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Cerebral manifestation of hydatid disease in a child

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Abstract. The case of a 9-year-old-boy with a left frontal hydatid cyst is reported. The infection was transmitted by a dog, the final host and a companion of the child. Recovery without complications was accomplished after puncturing, systemic antihelminthic therapy using mebendazole and, finally, total removal of the cyst.

Key words: Cerebral hydatid cyst – Child – Neuroradiology – Neurosurgery.

Hydatid disease is primarily a disease of the third world, which is spread in Europe by immigration and tourism [1]. Infected pigs, cattle and sheep transmit the disease. The proglottids of the dog tapeworm become mature in the intestines of the dog, the final host, whereupon humans, as intermediate host, ingest the eggs in contaminated food or by allowing the animal to lick their hands and face. Larvae of the tapeworm develop in the intestinal tract and penetrate through the wall of the intestine to reach the liver, kidney, lungs, or brain via the bloodstream.

The liver is the site of infection in 60% of cases (*Echinococcus cysticus*), while only in 2% is the central nervous system involved. In 1.4% of cases there is an intracerebral manifestation in which the cysts are mainly localized in the surface of the frontotemporoparietal region of the cortex. The cysts contain the infectious larval stage, the scolices [5, 6, 11]. Symptomas are intracerebral expansion with general indications of increased intracranial pressure and/or local brain alterations. Eosinophilia, the CBR, and the intracutaneous test are not relevant for a diagnosis [6, 10, 12]. Differential diagnosis should include a brain abscess. In this case there would commonly be general symptoms of infection and tumor of different histological characteristics that normally have perifocal edema [15, 20].

With regard to neuroradiology, the CT scan is the most useful method. Characteristic is a hypodense spheric cystic lesion that has a sharp demarcation, no perifocal edema, and does not have a ring structure after application of contrast medium [15, 18]. Angiography shows a non-specific avascular space corresponding to local displacement. Using magnetic resonance tomography, we obtained a signal hypointense zone, but this did not provide any additional information.

Therapy is total extirpation by the so called "no-touch technique," which leaves the cyst intact [6, 12]. In all cases, this should be followed by high-dose antihelminthic therapy with mebendazole (Vermox) in order to avoid systemic infection [3].

Case report

A 9-year-old boy presented for treatment with a few weeks' case history of symptoms of general tiredness, headache, vomiting, visual problems, and double vision. The neurological examination showed congested papillae; X-ray films of the skull revealed signs of high intracranial pressure with bursting of the lamdba coronoida (Fig. 1).

By CT, in the left frontal region, a hypodense, sharply defined lesion with a small perifocal edema was found, together with slight compression of the left anterior horn (compare Fig. 2). Angiography showed only an avascular space in the left frontal region. Standard chemical tests were at first inconclusive. Urine and feces did not appear to be unusual when examined microscopically.

Course

Initially, the cranium was opened in the left frontal region and the cyst punctured, whereupon 25 ml of clear, yellow, gel-like liquid was aspirated. The cyst was punctured because we, at that time, had no reason to consider parasitic disease. The cytopathological examination showed bluish concentric layers that were reminiscent of "Psammoma" bodies, with partly calcified spherical bodies laying on top of a circle of hooked structures. By microscopical examination, we discovered a large number of intact mature heads of the dog tapeworm partly within the wall of the cyst. Measurements of the hooks of the scolices were characteristic of *Echinococcus granulosus* (compare Fig. 3). We therefore closed the opening to allow the cyst to heal and await "no-touch" removal. Indirect fluorescence (IIF) and the ELISA gave a positive response of doubtful value. Both tests have low sensitivity for manifestations outside the liver, and there is no information

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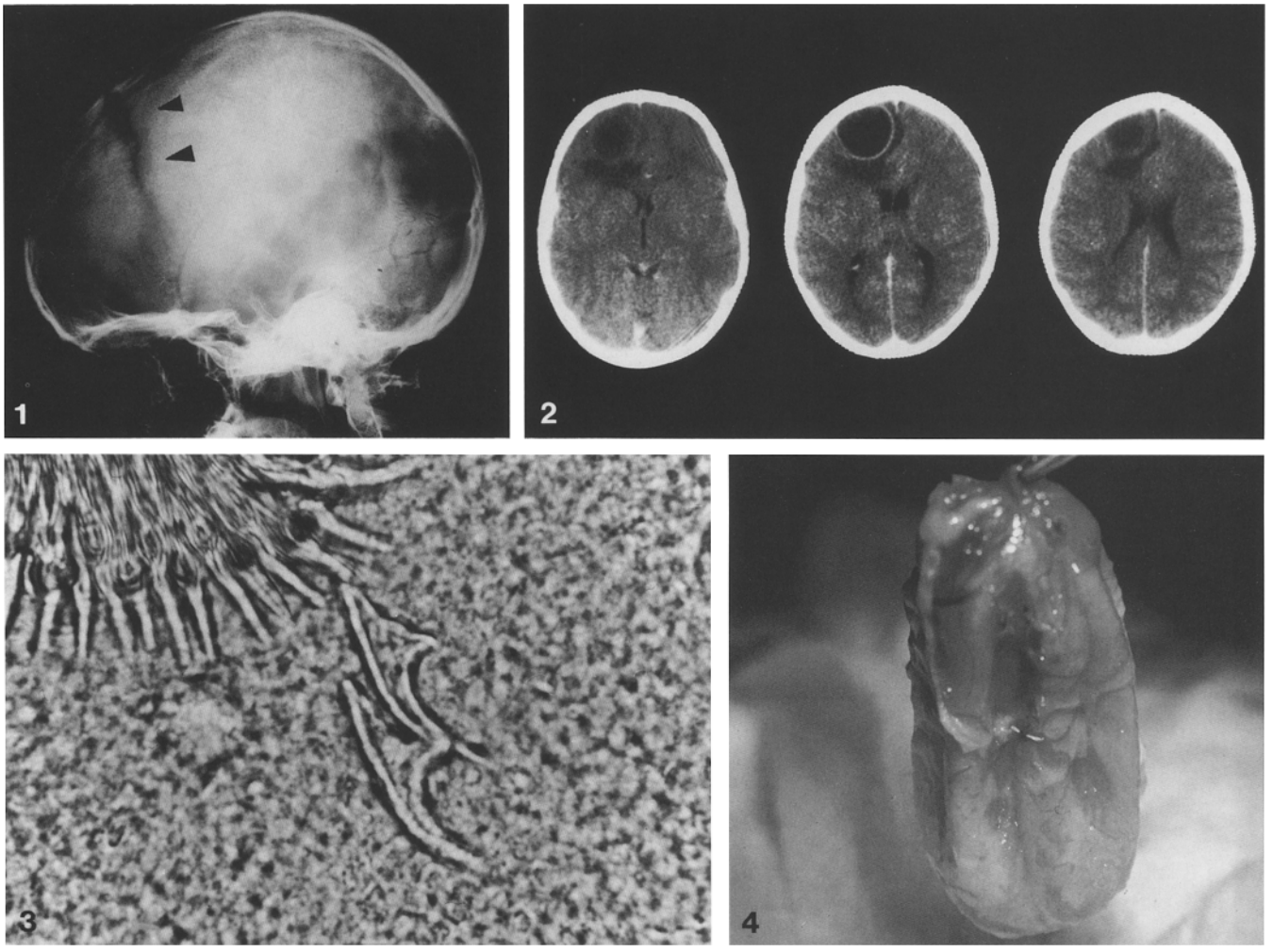

Fig. 1. CT scan of a left frontal hydatid, showing a hypodense spherical cystic lesion with sharp demarcation, a small amount of perifocal edema, and compression of the left anterior horn

Fig. 2. Angiography showed an avascular space corresponding to local displacement

Fig. 3. Microscopic examination revealed a crown of hooks from an intact head of the native dog tapeworm (*upper left*) and two detached hooks, showing the characteristic shape of *Echinococcus granulosus*

Fig. 4. Echinococcus cyst after removal by the "no-touch technique"

available on the cerebral manifestation (Professor Eckert, personal observations).

Accordingly, antihelminthic treatment with mebendazole was carried out. From day 4 to 13 after puncture of the cyst there was a minor increase in the number of eosinophils in the blood. Sonography of the epigastric region and magnetic resonance tomography of the abdomen and chest continued to show nothing unusual.

Clarifying the case history further, we then found the final host of the chain of infection in the household of the parents. They have a dog that had bitten the childs eyebrow and upper lip a few months earlier. Analysis of its feces revealed the presence of tapeworm.

Four weeks after puncturing the cyst, it was totally removed after opening the anterior of the cranium. The convolutions of the anterior-medial region of the cortex were discolored – yellowish white. Using the "no-touch technique", it was possible to remove the entire cyst (Fig. 4). Histopathological tests then found changes that were compatible with infection by tapeworm larvae. There were no postoperative complications – no neurological deficits appeared.

Discussion

The response of the nervous system to tapeworm larvae is rarely seen. There are only a few case reports of this type of manifestation in endemic regions of the third world [1, 4, 7, 9, 11]. In only 6 cases out of 3,000 intracerebral expanding processes could Bala Subramaniam et al. [9] prove a cerebral manifestation of this type. Reddy et al. [17] only did so in 4 out of 1,000 patients. Vasal et al. reported 1 case of a foramen jugularis syndrome [21], Assad and Lins [5] wrote about a patient with a mycotic aneurysm of the A. cerebri media, and Tuncalp et al. [22] reported 1 case of a hydatid cyst in the sella turcica.

Pathoanatomical examination reveals parasite cyst walls consisting of a chitin membrane and a parenchymic layer of small vesicles that contain the hooked heads of the

scolices [17]. Neuroradiological examination, in most cases, detects the cerebral and spinal lesion. It has the characteristics of a delimited encephalitis, so that on the whole the diagnosis of a brain abscess will stand. Only further investigations balanced against the information in the case history allow a definite differential preoperative diagnosis to be made. Should multifocal cysts be present (e.g., intraventricular, parenchymic, intracisternal or subarachnoidal), further parasite illnesses, in particular the so-called cysticercosis, must be considered.

If a parasite is assumed to be present preoperatively, the aim should be to remove the cyst directly using the "no-touch technique" in order to prevent the spread of infectious material through puncturing the cyst.

Our case described here of a child with a short case history and signs of increasing intracranial pressure indicates a neuroradiological diagnosis of a brain abscess. The neurosurgical measures, cytopathology and microscopical examinations, as described here, confirmed infection from *Echinococcus granulosus.* With the protection of antihelminthic treatment using mebendazole, it was possible to remove the cyst completely without postoperative illness.

In patients lacking the general signs of an infection, but having the typical characteristics of a brain abscess with an appropriate history, a parasitic disease can be included in the differential diagnosis.

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