Huge hemispheric intraparenchymal cyst caused by *Taenia multiceps* in a child

Case report

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The authors report an unusual case of a huge intraparenchymal cyst in a 4-year-old girl caused by *Taenia multiceps* infection. After surgical removal of the cyst, the child recovered completely. Brain infestation by coenurus is a rare disease, mainly reported in Africa, with a few case reports from patients in developed countries. Humans, especially young children, become intermediate hosts by ingesting eggs passed in the excrement of a definitive host, usually carnivores. In such cases, high mortality and morbidity rates have been reported. These rates decreased after the introduction of the modern neuroradiological techniques of computed tomography and magnetic resonance imaging. (DOI: 10.3171/PED-07/12/511)

KEY WORDS • coenurosis • intraparenchymal cyst • pediatric neurosurgery • *Taenia multiceps*

Parasitic infection must be considered in the differential diagnosis of an intraparenchymal brain cyst. Neurocysticercosis and echinococcosis are the most common parasitic infections of the central nervous system. In humans, cerebral coenurosis caused by *Taenia multiceps* is a rare disease reported mainly in developing countries. Dogs and sheep are the most common hosts, and humans become intermediate hosts by ingesting eggs, resulting in cerebral multilocular coenurus that are rarely intraparenchymal. The CT and MR imaging findings in patients with this infection are not specific and include an enhanced cyst capsule containing CSF-like fluid. Surgical removal of the cyst is indicated for establishing a diagnosis and for relief of symptoms. In the past, however, high mortality rates were reported with or without surgery, although more recent studies showed better outcomes.

In this report, we present an unusual case of a child with a huge intraparenchymal cyst due to *Taenia multiceps* infection. This case is the first case of cerebral coenurosis ever reported in Israel.

Abbreviations used in this paper: CSF = cerebrospinal fluid; CT = computed tomography; ICP = intracranial pressure; MR = magnetic resonance.

Case Report

**History and Examination.** This 4-year-old girl, previously healthy, presented to the emergency room with a 2-month history of morning headache and nausea. Her parents also reported that she had developed a right-sided limp during the preceding 2 weeks, immediately after she suffered a mild head injury. On neurological examination, she was noted to be irritable and exhibited paresis in both her right arm (Medical Research Council Grade 3/5) and leg (4/5). Funduscopic examination revealed bilateral papilledema. A complete blood count, including eosinophilic count, was within normal limits.

A head CT scan, followed by brain MR imaging, was performed (Fig. 1) and demonstrated a huge left hemispheric cystic lesion with no solid component and minimal enhancement in the medial aspect of the surrounding capsule. The main differential diagnosis at this point included low-grade primary brain tumor and parasitic infection.

**Operation and Postoperative Course.** A left frontoparieto-occipital craniotomy was performed the next day to resect...
the lesion. After a minimal corticectomy, a very thick, gray-brown capsule was found, which was initially very easily dissected from the brain (Fig. 2). A frozen section of a specimen from the superficial capsule was negative for any neoplastic process. Eventually, the capsule became very thin and adherent to the surrounding brain tissue, resulting in perforation of the outer capsule with spillage of its contents. An additional paper-thin transparent cyst was found, floating within the fibrous capsule. The inner cyst contained multiple white nodules measuring a few millimeters in diameter, which were later established to be the parasite protoscolices. Eventually, this cyst was removed intact (Fig. 3). What remained of the outer capsule was dissected from the brain, leading to gross-total resection of the lesion. The immediate postoperative course was uneventful.

**Histological Examination.** The results of histological examination of the outer cyst included typical components of cerebral chronic abscess: collagen fibers, dense acute and chronic inflammatory cells, and markedly reactive proliferation of glial cells (gliosis). The inflammatory infiltrate was rich in plasma cells and lymphocytes, but eosinophils were absent.

The inner cyst (Fig. 3) was unilocular, with a maximum diameter of approximately 6 to 7 centimeters, possessing a thin transparent and gelatinous wall, and exhibiting disseminated white nodules measuring only a few millimeters in diameter. Histological examination showed that these nodules were multiple protoscolices (Fig. 4). The germinative wall was thin, nonlaminated, and composed of a thin layer of tegumental cells surrounded by an amorphous pink tegument. The absence of a thick laminated wall, typically seen...
Cerebral coenurosis in a child

in hydatid cysts, excluded diagnosis of echinococcal infection. No daughter cysts were found.

The protoscolices of cysticercus and coenurus share very similar histological characteristics, and the presence of multiple protoscolices in one large unilocular cyst, typical for the parasite *Taenia multiceps*, established the diagnosis of coenurosis in this child. Examination of the cyst fluid did not reveal any free scolices or any hooklets. Furthermore, serological assays for echinococcal infection, performed pre- and postoperatively using enzyme-linked immunosorbent assay, immunoelectrophoresis, and immunoblot, yielded negative results.

A combined treatment with praziquantel (administered over 3 weeks until the final diagnosis could be established) and albendazole was given over an 8-week period. Six months after surgery the child had recovered completely, displaying no signs of neurological deficit or papilledema. No evidence of a residual lesion was detected on a subsequent MR image (Fig. 5).

Discussion

Coenurosis is a zoonotic cestode infection of humans caused by the larval stage of the *Taenia multiceps* species. Human brain infestation caused by coenurus is a rare phenomenon, mainly reported in Africa. Ing and colleagues reviewed the six cases of human coenurosis reported in North America; only three of them were associated with cerebral infection.

*Taenia* species require two hosts to complete their life cycle. The definitive host is a carnivore (such as dogs or foxes), whereas most of the intermediate hosts are herbivores (such as sheep, rabbits, rodents, and cows). The disease is moderately common in sheep in the Middle East. However, no information regarding current prevalence of the disease is available. Humans, especially young children, become intermediate hosts by ingesting eggs passed in the excrement of a definitive host. The ingested eggs release oncospheres in the host intestine that penetrate the intestinal wall and migrate toward target organs through the bloodstream, usually lodging in the brain, the spinal cord, and the eye. In the brain, coenurii tend to invade the parenchyma or spread along CSF pathways, eventually causing basal arachnoiditis or ependymitis. The most common signs and symptoms of coenurus infection are headache, vomiting, and papilledema, all caused by increased ICP from the mass effect of the cystic lesion. Focal neurological deficit, such as cranial nerve palsy and motor weakness, are also commonly seen.

The differential diagnosis of a supratentorial, intraaxial, cystic brain lesion includes low- and high-grade neoplastic lesions, abscesses, neuroglial cysts, and parasitic cysts, mainly neurocysticercosis and hydatid cysts. There are no reliable preoperative tests to establish a diagnosis of coenurosis. Hence, when the possibility of a parasitic infection involving the brain exists, coenurus should be considered after echinococcosis and cysticercosis infections are excluded. The final diagnosis of *T. multiceps* is primarily possible only after surgical removal and pathological examination of the cyst. The size of the cyst described in this case was approximately 13 cm in diameter, however, as compared with a cyst measuring less than 5 cm that might be found in both *T. solium* and *T. racemose* infections. Furthermore, so far there has not been any report of *T. solium* infection in Israel, and, since 1957, there have been no reports of a local infection of *T. multiceps* in sheep. In the present report, all the serological assays (which included enzyme-linked immunosorbent assay) from a commercial kit, as well as immunoelectrophoresis and immunoblot using hydatid cyst fluid, yielded negative results.

In the presurgical imaging studies of cerebral coenurosis, a CT scan usually demonstrates a contrast-enhanced peripheral rim surrounding a translucent lesion. The layer of fibrous tissue surrounding the viable cyst produces the enhancement. The cyst contents are displayed as a CSF-like pattern on MR imaging. Enhancement and edema are rare during the early stage and are more common during the degeneration and healing stages of the cyst. In contrast, only 2% of patients with hydatidosis will develop a cerebral cyst, generally unicellular, exhibiting a spherical shape and no contrast enhancement. In the past, surgery for this type of cyst was associated with high mortality and morbidity rates. The results were greatly improved after the introduction of the modern neuroradiological techniques of CT and MR imaging. If signs and symptoms of increased ICP are present and can be attributed to the brain cyst, total surgical removal is the treatment of choice. Surgical intervention for patients with multiple brain coenurii is not always indicated, and, as a result, these patients have a poor prognosis. There is no description of the outcome of intraoperative rupture of the outer (fibrous) capsule; however, it stands to reason that extra care should be taken to extract the inner cyst as a whole because it is the viable one and

![Fig. 5. Axial T2-weighted fluid attenuated inversion recovery image obtained 2 months postoperatively shows complete resection of the lesion with minimal gliosis in the cyst cavity.](image-url)
contains the parasite protoscolices. Complementary treatment with praziquantel should be administered, although caution should be exercised to prevent reported drug side effects such as blindness.

This case is the first reported case of an intracranial *Taenia multiceps* infection in Israel. This 4-year-old girl presented with signs and symptoms of increased ICP and focal neurological deficit due to a huge cystic lesion as demonstrated in imaging studies. The parents reported that the girl had had direct contact with dogs and possible contact with the dog’s stool. Surgical removal of both the outer and the inner cyst was performed: the outer capsule ruptured during the procedure with no clinical consequence. Complementary treatment with albendazole and praziquantel was administered, and the patient displayed complete neurological recovery, with no residual lesion demonstrated in postoperative MR imaging.

Although rare, cerebral coenurosis should be considered in the differential diagnosis of a brain cyst. Neurosurgical removal of the cyst is the treatment of choice, followed by complementary antibiotic administration.

**References**


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