

• CLINICAL RESEARCH •

Clinical features of human intestinal capillariasis in Taiwan

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Abstract

Human intestinal capillariasis is a rare parasitosis that was first recognized in the Philippines in the 1960 s. Parasitosis is a life threatening disease and has been reported from Thailand, Japan, South of Taiwan (Kaoh-Siung), Korea, Iran, Egypt, Italy and Spain. Its clinical symptoms are characterized by chronic diarrhea, abdominal pain, borborygmus, marked weight loss, protein and electrolyte loss and cachexia. Capillariasis may be fatal if early treatment is not given. We reported 14 cases living in rural areas of Taiwan. Three cases had histories of travelling to Thailand. They might have been infected in Thailand while stayed there. Two cases had the diet of raw freshwater fish before. Three cases received emergency laparotomy due to peritonitis and two cases were found of enteritis cystica profunda. According to the route of transmission, freshwater and brackish-water fish may act as the intermediate host of the parasite. The most simple and convenient method of diagnosing capillariasis is stool examination. Two cases were diagnosed by histology. Mebendazole or albendazole 200 mg orally twice a day for 20-30 d is the treatment of choice. All the patients were cured, and relapses were not observed within 12 mo.

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INTRODUCTION

Capillaria species parasitize many classes of vertebrates, although only 4 species described have been found in humans, namely *Capillaria philippinensis*, *Capillaria plica*, *Capillaria aerophila*, and *Capillaria hepatica*^[1]. *C. philippinensis* is a tiny nematode that was first described in the 1960 s as the causative agent of severe diarrheal syndromes in humans. In 1962, the first case of human intestinal capillariasis occurred in a previously healthy young man from Luzon (Philippines) who subsequently died. At autopsy, a large number of worms, later

described as *C. philippinensis*, were found in the large and small intestines^[2]. The disease was first reported by Chitwood *et al.* in 1964^[3]. During the Philippine epidemic from 1967 to 1968, more than 1300 persons acquired the illness and 90 patients with parasitologically confirmed infections died^[4]. In late 1978 and early 1979, another small outbreak was identified in northeastern Mindanao, the Philippines, and about 50 persons acquired the infection^[4]. Sporadic cases continued to appear in northern Luzon as well as in other areas where epidemics had occurred. The disease is also endemic in Thailand, and was first reported in 1973^[5]. Sporadic cases have also been found in Iran^[6], Egypt^[7,8], Taiwan^[9], Japan^[10,11], Indonesia^[12], Korea^[13], Spain (probably acquired in Colombia)^[14] and Italy (acquired in Indonesia)^[15], indicating that this infection is widespread. Because the infection can result in a severe disease with a high mortality when untreated, early diagnosis is very important. Here we described 14 cases of human intestinal capillariasis found in Taiwan from 1983 to 2001.

MATERIALS AND METHODS

Since 1983, 14 cases have been diagnosed as intestinal capillariasis in Taiwan, all with the symptoms of chronic diarrhea, abdominal pain, borborygmus and marked weight loss. All patients were hospitalized for examination and treatment. Their diagnosis was confirmed by eggs and/or larvae and/or adult *C. philippinensis* found in the feces of 5 patients. Two cases were recognized by a pathologist by histology of jejunum or ileum (Figures 1, 2) with negative stool examination. Bacterial cultures of stool specimens were negative in all patients. The stool specimens were examined by formalin-ether concentration method. *C. philippinensis* eggs were peanut-shaped with flattened bipolar plugs, 20×40 μm in size (Figure 3).

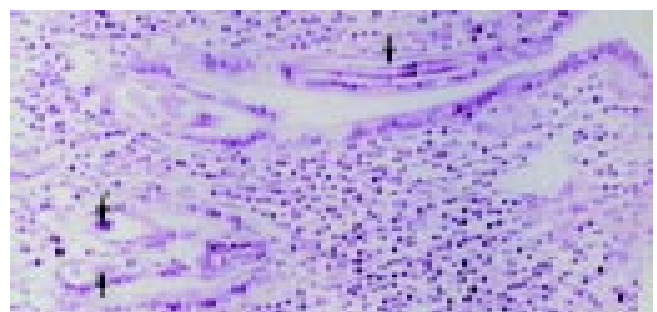


Figure 1 *C. philippinensis* worms embedded in intestinal mucosa (arrow). Hematoxylin and eosin, ×200.

RESULTS

Fourteen cases, nine males and five females, were 36 to 76 years old when they were diagnosed as intestinal capillariasis (Tables 1, 2). Three lived in Kaohsiung County and eleven in Taitung County. Seven of them were aborigines and two were brother and sister. Two of them had histories of travelling to Thailand. Two patients had history of eating raw or insufficiently cooked fresh-water fish. Four of 14 patients had mixed infection with *Clonorchis sinensis* or *Strongyloides stercoralis* whose eggs were also found in the feces.

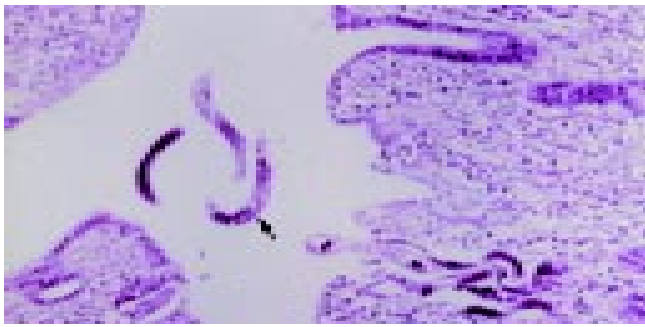


Figure 2 Multiple longitudinal sections of *C. philippinensis* on mucosal surface and lumen. The longitudinal sections shows a row of stichocytes (arrow). Hematoxylin and eosin, $\times 200$.

Three cases received emergency laparotomy due to peritonitis and two of them were found to have jejunitis cystica profunda. Small bowel series and colonoscopic study revealed mild dilatation and thickened mucosa of jejunum and ileum, which suggested malabsorption. Laboratory findings revealed anemia, malabsorption of fats and carbohydrates and low serum levels of potassium, sodium, calcium and total protein. Mebendazole 200 mg twice a day for 20 d was given to 12 patients, while albendazole was given to the other two patients. All of them were cured and relapses were not observed within 12 mo following chemotherapy and supportive treatment.

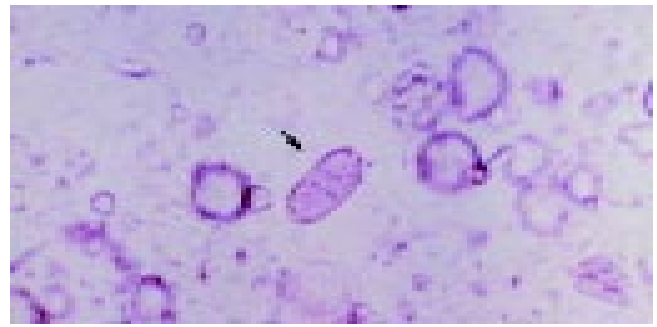


Figure 3 Peanut-shaped eggs of *C. philippinensis* in feces with flattened bipolar plugs. $\times 160$.

DISCUSSION

Capillaria species parasitize many classes of vertebrates, although only 4 species have been found in humans, namely *C. philippinensis*, *C. plica*, *C. aerophila*, and *C. hepatica*^[1]. *C. philippinensis* is a tiny nematode first described in the 1960s as a pathogen inducing severe diarrheal syndromes in humans. In 1962, the first reported case of human intestinal capillariasis occurred in a previously healthy young man from Luzon in the Philippines, who subsequently died. At autopsy, a large number of worms were found in the large and small intestines^[2]. Chitwood *et al.* described this case in 1964^[3]. There was an epidemic of

Table 1 Characteristics of seven patients with intestinal capillariasis reported in Tai-tung

Case No.	Year Occurred	Occupation	Age (yr)	Sex	Travel history	Associated parasite	Treatment	Outcome
1	1983	Farmer	76	M	-	<i>Clonorchis sinensis</i>	Albendazole	Cure
2	1987	Merchant	46	M	-	-	Albendazole	Cure
3	1988	Housewife	58	F	Thailand	-	Mebendazole	Cure
4	1990	Housewife	41	F	-	-	Mebendazole	Cure
5	1991	Housewife	36	F	-	-	Mebendazole	Cure
6	1991	Farmer	62	M	-	<i>Clonorchis sinensis</i>	Mebendazole	Cure
7	1991	Housewife	46	F	-	-	Mebendazole	Cure
8	1992	Merchant	53	M	Thailand	-	Mebendazole	Cure
9	1992	Farmer	45	M	-	-	Mebendazole	Cure
10	1995	Farmer	61	M	-	-	Mebendazole	Cure
11	1995	Housewife	45	F	-	<i>Strongyloides stercoralis</i>	Mebendazole	Cure
12	1999	Farmer	39	M	-	-	Mebendazole	Cure
13	2000	Fisher-man	69	M	-	-	Mebendazole	Cure
14	2001	Farmer	50	M	-	<i>Clonorchis sinensis</i>	Mebendazole	Cure

Table 2 Clinical features and diagnostic method of seven patients with intestinal capillariasis reported in Tai-tung

Case No.	Duration of onset to diagnosis	Diagnostic method	Chronic diarrhea	Abdominal Pain	Abdominal borborygmi	Body weight loss	Anemia	Hypoalbuminemia
1	119 d	Stool ova	+	+	+	+	+	+
2	2Y7 M	Stool ova	+	+	+	+	+	+
3	6 M	Stool ova	+	+	+	+	+	+
4	1Y3 M	Histology	+	+	+	+	+	+
5	120 d	Histology	+	+	+	+	+	+
6	70 d	Stool ova	+	-	-	+	-	+
7	7 d	Stool ova	+	+	-	-	+	+
8	65 d	Stool ova	+	+	+	+	+	+
9	34 d	Stool ova	+	+	+	+	+	-
10	103 d	Stool ova	+	+	+	-	+	+
11	66 d	Stool ova	+	+	+	-	-	+
12	37 d	Histology	+	+	+	+	-	+
13	25 d	Histology	+	-	-	+	-	+
14	17 d	Stool ova	+	+	+	+	-	+

the disease in the Philippines from 1967 to 1968, more than 1 300 persons acquired the illness, and 90 patients with parasitologically confirmed infection died^[4]. In late 1978 and early 1979, another small outbreak was identified in northeastern Mindanao, Philippines, with about 50 persons infected^[4]. Sporadic cases continued to appear in northern Luzon and also in other areas where epidemics had occurred. The disease is endemic in Thailand, where it was first reported in 1973^[5]. Sporadic cases have also been found in Iran^[6], Egypt^[7,8], Taiwan^[9], Japan^[10,11], Indonesia^[12], Korea^[13], Spain (probably acquired in Colombia)^[14], and Italy (acquired in Indonesia)^[12]. The parasite thus appears to be widespread. Because infection may result in severe disease with a high mortality when untreated, early diagnosis is very important. Infection with *C. philippinensis* should be considered in the differential diagnosis of malabsorption syndrome^[15]. There was often a delay in diagnosis, averaging 4 mo and even longer in Taiwan, especially in non-endemic areas^[16]. The delay was over a year in our cases.

It has been found that *Capillaria* species are closely related to *Trichuris* and *Trichinella* species^[1], and the eggs of *Trichuris trichiura* and *C. philippinensis* are similar in appearance, although they can be differentiated by experienced observers^[17]. Some individuals could be infected with both parasites, which further confuse the picture. In fact, 10 of the 11 patients described by Whaler *et al.*^[18] were infected with both *T. trichiura* and *C. philippinensis*. An inexperienced observer might confuse the eggs of *Capillaria* with those of *T. trichiura*^[1], although a correct parasitologic diagnosis could be made by finding characteristic peanut-shaped eggs with flattened bipolar plugs^[2].

The source of *C. philippinensis* infection in our two patients was unclear, particularly as they had no travel history. In Thailand and the Philippines, the infection has been attributed to eating raw or insufficiently cooked fish harboring the larvae^[2,19]. Hakka Chinese in Taiwan like to eat raw, freshwater fish, so they might be expected to have a significant incidence of infection if freshwater fish in Taiwan commonly host *C. philippinensis*. This is not the case, however. Our two patients lived in the southeastern part of Taiwan, closest to Luzon, so it was possible that the fish imported from the Philippines were the source of infection. Fish in markets in Taitung County, southeastern Taiwan, have been examined for *C. philippinensis* infection, but the results were negative. Recent findings suggested that fishing-eating birds might be the natural definitive hosts^[20], including *Bulbulcus ibis*, *Nycticorax nycticorax*, and *Ixobrychus sinensis*, all of which have been found in Taiwan^[21]. Therefore, the possibility of human infection acquired in Taiwan by direct or indirect ingestion of fresh-water fish with a larval stage of the parasite cannot be discounted.

Enteritis cystica profunda is a disorder with mucin-filled cystic spaces lined by non-neoplastic columnar epithelium in the wall of small intestine, predominantly the submucosa. The histology has been found to simulate mucinous carcinoma^[22]. It has also been shown to occur in the esophagus^[23] and stomach^[24,25]. The irregular distribution of the glands and cysts with normal-appearing glandular epithelium containing mucus and Paneth's cells were features suggestive of its benign nature^[26].

Albendazole was presently considered the drug of choice for the treatment of human intestinal capillariasis because it was effective against eggs, larvae, and adult worms^[1,26]. However, for a major infection, mebendazole (200 mg orally twice a day for 20 d) was recommended as the treatment of choice. Attempts to reduce the standard schedule of mebendazole treatment (400 mg daily for 3 wk) have failed in Thailand. Our patients responded well to a standard course of mebendazole and had no evidence of relapse.

Convenient international travel and commercial globalization

have facilitated the wide dissemination of infectious diseases, whether they are carried by human hosts or non-human vectors. Intestinal capillariasis needs to be considered in the differential diagnosis of patients with chronic diarrhea, borborygmus, abdominal pain, and marked weight loss.

REFERENCES

- 1 Cross JH. Intestinal capillariasis. *Clin Microbiol Rev* 1992; **5**: 120-129
- 2 Cross JH. Intestinal capillariasis. *Parasitol Today* 1990; **6**: 26-28
- 3 Chitwood MB, Valasquez C, Salazar NG. *Capillaria philippinensis* sp. N. (Nematoda: Trichinellida) from the intestine of man in the Philippines. *J Parasitol* 1968; **54**: 368-371
- 4 Cross JH, Singson CN, Battad S, Basaca-Sevilla V. Intestinal capillariasis: epidemiology, parasitology and treatment. *Proc R Soc Med Int Cong Symp Ser* 1979; **24**: 81-86
- 5 Pradatsundarasar A, Pecharanond K, Chintanawongs C, Ungthavorn P. The first case of intestinal capillariasis in Thailand. *Southeast Asian J Trop Med Pub Health* 1973; **4**: 131-134
- 6 Hoghooghi-Rad N, Maraghi S, Narenj-Zadeh A. *Capillaria philippinensis* infection in Khoozestan Province, Iran: case report. *Am J Trop Med Hyg* 1987; **37**: 135-136
- 7 Youssef FG, Mikhail EM, Mansour NS. Intestinal capillariasis in Egypt: a case report. *Am J Trop Med Hyg* 1989; **40**: 195-196
- 8 Mansour NS, Anis MH, Mikhail EM. Human intestinal capillariasis in Egypt. *Trans R Soc Trop Med Hyg* 1990; **84**: 114
- 9 Chen CY, Hsieh WC, Lin JT, Liu MC. Intestinal capillariasis: report of a case. *J Formosa Med Assoc* 1989; **88**: 617-620
- 10 Mukai T, Shimizu S, Yamamoto M. A case of intestinal capillariasis. *Jpn Arch Int Med* 1983; **3**: 163-169
- 11 Nawa Y, Imai JI, Abe T, Kisanuki H, Tsuda K. A case report of intestinal capillariasis. The second case found in Japan. *Jpn J Parasitol* 1988; **37**: 113-118
- 12 Chichino G, Bernuzzi AM, Bruno A. Intestinal capillariasis (*Capillaria philippinensis*) acquired in Indonesia: a case report. *Am J Trop Med Hyg* 1992; **47**: 10-12
- 13 Lee SH, Hong ST, Chai JY. A case of intestinal capillariasis in the Republic of Korea. *Am J Trop Med Hyg* 1993; **48**: 542-546
- 14 Drona F, Chaves F, Sanz A, Lopez-Velez R. Human intestinal capillariasis in an area of nonendemicity: case report and review. *Clin Infect Dis* 1993; **17**: 909-912
- 15 Paulino GB, Wittenberg J. Intestinal capillariasis: a new cause of a malabsorption pattern. *Am J Roentgenol Radiother Nucl Med* 1973; **117**: 340-345
- 16 Hwang KP. Human intestinal capillariasis in Taiwan. *Acta Paed Sin* 1998; **39**: 82-85
- 17 Zaman V, Keong LA. Helminths in: Handbook of medical parasitology. 2nd ed. *Edinburgh: Churchill Livingstone* 1990: 87-217
- 18 Whalen GE, Rosenberg EB, Strickland GT. Intestinal capillariasis: a new disease in man. *Lancet* 1969; **1**: 13-16
- 19 Bhahulaya M, Indra-Ngarm S, Anathapruit M. Freshwater fishes of Thailand as experimental intermediate host for *Capillaria philippinensis*. *Int J Parasitol* 1979; **9**: 105-108
- 20 Bhaibulaya M, Indra-Ngarm S. *Amaurornis phoeniceus* and *ardeola bacchus* as experimental definite hosts for *capillaria philippinensis* in Thailand. *Int J Parasitol* 1979; **9**: 321-322
- 21 Cross JH, Basaca-Sevilla V. Experimental transmission of capillaria philippinensis to birds. *Trans R Soc Trop Med Hyg* 1983; **77**: 511-514
- 22 Kyriakos M, Condon SC. Enteritis cystica profunda. *Am J Clin Pathol* 1978; **69**: 77-85
- 23 Volrol MW, Welsh RA, Genet EF. Esophagitis cystica. *Am J Gastroenterol* 1973; **59**: 446-453
- 24 Oberman HA, Lodmell JG, Sower ND. Diffuse heterotopic cystic malformation of the stomach. *N Engl J Med* 1963; **269**: 909-911
- 25 Fonde EC, Rodning CB. Gastritis cystica profunda. *Am J Gastroenterol* 1986; **81**: 459-464
- 26 Anderson NJ, Rivera ES, Flores DJ. Peutz-Jeghers syndrome with cervical adenocarcinoma and enteritis cystica profunda. *West J Med* 1984; **141**: 242-244