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Submission date: 20-Oct-2023 12:05PM (UTC+0700)

Submission ID: 2201546227

File name: ntation_of_Taenia_solium_control_measures_in_Bali,_Indonesia.pdf (743.98K)

Word count: 6672

Character count: 35306



Implementation of *Taenia solium* control measures in Bali, Indonesia: Survey findings and a historical overview

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ARTICLE INFO

Keywords:

Taenia solium taeniasis
Cysticercosis
Eradication
Bali
Indonesia

ABSTRACT

Historically, cysticercosis cases caused by infection with the larval stage of the *Taenia solium* tapeworm have occurred sporadically in Bali, with taeniasis carriers found primarily in villages located in the Kubu subdistrict of Karangasem. As Bali is a well-known tourist destination, living standards are relatively high on the island, except for an area located beneath the northeast slope of the active volcano Mt. Agung (Alt. 3031 m), which includes Kubu. Over the last 30 years, pigs originating from this area have been found with *T. solium* cysticerci, whereas pigs from other parts of the island have not been shown to be infected. Out of 108 individuals screened via fecal sample examination in Kubu during 2019, 3 cases of *T. solium* taeniasis (2.8%) were identified. There was no significant difference in taeniasis prevalence from surveys conducted in 2011–2016 (1.0%, 11/1089) ($p = 0.123$). Out of 110 humans and 140 pigs tested serologically in Kubu during 2019, no cases of cysticercosis were identified. This is in contrast to a seroprevalence of 4.1% (42/1025) in humans during 2011–2016 and a seroprevalence of 13.1% (43/329) in pigs during 2011–2013. Over the last decade, improved drinking water and sanitary systems have been employed in addition to health education targeting primary school children, including emphasis on washing hands before eating and after defecation. This review provides previously unpublished survey data and a historical overview of *T. solium* infection in Bali and offers guidance on best practices to ensure that remaining pockets of transmission are addressed.

1. Introduction

The pork tapeworm, *Taenia solium*, is the causative agent of human cysticercosis (Ito and Budke, 2021). This condition is acquired through ingestion of parasite eggs shed in the feces of a person with adult *T. solium* tapeworm(s) in their gastrointestinal tract (taeniasis). Acquisition of the adult worms by humans results from consumption of raw or undercooked pork containing larval cysts. The parasite commonly circulates in areas without sufficient meat inspection, and is often endemic in remote/rural areas where pigs are raised in conditions with low

biosecurity and low sanitation levels.

Infection with the larval stage of the parasite (cysticercosis) in humans can result in clinical manifestations related to cyst location. The condition is especially problematic when cysts enter the central nervous system, resulting in neurocysticercosis (NCC). Cysts in the brain can cause epileptic seizures and severe chronic headaches (Carabin et al., 2011). While cysticercosis and NCC are most commonly found in developing regions, tapeworm carriers may travel to non-endemic regions, resulting in a potential risk to others if appropriate hand hygiene and sanitation practices are not performed. Therefore, through

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<https://doi.org/10.1016/j.actatropica.2021.106297>

Received 24 December 2021; Accepted 25 December 2021

Available online 28 December 2021

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globalization, cysticercosis and NCC may have an impact on healthcare resources even in non-endemic regions due to the presence of visitors and immigrants from endemic countries (Schantz et al., 1992; Hira et al., 2004; O'Neal and Flecker, 2015; Ito and Budke, 2021).

Parts of Indonesia are known to be endemic for *T. solium*. Endemic areas are defined as regions where the full parasite life cycle is occurring (PAHO, 2021). In 1996, the Jayawijaya district of Papua was identified as an endemic area (Wandra et al., 2000, 2003; Subahar et al., 2001; Margono et al., 2003, 2006; Salim et al., 2009). In 2011, a second *T. solium* endemic area was identified in the Karangasem district of Bali (Fig. 1). This area was identified after a 9-year-old girl from the Kubu subdistrict of Karangasem was diagnosed with ocular cysticercosis at Indera Hospital, which is located in Denpasar, the capital city of Bali (Swastika et al., 2012; Wandra et al., 2013). Prior to 2011, NCC cases were diagnosed sporadically at Sanglah Hospital in Denpasar. However, the infection source was unknown, and *T. solium* taeniasis cases had not been detected via field surveys conducted from 2002 to 2010 in the 8 districts (regencies) of Bali or the capital city (Wandra et al., 2011, 2015) (Fig. 1).

During 2011–2019, *T. solium* control measures were implemented in Karangasem (Fig. 1) in collaboration with the central government of Indonesia, the Bali Provincial Health Office, the Karangasem District Health Office, public health centers, the local government, and health cadres. Activities included community studies to identify and treat cases of taeniasis as well as soil-transmitted helminth (STH) infections. These initiatives also had a strong health education component (MHRI, 2017). In addition, individuals with a history of epileptic seizures (suspected NCC) or individuals with subcutaneous nodules (suspected subcutaneous cysticercosis) were referred to higher levels of care for further

diagnostics and treatment. During nine field surveys conducted between 2011 and 2019, research teams also provided health education for communities in Karangasem (Fig. 2). One example was the use of a Snakes and Ladders type board game to improve children's knowledge about taeniasis (Wulanyani et al., 2019) (Fig. 2b.).

In addition to health education aimed at human infection, local control efforts included collaboration with the livestock sector to detect cysticercosis in pigs. As part of these efforts, which took place in January and September of 2011 and 2013, pigs that were seropositive for cysticercosis were slaughtered and necropsied in the field in order to show the cysticerci to the local population and describe the parasite life cycle and how to prevent infection in pigs (Fig. 3). The main messages being conveyed during these sessions were to prevent pigs from accessing human waste and to not consume raw or undercooked pork or any pork with cysticerci.

This paper provides information on previously unpublished epidemiological studies conducted between 2011 and 2019 and presents an overview of control processes put in place in Bali during 2011–2019, with the goal of eradicating local *T. solium* transmission.

2. Materials and methods

Community-based epidemiological surveys were conducted in the *T. solium* endemic subdistrict of Kubu during 2011, 2013, 2014, 2015, 2016, and 2019, and in the non-endemic subdistrict of Karangasem during 2017. Findings from the 2011–2016 surveys were previously published in Wandra et al. (2013, 2015), Swastika et al. (2017), and Sutisna et al. (2019), while findings from the 2017–2019 surveys have not been previously published.

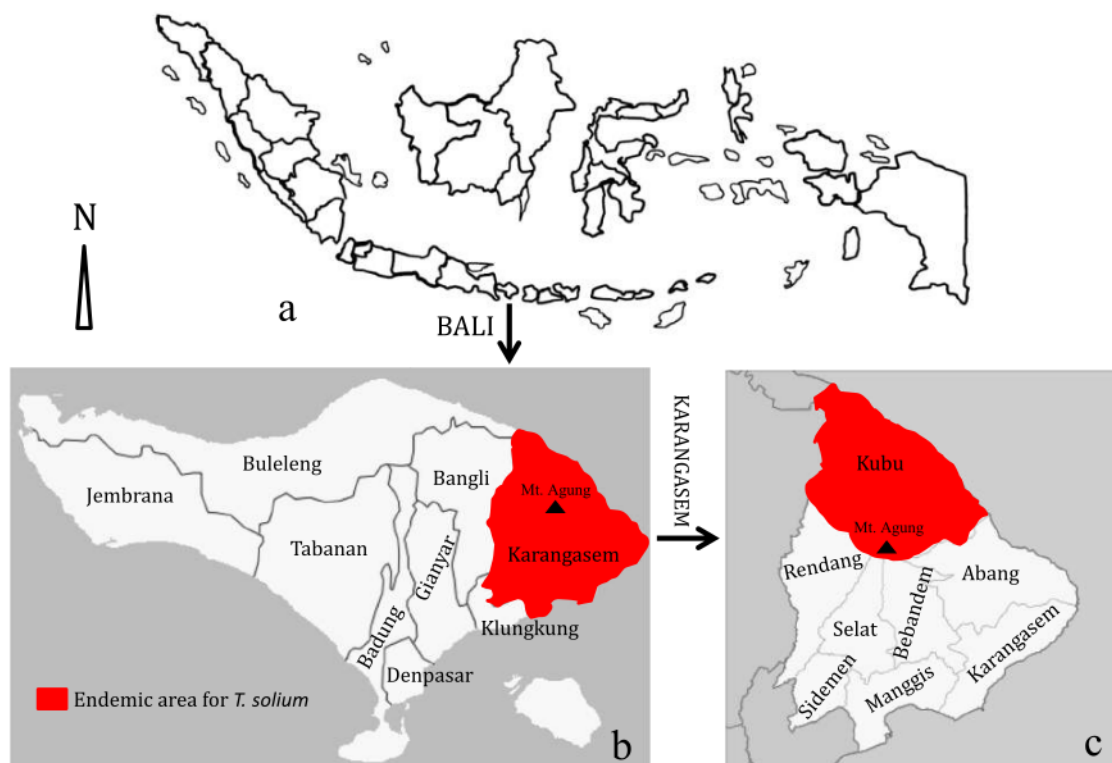


Fig. 1. Map of Indonesia (a) and a map of Bali (b) showing the endemic area for *Taenia solium* and (c) The *Taenia solium* endemic Kubu subdistrict of Karangasem. Kubu subdistrict consists of 9 villages: Ban, Baturringgit, Dukuh, Kubu, Sukadana, Tianyar Barat, Tianyar Tengah, Tianyar, and Tulamben.



Fig. 2. A banner displaying information about the *Taenia solium* life cycle (a), and school children playing a Snakes and Ladders board game containing information about *T. solium* transmission and prevention (Wulanyani et al., 2019) (b).



Fig. 3. Images showing the collection of blood samples from pigs to look for cysticercosis in collaboration with the Karangasem District and Kubu Subdistrict Livestock Offices (a, b, c), and necropsy of pigs with cysticercosis used as an opportunity to educate community members about the *Taenia solium* life cycle and control measures (d).

Kubu subdistrict is located in the Karangasem district of Bali and consists of nine villages (Fig. 1), with each village containing six banjars. A banjar is the smallest administrative unit (sub-village) with 159–165 families and 782–800 inhabitants (Wandra et al., 2006a). The study banjars were selected after identifying a locally acquired ocular cysticercosis case in 2010 and with the recommendations of local health centers. Studies were carried out in the village of Dukuh during January and September of 2011 (3 banjars), in Dukuh during January and

September of 2013 (3 banjars), in the villages of Dukuh and Tulamben during September of 2014 (4 banjars), in Dukuh and Tulamben during September of 2015 (4 banjars), in the villages of Dukuh and Kubu in September of 2016 (3 banjars), and in Dukuh during November of 2019 (3 banjars). Community-based surveys were also conducted in non-endemic areas of Karangasem subdistrict (villages of Bugbug, Seraya Barat, and Seraya Tengah) (one banjar in each village) during 2017 (Fig. 1).

For each community field survey (2011–2017 and 2019), after obtaining informed consent/assent, members of the study villages 2 years of age and older were invited to provide a fecal sample as well as obtain a free check-up at the local banjar's administrative office. A questionnaire was administered to identify any individuals with a history of epileptic seizures (suspected NCC). In addition, each participant was given a physical examination, which also included palpation for subcutaneous nodules (suspected subcutaneous cysticercosis). Fecal samples were examined microscopically using the Kato-Katz technique. Individuals who were found to have taeniid eggs in their feces were treated with niclosamide (Yomesan®, Bayer, 2 gr single dose) followed by 30 g of magnesium sulfate (Swastika et al., 2017). Expelled tapeworm material was collected for speciation. In addition, respondents with STH infections were treated with albendazole (400 mg single dose) for one day, except for those diagnosed with trichuriasis who received 400 mg of albendazole for 3 consecutive days. If a village was sampled over multiple years, fecal samples were only collected for an individual during a single sample year.

During the community surveys, a blood sample was also collected for ELISA and immunoblot testing for cysticercosis from survey participants. A questionnaire collecting information on possible risk factors for taeniasis (e.g., eating uncooked pork) and cysticercosis (e.g., sanitary facilities) was administered to survey participants 18 years of age and older by trained health workers and members of the researcher team. An adult or guardian responded to the questionnaire on behalf of survey participants less than 18 years of age. During 2019, researchers also went door-to-door to administer questionnaires to households not participating in testing for taeniasis or cysticercosis.

2.1. Identification of infecting species

All tapeworms expelled from tapeworm carriers were confirmed by mtDNA analysis using the cytochrome c oxidase subunit 1 (*cox 1*) gene to differentiate *T. solium*, *T. saginata*, and *T. asiatica* (Yamasaki et al., 2004).

2.2. ELISA and immunoblot

ELISA and immunoblot were carried out using glycoproteins (GPs) from a pH 8.1 fraction purified by preparative isoelectric focusing from *T. solium* cysts (Rotofor, BioRad, Hercules, CA, USA) (Ito et al., 1998, 1999) and a chimeric recombinant antigen (modified from Sako et al., 2000). Detection of antibodies by ELISA was performed as described by Ito et al. (1998, 1999) and Sako et al. (2000).

2.3. Pig surveys

During community surveys, serum samples were collected from all pigs 3 months to 3 years of age. Blood samples were collected from the jugular vein using a 5 ml syringe, with the samples kept at room temperature for three hours. After centrifugation, samples were assessed at a research team base camp in Karangasem using an ELISA that could be evaluated as a color change perceived by the naked eye. All serum samples were then stored at -20°C until they could be assessed using an ELISA reader at the Department of Parasitology, Faculty of Medicine, Udayana University in Denpasar, Bali and/or Asahikawa Medical University, Japan. Necropsies were performed on pigs with positive serology. The entire carcass was examined, with special attention given to muscle (including tongue), the heart, eyes, liver, lungs, kidney, and the brain.

2.4. Data analysis

Taenia solium taeniasis prevalence was calculated as the number of *T. solium* taeniasis cases (as determined by fecal examination) divided by the total number of individuals sampled $\times 100$ for each study year.

Cysticercosis prevalence was calculated as the number of serology positive cases divided by the total number sampled $\times 100$ for each study year. Comparison of infection prevalence by year(s) was conducted using the Pearson's chi square or Fisher's exact test, if one or more of the values in a 2×2 contingency table was less than 5. A *p*-value < 0.05 was considered statistically significant (Table 2).

3. Results

During the 2011–2016 community surveys, 1089 fecal samples and 1025 blood samples were collected from the inhabitants of study villages located in the endemic Kubu subdistrict of Karangasem. All 1089 participants were also given physical examinations and answered questions regarding possible symptoms associated with NCC. In 2019, 110 individuals, from the village of Dukuh located in the subdistrict of Kubu, were given physical examinations and answered screening questions. Of these 110 individuals, 108 provided fecal samples and 110 provided blood samples. In 2017, 123 individuals, from villages located in the non-endemic subdistrict of Karangasem, were given physical examinations and answered screening questions. Of these 123 participants, 68 provided fecal samples and 123 provided blood samples.

During the 2011–2019 surveys conducted in the Kubu subdistrict of Karangasem, 14 individuals had confirmed taeniid eggs on fecal examination and were treated with Yomesan®. All tapeworms collected from the Kubu subdistrict ($n = 14$) were confirmed as *T. solium* by multiplex PCR using the mitochondrial cytochrome c oxidase subunit 1 (*cox 1*) gene. None of the 68 individuals who provided fecal samples in the non-endemic subdistrict of Karangasem during 2017 were found to have taeniasis (Fig. 1).

The prevalence of *T. solium* taeniasis in Kubu subdistrict was 1.0% (11/1089) during 2011–2016 (data previously published in Swastika et al., 2017) and 2.8% (3/108) in the village of Dukuh in 2019 (Table 2). There was no significant difference in prevalence between studies conducted in 2011–2016 and those conducted in 2019 ($p = 0.123$). The largest number of taeniasis cases for all study years was found in the 31–40-years age group (50.0%, 7/14), with the youngest a 4-year-old girl and the oldest a 55-year-old man. Of the 14 confirmed *T. solium* taeniasis cases, 71.4% (10/14) were male (Table 1).

Based on physical examination and health survey findings, there were no participants with suspected NCC or suspected subcutaneous cysticercosis referred to higher level care in 2011–2019. The overall cysticercosis seroprevalence in humans in Kubu was 4.1% (42/1025) in 2011–2016 (Sutisna et al., 2019) and 0.0% (0/110) in the village of Dukuh in 2019. There was a significant difference in seroprevalence between 2011 and 2016 and 2019 ($p = 0.006$) (Table 2). There were no seropositive individuals found among the 123 survey participants in the non-endemic subdistrict of Karangasem during 2017 (Fig. 1).

In 2019, questionnaires were obtained from 110 respondents (from 110 families) who provided fecal or blood samples and 250 individuals (from 243 families) interviewed door to door. Questionnaire findings showed that 80.3% (289/360) of people consumed undercooked pork in 2019. There was no significant difference compared to 2011

Table 1

Age group and sex distribution of 14 *T. solium* taeniasis cases in the Kubu subdistrict of Karangasem, Bali, 2011–2019.

Age group (year)	Sex		Total
	Male	Female	
0–10	0	1	1
11–20	1	0	1
21–30	1	1	2
31–40	6	1	7
41–50	1	1	2
51–60	1	0	1
61–70	0	0	0
Total	10	4	14

Table 2
Prevalence of taeniasis/cysticercosis in the Kubu subdistrict of Karangasem, Bali during 2011–2019.

Taeniasis/cysticercosis	Year	Prevalence (%)	p-value*
<i>T. solium</i> taeniasis	2011–2016	1.0 (11/1089) ^a	0.123
	2019	2.8 (3/108)	
Cysticercosis (seropositive humans)	2011–2016	4.1 (42/1025) ^b	0.006
	2019	0.0 (0/110)	
		13.1 (43/329) ^b	
Porcine cysticercosis (seropositive pigs)	2011–2013	13.1 (43/329) ^b	<0.001
	2019	0.0 (0/140)	

^a Swastika et al. (2017).

^b Sutisna et al. (2019).

* P-values associated with Pearson's chi square or Fisher's exact test, if one or more of the values in a 2 × 2 contingency table was less than 5. Ap-value < 0.05 was considered statistically significant.

questionnaire results that also evaluated local pork consumption practices showing the 83.9% (52/62) of survey participants consumed undercooked pork ($p = 0.312$) (Wandra et al., 2013, 2015). Furthermore, in 2011, 71.0% (44/62) of individuals from 62 families had indoor toilets (Wandra et al., 2013; 2015) compared to 98.0% (346/353) (7 individuals were from the same families) in 2019 ($p < 0.001$).

The seroprevalence of pigs in Kubu was 13.1% (43/329) during 2011–2013 (Sutisna et al., 2019) and 0.0% (0/140) in the village of Dukuh in 2019. Unfortunately, data on the seroprevalence of pigs in 2014–2016 are not available due to technical errors encountered while using a new antigen product. There was a significant difference in seroprevalence between 2011 and 2013 and 2019 ($p < 0.001$) (Table 2). Of the 43 seropositive pigs, 9 (20.9%) had lesions consistent with cysticercosis on necropsy. No seropositive pigs (0/148) were found in the non-endemic subdistrict of Karangasem in 2017 (Fig. 1).

4. Discussion and historical overview

4.1. *T. solium* transmission and implementation of control measures in Bali during 2011–2019

The transmission of *T. solium* in remote areas of Indonesia has traditionally been facilitated by the local preference for outdoor defecation even though modern toilet facilities were being provided by the government. A variety of cultural and social factors likely contributed to this preference (Gajdusek and Zigas, 1957; Gajdusek, 1977; Desowitz et al., 1977; Steadman and Merbs, 1982). In addition, keeping of free-roaming pigs with access to human fecal material and a propensity for eating undercooked infected pork have helped maintain the local parasite life cycle. Cases of human cysticercosis are known to arise through ingestion of food or water contaminated with parasite eggs (Ito and Budke, 2021).

Overall, *T. solium* taeniasis is quite rare in Indonesia, where approximately 87% of the population is Muslim (Wandra et al., 2013). That being said, *T. solium* has long been recognized as being highly endemic in the mountainous, highland area of Papua, Indonesia (formerly known as Irian Jaya) (Gajdusek and Zigas, 1957; Tumada and Margono, 1973; Desowitz et al., 1977; Subianto et al., 1978; Gajdusek, 1978; Simanjuntak et al., 1997; Ito et al., 2002; Margono et al., 2003) where the population is predominantly Christian and eats pork.

In Papua, 6 of 16 *T. solium* taeniasis cases (37.5%) were serologically confirmed to have cysticercosis in 1998 (Margono et al., 2006). In addition, two adult dogs with high antibody titers died during community surveys. Necropsies were subsequently performed, and the dogs were found to have numerous cysticerci throughout their bodies, including in the brain and heart (Ito et al., 2002; Margono et al., 2003). The consumption of dog meat in Papua, Indonesia and some other Asian

countries may further contribute to the local role of dogs in the *T. solium* life cycle.

Transmission of *T. solium* has also been reported in Bali where the majority of the population is Hindu and pork is also commonly eaten (Le Coultré, 1928). It has been suggested that pigs from Bali were initially introduced to Papua in 1969 (Wandra et al., 2003). Therefore, it has been speculated that outbreaks of NCC in Papua can be traced back to this initial importation of pigs. However, further studies are needed to fully evaluate the parasite's local origin since re-evaluation of available data has shown that *T. solium* was likely circulating in Papua prior to 1973 (Tumada and Margono, 1973; Margono et al., 2003, 2006; Wandra et al., 2003; Yanagida et al., 2021).

Historically, NCC cases have occurred sporadically in Bali, with taeniasis carriers found primarily in villages located in the Kubu subdistrict of Karangasem (Wandra et al., 2006a, 2006b, 2013, 2015; Swastika et al., 2012, 2017). As Bali is a well-known tourist destination, living standards are relatively high on the island, except for an area located beneath the northeast slope of the active volcano Mt. Agung (Alt. 3031 m), which includes Kubu. Over the last 30 years, pigs originating from this area have been found with *T. solium* cysticerci, whereas pigs from other parts of the island have not been shown to be infected (Sutisna et al., 2019).

The geography of this *T. solium* endemic area is composed largely of open grasslands and is dry for half of the year. The rainy season is November through April, while the dry season is May through October. A 9-year-old girl with ocular cysticercosis was identified, from this area, in 2010. The larva was extracted, and histopathological examination confirmed *T. solium* infection. Serological testing using ELISA and immunoblot one week and again nine months post-surgery were negative for cysticercosis (Swastika et al., 2012; Sahu and Ito, 2015).

Besides the taeniasis cases found during the community studies conducted between 2011 and 2019 in Karangasem, a single case of *T. solium* taeniasis was identified during a field survey conducted in Ketewel village, Sukawati subdistrict, Gianyar district in 2016, which is considered an endemic area for *T. saginata* taeniasis. This individual was born and grew up in Kubu subdistrict, Karangasem district, but migrated to Sukawati 5 years prior. He was known to periodically return to Kubu and consume undercooked pork (Swastika et al., 2017).

The eruption of Mt. Agung in November 2017 had a large impact on Bali, including financial losses due to loss of tourism. In addition, approximately 43,000 people were evacuated from 22 villages near the volcano, but were able to return to their homes within a month. Most pigs from these villages were either evacuated with the villagers or sold cheaply to anyone who would buy them. However, during the evacuation period, it is known that some locals left their temporary housing outside of the evacuation zone in the morning to take care of their pigs (on their farms) and returned to the camp at night. Based on conversations with the local population, it is estimated that approximately 50% of pigs from the evacuated area were sold during the time period immediately after the eruption of Mt. Agung.

In 2018, as an adjunct to community surveys, two of the authors (AI and TY) and personnel from Udayana University visited Kubu subdistrict to introduce a newly developed health education board game into primary schools. At this time, improvements in the local living conditions were again noted, including installation of trash cans with lids along the road at regular intervals, decreasing waste deposited into the environment (Wulanyani et al., 2019). Overall, it appears that the eruption did not have a major impact on local human or animal health, but rather spurred improved health education in primary schools in addition to hygiene and infrastructure improvements.

During 2019, a total of three *T. solium* taeniasis cases (a 40-year-old man, a 36-year-old man, and a 28-year-old woman) were identified via fecal examination in the endemic Kubu subdistrict of Karangasem (Table 1). One additional individual (a 40-year-old woman) who reported expulsion of *T. solium* adult worms was reported by the health center in Kubu in 2020. Despite extensive health education programs,

survey findings show that there has been little to no change in food preparation or dietary habits in endemic communities, with *T. solium* transmission from pigs to humans continuing to occur at pre-control levels.

The consumption of traditional local dishes that contain undercooked pork, such as pork *lawar* commonly occurs at religious and other festivals (Ngoerah, 1975; Sutisna et al., 2019). Pork *lawar* consists of pork mixed with vegetables, coconut, herbs, and spices (Kardena et al., 2021). In traditional village communities in Bali, men often gather to enjoy *lawar* with *tuak* (palm wine) (Wandra et al., 2006a; Kardena et al., 2021). Since these traditions are not likely to change in the short term, measures need to be put in place to prevent pigs from accessing human feces in order to prevent *T. solium* transmission from humans to pigs. Since no humans or pigs were identified with cysticercosis during the 2019 community surveys and questionnaire findings showed an increased number of indoor toilets, this may indicate that measures to improve local hygiene and sanitation are having an impact.

4.2. Hospitalized neurocysticercosis cases in Bali during 2014–2019

From 2014 to 2018, 24 cases of NCC (average of 4.8 cases/year) were reported in Sanglah Hospital, Denpasar. Twenty-three of these patients reported currently living in non-endemic districts of Bali, with one case from the endemic district of Karangasem (Susilawathi et al., 2020). In 2019, 3 additional NCC cases were treated at Sanglah Hospital. All three of the patients were currently living in non-endemic districts of Bali (Raka Sudewi et al., unpublished).

5. Perspectives

Numerous infrastructure improvements have recently been made in Bali. During the last decade, improved drinking water and sanitary systems were employed in addition to health education targeting

primary school children, including emphasis on washing hands before eating and after defecation. Large water tanks were set up in each banjar to aid in improving hygiene practices. Many of these tanks had sedimentation or filtration systems for rainwater collection and storage. Furthermore, small water tanks were set up in each house to collect rain water. Prior to the incorporation of these water tanks, villagers largely relied on water trucks to provide fresh water to their households (Fig. 4). Along with improved sources of drinking water, continued public health education focusing on personal hygiene and environmental sanitation emphasising primary school-aged children, is needed (Wandra et al., 2015).

Due to socio-cultural and religious ties to consuming undercooked pork, implementing a preventive chemotherapy program may also be warranted in *T. solium* endemic regions of Bali. These programs should ensure that school-aged children are included. However, preventive chemotherapy should be only one facet of a more comprehensive One Health approach focusing on both humans and pigs (PAHO, 2021; OIE, 2021). A program combining human antiparasitic treatment along with treatment with oxfendazole and vaccination of pigs has been proposed as a means to control *T. solium* transmission in endemic areas such as Bali (Lightowlers, 2013; Wandra et al., 2015; WHO, 2021).

6. Conclusion

Based on recent studies and improved living conditions in Karangasem during the last decade, the goal of eradicating *T. solium* from Bali is believed to be attainable. While there is cause for optimism in Bali, the four cases of *T. solium* taeniasis found during 2019–2020 emphasize the need for vigilance to ensure that the parasite is not reintroduced, and any remaining pockets of transmission are destroyed through the implementation of a comprehensive surveillance and control program (Lightowlers, 2010; Wandra et al., 2015; Wulanyani et al., 2019). At this time, it is also not clear how the selling and movement of



Fig. 4. Images showing rainwater collection by a household (a), a public rainwater collection reservoir (b), a water delivery truck during the dry season (c), a water storage tank at a house (d), and a public water storage tank (e).

pigs out of *T. solium* endemic areas after the 2017 eruption of Mt. Agung may have impacted local parasite transmission. Therefore, further well-planned studies are needed to evaluate the impact of implementing control measures in the endemic subdistrict of Kubu. Regardless, improved biosecurity through limiting pigs' access to human feces, improved hygiene practices, and improvement of sanitary facilities overseen by the central and local government are needed to ensure a successful outcome.

7. Funding support

From April 1994 to March 2015, AI received funding from Grant-in-Aid from the Ministry of Education, Japan. From 2019 to 2020, Udayana University received funding from the Ministry of Education and Culture, Indonesia.

This paper is dedicated to Prof. Akira Ito, who was a true pioneer in

the study of *T. solium* cysticercosis.

Declaration of Competing Interest

No conflicts of interests.

Acknowledgments

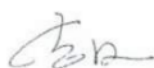
We sincerely thank personnel from the Bali Provincial and Karangasem District Health Offices, Sanglah Hospital, Denpasar, Bali, Karangasem District Livestock Office, local government, and the health centres in Kubu. We would also like to thank lecturers and researchers from Udayana University, who have contributed to this study, and John M. Saragih from the National Institute of Health Research and Development, Indonesian Ministry of Health.

Appendix

All authors have approved the content.



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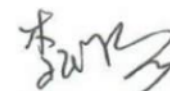
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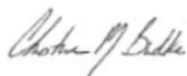
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