# **CASE REPORT**

# The first case of neurocysticercosis diagnosed ante-mortem in Malaysia

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#### Abstract

Cysticercosis had been exceedingly rare in Malaysia, with only one case of neurocysticercosis diagnosed during post-mortem in 1934. This is the report of the first case of neurocysticercosis diagnosed ante-mortem in Malaysia. An immigrant who was a 40-years old ethnic Indian housewife presented with recent onset of left upper limb weakness. Magnetic resonance imaging of the brain showed an enhancing right parietal cyst with a scolex. Serology for *Taenia solium* antigen was strongly positive. CT scan of brain showed multiple calcifications. There was long latency of more than 5 years when the patient left India, her original country of residence and the clinical disease. In view of the large immigrant population in Malaysia, the case emphasizes the importance of improved surveillance for cysticercosis.

#### INTRODUCTION

Human tapeworm infection by Taenia solium is endemic in areas where undercooked pork is consumed. Its prevalence is high in sub-Sahara Africa, Central and South America, India, China and parts of South East Asia.1 Cysticercosis refers to the human infection with the cysticercus stage (larva) of Taenia solium. It is acquired by accidental ingestion of eggs found in excreta of patients harbouring the adult worm as a result of contamination of food and water. Cysticercosis is one of the most common parasitic diseases of the nervous system in humans especially in developing countries.<sup>2</sup> The most common form is neurocysticercosis which commonly presents with epilepsy, headache, personality change, aphasia and limb weakness. About 50,000 deaths occur due to neurocysticercosis annually throughout the world, with no less than 20 million people infected by cysticerci.<sup>3</sup> Cysticercosis has been reported to be rare in Malaysia.<sup>4</sup> We describe the first case of an immigrant who was confirmed to have neurocysticercosis ante-mortem.

## CASE REPORT

A 40 years old ethnic Indian immigrant woman was previously living in New Delhi, India. At age 20 years, she had an acute episode of abdominal pain and severe vomiting. She passed out a tapeworm in her stool, which was confirmed to have proglottids and ova of *Taenia solium*. She was treated with niclosamide and saline purge, and has been asymptomatic till the recent illness. There was no other previous medical disorder.

She migrated to Malaysia at age 35 years and had not returned to India since then. Her recent admission to the Hospital was due to progressive weakness of left upper limb for one week. Other than weakness and hyperreflexia of the left upper limb, the physical examination was normal. Systemic investigations including full blood count, serum urea and electrolyte, chest X-ray were all normal. Enzyme immunoassay test using purified extracts of *Taenia solium* antigen was strongly positive for serum IgG with a ratio of 1.4 (positive if greater than 1.1). Serological tests were negative for amoebiasis, schistosomiasis, echinococcosis, toxocariasis and toxoplasmosis.

Computerised Tomographic (CT) study of the brain showed a large focal area of white matter hypodensity in the right parietal region, consistent with cerebral oedema. There were several calcified spots in other parts of the brain without surrounding oedema (Figure 1). T1 sequence with gadolinium of MRI brain showed a rounded cystic area in the right parietal region with signal intensity slightly higher than cerebrospinal fluid.

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There was marked ring enhancement and surrounding oedema (Figure 2). Several low signal intensity lesions on T1 and T2-weighted images corresponding to the calcified lesions on CT were also seen. On fluid-attenuated inversion recovery (FLAIR) sequence, a high-signal nodule, appearance of the scolex was seen at the center of the right parietal cyst (Figure 3). There were two other smaller isodense cysts with ringenhancement at the right parietal region without scolex or surrounding oedema. Lumbar puncture was not carried out.

Following treatment with albendazole 15 mg / kg in divided doses and oral prednisolone for four weeks, she had complete clinical recovery. A follow-up gadolinium MRI brain four months later showed that the original contrast enhancing right parietal lesion with scolex has become much smaller, the contrast enhancement less prominent, with no scolex and oedema. There was no other enhancing lesion while the calcified spots remained unchanged.

## DISCUSSION

Cysticercosis has been said to be rare in Malaysia despite considerable consumption of pork particularly among the ethnic Chinese population.<sup>4</sup> This was attributed to the systematic examination of carcasses in the abattoirs, and the local practice of thorough cooking of pork before human December 2001



Figure 2. MRI brain (T1-weighted sequence with gadolinium) showing rounded cystic area with marked ring enhancement in the right parietal region. There was significant surrounding oedema. This corresponded with the "colloidal vesicular stage".



Figure 1. Brain CT scan showing an area of focal hypodensity in the right parietal region consistent with cerebral oedema. No scolex was visible. The calcified spots at other regions represented dead and calcified larva.



Figure 3. MRI brain with FLAIR sequence. Arrow showing high-signal nodule representing the scolex at the center of the cyst in the right parietal region.

consumption. In 1937, the Director of Veterinary Research reported that of the 744,379 pigs slaughtered in Malaya, 1235 carcasses were positive for Cysticercus cellulosae. Of these only nine carcasses were pigs bred locally. Since then, the pig-farming industry has expanded with the pork consumed supplied locally. As for human disease from cysticercosis, so far there has been only three cases reported in the medical literature involving the brain, muscle and larynx.5-7 The only case of neucysticercosis was diagnosed in a Malayan Chinese at necropsy close to seven decades earlier in 1934.<sup>5</sup> The present case is remarkable as it is the first case of neurocysticercosis diagnosed ante-mortem, after such a long interval.

The social situation in Malaysia has changed over the past two decades with massive influx of immigrants, some from countries endemic for cysticercosis such as India<sup>1</sup> and Indonesia.<sup>8</sup> Currently, there are about 1.5 million registered immigrants working in Malaysia, excluding the illegal immigrants who usually do not undergo medical examinations prior to entry into the country. Serological survey of registered immigrants showed that 0.05% was positive for Taenia solium (personal communication: Zaini A and Anuar K, University Malaya Medical Centre). Some of the immigrants may thus be tapeworm carriers who can transmit the infection to the local population, including the Muslims population that does not consume pork. This is illustrated by the report of neurocysticercosis in four Orthodox Jews in New York who did not consume pork. The infection was attributed to transmission from immigrants employed as housekeepers.9 The apparent scarcity of cysticercosis in Malaysia may be partly due to lack of awareness and under-diagnosis. Furthermore, neurocysticercosis have protean neurological manifestations and is a great imitator of many neurological disorders.<sup>10</sup>

The diagnosis of neurocysticercosis in this patient is supported by the demonstration of scolex within a cyst by imaging which was said to be pathogomonic of neurocysticercosis.<sup>11</sup> The recently proposed diagnostic criteria by Brutto et al also listed the same imaging change as absolute definitive criteria for diagnosis of neurocysticercosis.12 This patient also fulfill other criteria by Brutto et al12, they are: MRI brain showing enhancing cystic lesions without scolex, brain parenchymal calcifications, positive serological test for Taenia solium antigen, limb weakness, and previously living in an endemic

area. The serological test used in this patient was enzyme-immunoassay\_which has been reported to have a sensitivity of 100% and specificity of 80%.<sup>13</sup> The specificity increases if the patient is from an endemic area and is said to reach 100% with supporting evidence of MRI. One of the limitations of the test was the cross-reactivity with *Echinoccoccus spp*. However, infection with *Echinococcus spp* has not been reported in Malaysia.

As the patient migrated to Malaysia from India five years earlier without returning to India in the interval period, the most likely source of infection was consumption of food or water contaminated by the parasitic eggs when she was in India. In view of the severe vomiting that she had at age 20 years, another possibility was auto infection whereby she had regurgitated the proglottids into her stomach, resulting in liberation of eggs. The long interval between the infection and clinical symptoms of at least five years was consistent with a previous study, which reported a latency varying between a few months to 30 years, average 4.8 years.<sup>14</sup> The invasion of the brain by larva of Taenia solium has been known to result in a unique host-parasite immune response.<sup>15</sup> By the time the host has generated an antibody response, the parasite could develop the ability to evade the complement and antibodymediated destruction by the host. It could also modulate the host immune system by interfering with the lymphocyte proliferation and macrophage function. This effect on the host immune response could be localized to the area immediately surrounding the larva, as it has been observed that inflammatory response in a cyst could coexists with another viable cyst that has no evidence of inflammation. As the larva dies, the immunosuppression faded, resulting in destruction of larva by host immune response (the "colloidal vesicular stage"). The mechanism of death of the larva remains speculative. It may have resulted from either failure of protective mechanism in that particular cyst or simply senescence.<sup>16</sup>

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