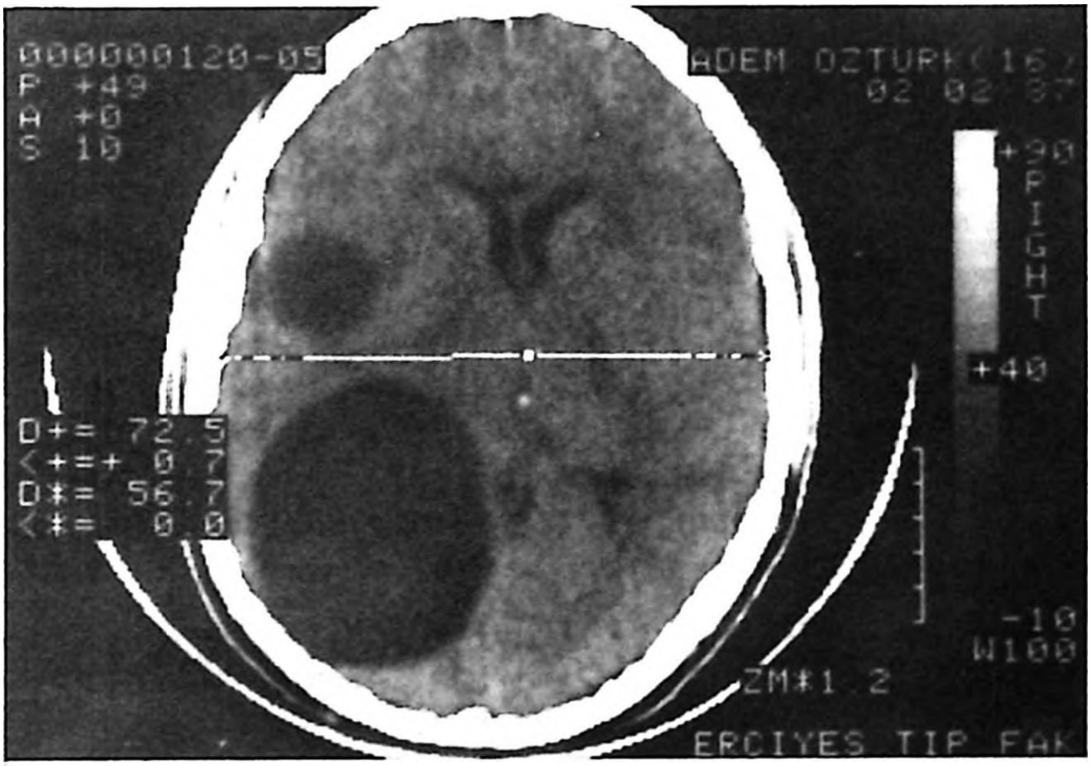

Fig. 2: Follow-up CT scan seven months later showing a large recurrent cyst in the operated area and extremely rapid growth of the small cyst seen in the first scan.

Discussion

The incidence of hydatid cysts among intracranial space-occupying lesions varies between 1.6 and 5.2 percent[3] in different countries. In Turkey, this rate was found to vary between 3.4 and 2.4 percent[4,8]. Hydatid cysts of the brain are more common in children than in adults[1,2,5], and the cysts are always solitary when the primary localization is in the brain[3,4,9]. Multiple cerebral hydatid cysts are rather rare[2,3], and result from spontaneous, traumatic or surgical rupture of a primary solitary cerebral cyst or as a consequence of a cyst rupture elsewhere and embolization of hydatids to the brain[1-6,10-12]. These secondary cysts which are acephaloceles and infertile lack a broad capsule[3,5].

Although Gordillo et al[13] recently described a case of multiple cerebral hydatid cysts with no previous history of trauma or rupture of cysts elsewhere, there was no detailed information confirming that all were primary cysts. The only case of multiple primary hydatid cysts of the brain has been reported by Sharma and

Abraham[7]. In our case, the second case of multiple primary hydatid cysts of the brain, all the cysts contained broad capsules and scolices, and repeated postoperative follow-ups showed no evidence of hydatid disease elsewhere in the body.

Cerebral hydatid cysts are often very large, especially in children. They are tolerated for a long period, and are usually huge when diagnosed[2,3,5]. An average growth of one cm per year for intracranial hydatid cysts in the adult has been suggested[6,12,14]. This rate can be much faster in children, as in the case reported by Gordillo et al[13], and as in our case. The growth rate in our case was more than ten cm per year. We think that such extreme rapid growth of hydatid cysts must be considered in children and this aspect of the disease deserves detailed studies.

CT scanning can be relied on in the differential diagnosis of cerebral hydatid cysts. There is a spherical well-defined intracerebral cystic lesion containing fluid with a density value similar to that of CSF, with no ring enhancement[15,16]. Though multiple hydatid cysts can also be differentiated from other cystic lesions by these characteristic features, it should be remembered that contrast enhancement of the periphery of the cyst, probably due to compression of the surrounding parenchyma may be found[17], as was seen in our case.

The only treatment for hydatid cysts is surgery, with removal of the cysts intact and avoidance of spillage of the hydatid fluid. Nevertheless, deep-seated and tightly surrounded small cysts may not be able to be delivered intact and a ruptured cyst always carries a high risk of recurrence. Research into therapy with benzimidazole carbamates is in progress. The clinical trials of mebendazole and flubendazole have not been well-coordinated, and the results which mostly concern patients with liver, peritoneal, bone, lung, and spinal or paraspinal cysts, are inconclusive[18-22]. At present, there is not sufficient evidence to justify the routine pre-and postoperative use of these agents in patients with hydatid cysts of the brain.

We believe that surgical intervention should be considered in every case, even in the event of recurrence.

Summary

A rare case of multiple primary hydatid cysts of the brain is presented in a 15-year-old boy. The largest of the seven cysts located on the right hemisphere measured six cm across, while the smallest one was one cm in diameter. Detailed studies revealed no evidence of hydatid disease elsewhere in the body. The growth rate of an untreated cyst was about ten cm per year determined by follow-up computerized tomography. Since the only treatment of cerebral hydatid cysts is surgery, with removal of the cysts intact, surgical intervention should be considered in every case, even in the event of multiple recurrences.

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