

CASE REPORT

새인두흡충에 의한 인후두염을 내시경으로 치료한 1예

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A Case of Endoscopically Treated Laryngopharyngitis Resulting from *Clinostomum complanatum* Infection

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A 46-year-old woman visited our hospital presenting throat pain and globus sensation. The symptoms occurred seven days after eating raw perch and mullet. An endoscopy under sedation showed a fluke—with an approximate length of 4.8 mm and width of 1.5 mm—on the left aryepiglottic fold, with active motility on the mucosa. It was extracted from the larynx using biopsy forceps and was identified as *Clinostomum complanatum*. To the best of our knowledge, this is the second reported case of human infection with *Clinostomum complanatum* diagnosed and treated by an endoscopy in Korea. Endoscopy is a useful tool in the diagnosis and treatment of patients at risk for parasitic infections complaining of throat pain. (*Korean J Gastroenterol* 2017;69:177-180)

Key Words: Food-borne diseases; Parasites; Endoscopy; Digenea; *Clinostomum complanatum*

INTRODUCTION

Clinostomum complanatum (*C. complanatum*) commonly infects the pharynx, larynx, or upper esophagus of piscivorous birds.¹ Cases of human infection with *C. complanatum* have been reported mainly in Japan.^{2,3} The first Korean case of human infection was published in 1995; two additional cases have been reported to date.⁴⁻⁶ *C. complanatum* is a parasitic flatworm—known as a fluke—that remains in an encysted resting state (metacercaria) inside its second intermediate host, causing an infection in the final host.^{7,8} Although an infection by *C. complanatum* is extremely rare in

humans, laryngopharyngitis can occur when raw fish (i.e., the second intermediate) is ingested (i.e., the final host).

CASE REPORT

A 46-year-old woman, without any underlying disease, visited our outpatient clinic presenting throat pain and globus sensation for the past two days. Her vital signs were stable, and her initial physical examination revealed no abnormal findings. She reported eating raw perch and mullet one week prior to the onset of these symptoms. A laryngoscopic study conducted in the ENT department of our hospital showed no

Received November 10, 2016. Revised December 29, 2016. Accepted January 17, 2017.

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Financial support: None. Conflict of interest: None.

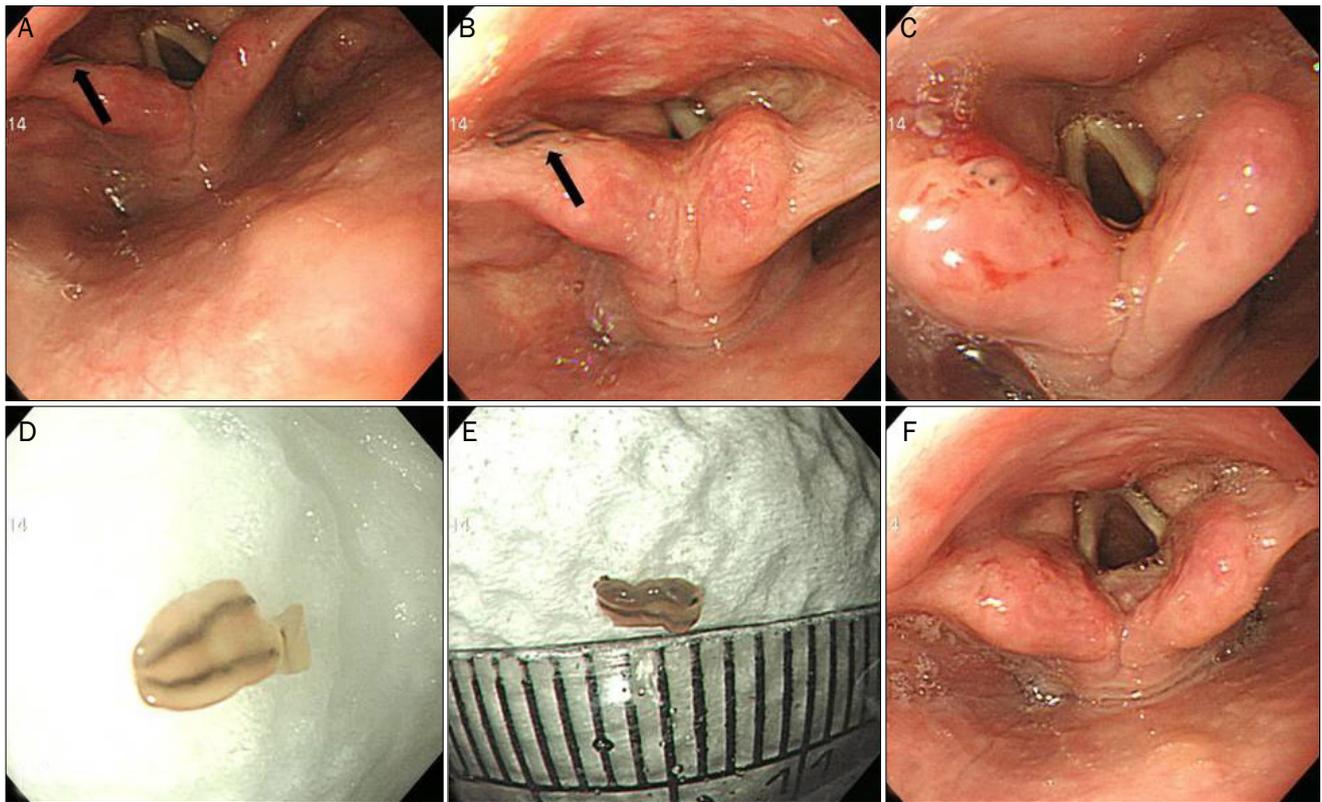


Fig. 1. (A, B) Endoscopy shows the entire *Clinostomum complanatum* organism (black arrow) on the lateral side of the left aryepiglottic fold. (B–E) The fluke is approximately 5 mm in length and 2 mm in width, and it was torn during the removal procedure. (F) After removal, small mucosal petechiae are observed in the affected area, but neither active bleeding nor mucosal defects are observed.

abnormal findings in the pharyngolarynx. An endoscopy was performed under sedation, and a 5-mm-long motile fluke was observed on the lateral side of the left aryepiglottic fold (Fig. 1A, B). Inside the fluke, which was translucent, two black-colored lines were observed (Fig. 1B, D, E). The fluke was successfully extracted out of the larynx using biopsy forceps. After removal, small mucosal petechiae were observed on the affected mucosa. However, no active bleeding or mucosal defects were observed (Fig. 1C, F).

The fluke was torn in half during the removal procedure (Fig. 1C). To identify its morphology, longitudinal sections of the parasite were made and stained with hematoxylin and eosin (Fig. 2A). The internal structures recognized in the sections were the testis, ovary, uterus, vitellaria, and intestinal ceca. The two testes were parallel to one another and divided by the uterus. On the outer surface of the tegument, many filamentous materials were observed (Fig. 2B). Moreover, no tegumental spine was observed. Based on these findings, we concluded that the removed fluke was *C. complanatum*. Upon removal of the fluke, the symptoms improved and she

was discharged.

DISCUSSION

The incidence of parasitic diseases in Korea has been decreasing rapidly in the past several decades. However, food-borne parasites, including those carried by freshwater fish and brackish-water fish, crayfish, oysters, and shellfish, remain to be a problem.⁹ Moreover, parasites that did not previously infect humans have emerged as new infective agents due to changes in human living environments and lifestyles. Moreover, parasitic infections that could not be diagnosed in the past—due to a lack of diagnostic techniques or equipment—can now be diagnosed due to recent advancement in medicine.

C. complanatum mainly infects freshwater fish or brackish-water fish, and humans can become accidental hosts when they ingest an affected raw fish. Laryngopharyngitis, caused by *C. complanatum*, was first reported by Yamashita in 1938, and 19 cases of human infection have been reported

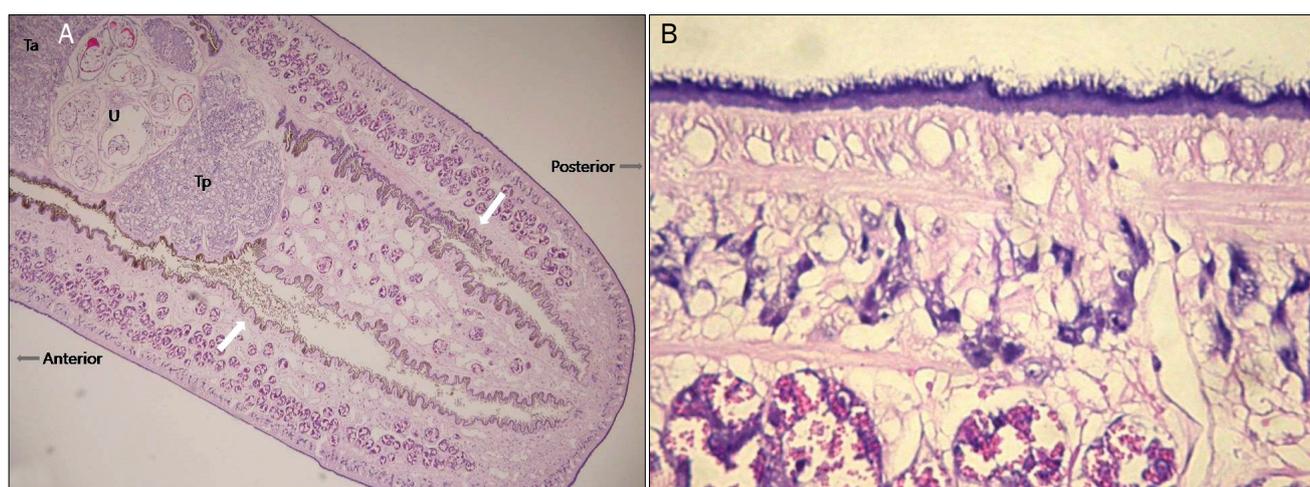


Fig. 2. (A) Histological sections of the fluke show a characteristic distribution of internal organs for *Clinostomum complanatum*. The two testes are roughly triangular. The posterior testis is a rounded triangular shape and anteriorly concave. A part of the uterus containing eggs is positioned between the anterior and posterior testes. The ovary is placed laterally to the uterus. The numerous follicular vitellaria are distributed along the lateral margins of the posterior half of the body. Two intestinal ceca (white arrows) run posteriorly, containing digested materials that are stained brown and dark gray in color (H&E stain, x40). (B) Histological examination demonstrates the tegument with the many filamentous materials (H&E stain, x400). Ta, anterior testis; U, uterus; Tp, posterior testis.

Table 1. Summary of *Clinostomum complanatum* Infections in Korea

Case no.	Study	Age(yr)/ Sex	Site of infection	Symptoms (Duration, day)	Food	Removal method
1	Chung et al. (1995) ⁴	56/M	Pharynx	Throat pain (3-4 d)	Fresh-water fish	Otolaryngological removal
2	Park et al. (2009) ⁵	33/M	Larynx	Throat pain (7-8 d)	Perch	Endoscopic removal
3	Jung et al. (2015) ⁶	13/F	Nasopharynx	Globus sensation (4 d)	Mullet	Surgical removal
4	Present case	46/F	Larynx (AEF)	Throat pain, globus sensation (7d)	Perch, Mullet	Endoscopic removal

AEF, aryepiglottic fold.

in Japan since then.^{2,3} *C. complanatum* is a fluke that is indigenous to Korea.^{7,8} Four human cases have been reported in Korea to date, including the present case (Table 1).⁴⁻⁶ Perch, which is a brackish-water fish, is a medically important intermediate host for *C. complanatum*, and human infections may occur if an affected fish is consumed raw.^{7,8} This fluke attaches to the mucosa, causing a parasite-induced laryngopharyngitis in humans; however, to the best of our knowledge, there has not been any cases involving serious complications associated with *C. complanatum* infection.³

The only known treatment for this kind of infection is removing the fluke. In previously reported cases, *C. complanatum* was removed under general anesthesia using a laryngoscopic method.^{6,10} In this case, we successfully removed the fluke using an endoscopy under sedation. We used an endoscopy to confirm that no flukes remained in the larynx, pharynx, esophagus, or stomach. Treatment with endoscopy un-

der sedation presents several advantages. First, parasites can be extracted without causing pain or eliciting a gag reflex. Second, no additional tests are necessary to confirm whether parasites are present in the esophagus or the stomach. Third, it is a cost-effective method for an effective removal of the parasites without general anesthesia or surgery.

In the present case, the parasite was strongly attached to the mucosa and was torn during extraction, as shown in Fig. 1. The parasite can be removed using a lidocaine spray. The removal process can be made easy by using lidocaine, which can reduce the movement of parasites and weakening their attachment to the mucosa.¹⁰

New parasitic diseases have recently been reported, as freshwater fish, saltwater fish, and brackish-water fish are more commonly ingested raw due to the increasing popularity of certain types of diets.⁹ Since the first report of *C. complanatum* infection in humans in 1995, only three cases have

been reported in Korea.^{4,6} However, the number of cases may be underestimated because this type of parasite may not be well known or as frequently reported; it is possible for *C. complanatum* infection to be misdiagnosed as anisakiasis.^{11,12}

In conclusion, laryngopharyngitis resulting from *C. complanatum* infection is a rare occurrence. This case demonstrates that collecting the history of sashimi ingestion is important for patients with acute laryngopharyngitis. In particular, raw perch consumption can result in human infection with *C. complanatum*. An endoscopy is helpful for those presenting a globus sensation several days after consuming raw fish. During an endoscopy, it is important to consider the possibility of fluke at the border between the epiglottis and aryepiglottic fold, as well as in the oral cavity.

REFERENCES

1. Aohagi Y, Shibahara T, Machida N, Yamaga Y, Kagota K, Hayashi T. Natural infections of *Clinostomum complanatum* (Trematoda: Clinostomatidae) in wild herons and egrets, Tottori Prefecture, Japan. *J Wildl Dis* 1992;28:470-471.
2. Yamashita J. *Clinostomum complanatum*, a trematode parasite new to man. *Annot Zool Japan* 1938;17:563-566.
3. Hara H, Miyauchi Y, Tahara S, Yamashita H. Human laryngitis caused by *Clinostomum complanatum*. *Nagoya J Med Sci* 2014;76:181-185.
4. 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.
5. Park CW, Kim JS, Joo HS, Kim J. A human case of *Clinostomum complanatum* infection in Korea. *Korean J Parasitol* 2009;47:401-404.
6. Jung SC, Oh HJ, Kim DM, Park JH. A case of pharyngitis caused by *Clinostomum complanatum*. *Korean J Otorhinolaryngol-Head Neck Surg* 2015;58:61-63.
7. Sohn WM, Na BK, Cho SH, Lee SW, Choi SB, Seok WS. Trematode metacercariae in freshwater fish from Water Systems of Hantangang and Imjingang in Republic of Korea. *Korean J Parasitol* 2015;53:289-298.
8. Chung DI, Kong HH, Moon CH. Demonstration of the second intermediate hosts of *Clinostomum complanatum* in Korea. *Korean J Parasitol* 1995;33:305-312.
9. Chai JY. Fish-borne parasitic diseases. *Hanyang Med Rev* 2010;30:223-231.
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.
11. Choi JH, Kim EJ, Kim DM, Park SH, Kim SW. A case of anisakiasis invading the oropharynx. *Korean J Otorhinolaryngol-Head Neck Surg* 2015;58:61-63.
12. Lee EJ, Kim YC, Jeong HG, Lee OJ. The mucosal changes and influencing factors in upper gastrointestinal anisakiasis: analysis of 141 cases. *Korean J Gastroenterol* 2009;53:90-97.