



Neurocysticercosis Diagnosed in a Patient with *Taenia saginata* Taeniasis after Administration of Praziquantel: A Case Study and Review of the Literature

Toni Wandra¹, Raka Sudewi², Ni Made Susilawati², Kadek Swastika³, I Made Sudarmaja³, Luh Putu Eka Diarthini³, Ivan Elisabeth Purba¹, Munehiro Okamoto⁴, Christine M. Budke⁵ and Akira Ito^{6*}

¹Sari Mutiara Indonesia University, North Sumatra, Indonesia

²Department of Neurology, Sanglah Hospital, Udayana University, Denpasar, Bali, Indonesia

³Department of Parasitology, Udayana University, Denpasar, Bali, Indonesia

⁴Section of Wildlife Diversity, Center for Human Evolution Modeling Research, Primate Research Institute, Kyoto University, Inuyama, Japan

⁵Department of Veterinary Integrative Biosciences, College of Veterinary Medicine & Biomedical Sciences, Texas A&M University, College Station, TX, USA

⁶Department of Parasitology, Asahikawa Medical University, Asahikawa, Japan

Abstract

Taeniasis, caused by infection with *Taenia saginata* or *Taenia solium*, occurs on Bali due to the consumption of undercooked beef and pork, respectively. Fieldwork conducted on Bali from 2002-2007, identified 69 taeniasis cases due to *T. saginata*. In August 2007, three *T. saginata* tapeworm carriers in the Gianyar district of Bali were treated with a single dose of praziquantel. Within a few hours of treatment, a 47 year old man had a seizure and was admitted to a hospital in the city of Denpasar. A computed tomography (CT) scan revealed two cystic lesions in the man's brain. Serology showed specific antibody responses to *T. solium* metacestode antigens. The patient was, therefore, diagnosed with a dual infection of *T. solium* neurocysticercosis (NCC) and *T. saginata* taeniasis. This case report is illustrative of the risk of veiled NCC in areas where *T. saginata* and *T. solium* are co-endemic. As globalization increases, the possibility of co-infections will continue to rise and researchers and physicians participating in mass drug administration programs will need to be vigilant in their monitoring for adverse effects.

Keywords: Taeniasis; *Taenia saginata*; *Taenia asiatica*; *Taenia solium*; Neurocysticercosis; Praziquantel; Niclosamide; Bali; Indonesia; Asia

Introduction

Taeniasis/cysticercosis, caused by the pork tapeworm, *Taenia solium*, is a parasitic zoonosis found primarily in countries where pigs are raised and meat inspection is lacking [1,2]. The potential for a tapeworm carrier to infect another individual (or themselves via autoinfection), resulting in cysticercosis or neurocysticercosis (NCC), is a risk not only in endemic areas, but also in areas with immigrants from endemic countries [1,3-5]. An example of such an outbreak occurred in an Orthodox Jewish community in New York City when family members were infected by a domestic employee from a *T. solium* endemic country [6]. Such NCC outbreaks have occurred in other developed countries, including countries in the Middle East where people do not consume pork [4,5,7-13].

In Asia, there are three *Taenia* tapeworms that infect humans; *T. solium*, *Taenia saginata* and *Taenia asiatica* [4,5,14-24]. Although adult *T. solium* worms with scoleces are easily differentiated morphologically from other taeniids, it is almost impossible to differentiate adult *T. asiatica* and *T. saginata* [5,22,25,26]. Therefore, molecular differentiation is often required to correctly identify the infecting species [27]. Recent studies have also reported hybrids of *T. saginata* and *T. asiatica* in Thailand and China [5,25,28-32]. This makes mitochondrial and nuclear gene analyses even more important for the differentiation of *T. saginata*, *T. asiatica*, and hybrids of these two species [30-32].

In Indonesia, *T. solium*, *T. saginata* and *T. asiatica* are distributed according to livestock rearing and local food consumption practices [33-40]. Both *T. saginata* and *T. solium* have been confirmed from the island of Bali, where the local inhabitants are known to eat dishes that contain undercooked beef and pork, such as the traditional dish Lawar. NCC was considered endemic on Bali more than 20 years ago [41,42]. However, education programs aimed at keeping pigs indoors to prevent access to human faeces and the introduction of improved sanitation

systems and practices have substantially reduced the prevalence of NCC during the past 2 decades [35,37,43]. Nevertheless, there are still sporadic NCC cases identified on Bali [40,42-45].

Case Report

Fieldwork conducted on Bali from 2002-2007 resulted in the identification of 69 taeniasis carriers who were treated with a single dose of praziquantel (PZQ) at 15 mg/kg body weight (Table 1). All worms were confirmed to be *T. saginata* by multiplex PCR [36]. In August 2007, a 47-year-old male taeniasis carrier from the Gianyar district suffered a single tonic-clonic seizure within a few hours of PZQ administration. The patient was subsequently admitted to the Sanglah Hospital in Denpasar. A computed tomography (CT) scan revealed one viable cystic lesion and one calcified lesion in the right frontal lobe of the brain. At this time, serology (ELISA) was also carried out using partially purified antigens by cation exchange chromatography [46]. The patient was subsequently diagnosed with NCC and treated with albendazole for one month. He was followed serologically through May 2015 (Table 2).

Discussion

Although PZQ has been recommended for the treatment of taeniasis, there are reports of seizures triggered within a few hours of

*Corresponding author: Akira Ito, Department of Parasitology, Asahikawa Medical University, Asahikawa, Japan, E-mail: akiraito@asahikawa-med.ac.jp

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District* (Year)	No. of <i>T. saginata</i> taeniasis cases	No. of <i>T. solium</i> taeniasis cases
Gianyar (2002) [#]	32	0
Gianyar (2004) [#]	14	0
Gianyar (2005) [#]	5	0
Gianyar (2006) [#]	2	0
Gianyar (2007) [#]	3	0
Gianyar (2008)	4	0
Gianyar (2009)	7	0
Gianyar (2010)	18	0
Gianyar (2011)	9	0
Gianyar (Jan 2013)	6	0
Gianyar (Sept 2013)	9	0
Gianyar (2014)	4	0
Badung (2004) [#]	1	0
Denpasar (2004) [#]	9	0
Denpasar (2005) [#]	2	0
Denpasar (2010)	3	0
Karangasem (urban area, 2006) [#]	1	0
Bangli (2007) [#]	0	0
Tabanan (2008)	0	0
Jembrana (2008)	0	0
Klungkung (2009)	0	0
Buleleng (2009)	0	0
Karangasem (rural area, 2011)	0	3
Karangasem (rural area, Jan. 2013)	0	6
Karangasem (rural area, Sep. 2013)	0	2 [§]
Karangasem (rural area, 2014)	0	2 [§]
Total	129	13

* See Figure 1.

[#] Treated with Praziquantel.

[§] Swastika et al., unpublished.

Table 1: Number of taeniasis cases due to *T. saginata* and *T. solium* on Bali 2002-2014 [11].

drug administration. The occurrence of seizures is often accompanied by an antibody response that can remain for months [2,9,47-52]. PZQ is also the first choice for treatment of trematodiasis, including schistosomiasis and food-borne trematodiasis [53-56]. Therefore, in regions such as Asia where trematode and *T. solium* infections both occur, there is also the risk of a negative reaction if an individual treated for trematodiasis has silent NCC.

Previously asymptomatic NCC may become symptomatic when the parasite(s) are damaged by the host's immune response or by PZQ treatment. Therefore, there is a debate whether NCC patients should be treated with an anthelmintic, such as albendazole, to accelerate the death of the parasite or solely with a steroid to control the inflammatory response [2,9,47-53]. The patient described in this case report had no history of epileptic seizures before PZQ treatment. A single dose of PZQ at 40 mg/kg is typically used for mass drug treatment of trematodiasis, whereas a single dose of PZQ at 10 mg/kg is considered >95% efficacious for treating taeniasis [2,57]. In comparison, a study conducted in Mexico reported that symptoms associate with NCC could be induced with a single dose of PZQ at 5 mg/kg [58,59].

In Indonesia, taeniasis is primarily caused by *T. saginata*. Even on Bali where the local inhabitants eat both pork and beef, the majority of tapeworms are identified as *T. saginata* (Table 1 and Figure 1). However, since January 2011, a total of 13 *T. solium* tapeworms have been confirmed from 13 villagers from small remote villages located on the

Date	Absorbance value	Cut off
August 2007 (before treatment)	0.103	0.022
December 2007 (after treatment)	0.578	
February 2009	0.070	0.038
May 2015	0.016	

Table 2: ELISA results from 2007 until 2015.



Figure 1: Map of Indonesia (upper) showing endemic areas for the three human *Taenia* species (North Sumatra: *T. asiatica*; Bali: *T. saginata* and *T. solium*; and Papua: *T. solium*). The lower map depicts Bali's nine districts: on Bali (lower). Jembrana (01), Tabanan (02), Badung (03), Denpasar (04), Gianyar (05), Bangli (06), Klungkung (07), Karangasem (08), and Buleleng (09). Denpasar is the capital city of Bali [43].

eastern slope of Mt. Agung in the Karangasem district of northeastern Bali (Table 1). The patient described in this case report had no history of visiting Karangasem. Therefore, it is likely that he became infected with *T. solium* eggs in Gianyar or the surrounding area. Additional studies are needed to better elaborate the epidemiology of *T. solium* in Gianyar.

On Bali, *T. saginata* taeniasis is rather common in communities where people like to eat dishes prepared with undercooked beef (Table 1) [11,36,43]. While *T. saginata* taeniasis carriers are not directly a public health threat, *T. solium* taeniasis carriers can potentially infect others (or themselves via autoinfection). This is especially worrisome when considering that populations from known *T. solium* endemic regions of Bali, such as Karangasem, often travel to other parts of the island to find work [11].

If PZQ is recommended for mass drug treatment of trematodiasis or taeniasis where *T. solium* is distributed, seizures related to previously asymptomatic NCC should be anticipated. There are currently two choices for anthelmintic treatment of tapeworm carriers. The first is treatment with PZQ and the second is treatment with niclosamide. Since 2008, all *T. saginata* taeniasis cases (n=60) on Bali have been treated with niclosamide (single dose of 2,000 mg). Thus far, no NCC related clinical manifestations have been reported in response to niclosamide administration, including in 3 patients with confirmed dual infections [40]. Chinese traditional medicine (pumpkin seeds plus areca nut extract) has also been used to expel tapeworms without the risk of causing seizures [26,54,60].

In order to avoid inducing seizures in previously asymptomatic NCC patients, it would be beneficial to test taeniasis carriers for a *T. solium* cysticercosis antibody response prior to treatment in areas where *T. solium* is sympatrically distributed with other human *Taenia* species [60]. For such real-time serodiagnosis in the field, more rapid field-based tests are needed [4]. In the time being, researchers and physicians participating in mass drug administration programs using PZQ should be vigilant in their monitoring for adverse effects of drug administration that may be attributable to previously silent NCC.

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