A Case of Chronic Diarrhea Secondary to *Capillaria philippinensis* in Occidental Mindoro: Possibly a Newly-Described Endemic Area?

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ABSTRACT

A 41-year-old man from Occidental Mindoro was diagnosed with intestinal capillariasis at the Philippine General Hospital after a year of diarrhea and multiple hospital admissions. The patient was noted to be harboring *Capillaria philippinensis*, *Strongyloides stercoralis*, *Entamoeba histolytica*, *Mycobacterium tuberculosis*, and *Candida albicans* in his stool. He was treated with albendazole, metronidazole, diloxanide, fluconazole, and anti-Koch's. This case was reported to the Department of Health to pursue this site as a potential area for epidemiologic investigation.

Key Words: Capillaria, Capillaria philippinensis, chronic diarrhea, intestinal parasitism, enteric pathogen, polyparasitism, Philippines

Introduction

Intestinal capillariasis or *capillariasis philippinensis* is caused by a small nematode known as *Capillaria philippinensis.* Symptoms of this illness are diarrhea, borborygmi, and abdominal pain. This helminthic infection was virtually unknown until 1963 when Chitwood reported the first case, a schoolteacher from Bacarra, Ilocos Norte. He had a history of intractable diarrhea for 3 weeks with ascites, emaciation and cachexia and later died at the Philippine General Hospital in Manila.[1,2]

In the 1960s, a capillariasis outbreak occurred in Tagudin, Ilocos Sur where almost a hundred died.[3] Another epidemic was recorded in Southern Leyte in the Philippines in the early 1980s.[4] In the recent past, this parasite was also isolated in Compostela Valley Province[2] and Zamboanga del Norte in Western Mindanao.[5]

We present a case of chronic diarrhea secondary to multiple enteric pathogens, including *Capillaria philippinensis* in Occidental Mindoro, a possible newly-described endemic area.

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Case

A 41-year-old male from the town of Alacaak, Sta. Cruz, Occidental Mindoro presented with a one-year history of colicky abdominal pain and watery diarrhea. Multiple consults were done in their region that revealed "unremarkable fecalyses". He was given various antibiotics but this did not resolve his symptoms.

Six months prior to admission, four men in their barangay died from chronic diarrhea and this prompted the patient to seek consult in Manila. He was admitted in a private hospital for work-up where fecalysis again was "unremarkable", abdominal CT that revealed no masses, and colonoscopy with biopsy that revealed mild nonspecific colitis. During this time, the patient had lost more than a third of his weight and developed bipedal edema. Patient was repeatedly readmitted but no definite diagnosis was established. He was never febrile.

The patient had a history of pulmonary tuberculosis treated for only a week. He was fond of eating dogs and *kilawin*. He knew the four people who died in their locality. He admitted to having been in a drinking binge with at least one of them and ate *kilawin* during their binges. He worked as a farmer, as a fisherman, and as a tricycle driver. He would catch fish in Pagbahan River, but would sometimes buy from the local market. No other family member presented with diarrhea. He had no history of travel.

The patient presented at the Emergency Room of the Philippine General Hospital severely emaciated, unable to ambulate and with palpatory BP (Figure 1). He weighed 42 kilograms (BMI=16), with bronchovesicular breath sounds, normal cardiac rate and regular rhythm, scaphoid abdomen which was normoactive, grade 2 bipedal non-pitting edema, and unremarkable digital rectal exam. Neurologic findings were normal. He was hydrated and started on inotropes.

Initial investigations revealed the following: severe hypoalbuminemia (2g/L) with no proteinuria, hemoglobin 142g/L, wbc 7.4 x 10^9/L (neut 0.64 lymph 0.32 eos 0.013), and low electrolytes (sodium, potassium, calcium, magnesium, and phosphorus). He had normal creatinine, bilirubins, and liver enzymes. His ECG had low voltage complexes. His initial stool examination showed abundant white blood cells, without any RBC, fat globules, ova, or parasites. Fecal

occult blood was likewise negative. Investigations for pulmonary tuberculosis including a chest film and sputum AFB were normal. No enteric pathogen was isolated on initial stool culture. Stool was sent to the College of Public Health for FECT which revealed no parasites. A 2D-echo was requested to rule out a cardiogenic cause for the shock and it revealed a small pericardial effusion with an ejection fraction of 33%.

Figure 1. Appearance of the patient prior to treatment (BMI=16 kg/m^2).

Since no enteric pathogen was initially isolated, the investigator initially pursued work-up for non-infectious causes like malabsorption syndromes, irritable bowel disease, malignancy, adrenal insufficiency, hyperthyroidism, and carcinoid. Infectious causes were likewise pursued. Cortisol (560 pmol/L) and thyroid function tests were both normal. Irritable bowel disease and malignancy were ruled out because of the CT scan and the colonoscopy findings. Another stool culture was done and this revealed growth of *Candida* species. Salmonella IgG/IgM were non-reactive. HIV screening was negative. Stool AFB was +1 on day 1 and day 2. However, the investigator was not entirely convinced that this was just TB and stool was sent almost every day for fecalysis.

The patient was on inotropes and colloids during the first week of his admission. He was placed on nothing per orem, except medications, to allow time for the patient's intestinal villi to regenerate. Gastroenterology consult was sought and the patient was treated as if he had short gut syndrome. He was started on ranitidine, loperamide, and racecadotril. Fluconazole and Category I TB treatment were started as well. He was placed on total parenteral nutrition. By the end of the second week, patient was off inotropes and was started on clear liquids. When this was tolerated, he was started on elemental feeding.

However, diarrhea persisted. By the third week, serial fecalyses revealed the presence of the following: *Entamoeba histolytica* cysts, *Strongyloides stercoralis* adult and rhabditiform larvae. First line treatment for strongyloidiasis is Ivermectin but this was not available in the Philippines. Second line drug is albendazole 400 mg bid x 5 days every 2 weeks x 2 cycles and this was started on the patient the same day. Since the patient had been treated multiple times for amoebiasis, diloxanide furoate, a luminal agent, was added on top of metronidazole.

The next stool sample had suspected *Capillaria philippinensis* ova, which were atypical because they did not possess bipolar plugs. Another stool sample was sent the following day and the diagnosis of capillariasis was established when it revealed typical *Capillaria* ova with bipolar plugs (Figure 2). The patient was then treated with the following drugs: albendazole 400 mg bid x 10 days then repeated after 2 weeks (this covered for both *C. philippinensis* and *S. stercoralis*); metronidazole 500 mg tabs q6 x 14 days; diloxanide furoate 500 mg tab tid x 14 days; fluconazole 200 mg cap x 10 days; and quadruple anti-Koch's. By this time, the chronic diarrhea of the patient was attributed to: 1.) *TB colitis,* 2.) *C. philippinensis*, 3.) *S. stercoralis,* 4.) *E. histolytica,* and 5.) *C. albicans.*

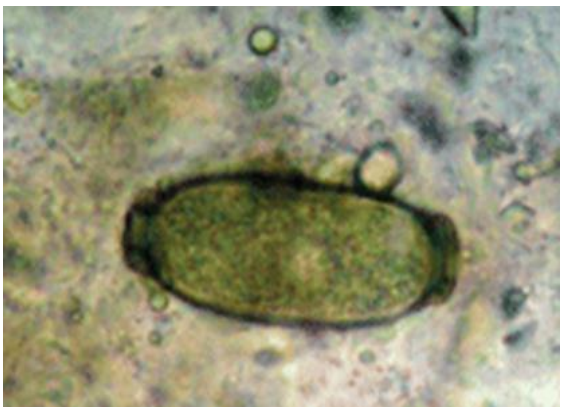

Figure 2. *C. philippinensis* ova recovered from the stool of the patient.

Three days after being given those drugs, diarrhea resolved. A repeat 2D-Echo showed an improved ejection fraction of 72%, consistent with nutritional cardiomyopathy. After another week, the patient was discharged wheelchair-borne. He was educated on how he acquired his illness. Avoidance of eating raw fish and walking barefooted was emphasized. A month after, the patient followed up weighing 49 kg (Figure 3). He was already ambulant and had good appetite. Albumin improved from 2 g/L to 24 g/L after just one month. Repeat FECT revealed no organisms. By the time he completed his 6 months of anti-Koch's, he was already back in Occidental Mindoro where he now drives his tricycle for a living.

Figure 3. The patient after treatment (BMI=20 kg/m^2).

Discussion

Capillariasis is a parasitic disease caused by infection with any of the four Capillaria species found to infect humans: *Capillaria hepatica, C. aerophila, C. plica,* and *C. philippinensis*.[3] *Capillaria philippinensis* inhabits the small bowel of humans, causing mostly diarrhea and malabsorption. The life cycle is incompletely understood, although freshwater fish contain larvae infectious for humans and birds. The latter is an important reservoir host, especially migratory birds that could drop their feces in any body of water. Laboratory examinations document the typical findings of protein-losing enteropathy, malabsorption, and electrolyte loss. The diagnosis is established by detecting characteristic ova or larvae in the stool. In untreated patients, mortality rate is 33%, but specific anti-helminthic therapy is effective and life-saving. Infection is prevented by eating only properly cooked freshwater fish.[6]

C. philippinensis was first reported in 1963 in Bacarra, Ilocos Norte Province and has been endemic in that area ever since, particularly in Pudoc Lake where it was isolated.[1] Since then, additional endemic foci of *C. philippinensis* have been identified in Southern Leyte,[3] in Compostela Valley, in Southern Mindanao,[2] and in Zamboanga del Norte, Western Mindanao.[5]

To date, this is the first reported case of intestinal capillariasis from Occidental Mindoro, an island bounded by the Mindoro Strait and the Mamburao reef (Figure 4). There are 11 barangays in Sta. Cruz, and in Alacaak, where the patient resides, there is a freshwater reservoir called the Pagbahan River where he sometimes catches fish. People here are fond of eating *kilawin*, which is raw fish soaked in vinegar and seasoning.

The lacustrine goby or *dulong* (*Gobiopterus lacustris*) and the Philippine anchovy or *bolinaw* (*Encrasicholina oligobranchus*) are the two most common species eaten raw in their locality because they are cheap. In fact, they are sometimes called the "poorman's fish". However, when there is a festivity, they would sometimes use chopped and pounded milkfish (*Chanos chanos*) or *tanigue* (*Acanthocybium solandri*) in their *kilawin*. Any of these species of fishes could have been harboring the infective larvae and could have been ingested by migratory birds as the definitive host or by men as the accidental host.

Figure 4. The Municipality of Sta. Cruz, Occidental Mindoro (Source: www.wikipilipinas.org, reproduced with permission).

According to the patient, people living here have water-sealed toilets, although he is aware of some families using pit privies. He also attested that some people in their locality resort to open and indiscriminate defecation. Children sometimes defecate near the shoreline when they are playing and swimming.

In this case, we believe that the patient may be an "indigenous case" in that he acquired his infection from his locality (rather than an "imported case", where a patient got the infection from an already established endemic area). He had no history of travel, and there were already 4 deaths in their area attributed to diarrhea secondary to an undetermined cause. Since migratory birds are in part responsible for dissemination, the bodies of water surrounding Sta. Cruz may have been part of their usual stops. The cycle of fish eating infected feces, then man eating infected fish could have easily been perpetuated in this area because eating raw fish is a common practice. Furthermore, since open and indiscriminate defecation has been described by the patient, fish eating not just infected bird feces, but infected human feces as well, could have been part of the cycle.

This case presented with diagnostic difficulties since the patient presented with almost a year of "unremarkable"

laboratory results. Capillariasis could possibly be endemic in this barangay for quite some time now, but was left undiagnosed. In Compostela Valley, there have been 12 deaths before the diagnosis of capillariasis was established. This was attributed to the lack of proficiency of the local health staff in laboratory diagnosis and a general lack of awareness of this potentially fatal parasitic infection.[2] An inexperienced observer may confuse the eggs with those of *Trichiuris trichiura*, which are larger (50 to 54 by 22 to 23 μm) and barrel-shaped, with more prominent mucoid bipolar plugs.[3]

According to a study by Belizario et al., 54.7% of patients examined at Compostela Valley were infected with ≥1 organism, including *C. philippinensis*. Most of the patients were co-infected with hookworms, *Endolimax nana*, *Entamoeba histolytica, Entamoeba coli*, and *Heterphyes* sp.[2] This is called *polyparasitism*, and the study done in Mindanao reflected the same problem in Thailand where it is a common practice to eat *koi pla* (chopped raw fish mixed with lemon juice, finely cut red onion and chili) and *tab kaab* (pounded raw whole fish mixed with lemon juice, finely cut red onion and chili). *C. philippinensis* has been identified in 25 provinces in Thailand and majority of infected patients also harbored hookworms.[7,8] In addition, cases of polyparasitism with *C. philippinensis* have also been reported in Taiwan,[9] Lao Republic,[10] Egypt,[11-12] Iran,[13] Southern Brazil[14] and Africa.[15]

In our patient, the chronic diarrhea could have easily been caused by *C. philippinensis, S. stercoralis,* and *E. histolytica*. All these organisms are related to poor environmental sanitation, possibly stemming from indiscriminate defecation. Our patient was unique because the *Capillaria* eggs only surfaced after more than 10 serial fecalyses done at our Institution. This is odd, since *Capillaria* infections are usually prolific. Eggs, larvae, and adults appear in great numbers in stool specimens.[3] This is in contrast to *S. stercoralis* wherein worm load is low. In these cases, chances of finding the parasite is proportional to the number of occasions in which the stool is examined, or if available, stool should be analyzed using concentration techniques. It is also important to note that occurrence of *autoinfection*, or the ability of a parasite to multiply and complete one's life cycle inside humans, is recognized only in *Strongyloides stercoralis* and to a lesser extent, in *Capillaria philippinensis* infections.[16] In these two infections, complete eradication of the parasite is necessary for cure.

The patient also had TB colitis, which is permissive for infection with *C. philippinensis* and *S. stercoralis*.[16] Parasitic overgrowth rendered him susceptible to *Candida* as well. In these cases of polyparasitism, morbidity becomes additive and mortality subsequently is higher. The key then to the cure of our patient was clinching the correct diagnosis that facilitated correct treatment.

Conclusion

In summary, we have presented a case of intestinal capillariasis from Occidental Mindoro, left undiagnosed for more than a year in different institutions. Although capillariasis contributed to his disease, polyparasitism with *Strongyloides stercoralis* and *Entamoeba histolytica* could account for the patient's overall condition. Presently, there is still a need to identify the possible occurrence of intestinal capillariasis in other unrecognized foci and to define its geographic distribution here and abroad.

Recommendations

This case was reported to the Department of Health to pursue this site as a potential area for epidemiologic investigation. Accurate diagnosis will result in the appropriate treatment and provide information and evidence as basis for the control and prevention of this potentially fatal disease in the community.

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