Detection of an Aberrant Motile Larval Form in the Brain of a Patient with Neurocysticercosis

M. E. Hellard, A. C. Street, P. D. R. Johnson, E. A. Popovic, and G. V. Brown

Neurocysticercosis is the most common parasitic infection of the central nervous system in humans. The adult pork tape worm, *Taenia solium*, resides in the lumen of the intestine, and the only structural element of an adult worm usually found within a tissue cyst in the brain or in muscle is a single invaginated scolex. We report a highly unusual, and perhaps unique, occurrence of neurocysticercosis in which an aberrant worm-like larval form, 12.5 cm in length, was found within a cyst in brain parenchyma.

Neurocysticercosis is an endemic disease in many countries [1, 2], in which the larvae of the pork tape worm, *Taenia solium*, develop into cysts within the brain. We describe a case of neurocysticercosis in a diabetic Australian man in whom a live worm-like larval form was found within a cyst in the brain parenchyma. We are unaware of any other similar reports in the worldwide literature.

Case Report

A 50-year-old Australian man was admitted to the Royal Melbourne Hospital (Parkville, Victoria, Australia) 1 week after returning from Tanzania, where he had lived for 5 years. He had been ill for 6 weeks with intermittent fevers and diarrhea, a worsening headache, and a brief episode of left-hand weakness and clumsiness. His type II diabetes, which was usually well controlled with diet and oral hypoglycemic agents, had become difficult to manage. In Tanzania he received chloramphenicol for presumed typhoid fever, but his headaches persisted and he returned to Australia. After his return, his left-sided weakness recurred, and he sought medical attention. On physical examination he had mild weakness of the left upper and lower limbs. A cerebral CT scan revealed multiple space-occupying, ring-enhancing lesions with surrounding edema involving the right frontoparietal, right parieto-occipital and left occipital regions (figure 1). He had a total WBC count of 8.3 × 10⁹/L, with a normal differential. No organisms were isolated from blood and fecal cultures. No cysts, ova, or parasites were detected in feces. The results of other investigations were normal, and because of the patient’s apparent response to antibiotics, with relapse on cessation of treatment, a provisional diagnosis of multiple cerebral abscesses was made.

A stereotactic biopsy of a right parieto-occipital lesion was performed to identify the infecting microorganism. Turbid fluid containing polymorphonuclear leukocytes was obtained, but a gram stain and cultures were negative, and there was no evidence of a cyst on histology. The patient was treated empirically with penicillin, cefotaxime, metronidazole, and dexamethasone. His postoperative course was complicated by intermittent facial weakness and slurred speech, persistent left-arm weakness, low-grade fever, and poorly controlled diabetes. A repeated CT scan showed a slight increase in the size of the right frontoparietal lesion. Three weeks after admission he underwent open craniotomy. The right frontoparietal lesion was incised and pus-like fluid was aspirated. A thin ribbon-like structure, which did not resemble brain tissue, was adherent to the end of a sucker. It moved periodically after placement in a specimen jar.

The specimen was 12.5 cm in length, ~2 mm in width, white, and bulbous at one end. Under a dissecting microscope, a live worm comprising a scolex with suckers and hooks and a segmented body was seen. When stained and sectioned, it was confirmed to be an aberrant larval form of *T. solium*. The scolex had four suckers and 28 hooks in two rows. The body had pseudosegments but, unlike an adult worm, had no identifiable sex organs (figure 2).

The patient was treated with albendazole, 15 mg/kg in three divided doses, for 8 days, without complications [3]. Serology for cysticercosis (Immunoelectrotransfer blot, Centers for Disease Control and Prevention, Atlanta), requested before the first operation, was positive [4]. Three months after discharge from the hospital, the patient had minimal left-arm weakness, and a CT scan showed that the lesions had resolved.

Discussion

Neurocysticercosis is a disease of the CNS that is endemic in many countries but uncommon in Australia. When it occurs in Australia, it is most often found in immigrants from countries where it is endemic or in long-term travelers returning from regions of endemicity [1, 2, 5–7]. The disease rarely occurs in short-term travelers.
and perhaps unique. Aberrant larval development has been described previously only in necrotic cysts, within the human eye, and, most recently, in the fourth ventricle of a brain visualized by MRI [11]. Autopsies of patients with neurocysticercosis who die suddenly have revealed cysts containing scolices with thin necks extending into the entrance canal or beyond the bladder wall [10]. In a study of the beef tapeworm *Taenia bovis*, live cysticerci were seen with the scoleces extending beyond the bladder wall, but these larval forms measured <10 mm [10]. The lack of previous reports describing aberrant larval development within brain parenchyma suggests that the occurrence is unusual, but biopsies are reserved for unusual or difficult cases in countries where the tapeworm is endemic. It is possible that aberrant larval formations occur but are not recognized because biopsies are not performed.

We can only speculate on the factors responsible for transformation from the larval form into the adult form. The CT findings were consistent with cyst degeneration, and the aberrant

Humans are the definitive host for *T. solium*, harboring the adult tapeworm in the intestines. The adult worm produces eggs that are shed into the environment. Pigs, the usual intermediate host, ingest the fertilized eggs and develop cysts within tissues such as muscle. The cycle is completed when humans ingest undercooked pork containing cysts that mature into adult tapeworms in the small intestine. Humans can also act as the intermediate host if they ingest eggs excreted by the tapeworm. After ingestion, the eggs undergo peptic digestion and develop into oncospheres that pass through the bowel wall into the bloodstream and are carried to peripheral tissues. In humans, the oncospheres have a predilection for the brain, the muscles, and the eye [8, 9]. The oncosphere develops into a cystic cavity, usually containing an invaginated scolex with suckers and hooks and a neck sitting within the spiral canal [10]. Scolex formation in the human brain is usually considered a “dead-end” in the life cycle of *T. solium*. The cysts degenerate over time and should not develop into a live tapeworm.

The finding of a live larval form of *T. solium*, measuring 12.5 cm, within our patient’s brain parenchyma is most unusual
larval growth may have occurred as an adaptive response to imminent death, similar to transformation of larvae observed in vitro in response to stress. Our patient’s recent operation, postoperative steroid therapy, and poorly controlled diabetes may have been contributing factors. Antibiotics should have had no effect on the cyst.

The management of cysticercosis and the role of drug therapy is uncertain [12]. Albendazole or praziquantel with steroids were considered the therapies of choice at the time our patient was in the hospital [3]. Recent randomized controlled trials have shown no clinical or radiological benefit from the addition of anticestodial therapy to symptomatic care in selected patients [12]. Uncertainty remains as to which patients require supportive therapy alone, which patients would benefit from anticestodial drugs, and the timing of the introduction of these drugs. Further randomized trials should help to resolve these questions.

Acknowledgments

The authors thank Dr. Joc Forsyth (Department of Microbiology, University of Melbourne) and Dr. Robin Gasser (Department of Veterinary Science, University of Melbourne), who helped identify the larval form of *T. solium*.

References