



# Gongylonema pulchrum infection in the human oral cavity: A case report and literature review

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*Gongylonema* infection is a zoonotic disease occurring throughout the world and is mainly caused by consumption of contaminated water and raw food. Adult *Gongylonema* worms can exist as parasites in the human body for up to 10 years and cause symptoms of local irritation in the oral cavity, esophagus, and pharynx. Herein, we report a rare case in which live *Gongylonema pulchrum* was detected and extracted from the oral cavity of a woman. The pathogen was confirmed as *G. pulchrum* on the basis of microscopic examination and morphologic analysis. The patient's symptoms resolved immediately after surgical removal of the parasite, and the patient has been advised not to drink water that has not been boiled and to avoid consuming unwashed raw vegetables. (Oral Surg Oral Med Oral Pathol Oral Radiol 2018;125:e49–e53)

*Gongylonema pulchrum*, which has been detected in the upper digestive tract of mammals worldwide, is a widespread spiruroid nematode responsible for gongylonemiasis.<sup>1</sup> *G. pulchrum* can infect a wide range of domestic and wild animal hosts and is commonly found in ruminants but also in rodents,<sup>2-4</sup> bears,<sup>5-7</sup> nonhuman primates,<sup>8</sup> and pigs.<sup>9</sup>

Gongylonemiasis in humans has been reported in many countries. Although in most of the reported cases the source of infection is unknown, insects such as cockroaches and beetles are considered to act as intermediate hosts. In most identified cases of human infection with *G. pulchrum*, clinical signs include local irritation of the oral mucosa as well as occasional blood expectoration, tarry stools, numbness of the tongue, pain in the chest and abdomen, vomiting, bloating, pharyngitis, and stomatitis.<sup>10</sup> Diagnosis is difficult, and there are reports of gongylonemiasis being misdiagnosed as oral candidiasis,<sup>11</sup> burning mouth syndrome, and even psychosis.<sup>12</sup>

Clinical diagnosis of gongylonemiasis is challenging because of the rarity of the infection, the transient nature

of the symptoms, and the uncertainty of the location of the worms within the oral cavity. The chief symptom of the patient described in this report was an intermittent itchy sensation in the oral mucosa. The patient was examined thoroughly, and live *G. pulchrum* was detected in, and extracted from, the buccal mucosa. The pathogen was confirmed as *G. pulchrum* based on morphologic analysis.

## CASE REPORT

### History review

A 30-year-old woman presented to our clinic chiefly for an itchy sensation that was sometimes accompanied by a feeling that a worm was crawling over the oral mucosa of her upper and lower lip, gingiva, and buccal mucosa. The patient had been experiencing these symptoms for 2 years and reported that they usually disappeared spontaneously within a few hours without any associated systemic symptoms. On first experiencing the symptoms, the patient had been seen by medical professionals from several departments; however, a diagnosis could not be confirmed and therefore no treatment was initiated.

The patient denied experiencing weight loss, rash, fever, chills, or shortness of breath. She reported that she did not have a history of contact with livestock, walking barefoot on wetlands (e.g., paddy fields), or visiting endemic areas. She had been living in a rural area of northern China

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## Statement of Clinical Relevance

Gongylonemiasis is a disease caused by *Gongylonema* infection, which affects individuals who consume contaminated water and raw food. Clinicians should consider the possibility of gongylonemiasis if patients report a creeping sensation within the oral cavity. This article provides information that could help clinicians in the diagnosis and management of this condition.

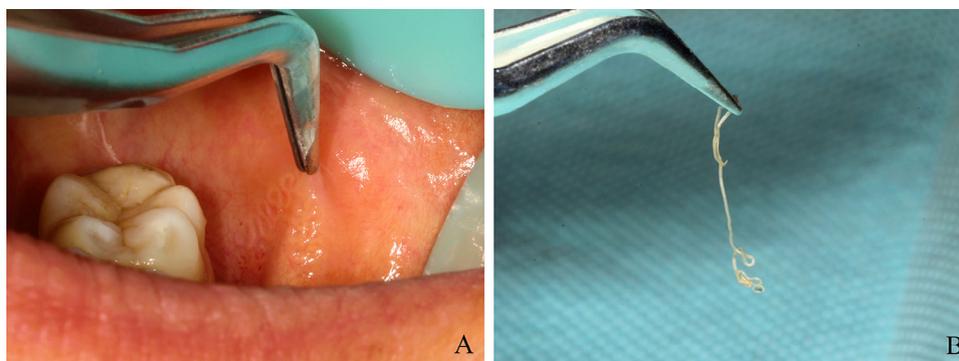


Fig. 1. (A) The serpentine path of *Gongylonema pulchrum* along the left buccal mucosa of the patient. (B) The living nematode that migrated.

and had no related medical history. However, she did report that she liked to eat romaine lettuce and occasionally drank tap water.

A complete blood count revealed no abnormalities. On oral examination, a filamentous, submucosal lump was faintly visible on the posterior left buccal mucosa (Figure 1 A). On palpation, the lump moved toward the back of the oral cavity, making it difficult to inspect visually and remove easily. On more thorough examination, the lump migrated to a more anterior superficial position. The organism was then extracted alive using forceps (Figure 1 B) under surface anesthesia with 2% tetracaine and was immediately placed in 75% ethanol solution for identification. The patient's symptoms resolved immediately after removal of the pathogen.

### Description of specimen

The specimen was transferred to the Department of Microbiology and Parasitology, Peking Union Medical College, for identification. It was a nematode of thread-like form and was coiled while soaked in 75% alcohol. It was approximately 35 mm in length and approximately 0.3 mm in width. The anterior end was covered by numerous round or oval cuticular thickenings. The uterine branches joined with the vagina near the middle of the body, and eggs were found in the vagina. There was a conical and obtuse tail at the posterior end. The morphologic characteristics (size, color, shape, microscopic features of the mouth opening, anterior end, and internal structure) enabled eventual identification of the specimen as an adult female *G. pulchrum*<sup>13,14</sup> (Figure 2).

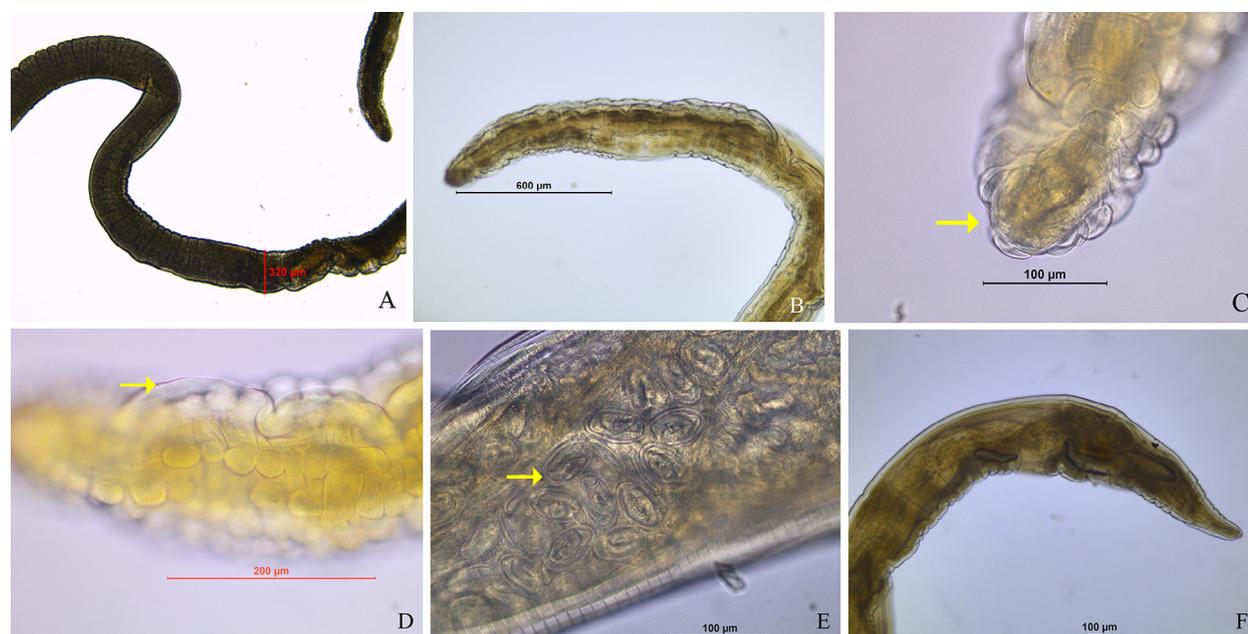


Fig. 2. Morphologic features of *Gongylonema pulchrum*: (A) Body length of 35 mm and width of 0.3 mm; (B) anterior end; (C) mouth opening (arrow, original magnification  $\times 200$ ); (D) cuticular interstitial ridges in the anterior part of the body (arrow,  $\times 200$ ); (E) eggs in the vagina (arrow); (F) posterior end (arrow).

**Table I.** Reported cases of human gongylonemiasis (2001-2016)\*

No.	Year	Demographic characteristics			Medical history			Lab test	Diagnostic method	Treatment	Follow-up	Recurrence	Misdiagnosis
		Region	Age	Sex	Chief complaints	Duration	Site(s)						
1 <sup>a27</sup>	2001	US	F	38	A worm in the mouth	0.5 years	Right side of the buccal mucosa to the midline of the lower lip; mandibular buccal mucosa	Normal	Morphologic examination	1. Removal 2. Albendazole (200 mg bid) for 3 days	—	—	—
2 <sup>30</sup>	2005	Japan	M	34	Irritable stomatitis	—	Left lower lip	Normal	Morphologic examination	—	—	—	Stomatitis
3 <sup>12</sup>	2006	Iran	F	35	A migratory creeping sensation in the neck region and upper part of the digestive tract	1 year	The squamous epithelium on the right side of the lingual frenulum; neck region and upper part of the digestive tract	—	Morphologic examination	1. Removal 2. Albendazole (400 mg)	3 M	N	Psychotic symptoms
4 <sup>11</sup>	2012	US	M	26	A sensation of a migrating, white worm-like lesion in the oral cavity	10 years	Left soft palate	—	Morphologic examination	1. Removal 2. Albendazole (400 mg/d) for 3 days	2 M	N	Oral candidiasis
5 <sup>17</sup>	2013	France	M	48	A moving, wormlike organism in the mouth	—	Mouth	Normal	Morphologic examination	Removal	—	—	—
6 <sup>28</sup>	2013	US	M	36	Removed an intact, living, parasitic nematode from the mucosa of the cheek	3 months	Lower lip	—	PCR	—	—	—	Normal discoloration
7 <sup>29</sup>	2016	China	M	42	Chest congestion	1 month	Esophageal mucosa	Normal	Morphologic examination	Removal	3 M	N	—
8 <sup>†</sup>	2016	China	F	30	A worm in the mouth	2 years	Left buccal mucosa	Normal	Morphologic examination	Removal	3 M	N	—

*bid*, twice daily; *PCR*, polymerase chain reaction.

\*Travel-related case.

<sup>†</sup>Consumed contaminated water or food; —, not mentioned.

**Table II.** Morphologic features of *Gongylonema* spp

No.	Year	Number	Sex	Species	Length (mm)	Width (mm)	Alive or dead
1 <sup>27</sup>	2001	1	F	<i>G. pulchrum</i>	25	0.2	—
2 <sup>30</sup>	2005	1	F	<i>G. pulchrum</i>	46.12	0.19	—
3 <sup>12</sup>	2006	2	1 M, 1 F	<i>G. pulchrum</i>	M17, F30	M0.13, F0.13	—
4 <sup>11</sup>	2012	1	—	<i>Gongylonema</i>	34	—	—
5 <sup>17</sup>	2013	1	M	<i>G. pulchrum</i>	39	0.25	—
6 <sup>28</sup>	2013	1	—	<i>G. pulchrum</i>	20	0.12	—
7 <sup>29</sup>	2016	2	—	<i>G. pulchrum</i>	30	—	Alive
8	2016	1	F	<i>G. pulchrum</i>	35	0.3	Alive

—, not mentioned.

### Treatment and follow-up

After the nematode was removed, the patient's symptoms resolved completely and she was advised to take albendazole 400 mg daily for 3 days, not to drink water that had not been boiled, and to avoid consuming unwashed raw vegetables. At her 1-year follow-up, the patient was completely asymptomatic.

### DISCUSSION AND LITERATURE REVIEW

*G. pulchrum* Molin, 1857, is an ematode parasite of the superfamily Spiruriodea within the order Spirurida, which also includes 10 parasitic species identified in birds and approximately 25 species identified in several kinds of domestic and wild mammals worldwide.<sup>15</sup> Usually, humans are accidental spiruroid hosts. To date, nematodes of 7 spiruroid genera have been reported to infect humans; these include *Gnathostoma*, *Thelazia*, *Gongylonema*, *Physaloptera*, *Spirocerca*, and *Rictularia*.<sup>16</sup> *G. pulchrum*, also known as gullet worm or scutate threadworm, is most often responsible for human spiruroid infections.<sup>10</sup>

To date, human gongylonemiasis has been reported in several countries (Austria,<sup>17</sup> former Soviet Union,<sup>18</sup> Ceylon,<sup>19</sup> China,<sup>20</sup> France,<sup>21</sup> Germany,<sup>22</sup> Hungary,<sup>23</sup> Iran,<sup>12</sup> Japan,<sup>24</sup> Laos,<sup>16</sup> Morocco,<sup>25</sup> New Zealand,<sup>26</sup> the Soviet Union,<sup>27</sup> Spain,<sup>28</sup> Thailand,<sup>29</sup> and the United States<sup>30</sup>) since the first case was reported in the United States in 1850.<sup>15</sup> In the last decade, most cases have been reported in the United States,<sup>31</sup> France,<sup>21</sup> and China.<sup>32</sup> In 2005, Haruki et al.<sup>33</sup> reviewed 48 cases reported from across the world from 1850 to 2001, and we have reviewed the cases of human *Gongylonema* infection published in English since 2001. Clinical information and the morphologic features of *Gongylonema* spp. are summarized in Tables I and II.

The mode of transmission of infection is similar in both humans and animals. The final host, whether a human or an animal, becomes infected by ingestion of the intermediate host containing third-stage larvae that fall into water or adhere to vegetables.<sup>23,24</sup> On ingestion by the final host, the larvae are freed of their capsules and possibly migrate within the wall of the stomach or duodenum

up into the esophagus or oral cavity.<sup>28</sup> In the present case, the patient had a habit of drinking tap water and eating raw vegetables, both of which are risk factors for infection.

Clinically, *Gongylonema* spp. can be found in different areas of the mucous membrane of the upper digestive tract, such as the lips, hard palate, buccal mucosa, gums, and esophagus.<sup>24,32</sup> Clinical manifestations usually include a sensation of transient inflammatory migrating or moving blisters or nodules in the mucosa. Symptoms of pruritus, salivation, toothache, pharyngitis, and esophagitis have also been described,<sup>21</sup> and these symptoms may be misdiagnosed because it is difficult to detect the causative organism. In the present case, it is fortunate that the live organism was detected and extracted, resulting in complete resolution of the patient's symptoms.

There are a few reports of antihelminthic treatment for human gongylonemiasis.<sup>23,33</sup> In several cases, albendazole was administered, although comprehensive data regarding the drug's efficacy and an associated decrease in the number of worms are not conclusive.<sup>11,12,30</sup> No in vitro or in vivo studies on the susceptibility of *G. pulchrum* to antihelminthic medication have been published.<sup>1</sup> However, the prognosis is generally good after extraction of the nematode.<sup>29</sup>

In summary, clinical awareness is critical given the rarity of *Gongylonema* infection in humans and the fact that a confirmed diagnosis is difficult to make without extraction of the parasite. Clinicians should consider the possibility of gongylonemiasis if patients report a creeping sensation within the oral cavity.

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