

## *Gongylonema* infection in man: A first case of gongylonemosis in Japan

K. HARUKI, H. FURUYA<sup>1</sup>, S. SAITO<sup>1</sup>, S. KAMIYA<sup>2</sup>, N. KAGEI<sup>3</sup>

Department of Hygiene, National Defence Medical College, 3-2 Namiki, Tokorozawa, Saitama, 359-8513, Japan; <sup>1</sup><sup>3</sup>rd Department of Internal Medicine, Kyorin University School of Medicine; <sup>2</sup>Department of Infectious Diseases, 6-20-2 Shinkawa, Mitaka, Tokyo 181-8611, Japan; <sup>3</sup>Department of Parasitology, National Institute of Infectious Diseases, 1-23-1 Toyama, Shinjuku Tokyo 186-0052, Japan, E-mail: [malaria@cc.ndmc.ac.jp](mailto:malaria@cc.ndmc.ac.jp)

### Summary

*Gongylonema* spp. is a nematoda parasite belonging to the subfamily Spiruroidea. This thread like worm usually affects localising in their oesophagus and stomach. Intermediate hosts are thought to be beetles and cockroaches. Infection is established when the animals ingest the insect carrying third-stage larvae in their muscles or haemoceles. Usually, the insect is ingested accidentally with grass or in contaminated water. Larvae penetrate the intestinal wall and migrate toward the oesophagus or stomach where they mature and produce eggs. Human gongylonemosis is a rare infection, only 52 cases world wide have been reported since the first report by Leidy in 1850 (Belding, 1965; Jelinck & Loescher, 1994).

We report the first case of human gongylonemosis in Japan and reviewed 48 cases by reference.

Key words: *Gongylonema* spp.; Spiruroidea; human gongylemiosis; Japan

### Case report

A man aged 34 came to see a doctor complaining of irritable stomatitis on his left lower lip in the August 1996. He was treated with local ointment having been diagnosed with simple stomatitis. His stomatitis subsided spontaneously, but recurrent stomatitis occurred several times until December 1996. He had no other symptoms and had no history of travelling abroad. He was born in the Tokyo metropolitan area and had grown up in the same place. He had no history of eating insects or unusual foods. When he visited the same doctor in December, a thread-like organism was found protruding from his ulcer, localised on his lower lip. The organism was extracted and sent with the patient to the 3<sup>rd</sup> Department of Internal Medicine, Kyorin University School of Medicine. Chemical and haematological examinations were performed and the results showed no

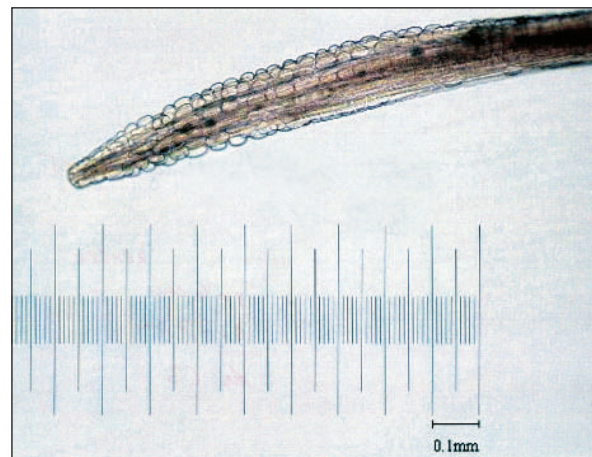


Fig. 1. The anterior part of *Gongylonema pulchrum*: prominent oval cuticular thickenings. x 200

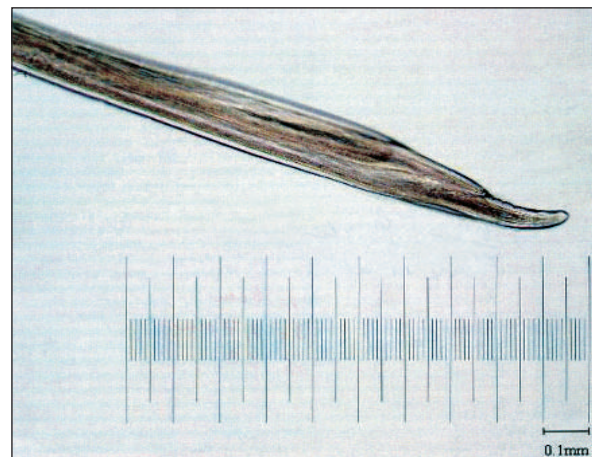


Fig. 2. The posterior end of *Gongylonema pulchrum*: conical, obtuse tail. x 200

Table 1. Reported forty eight cases of *Gongylonema* infection in man

No	Year	Country	Species	Number and Sex of Parasite	Localization	Patient M/F & Age of Patient	Reference
1	1850	USA	<i>G. pulchrum</i>	3 ?	Oral cavity	Infant	Gefter & Nemirovskaya, 1964
2	1864	Italy	<i>G. pulchrum</i>	? ?	Upper lip	M Student	Gefter & Nemirovskaya, 1964
3	1914	Italy	<i>G. sp.</i>	6 ?	Oral cavity	F 17	Gefter & Nemirovskaya, 1964
4	1916	USA	<i>G. pulchrum</i>	1 F	Lower lip	F 16	Ward, 1916
5	1920	USA	<i>G. pulchrum</i>	? ?	Lower lip, oral cavity	F 13	Stiles, 1920
6	1921	USA	<i>G. pulchrum</i>	? ?	Tonsil	F 50	Stiles, 1921
7	1923	USA	<i>G. pulchrum</i>	1 F	Oral cavity	M ?	Ransom, 1923
8	1928	USA	<i>G. pulchrum</i>	1 F	Left lip, sublingual	F 18	Stiles, 1928
9	1934	Ukraine	<i>G. pulchrum</i>	? ?	Oral cavity	F ?	Gefter & Nemirovskaya, 1964
10	1935	USA	<i>G. pulchrum</i>	1 M	Maxilla	M 30	Waite & Gorrie, 1935
11	1936	New Zealand*	<i>G. pulchrum</i>	1 F	Lower lip	M Young	Johnston, 1936
12	1941	Bulgaria	<i>G. pulchrum</i>	4 (1M)	?	F Young	Gefter & Nemirovskaya, 1964
13	1941	Bulgaria	<i>G. pulchrum</i>	1 ?	?	F Young	Gefter & Nemirovskaya, 1964
14	1941	Bulgaria	<i>G. pulchrum</i>	1 ?	?	M Young	Gefter & Nemirovskaya, 1964
15	1950	Sri Lanka	<i>G. pulchrum</i>	1 F	Left buccal mucosa	M 32	Crusz, 1950
16	1951	Morocco	<i>G. pulchrum</i>	2 F	Buccal mucosa	F 24	Gaud & Chabaud, 1951
17	1952	USA*	<i>G. pulchrum</i>	1 F	Lower lip	M 20	Thomas, 1952
18	1952	Morocco	<i>G. pulchrum</i>	1 M	Lower lip	F 21	Gefter & Nemirovskaya, 1964
19	1953	Georgia	<i>G. pulchrum</i>	6 (1M2F)	Lower lip	M 22	Gefter & Nemirovskaya, 1964
20	1953	USA	<i>G. pulchrum</i>	1 F	Sublingual	F 26	Young & Hayne, 1953
21	1955	Italy	<i>G. pulchrum</i>	1 ?	Upper lip	F 10	Gefter & Nemirovskaya, 1964
22	1955	China	<i>G. pulchrum</i>	1 M	Sublingual	F Young	Gefter & Nemirovskaya, 1964
23	1955	China	<i>G. pulchrum</i>	2 ?	Sublingual, oesophagus	F Young	Gefter & Nemirovskaya, 1964
24	1956	USA	<i>G. pulchrum</i>	1 F	Right buccal mucosa	F ?	Burrill <i>et al.</i> , 1957
25	1958	China	<i>G. pulchrum</i>	3 (2F1M)	Buccal mucosa, tongue	M 28	Ch'en <i>et al.</i> , 1958
26	1960	former USSR	<i>G. pulchrum</i>	6 F	Oral cavity	F 21	Bulycheva & Augustina, 1962
27	1961	Bulgaria	<i>G. pulchrum</i>	2 (1M)	Lower lip	F 30	Gefter & Nemirovskaya, 1964
28	1962	former USSR	<i>G. pulchrum</i>	2 (1M)	Lower lip	F ?	Gefter & Nemirovskaya, 1964
29	1963	USA	<i>G. pulchrum</i>	1 M	Lower lip	M 31	Dismuke & Routh, 1963
30	1964	former USSR	<i>G. pulchrum</i>	? ?	Sub lingua	? 23	Engelstein & Kiegel, 1965
31	1964	former USSR	<i>G. pulchrum</i>	1 M	Gingiva	F 25	Gefter & Nemirovskaya, 1964
32	1966	Moldavia	<i>G. pulchrum</i>	? ?	Oral cavity, tongue	M 26	Grinberg, 1968
33	1967	Uzbekistan	<i>G. pulchrum</i>	? ?	?	? ?	Zhdanova, 1967
34	1969	former * Czechoslovakia	<i>G. pulchrum</i>	4 (1M3F)	Submandibulla, tongue	M ?	Ryšavý <i>et al.</i> , 1969
35	1973	former USSR	<i>G. sp.</i>	? ?	?	? ?	Maionova, 1975
36	1972	Hungary	<i>G. pulchrum</i>	2 (1F)	Upper lip	F 20	Amasza <i>et al.</i> , 1972
37	1973	Germany*	<i>G. pulchrum</i>	1 F	Maxilla, gingiva	F 22	Weber & Mache, 1973
38	1976	Moldavia	<i>G. pulchrum</i>	? ?	?	? ?	Shumilo & Spasskii, 1976
39	1983	China	<i>G. sp.</i>	1 M	Right tongue	F 26	Wen <i>et al.</i> , 1983
40	1983	China	<i>G. sp.</i>	1 ?	Lower lip	F 38	Zhang <i>et al.</i> , 1983
41	1983	China	<i>G. sp.</i>	1 ?	Lower lip	M 37	Zhang <i>et al.</i> , 1983
42	1988	Spain	<i>G. sp.</i>	1 F	Sublingua	F 31	Illescas-Gomez <i>et al.</i> , 1988
43	1991	former USSR	<i>G. pulchrum</i>	? ?	?	? ?	Maksimova <i>et al.</i> , 1991
44	1994	Germany*	<i>G. pulchrum</i>	1 F	Left buccal mucosa	F 43	Jelinek & Loescher, 1994
45	1996	Japan	<i>G. pulchrum</i>	1 F	Left lower lip	M 34	This report
46	1999	USA*	<i>G. pulchrum</i>	2 F	Lower lip, gum	F 41	Ebenhard & Busillo, 1999
47	2001	USA*	<i>G. pulchrum</i>	F	Buccal mucosa, lower lip	F 38	Wilson, <i>et al.</i> , 2001
48	2001	Thailand*	<i>G. sp.</i>	1 F	?	M middle age	Wilde, <i>et al.</i> , 2001

\* – Travel related cases

particular abnormality such as eosinophilia inclusive. The stomatitis was itself cured completely without any scar or sequels.

The organism was transferred to the department for identification. It was a nematoda of thread-like form and coiled while floating in saline. The length of worm was 46.12 mm with a maximum thickness of 0.19 mm. The two (dorsal and ventral) lips were fully extruded. The anterior end has a number of round or oval cuticular thickenings or plaques irregularly arranged in longitudinal rows which extend to a distance of 1.22 mm from the anterior end; cuticular cross striations are present at intervals of 3.9  $\mu$ m. The pharynx is short (0.41 mm), narrow, cylindrical and thick walled. The oesophagus is long (6.42 mm) and consists of two parts. The anterior part is short and slender, while the posterior part is long and thick (Fig. 1). The posterior end is bluntly rounded; there is a conical, obtuse tail. The tail measures 0.22 mm. The vulva is situated ventrally at a distance of 2.34 mm from the tip of the tail (94.9 % from anterior end). The vagina leads into the vulva from an anterior direction; the opposed uterine branches join with the vagina nearer the middle of the body (Fig. 2). The eggs are not fully developed, being devoid of shells. The state of development of the eggs and the relatively small size of the worm indicates that the specimen is an immature female of *Gongylonema pulchrum*, Molin 1857 (Baylis, 1925 and Kudo *et al.*, 1992).

## Review

A total of 52 cases have been described from all over the world since Leidy's first report in late nineteenth century (Belding, 1965; Jelinek & Loescher, 1994). We analysed a total of 48 reported cases of human *Gongylonema* infection by references, including our case. The other countries where incidences of gongylonemosis have been reported are USA, former USSR, China, Bulgaria, Italy, New Zealand, Sri Lanka, Morocco, former Czechoslovakia, Hungary, Germany, Spain, Thailand and present case Japan. The distributions of the countries are worldwide. Many of the reported cases involve immature female worms from the morphological point of view. Species and genders of the parasite are shown in Table 1. The main genders of the hosts are young females and the affected part of the oral apparatus is the lower lip. The oesophagus is not a major route of infection in man, although parasites prefer to involve upper gastrointestinal tracts in definitive hosts. Only two cases of oesophageal involvement have been reported from New Zealand Johnston (1936).

The transmission of the parasite mainly occurs by accidental ingestion of an intermediate host or 3<sup>rd</sup> larva of the worm. The intermediate host are recognised as some species of beetles, such as *Aphodius rectus* (infection density 0.523 larvae / examined *A. rectus*), *A. sordidus* (0.224) and *Copris ochus* (3.788) in Aomori prefecture northern Japan (Kudo *et al.*, 1996). Some case reports mentioned that transmission possibly occurred by drinking water in ende-

mic areas. A German patient may have contracted it by drinking water from a well in rural area of Hungary (Jelinek & Loescher, 1994) and one American infected by also drinking water in Mexico (Thomas, 1952). There are at least eight cases related travel and immigration in references. The route of transmission in our case is still unknown. The patient had no history of travelling abroad and had stayed in the metropolitan area a long time. He never drank water from well. There was no history of insect contact. One suggestion was given in a paper and we can only speculate what happened to him. The larva of *Gongylonema* emerged from the intermediate host when the insect dropped into water, such as a well. The third stage of larva can survive for over 30 days in (Kudo *et al.*, 1996). If the patient drank the water contaminated with the larva, infection might have occurred. But in fact, there is no evidence of any beetles living in Tokyo metropolitan, having larva of *Gongylonema*. In addition there are very few domestic animals around the city. The life cycle may not be able to be maintained as a natural infection. Therefore, infection may have occurred, when the patient ate something that had been washed with contaminated water in an endemic area with living larva.

Because of the development of transportation, our daily consumption of food is now not country wide but worldwide. We transport food from all over the country, possibly from endemic areas and also import from endemic areas abroad. This parasitic disease is not definitely localised in endemic areas but is a globalised disease. At least eight cases out of 48 cases are related travel in reference. *Gongylonema* itself is not a dangerous parasite from a clinical point of view, in spite of some cases having reported neurosis due to irritable, recurrent, chronic stomatitis and ulcer (Burrill *et al.*, 1957) and it is often misdiagnosed as a simple ulcer. Parasite infection might be also persistent several years ulcer (Burrill *et al.*, 1957; Ch'en *et al.*, 1958). The differential diagnosis of chronic and recurrent oral ulcer could be Bechet disease, some kind of collagen diseases and cancer. Presumably, a physician or a dentist does not to consider the parasite as a differential diagnosis on a patient with chronic and recurrent oral ulcer in daily consultation, because of that it is a very rarely diagnosed and there are few descriptions of it in medical text books. Therefore, it is feasible to be expected that some cases may not be diagnosed as gongylonemosis in the past or present. When the clinicians have an opportunity to see patients with recurrent, chronic stomatitis and/or ulcer, they should consider that gongylonemosis is one of the possibilities in differential diagnosis. Especially returning the countries where gongylonemosis have been reported.

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