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—approximately 4.8 mm in length and 1.5 mm in width—on the left aryepiglottic fold, with active motility on the mucosa. It was extracted from the larynx using biopsy forceps and 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. Cases of human infection with C. complanatum have been reported mainly in Japan. The first Korean case of human infection was published in 1995; two additional cases have been reported to date. 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. 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
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 *Clinostomum 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. 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

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

AEF, aryepiglottic fold.

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.⁵,¹⁰ 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 under 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. 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.

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