

CASE REPORT

Nagoya J. Med. Sci. 76. 181 ~ 185, 2014

HUMAN LARYNGITIS CAUSED BY *CLINOSTOMUM COMPLANATUM*

HIROTAKA HARA¹, MD; YUJI MIYAUCHI², MD; SHINSAKU TAHARA¹, MD;
and HIROSHI YAMASHITA¹, MD

¹Department of Otolaryngology, Yamaguchi University Graduate School of Medicine, Ube, Yamaguchi, Japan
²Yamaguchi Grand Medical Center, Hofu, Yamaguchi, Japan

ABSTRACT

A 64-year-old Japanese man visited our outpatient department complaining of an irritable sensation in the throat, occurring two days after eating raw freshwater fish (carp sashimi) at a Japanese-style inn. During laryngeal endoscopy, a slow-moving worm (fluke) was found attached to the surface of the right aryepiglottic fold. After inhalation of 4% lidocaine, the fluke was removed using endoscopic forceps. Patient's throat symptoms immediately improved. The worm was microscopically identified as *Clinostomum complanatum*. *C. complanatum* is a digenetic trematode that usually infects fish-eating water birds. *Clinostomum* infections in humans are rare, and only 21 cases have been described in Japan and Korea. *C. complanatum* infection is known to occur after eating raw freshwater fish, which is a secondary intermediate host. In humans, the metacercariae are released into the stomach and migrate through the esophagus before lodging in the throat. Primary therapy involves endoscopic removal of the worm.

Key Words: *Clinostomum complanatum*; parasite; laryngitis

INTRODUCTION

Clinostomum complanatum is a digenetic trematode. The primary hosts of this parasite are birds such as herons or ducks, and its second intermediate hosts are freshwater fish such as carp, crucian carp, or loach^{1, 2)}. The parasite becomes a metacercaria inside the second intermediate host by forming a globular film around its body and waits for an opportunity to infect the final host^{1, 3, 4)}. Although infections in humans are extremely rare, if a person consumes a raw freshwater fish infected by the larvae of this parasite, the larvae will excyst in the stomach, migrate through the esophagus, and attach to the throat and mature, causing parasitic pharyngitis or laryngitis. Human laryngitis caused by *C. complanatum* is a rare disease; however, in Japan, Korea, and other Asian countries where eating raw fish is a tradition, it is possible for individuals to become infected by this intestinal parasite^{5, 6, 7)}. In this study, we describe a patient with laryngitis caused by *C. complanatum* that occurred after the patient ate freshwater fish sashimi.

Received: September 24, 2013; accepted: October 29, 2013

Corresponding author: Hirotaka Hara, MD

Department of Otolaryngology, Yamaguchi University Graduate School of Medicine,
Ube, Yamaguchi 755-8505, Japan

Tel and Fax: 81-836-22-2280, E-mail: harahiro@yamaguchi-u.ac.jp

CASE REPORT

A 64-year-old Japanese man visited the outpatient's department of otolaryngology, Yamaguchi University Hospital, Ube, Japan. The patient complained of suddenly developed violent cough and irritable sensation in the throat. He also experienced intermittent throat soreness. His symptoms occurred two days after he had eaten raw carp sashimi for dinner at a Japanese-inn situated at a hot spring.

He exhibited no symptoms of hoarseness, dyspnea or swallowing disturbance. Furthermore, a blood test showed no increase in white blood cell count or C-reactive protein level, and a differential white blood cell count showed no eosinophilia. A fiberoptic examination of the larynx revealed no clear signs of inflammation, such as redness or swelling of the pharynx or larynx. However, there was a foreign object attached to the surface of the right aryepiglottic fold that moved slowly during observation (Figure 1). Considering that the patient had consumed raw carp and that the shape of the object resembled a fluke when more closely observed, the probable diagnosis was infection by a parasitic fluke.

After explaining the procedure to the patient and obtaining his consent, an endoscopic removal of the fluke was performed under local anesthesia because he did not have a strong gag reflex. To stop the parasite moving and to make the removal easier, 1 ml of 4% lidocaine via inhalation was administered to the patient; with the patient in the right lateral decubitus position, a parasite of approximately 6-mm length was removed using biopsy forceps with a laryngeal endoscopy. The parasite was almost completely motionless during removal because of the effects of the lidocaine. Removal was easy because the parasite was not strongly attached to the mucosa, too. The patient's throat discomfort disappeared almost immediately after the parasite was removed.

The patient was consulted and visited the Gastroenterology department of our hospital for an upper gastrointestinal endoscopy to confirm that there were no residual parasites in the esophagus or stomach, and his treatment was completed.

The overall length of the parasite was approximately 6 mm (Figure 2). Microscopic findings showed that the frontal end consisted of an oral sucker with a ventral sucker slightly caudal to it. There were black ovaries on both sides of the body, which was identified as the digenetic trematode *C. complanatum*.

DISCUSSION

The frequency of parasitic infections in humans is decreasing because of changes in lifestyle and environments where food is cultivated. For example, roundworm infections spread because of the agricultural use of human waste, but their incidence has been greatly reduced with the widespread use of chemical fertilizers. Similarly, in Japan, improvements in sanitation have decreased the incidence of parasitic diseases, such as tapeworms and pinworms. Sometimes overseas travelers carry parasitic diseases such as malaria.

However, infections from parasites in food are still observed in areas where people consume raw meat or fish. People in Japan, Korea, and other Asian countries have consumed raw fish for long. One traditional dish is sashimi and slices of raw fish that are now consumed all over the world as typical Japanese food such as sushi. Sashimi prepared from fresh marine fish can be safely consumed, but not completely safe, because parasite such as *Anisakis* can infect human, and other bacteria or virus also can cause gastroenteritis. However, wild fish that live in brackish water or fresh water may be infected with parasites; thus, they are inappropriate to be consumed as sashimi.

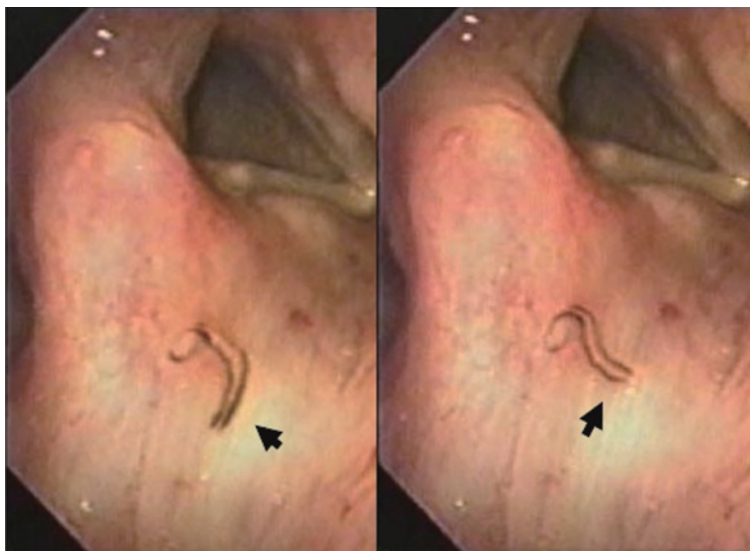
LARYNGITIS CAUSED BY *C. COMPLANATUM*

Fig. 1 Endoscopic findings of *Clinostomum complanatum* infection
A slow-moving worm attached to the surface of the larynx (arrow).

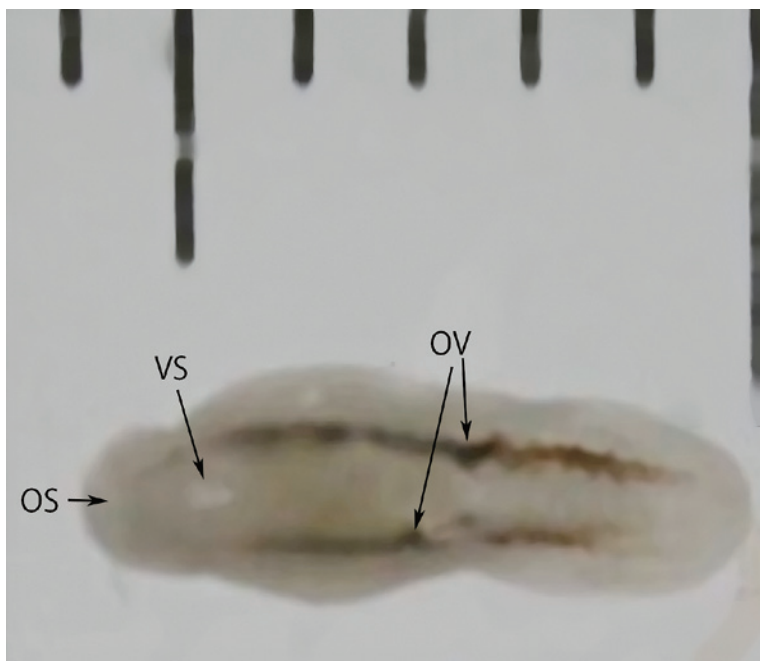


Fig. 2 Gross image of *Clinostomum complanatum* (6.0 mm in length)
There were black ovaries on both sides of the body.
OS: oral sucker, VS: ventral sucker, OV: ovary

Table 1 A list of human cases of *Clinostomum* infection in Japan.

Case No.	Age	Sex	Address	Site of infection	Symptoms	Author(s)
1	22	F	Osaka	Pharynx	Unknown	Yamashita (1938)
2	38	F	Toyama	Pharynx	Throat pain globus sensation	Hori (1942)
3	30	F	Shimane	Larynx (arytenoid)	globus sensation	Kamo <i>et al.</i> (1962)
4	34	F	Nagasaki	Pharynx	globus sensation swallowing pain	Sakaguchi <i>et al.</i> (1966)
5	53	F	Gifu	Pharynx (posterior wall)	globus sensation	Sano <i>et al.</i> (1980)
6	31	F	Aichi	Pharynx~lower airway	Cough Hemoptum throat pain low grade fever	Kumada <i>et al.</i> (1983)
7	35	F	Kumamoto	Pharynx (posterior wall)	globus sensation	Hirai <i>et al.</i> (1987)
8	57	F	Shiga	Pharynx (posterior wall)	globus sensation swallowing pain	Furukawa <i>et al.</i> (1987)
9	15	F	Shimane	Pharynx	globus sensation	Yamane <i>et al.</i> (1990)
10	54	F	Saga	Larynx (arytenoid)	globus sensation	Umezaki <i>et al.</i> (1990)
11	70	F	Akita	Pharynx	Throat pain	Yoshimura <i>et al.</i> (1991)
12	68	F	Shimane	Pharynx (posterior wall)	globus sensation	Isobe <i>et al.</i> (1993)
13	37	F	Saga	Pharynx	globus sensation	Kifune <i>et al.</i> (1994)
14	40	F	Saga	Pharynx	Throat pain	Kifune <i>et al.</i> (1994)
15	27	F	Shimane	Pharynx (posterior wall)	globus sensation	Maejima <i>et al.</i> (1996)
16	27	F	Hiroshima	Unknown	globus sensation	Shirai <i>et al.</i> (1998)
17	20	F	Yamaguchi	Larynx (arytenoid)	globus sensation	Kifune <i>et al.</i> (2000)
18	27	M	Saga	Larynx (arytenoid)	Throat pain globus sensation	Kakizoe <i>et al.</i> (2004)
19	64	M	Yamaguchi	Larynx (arytenoid)	globus sensation	Present record

Sashimi made from carp, a freshwater fish, is a traditional dinner item that is sometimes served at Japanese-style hot spring inns near lakes or streams. Currently, only carp farmed in spring water that cannot be infected by parasites are used to make sashimi because cooking wild carp is an issue. To consume the wild fresh-water fish without heating it, it would normally have to be frozen for 24 h at -20°C (-4°F) in order to kill the parasites. As far as we searched previous reports extensively, this case constitutes the 19th documented case of *Clinostomum* infection in Japan (Table 1). There are many reports from western Japan, and the infection is prevalent in areas along the Sea of Japan⁷. Interestingly, there have also been two recent reports from Korea^{6, 8}. Sites of parasitic infection include the pharynx, arytenoid region, posterior oropharyngeal wall, and lateral lymphatic band. Typically, the only symptom is throat discomfort. However, some case reports also mention a sore throat or swallowing pain, while others report bloody phlegm and fever⁹.

A diagnosis is easily made by finding the parasite. If the patient has consumed raw freshwater fish, the physician can search for a parasite while carefully inspecting the symptomatic area.

There is no effective antiparasitic drug, which makes mechanical removal necessary. Reported methods of removal include endoscopic removal under local anesthesia and removal using a direct laryngoscope under general anesthesia. The alleged reason for choosing to remove the parasite under general anesthesia is to ensure a proper grip on the fluke^{9, 10}, but the fluke can almost be completely removed under local anesthesia^{7, 8}. In our case, the patient's gag reflex was not strong; therefore, we had performed the endoscopic removal of the fluke under local anesthesia. If a direct laryngoscope was required to remove the fluke, the position of the laryngoscope should be changed in case the fluke moved, which would probably make the procedure more complicated. In contrast, with an endoscope, even if the fluke moves the endoscope can easily bend and follow it.

LARYNGITIS CAUSED BY *C. COMPLANATUM*

Kitagawa *et al.*¹⁰⁾ reported that when they were removing *C. complanatum*, the fluke was quickly moving; therefore, they had to apply directly spraying it with 8% lidocaine solution to decrease its movements. In our case, the patient inhaled 1 ml of 4% lidocaine to stop the parasite's movements before safely removing the parasite using biopsy forceps. In the report described above, a minimal amount of lidocaine was used to effectively stop the parasite's movements, and we found that inhaled only 1 ml of 4% lidocaine was also effective.

In the life cycle of *C. complanatum*, the metacercariae enter a patient's body, excyst in the stomach, migrate through the esophagus, and then attach to the throat and mature, causing parasitic laryngitis. Therefore, there may be a delay of several days between oral ingestion and emergence of symptoms. Even when a fluke is removed from the throat, the metacercariae may remain in the digestive tract or there may be more than one parasite, making it necessary to not only examine the throat but also perform an upper gastrointestinal endoscopy. We also performed an upper gastrointestinal endoscopy in this case the day after removing the parasite and confirmed that there were no residual parasites.

CONCLUSION

We described a rare case of laryngitis caused by *C. complanatum*.

In cases where patients develop throat discomfort after consuming raw freshwater fish, this disease should be considered as a possible diagnosis, and a fiberoptic examination of the laryngopharynx should be performed.

Conflict of Interest: None

REFERENCES

- 1) Yamaguti S. Studies on the helminth fauna of Japan. Part 1. Trematodes of birds, reptiles and mammals. *Jpn J Zool* 1933; 5: 1–134.
- 2) Yamaguti S. A Synoptical Review of Life Histories of Digenetic Trematodes of Vertebrates, with Special Reference to the Morphology of Their Larval Forms. Tokyo: Keigaku publ. Co; 1975. p.304–310.
- 3) Kagei N, Yanohara Y, Uchikawa R, Sato A: On the yellow grubs, metacercariae of *Clinostomum complanatum* (Rudolphi,1819), found in the cultured loach. *Jpn J Parasitol* 1984; 33: 59–62.
- 4) Kagei N, Yanohara Y, Uchikawa R, Sato A. Natural infection with *Clinostomum complanatum* (Rud., 1819) in the birds of southern Japan. *Jpn J Parasitol* 1988; 37: 254–257.
- 5) Yamashita J. *Clinostomum complanatum*, a trematode parasite new to man. *Annot Zool Japan* 1938; 17: 563–566.
- 6) Chung DI, Moon CH, Kong HH, Choi DW, Lim DK. The first human case of *Clinostomum complanatum* (Trematoda: Clinostomidae) infection in Korea. *Korean J Parasitol* 1995; 33: 219–223.
- 7) Kifune T, Ogata M, Miyahara M. The first case of human infection with *Clinostomum* (Trematoda: Clinostomidae) in Yamaguchi Prefecture, Japan (in Japanese). *Med Bull Fukuoka Univ* 2000; 27: 101–105.
- 8) Park CW, Kim JS, Joo HS, Kim J. A human case of *Clinostomum complanatum* infection in Korea. *Korean J Parasitol* 2009; 47: 401–4.
- 9) Umezaki T, Shin T, Osa M, Kifune T and Mogi M. A case of acute laryngitis caused by *Clinostomum complanatum* with a complaint of throat irritation (in Japanese). *Otologica Fukuoka* 1990; 36: 665–668.
- 10) Kitagawa N, Oda M, Totoki T, Washizaki S, Oda M, Kifune T. Lidocaine spray used to capture a live *Clinostomum* parasite causing human laryngitis. *Am J Otolaryngol* 2003; 24: 341–343.