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☐ **Case Report** ☐

Two cases of *Gymnophalloides seoi* infection accompanied by diabetes mellitus

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Abstract: *Gymnophalloides seoi*, a new intestinal trematode of humans transmitted by oysters on a southwestern island of Korea, drew medical attentions because of its possible relationship with evoking pancreatitis or other pancreatic diseases. We experienced two interesting cases of *G. seoi* infection who were accompanied with diabetes mellitus. In routine stool examination, eggs of a gymnophallid were detected from two patients, and after treatment with praziquantel and purgation, 772 and 10 adult flukes were recovered respectively. They were identified as *G. seoi*. The first patient was a 62-year old man who lived in Mokpo, nearby the known endemic area, and the second patient, a 54-year old woman who lived in Inchon. They used to eat raw oysters. It should be ruled out that *G. seoi* infection has some relationship with pancreatic diseases.

Key words: *Gymnophalloides seoi*, diabetes mellitus, human, case report

INTRODUCTION

Trematodes belonging to the family Gymnophallidae had been known as parasites of avian hosts (James, 1964; Ching, 1965 & 1973), but it began to draw medical attentions after the discovery of a patient infected with *Gymnophalloides seoi*, a new human intestinal fluke in Korea (Lee *et al.*, 1993). Subsequently this gymnophallid was shown to be highly prevalent on a southwestern coastal island (Lee *et al.*, 1994). The infection source was proved to be locally produced oysters which are favoured by people under raw conditions (Lee *et al.*, 1995). The 1st intermediate host and natural final hosts other than humans have not yet been discovered.

Because the first patient infected with *G. seoi* suffered from acute pancreatitis with severe abdominal discomfort and he was recovered from these symptoms after anthelmintic treatment (Lee *et al.*, 1993), a possible relationship of *G. seoi* infection with pancreatic duct involvement was strongly suggested. We present here two interesting cases of *G. seoi* infection who were accompanied with diabetes mellitus.

CASES DESCRIPTION

Case I:

A 62-year old Korean man was admitted to the Seoul National University Hospital (SNUH) for management of diabetes mellitus (DM) and essential thrombocythemia. He complained of easy fatiguability due to DM, but no significant gastrointestinal symptoms were accompanied.

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He lived in Mokpo-shi, Chollanam-do, for more than 60 years and had eaten freshwater fishes including the minnow and eel, and marine bivalves such as oysters. In fecal examination eggs of a gymnophallid and *Heterophyes nocens* were detected. The gymnophallid eggs measured 19-21 μm (mean 20) long and 14-16 μm (15) wide, and were characterized by elliptical, transparent, and thin egg shell.

Laboratory data showed mild increase of the total number of WBC(20,000/mm^3) and 10% eosinophilia. The platelet number was significantly increased, 1.35 million/mm^3. Serum albumin level was as low as 0.7 g/dl, and urinalysis revealed 3+ of glucose, but serum amylase was within normal limit. The patient was treated with 10 mg/kg of praziquantel and given 30 g of $MgSO_4$ twice 30 minutes after the treatment. The worms discharged were collected from the diarrheic stool, and 772 *G. seoi* and 98 *H. nocens* worms were recovered. In laboratory examination after the treatment the number of WBC was lowered to 6,000/mm^3, eosinophil count 2%, and serum albumin 3.9 g/dl.

The recovered gymnophallid flukes were 0.35-0.45 mm long and 0.25-0.32 mm wide, and identified as *Gymnophalloides seoi* Lee, Chai and Hong, 1993 based on the following morphological characters: presence of the ventral pit surrounded by strong muscle fibers, and small inconspicuous genital pore opened at the anterior margin of the ventral sucker not surrounded by prominent muscle fibers (Lee *et al.*, 1993).

Case II:

A 54-year old woman was admitted to the Seoul Paik Hospital for management of DM. She suffered from DM for 4 years, and experienced various symptoms of DM including 8 kg weight loss during the period of her illness. Physical examination revealed no abnormalities except limitation of motion of the left shoulder. She complained of moderate degrees of gastrointestinal symptoms such as epigastric discomfort, indigestion, and diarrhea.

She had lived in Inchon City, and recalled she had eaten raw oysters there. In laboratory examination the total number of WBC was increased to 16,700/mm^3 but the eosinophil count(2%) and serum amylase level were within normal limits. At stool examination eggs of a gymnophallid were detected. After a treatment with praziquantel and $MgSO_4$ administration, 10 adult *G. seoi* worms were recovered.

DISCUSSION

Since the eggs of *G. seoi* are very small in size, not so peculiar in their morphology, and produced only in small numbers (unpublished data), correct diagnosis of this trematode infection is very difficult unless the adult worms are recovered. Physicians and laboratory technicians generally do not know about this parasite, so many of *G. seoi* infected cases seem unnoticed or misdiagnosed as other parasite infections.

The clinical symptoms and pathologic features due to *G. seoi* infection are not precisely known yet. However, the first human case (Lee *et al.*, 1993) suffered from acute pancreatitis with elevated serum amylase level. It was also notifiable that many of the infected inhabitants in a highly endemic area complained of variable degrees of manifestations from mild indigestion to severe colicky pain (Lee *et al.*, 1994), just as seen in other intestinal trematode infections (Chai and Lee, 1990).

The reason for such a variability of clinical symptoms due to intestinal fluke infections in general is not clear, although mild symptoms might be due to a development of resistance by repeated infections in endemic areas. In fact, a man who was residing in an endemic area of *M. yokogawai* and harbouring as many as 63,587 worms complained of only minor gastrointestinal troubles (Seo *et al.*, 1985). Conversely a patient infected with only 110 *M. yokogawai*, who was a visitor to an endemic area, suffered from severe abdominal pain, diarrhea, and fatigue (Chai *et al.*, 1989). On this account, Chai *et al.* (1989) suggested that the severity of clinical symptoms in intestinal fluke infections seems not necessarily correlated with individual worm burden, but dependent upon the immune status and susceptibility of the host.

It is of special interest, however, that the present two cases of *G. seoi* infection were accompanied by DM. It is of course well known that DM patients reveal increased susceptibility to infection by various agents such as virus, bacteria and parasites. However, since the first patient of *G. seoi* infection suffered from acute pancreatitis (Lee *et al.*, 1993), a possibility for *G. seoi* infection to cause or facilitate development of pancreatic duct inflammation and DM should also be considered.

Generally DM is subdivided into two groups, i.e. insulin-dependent and insulin-independent types. Insulin-dependent DM classically occurs in children and juvenile ages, and is known to be a consequence of severe pancreatic β-cell loss. Insulin-independent DM occurs in adult age, and is related with obesity (Kovacs and Asa, 1991). Possible causes of insulin-dependent DM that have been considered include viral infection and anti-islet cell antibodies, and is associated with HLA subtype (ReGroof, 1989). However, most of the insulin-dependent DM cases are from unknown etiology.

In Case I of this study, the patient has been ill with insulin-dependent DM after the age of thirty, at which age insulin-independent DM is more common. He might have been suffering from *G. seoi* infection for a long time. It is further speculated with no evidence that *G. seoi* migrated to the pancreatic duct and promoted destruction of pancreatic islet cells. At present, however, this possibility in humans and animals remains uncertain. In other trematodes belonging to the family Gymnophallidae they can parasitize extraintestinal organs (Yamaguti, 1939). James (1964) observed that gymnophallid flukes were parasitizing in the bursa of Fabricii, gall bladder, and intestine of their hosts. Cable (1953) also discovered adults of a gymnophallid in the intestine and gallbladder of aquatic birds. So it should be ruled out that *G. seoi* could migrate into the pancreatic duct of humans, and possibly have some relationship with DM.

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=초록=

당뇨병과 동반된 참굴큰입흡충증 2례

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참굴큰입흡충(*Gymnophalloides seoi*)은 참굴이 매개하는 우리 나라 고유의 인체기생 장흡충의 하나로서, 전남 신안군 도서 지방에서 큰 유행지가 발견된 바 있다. 이 흡충은 췌장 질환과의 관계가 의심되어 의학적 관심의 대상이 되고 있다. 이번 증례 보고에서는 당뇨병과 동반된 흥미있는 *G. seoi* 감염 2례를 발견하여 보고하고자 한다. 환자들은 당뇨병 치료를 위해 내원하였으며 내원시 대변검사에서 참굴큰입흡충류(gymnophallid)의 충란이 검출되었다. 프라지콴텔 및 하제를 투여한 후 충체를 수집한 결과 *G. seoi* 충체 772마리 및 10마리가 각각 검출되었다. 증례 I은 62세 남자로 *G. seoi*의 유행지 부근인 목포에 거주하고 있었고, 증례 II는 인천에 거주하고 있으면서 참굴을 날로 즐겨 먹었다고 하였다. 이 증례들을 볼 때 *G. seoi* 감염이 당뇨병과 어떠한 상관관계를 가지고 있을 가능성을 시사하였다.

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