[Jpn. J. Parasitol., Vol. 45, No. 2, 157–161, April, 1996]

Case Report

The First Human Case of Diplogonoporiasis in Okinawa Prefecture, Japan

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(Accepted March 12, 1996)

Abstract

An immature diphyllobothriid cestode with a scolex was expelled by Gastrografin administration from a 29-year-old man residing in Naha, Okinawa Prefecture, Japan. The strobila was identified as *Diplogonoporus grandis* based on the morphological characteristics. The source of infection was estimated to be raw sardine ingested during his stay in the mainland of Japan. This is the first case of diplogonoporiasis in Okinawa Prefecture.

Key words: *Diplogonoporus grandis*; Cestoda; human case; Gastrografin; morphology; Okinawa.

In Japan, *Diplogonoporus grandis* (Blanchard, 1894) is well known as a human parasite, and more than 200 human cases have been recorded (Hatsushika *et al.*, 1994). The geographical distribution of diplogonoporiasis shows an aggregation in the areas west of Kanto District although sporadic cases have been reported from northeastern Japan up to Aomori Prefecture (see Suzuki *et al.*, 1993; Hatsushika *et al.*, 1994). From Okinawa, the westernmost prefecture of Japan, diplogonoporiasis has not been reported. Recently we found a case of *Diplogonoporus* infection in a man of Okinawa as described herein.

The Case

The patient was 29-year-old male, a company employee, residing in Naha, Okinawa Prefecture, Japan. His family and former histories were not remarkable. On September 2, 1995, he had diarrhea after drinking liquor. Next morning, he found that a whitish tape-like material was expelled with feces, and he visited the Izumizaki Hospital for consultation. On admission, he was 175.5 cm in height and 83.7 kg in weight. Blood pressure was 144/94 mmHg. No abnormal sign was noticed on physical examination. The tape-like material was revealed to be a cestode strobila. Under the diagnosis of cestode infection, anthelmintic treatment with Gastrografin® (Meglumine sodium amidotrizoate, Schering AG, Germany) was performed. When 200 ml of Gastrografin was introduced into the duodenal lumen through a tube, roentgenography revealed that a tape-like worm moved rapidly downwards through the intestinal lumen and finally reached at the end of the descending colon. Subsequently, a living cestode with a scolex was expelled.

The patient was fond of sushi and raw fish, and he ingested raw sardines and saurels when he visited the mainland of Japan at 2 or 3 months interval. About 1 month preceding to the worm expulsion, he ate raw sardine in Tokyo. He also visited Thailand, Philippines, Mongolia and Hawaii, where he in-

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gested raw salmon, masou salmon and squids. He was born in Nagoya, and then lived in Tokyo, Hakodate, Iwate, Osaka, Fukuoka and Kagoshima Prefectures before he moved to Okinawa.

The Worm

The strobilae, both passed spontaneously and expelled by Gastrografin administration, were fixed in 10% formalin solution without relaxation or pressure. Then, the strobilae were submitted to the Department of Parasitology, Faculty of Medicine, University of the Ryukyus, for identification. The worm was cleared in a glycerol aqueous solution of ascending concentration, and observed and measured under a stereomicroscope. In order to make detailed observation of internal structures, proglottids near posterior end of the strobila expelled by Gastrografin administration were embedded in paraffin, sectioned serially at thickness of 8 μm, and stained by Gomori's one-step trichrome method.

The worm is white, flat, and irregularly shrunken (Fig. 1). The number of proglottids is not countable. The strobila submitted by the patient is 72 cm long and 2.9 mm wide, and lacks a scolex. The strobila expelled by Gastrografin is 165 cm long and 5.7 mm wide at thickest portion, and has a scolex (Fig. 1). The scolex is approximately oblong with overlapped bothrial rims, and 1.06 mm long by 0.68 mm wide (Fig. 2). The neck is 1.71 mm long and 0.46 mm wide near anterior end. The proglottids at about 5 cm posterior to the scolex is relatively relaxed and show secondary segmentation (Fig. 3). The worm width increases posteriorly and the posterior end is round. The proglottids are much wider than long usually: the width is more than 15 times as long as the length in the posterior portion of the strobila (Figs. 4, 5). The genital pores, uterine pores and eggs are not observed in any proglottids (Fig. 4). In cross section, the longitudinal musculature is thick, forming a laterally-elongated circle, and the transverse musculature forms two thin bands inside of the circle (Fig. 5). The genital primordia are double and situated midlaterally in the medulla (Fig. 5). In the horizontal section, the genital primordia forms two midlateral longitudinal rows of which outer sides the collecting tubes of excretory system and the nerve trunks are running (Figs. 6 and 7). The genital primordium is an elliptical mass of many nuclei stained densely with hematoxylin, and is slightly enlarged anteriorly and posteriorly (Fig. 7). The ovary, testes and cirrus sac are not discernible (Figs. 5–8). The two genital primordia in a proglottid are separated by distance of about 30% of the proglottid width, and no longitudinal muscle bundle is observed between them (Figs. 5, 6). Osmoregulatory canals are distributed sparsely cortically.

Discussion

Because the present worm is strongly contracted and immature, it is difficult to identify the species. However, the presence of double sets of genital primordia in each proglottid and the secondary segmentation of the proglottids indicate that this worm belongs to the genus *Diplogonoporus* Lönnberg, 1892 (Schmidt, 1986; Bray *et al.*, 1994). Schmidt (1986) described that *Diplogonoporus* lacks a neck, and Rausch (1964) also mentioned the absence of neck in *D. balaenopterae* Lönnberg, 1892 and *D. tetrapterus* (von Siebold, 1848). Nevertheless, Delyamure (1955) recorded the length of the neck in *D. balaenopterae*. Also, in the descriptions of *D. grandis* and *D. fukuokaensis* Kamo and Miyazaki, 1970 collected in Japan, the length of the neck has been recorded (Kamo and Miyazaki, 1970). It is therefore considered that a *Diplogonoporus* cestode has a distinct neck.

Schmidt (1986) listed seven species in the genus *Diplogonoporus*, and 3 of them; i.e., *D. grandis*, *D. brauni* Leon, 1907 and *D. fukuokaensis*, have been recorded from humans. Besides these species, *Diplogonoporus* sp. was recorded from a human in Kagoshima Prefecture, Japan (Kamo and Miyazaki, 1971). The present species differs from *D. brauni*, a much smaller cestode (12 cm in total length) that has been recorded only from Rumania and suggested to belong to the genus *Digramma* Cholodkovsky, 1915 (Joyeux and Baer, 1929). *Diplogonoporus fukuokaensis* has a longitudinal muscle bundle running between the genital organ rows, and *Diplogonoporus* sp. of Kamo and Miyazaki, 1971 has the distance between the genital organ rows of only less than 20% of the proglottid width, and thus both differ from the present worm (Kamo and Miyazaki, 1970, 1971).

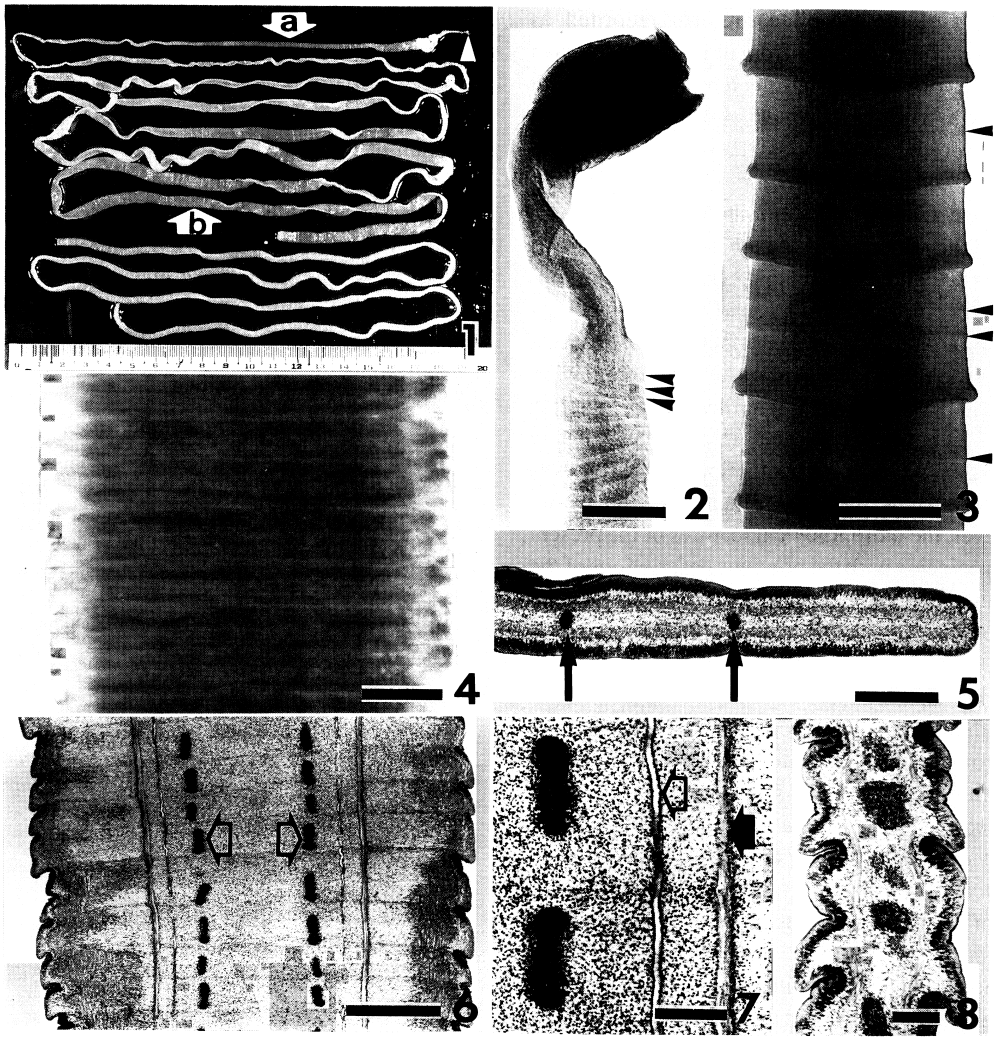

Fig. 1 Strobila with scolex (arrowhead) expelled by Gastrografin (above) and strobila discharged spontaneously prior to the treatment (below). Arrows a and b indicate the portions of which enlarged views are presented in Figs. 3 and 4, respectively.

Fig. 2 Anterior portion of the strobila showing the scolex with bothria and the neck. Arrowheads indicate proglottid formation. (Scale=0.5 mm)

Fig. 3 Enlarged view of the portion a in Fig. 1 showing secondary segmentation in proglottids (arrowheads). (Scale=1 mm)

Fig. 4 Enlarged view of the portion b in Fig. 1 showing laterally-elongated proglottids. (Scale=1 mm)

Figs. 5–8 Histological sections of proglottids of portion b presented in Fig. 1. 5, Cross section showing two genital primordia (arrows). (Scale=0.2 mm); 6, Horizontal section showing two longitudinal rows of genital primordia (open arrows). (Scale=1 mm); 7, Enlarged view of Fig. 6 showing genital primordia, protonephridial collecting canal (open arrow) and nerve trunk (closed arrow). (Scale=0.2 mm); 8, Parasagittal section through the longitudinal row of genital primordia. (Scale=0.2 mm)

Some of the strobilae hitherto recorded as *Diplogonoporus grandis* from humans were gravid attaining to more than 10 m in length and 25 mm in width, whereas some were immature having a length less than 1 m and a width less than 5 mm (see Suzuki *et al.*, 1985; Suzuki and Imamura, 1991). Moreover, there are considerable morphological variations among the strobilae. There are still disagreements on the taxonomic status of *D. grandis*. Some researchers consider that *D. grandis* is a junior synonym of *D. balaenopterae* Lonnberg, 1892 (Rausch, 1964; Iwata, 1967; Delyamure and Skrjabin, 1986), whereas the others suspected that the so-called *D. grandis* is not a single species (Kamo, 1969; Kamo *et al.*, 1971). Under such situation, it is difficult to determine the species of the present immature worm. However, the morphological features of the present worm coincide with the previous descriptions of the immature strobilae recorded as *D. grandis* (see Kamo *et al.*, 1968; Kifune *et al.*, 1981; Kumazawa, 1981; Kanazawa *et al.*, 1986). Thus, we identified it as *D. grandis*.

Although the plerocercoid of *D. grandis* has not been found and the life history of this cestode has not been fully elucidated, its second intermediate host has been surmised to be sardines (Kamo, 1969; Kamo *et al.*, 1971). Because the present patient ingested raw sardines during his stay in the mainland of Japan, he might acquire the infection from the fish.

The Okinawan inhabitants lacked the habit of eating raw fish in the past, and hence number of cases with diphyllobothriid infection was quite few. Before 1980, only three cases have been recorded from Okinawa and Miyako Islands. The two cases on Okinawa Island were due to *Diphyllobothrium pacificum* (Nybelin, 1931) and *Diphyllobothrium yonagoense* Yamane *et al.* 1981 (Kamo *et al.*, 1982, Hasegawa, unpublished data). The patients of these two cases have been estimated to have acquired the infections by ingesting some inshore fishes of Okinawa. After 1984, human infection with *Diphyllobothrium nihonkaiense* Yamane *et al.*, 1986 became to occur in succession. This outbreak was apparently due to (1) the start of transportation of fresh fishes including masou salmon, the intermediate host of *D. nihonkaiense*, from the mainland of Japan to Okinawa, (2) the changes in food habits of the Okinawan inhabitants and (3) the increased opportunity to travel to the mainland of Japan by the Okinawan people (Hasegawa *et al.*, 1994). Raw sardines have not been ingested in Okinawa Prefecture until recently. However, in the 1990s, "sashimi" sardines transported from the mainland of Japan became to be served in Japanese food restaurants, and sardines for raw eating are sold in the large markets nowadays. Thus, it may be predicted that infection with *Diplogonoporus grandis* acquired from fishes sold in Okinawa Prefecture will occur in near future.

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