

Case Report

First Two Cases of Diplogonoporiasis Grandis from Nagano Prefecture, Japan

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Abstract

Two cases of diplogonoporiasis grandis were noted for the first time from Nagano Prefecture in the Central Honshu, Japan. These cases indicate it necessary to make a definite differential diagnosis for each pseudophyllidean tapeworm infection, based on the understanding that diplogonoporid infection occurs in inland inhabitants as well as in coastal dwellers.

Key words: diplogonoporiasis grandis, geographical distribution, inland case

Human cases of pseudophyllidean tapeworm infections have been increasing in these two decades in Nagano Prefecture, remote from the sea coasts (Oshima, 1976). In the majority of these cases, *Diphyllobothrium nihonkaiense* Yamane *et al.*, 1986 has been incriminated as the causative tapeworm. Further, we confirmed the two cases of *Diplogonoporus grandis* infection in the inhabitants of this inland and mountainous prefecture, one in 1984 and the other in 1991. Diplogonoporiasis caused by eating some marine fishes raw has long been observed to occur in the coastal regions of Kyushu,

Sikoku, and Honshu east to Chiba Prefecture along the Pacific coast and north to Kyoto Prefecture along the Japan Sea coast (Kagei, 1989; Kamo *et al.*, 1968; Kamo, 1969; Kamo *et al.*, 1971; Suzuki, 1989). Its range has recently been extending north or west beyond the above regions as shown by a few cases that have been encountered mainly in Kanto District (Akao, 1990; Uranaka *et al.*, 1990; Yamamoto *et al.*, 1992) and even in Aomori Prefecture (Tsuge *et al.*, 1991). Although diplogonoporiasis among inland inhabitants have been seen in Tochigi (Uranaka *et al.*, 1990) or Saitama Prefecture (Yamamoto *et al.*, 1992), we report here our cases as the first and second ones from Nagano Prefecture.

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Case Report

Case 1: The patient was a 57-year-old male, local public service worker, living in Matsumoto City. He suddenly had abdominal pain with diarrhea on April 15, 1984, and observed a worm of some 1 m long having been evacuated from him. On April 19, he discharged a piece of worm with diarrhea again. His home doctor prescribed an antidiarrheal drug and, under a diagnosis of cestoidiasis, immediately introduced him to the Department of Parasitology, Shinshu University School of Medicine, for treatment. Paromomycin® (Aminosidine), 50 mg/kg, followed

by saline purgation was given to him on April 20, although neither parasites nor parasite eggs had been seen in his stool. Since no worm was expelled by this medication, the parasite was thought to have been naturally evacuated during diarrhea.

Dietary history and life style of the patient were not well known, but marine fishes were among favorite foods. He also seemed to have official trips frequently to many places outside the prefecture.

Delineation of worm: A small part of the strobila was stained with alum carmine and mounted in Canada balsam. Paraffin serial sections (10 μ m thick) of some proglottids were stained with Delafield's hematoxylin and eosin.

Scolex lacking. Strobila flat, thin, snow white in color, and about 120 cm long by evenly 15 mm wide, with longitudinal striae on both dorsum and venter; no mature proglottids included. Tegument disappeared (Fig. 1C); proglottids much wider than long; regular and irregular segmentations and multiplication of each one into two discernible (Figs. 1A, B). Basically two genital anlagen in each proglottid which were apart from each other a little more than one-third of proglottid breadth; an additional genital anlage in many proglottids interiorly closer to either one of the paired ones (Fig. 1B). Cortical outer cellular zone, longitudinal and circular muscle fibers, genital anlagen, nerve trunks, and excretory canals considerably degenerated (Fig. 1C).

As seen above, only the paired genital anlagen in a proglottid were indicative of the specific character for a pseudophyllidean tapeworm. Dr. Hajime Kamo, the then professor of the Department of Medical Zoology, Tottori University School of Medicine, kindly examined our specimen and suggested that the worm was similar to the so-called *Diplogonoporus grandis* (Blanchard, 1894) Lühe, 1899 (in lit., Sept. 3, 1984).

Case 2: The patient was a 31-year-old male, brewer, dwelling in Nakano City. He noticed the onset of loose stools on the night of July 11, 1991. On the midnight of the following day, he developed abdominal pain and diarrhea occurring 6 to 7 times a day. On the evening of July 14, his diarrhea became serious with drinking beer. He noticed something tape-like hanging through the anus and pulled it out until he thought it better to cut it by

himself. On July 15, he was diagnosed as having diphyllbothiriasis based on his complains and a positive stool examination at Iiyama Red Cross Hospital. He was hospitalized instantly for a Gastrografin[®] treatment. On the day of admission, his physical and laboratory examinations revealed no abnormalities, except for a stool examination positive for the parasite eggs. He also denied the history of nausea, vomiting, hematochezia or fever.

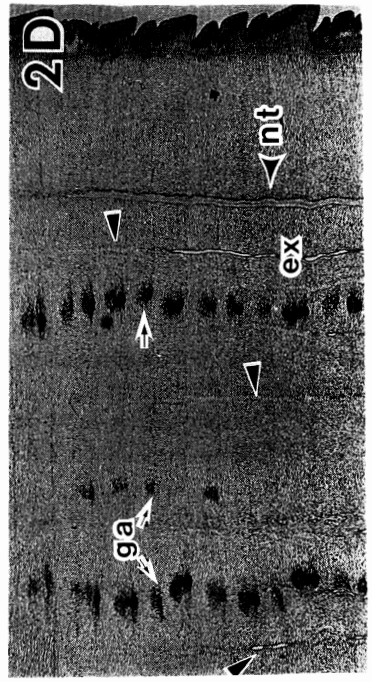
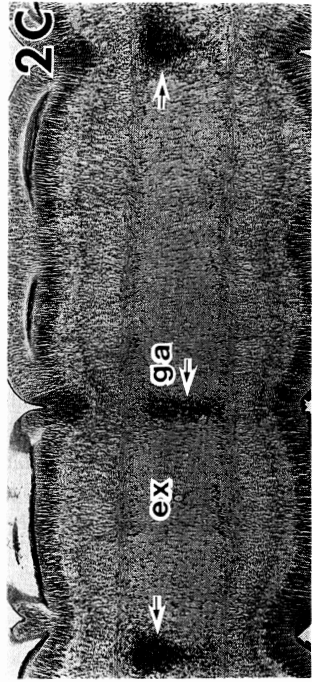
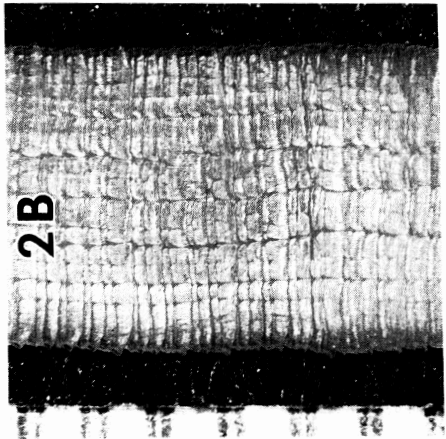
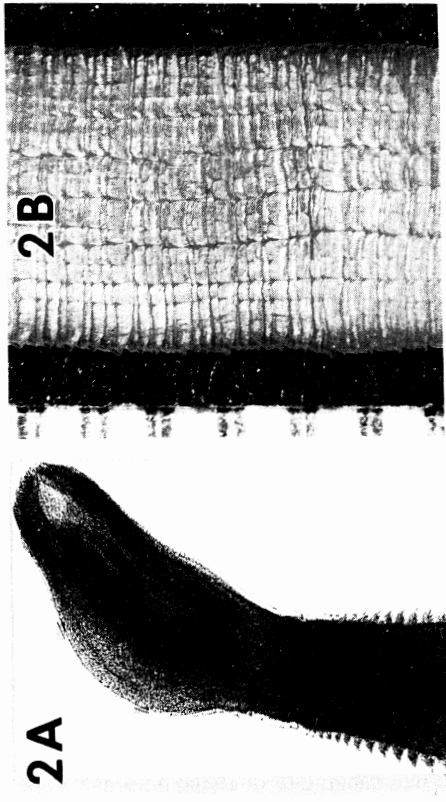
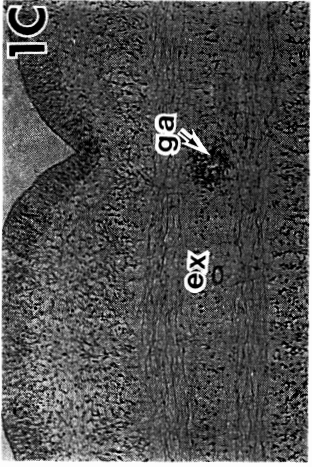
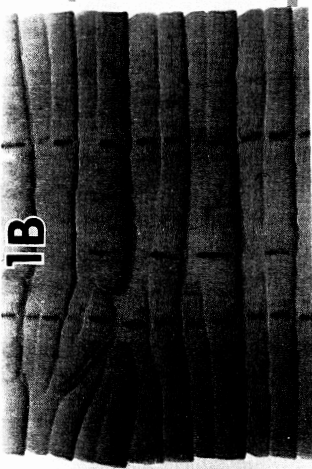
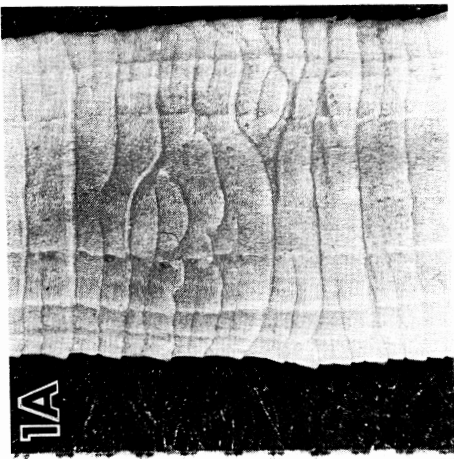
On July 16, he was given 100 ml of Gastrografin[®] rapidly through a duodenal tube. The worm was not detectable at fluoroscopy, but was expelled successfully with its scolex half an hour later.

He had habitually eaten raw marine fishes, including masu salmon sushi in Kanazawa City in early April, 1991, and raw sardine viscera twice, once in Niigata City on May 13 and again at his own home in the middle of June, 1991.

Delineation of worm: The preparation of microscopic specimens as in Case 1. Strobila creamy in color, thick, closely segmented, and about 180 cm long by 5 mm in maximum width, with finely serrate lateral margins and remarkable striae on either side. Scolex broad leaf-like in form, and 1.32 mm long by 0.70 mm wide on the mounted specimen, bothria extending nearly whole length of scolex and enlarged anteriorly, foliate structures overlapping over each bothrium; and neck not well defined (Fig. 2A). Immature proglottids much wider than long; multiplication of each proglottid frequently recognizable (Fig. 2B); and no mature proglottid available. Cortical cellular zone, longitudinal and circular muscles well developed; two or three genital anlagen in each proglottid (Figs. 2C and D).

Feature of the scolex, remarkable multiplication of proglottids, paired genital anlagen apart from each other by a distance of one-third width of proglottid, an additional genital anlage in some proglottids, and well developed longitudinal muscles were similar to those of *D. grandis* redescribed by Kamo *et al.* (1968, 1971).

Remarks. Kamo *et al.* (1968) were of the opinion that diplogonoporids infecting man in Japan should better be treated as *D. grandis* for avoiding confusion in systematics of this group of tapeworms. No mature proglottids were available for our study so that it was difficult to identify the two specimens



exactly. These specimens seemed to have no specific characters that discriminate them from *D. grandis*. Thus, we thought that both specimens in Cases 1 and 2 were those of the so-called *D. grandis* despite their great differences in color, thickness and breadth of the strobila.

The present two cases are significant in that both occurred in the inhabitants of the inland regions. However, it is difficult to know where and how the patients did acquire the infection. They might have this chance either at coastal regions during their trips or at their own living areas from marine fishes transported there in suitable conditions for fishes themselves and, therefore, for their parasites. In the case of an inland patient of Fushimi, Kyoto, a raw sardine that had been brought privately by a fisherman from Fukui on the Japan Sea coast to Fushimi was suspected as the source of infection of *D. grandis* (Nagahana *et al.*, 1967). In addition to diphylobothriid tapeworm infections as mentioned above, anisakid nematode infections have been remarkably increasing in Nagano Prefecture in these two decades (Omachi *et al.*, 1985; Omachi and Omachi, 1992). Subsequent to these parasites, it can not be denied that diplogonoporida tapeworms are infectious to inland inhabitants at their own living areas provided with an excellent transportation system for marine fishes as foods. Consequently, we should pay much attention to diplogonoporiasis in a wider range of Japan, inclusive of both coastal and inland regions. It is also necessary for us to think of diplogonoporiasis that has been possibly lumped under diphylobothriasis in some regions where the latter is far more prevalent than the former.

It is rather easy to discriminate diplogonoporida tapeworms from diphylobothriid ones when mature proglottids are available. However, only immature proglottids of pseudophyllidean tapeworms are sometimes provided for physicians in identifying species as in our two cases. In such a case, the understanding that diplogonoporida tapeworms may be found in inhabitants of a wider range is required for stepping up to a differential diagnosis.

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Fig. 1 Tapeworm from Case 1. A: Strobila (scale = 1 mm); B: Stained proglottids; C: Transverse section of proglottid.

Fig. 2 Tapeworm from Case 2. A: Scolex; B: Strobila (scale = 1 mm); C: Transverse section of proglottid; D: Frontal section of proglottids. Abbreviations: ga-genital anlage, ex-excretory canal, nt-nerve trunk.