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Review
Introduction to the symposium on cestode zoonoses in Asia and the Pacific at the 21st Pacific Science Congress

Diphyllobothriasis and sparganosis in Indonesia

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Abstract: Food-borne zoonotic cestode infections by *Diphyllobothrium* spp. and *Spirometra* spp. are relatively uncommon in Indonesia. So far, only one case of diphyllobothriasis was confirmed in 2004 in Jakarta, whereas there were 4 sparganosis cases in Indonesia. Morphology of eggs and gravid proglottids revealed the first case to be caused by *Diphyllobothrium* species. However, molecular identification of the species was not successful. Sparganosis may not be particularly rare in Indonesia, since *Spirometra* species have often been found in cats and other animals. These topics in Indonesia are briefly overviewed with reference to historical records and socio-cultural background information.
Key words: Diphyllobothriasis, Sparganosis, Indonesia

INTRODUCTION

Diphyllobothriasis is a food- or fish-borne tapeworm infection caused in human by Diphyllobothriid cestodes recognized mostly in temperate and sub-arctic areas. It was first reported from areas of Europe and North America with a high latitude, and later from South America and Asia, mainly Japan and Korea, as well [1-5]. Fish such as salmonids, pike, burbot, perch and ruff have been reported as the most important transmitters of *Diphyllobothrium latum* on the Eurasian continent. Humans are thought to get the infection through consumption of raw or under-cooked fish harboring plerocercoids, the larval stage infective to human. The custom of eating raw or under-cooked fish is now rather common and has become part of the cosmopolitan life style throughout the world, but it may contribute to the increase in the number of human diphyllobothriasis cases, even unexpected cases in tropical countries including Indonesia [3]. The diphyllobothriasis in Indonesia might reflect the "globalization" of food transportation.

There are many *Diphyllobothrium* species difficult to distinguish morphologically. Historically, most of the species infecting humans have been identified as *D. latum*. Besides domestic animals like dogs, cats and pigs, many wild animals have been listed as the definitive host, i.e. jackal, puma, tiger-cat, red fox, arctic fox, polar bear, brown bear, seals, walrus, sea-lion, otter and porpoise [6]. By contrast, sparganosis in humans is thought to be caused through somewhat different routes of infection such as accidental drinking of water contaminated with copepods, the first intermediate host harboring procercoids of *Spirometra* spp. De Hartogh [7] concluded that human infection with spargana (= plerocercoids) was possibly due to ingestion of spargana in under-cooked frog meat. Copepods harboring procercoids are thought to be the main source of infection for the second intermediate host or parathenic host. Patients suffering from sparganosis have been reported in several countries, mainly in Southeast Asia including China and Japan. The sparganum has been reported from frogs, monkeys and pigs in Indonesia [8-10].

It is expected that more human cases of diphyllobothriasis and sparganosis can be detected if clinicians as well as pathologists and parasitologists gain greater awareness of the possibility of infection with *Diphyllobothrium* spp. and *Spirometra* spp. in Indonesia.

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DIPHYLLOBOTHRIASIS

Human diphyllobothriasis had never been reported in Indonesia until the first case was detected in 2004 in Jakarta, the capital city of Indonesia.

The first case of diphyllobothriasis in Indonesia:

The patient was an Indonesian business man, 52 years old, who traveled to Japan, Australia, China, Singapore many times and once to Switzerland, and ate raw or undercooked fish even in Indonesia. He complained regularly of shedding a "ribbon-like" organism for 4 months. This was identified as *Diphyllobothrium* sp. based on the morphology of eggs and gravid proglottids (Fig. 1). The patient suffered from soft stools and mild diarrhea, especially when the very long ribbon-like organism crawled out of his body. He had no abdominal pain. He was at first treated with mebendazole, single dose, 1 tablet of 500 mg, for 2 consecutive days. He had been consuming *sashimi* and *sushi*, Japanese raw or half cooked dish which contained fresh-water as well as marine fish like salmon, tuna, *toro* and *gindara* for two years before 2004. Other complaints, such as feeling hungry, weak, colic pain etc. were absent. Two months afterwards, he removed an organism in length of approximately a half meter from his anus. The whole "ribbon" was thrown away. Three months after the first expulsion of the moving organism, another half meter-long "ribbon" came out without any treatment. During the fourth month, before visiting our laboratory at the University of Indonesia, he had suffered more than 10 bouts of diarrhea over 24 hours accompanied by abdominal discomfort. In the morning before the diarrhea started, he consumed juice containing ginseng leaves and guava fruit with honey. After the rest of the "ribbon" was expelled, his stool was not loose anymore. The organism was morphologically identified as *Diphyllobothrium* sp.

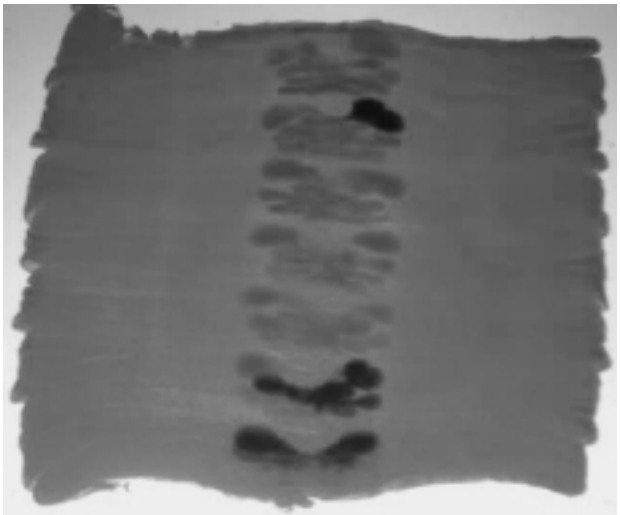

Fig. 1. Gravid proglottids of *Diphyllobothrium* spp. expelled from the first case in Indonesia

When the patient visited the hospital laboratory (Laboratory Mitra Keluarga, Jakarta), he brought a worm-like organism, approximately 30 cm long, already fixed in 10% formalin. Then he visited our laboratory at the University of Indonesia with a specimen approximately 10 cm long. No egg was found from the stool sample. Morphological observation of eggs pushed out from gravid proglottids and gravid proglottids themselves revealed the organism to be *Diphyllobothrium* sp., a broad fish tapeworm. However, we could not obtain DNA from the tapeworm fixed in formalin and failed to identify the species.

SPARGANOSIS

Adult *Diphyllobothrium* and *Spirometra erinacei* were detected from cats and dogs by de Hartogh [7] and Meijer [12] in 1932 and 1937, respectively. Since then there have been several reports on experimental infection with *Diphyllobothrium* spp. in domestic animals, mainly cats [7,8,12].

The first case of human sparganosis was a navy officer settled in Ambon, Mollucs, East Indonesia. The sparganum was found in the urinary bladder by von Römer [13] (quoted by Bonne [11]). A report on an autopsy case at the Institute of Pathology in Batavia (Jakarta) described a very large sparganum in the right pulmonary artery of a Malayan (Indonesian), who died in a mental asylum. Histological examination revealed an extensive hemorrhagic infarction of the right lung. Infarctions were also found in the kidneys, as well as hemorrhage in the cerebral cortex and basal ganglia and slight fibrinous peritonitis in the region of the ascending colon. It was speculated that the sparganum migrated through the body, resulting in multiple lesions. The location in the pulmonary artery was unusual for a sparganum, as was the size of about 40-50 cm [9]. Two spargana were obtained from the small intestine of the second patient, an insane 45 years old Malayan (Indonesian) who died of tuberculosis. Numerous calcareous corpuscles were found inside the body. The structure of the organism was not clear. However, eggs were obtained from the feces of a kitten after 27 days of experimental infection with these two spargana. Two adult tapeworms were recovered from the kitten 42 days after infection and identified as *Diphyllobothrium* sp. The worms were morphologically different from *D. latum*. The authors concluded that the adult worms were *D. erinacei* (Rudolphi 1819) according to Neveu Lemaire who stressed that *D. mansoni* and *D. erinacei* were the same species [10].

In the early 1970's, the first case of ocular sparganosis

Table 1. Seroprevalence of sparganosis in Papua (Manokwari and Nabire Districts), North Sumatra (Samosir island) and Bali (Denpasar and Ginayar Districts), Indonesia, 2005. Serum samples were obtained from the field survey for taeniasis/cysticercosis and other helminthic infections [14-17].

Province	No. of serum sample examined	Seroprevalence (%)
Papua	257	2.7 (7/257)
North Sumatra	105	2.9 (3/105)
Bali	29	6.9 (2/29)
Total	391	3.1 (12/391)

was diagnosed in Purwodadi, Semarang by Lokollo and Wilardjo. The patient, a man of about 30 of age, complained of irritation and a decrease in acuity of the left eye. Examination revealed that the conjunctiva was reddish and the acuity of the left eye somewhat decreased. A moving organism, about 1 cm in size, was detected in the anterior eye chamber. The patient refused invasive treatment but came back at the ophthalmology department a few days later. The organism was breaking into fragments. The diagnosis of sparganosis oculi was determined using a slit lamp (Wilardjo, personal communication).

Serological surveys on sparganosis, toxocariasis and cysticercosis were carried out in the provinces of Papua (Irian Jaya), North Sumatra and Bali [14-17] in 2005 (Wandra *et al*., unpublished data). With regard to sparganosis, crude extracts of *S. erinaceieuropaei* plerocercoids were used for dot-ELISA. Sparganosis was serologically detected in these provinces (Table 1). In Indonesia, sparganosis should be kept in mind in areas where the custom of eating frog meat is common.

Spargana were frequently found in frogs living in rice fields around Jakarta (formerly Batavia). Experimental infection of kittens with spargana obtained from frogs resulted in the observation of adult *D. ranarum* [7]. Spargana were also occasionally detected from monkeys [9] and pigs [12]. Adult *D. ranarum* were recovered from a cat infected with spargana from monkeys, whereas spargana from pigs developed into *Diphyllobothrium* spp. in cats but were diagnosed under different names by different researchers (*D. raillieti* and *D. erinacei*). However, the worms were probably identical to *D. ranarum*. Spargana were often found in toads [11]. The life cycle of *D. ranarum* was studied by Gan [18]. Tadpoles infected with spargana of *D. ranarum* were fed to cats which later harbored adult worms.

DISCUSSION

***Diphyllobothriasis*:** This is an anthropozoonoses and a food- or fish-born parasitic disease, involving many mammals including humans as the definite host [6,19,20]. Mammals such as mongoose, walrus, seal, sea lion, bear, fox, mink and hog have been reported as reservoir hosts. *Diphyllobothrium* spps. have been detected in humans and reported as *D. latum*, *D. houghtoni*, *D. parvum*, *D. cordatum*, *D. minus*, *D. pacificum*, *D. ursi*, *D. dendriticum*, *D. lanceolatum*, *D. yonagoensis*, *D. alascense*, *D. dalliae*, *D. erinacei*, *D. nihonkaiense*, *Diplogonoporus grandis*, *Digramma brauni*, *Ligula intestinalis*, *Pyramicocephalus phocarum* etc. [4,19-23]. However, these are now divided into three genera, *Diphyllobothrium*, *Spirometra* and *Diplogonoporus* [24,25].

Diphyllobothriasis is nowadays found not only in “traditional” areas such as the Scandinavian countries and other Eurasian countries including Russia and China, and Canada and the USA, but also in Japan and Korea in Asia, and in Chili, Peru, Argentina and other South American countries. An excellent review was conducted by von Bonsdorff [26]. However, it was not clear if the source of infection in Japan and Korea, where people tend to eat marine fish, was the same as that in the “traditional countries” where the infection is caused by consumption of raw or under-cooked fresh water fish [2]. Recent cases reported in tropical countries are thought to be due to the consumption of imported fish contaminated with *Diphyllobothrium* sp. However, infection through consumption of local, raw fish infected with unknown local *Diphyllobothrium* sp. is an alternative possibility.

Diphyllobothriasis is common in northern Japan where more than 100 cases a year are reported [27]. There are many reports describing *D. latum* as the cause of diphyllobothriasis in Japan. However, the view that the infection was acquired by eating *sashimi* made from Pacific salmonoids and other marine fish should have alerted investigators to the possibility that the diphyllobothriasis in Japan was not caused by *D. latum* [2]. According to Ando and others [22], *D. nihonkaiense* was the dominant species found in humans throughout Japan. Yamane and others [4] described *D. nihonkaiense* and revised the taxonomy for Japanese *Diphyllobothrium* spp. In Korea, a total of 32 human diphyllobothriasis cases were identified up to 1997 [28]. In Malaysia, two cases have recently been reported [29].

Identification of the species of genus *Diphyllobothrium* seems to cause confusion. Re-evaluation of the species of genus *Diphyllobothrium* should be carried out on the molecular level as well as with regard to morphology, life cycle etc. In the present case from Jakarta, the morphological findings implied neither *D. latum* nor *D. nihonkaiense*. An effort to identify the species through mitochondria DNA examination was not successful due to tech-

nical problems. In this case, it was only confirmed that the worm belonged to the genus *Diphyllobothrium*. Recently, mitochondrial DNA sequences of *D. latum* and *D. nihonkaiense* have been completely determined [30]. As far as we know, however, mitochondrial sequences have not yet been fully elucidated for other Diphyllobothriids. In the future, when investigations on DNA structure of these worms become more advanced, different species can be accurately distinguished.

In this first reported case of human diphyllobothriasis in Indonesia, it was difficult to state definitively where the infection took place or what kind of fish was the source of infection, because the patient consumed raw fish in many countries including Indonesia. It was most likely that the infection resulted from consumption of a Japanese food. However, fish caught in Indonesian seas should also be investigated for the presence of plerocercoids as local risk factors. It is interesting to note that the patient was cured after a bout of heavy diarrhea induced by juice consisting of ginseng leaves and guava fruit with honey.

***Sparganosis*:** Historically, spargana have been considered to be the plerocercoids of genus *Diphyllobothrium* (=*D. ranarum*). However, the question of whether or not it is the same as *Spirometra erinaceieuropaei* should be resolved, since Okamoto and others identified dogs in Sumatra infected with *S. erinaceieuropaei* and stressed that genus *Spirometra* is crucially different from genus *Diphyllobothrium* [24]. In Indonesia, two of the three sparganosis cases were men with mental disorders [10,11]. The presence of spargana was suspected to be associated with unhygienic behavior such as drinking water contaminated with infected cyclops or consuming raw or insufficiently cooked frog meat or other animals infected with spargana. The custom of eating frog and snake meat exists especially among Chinese descendents in Indonesia. However, frog and snake meat are fried or grilled. Therefore, the risk of acquiring infection from contaminated frog and snake meat with spargana is not so high. Occasionally fresh cobra blood is drunk as an aphrodisiac. However, more than 90% of people in Indonesia are Muslims, who are prohibited to consume amphibians and reptiles as well as pork. We can assume, therefore, that sparganosis is obtained mostly by drinking unboiled water contaminated with cyclops infected with procercoids. In a report on 34 cases of sparganosis in Thailand, it was found that 12 patients had a history of drinking impure water. Among these persons, five consumed frog or snake meat and two used frog or snake meat as a poultice [31].

CONCLUSION

The first case of diphyllobothriasis from Indonesia was briefly described. It is also expected that more diphyllobothriasis cases will be reported in Indonesia due to the global trend of consuming raw fish.

Sparganosis cases in Indonesia were briefly overviewed. The parasite might be detected more frequently in Indonesia if serological surveys are introduced for diagnosis and epidemiological investigations.

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