# An Unusual Case of Echinostomiasis in a Retropositive Patient: A Case Report



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**ABSTRACT:** This report describes an unusual case of echinostomiasis in a retropositive patient with swelling, pain, and purulent discharge over back and right scapular region for 10 days. Stool examination revealed the presence of eggs of hookworm and echinostome. Diagnosis is challenging due to few characteristic features of eggs of echinostomes. They usually infect birds and mammals, and only limited species are implicated in the causation of human disease. Mild infections are of little significance, but heavy fluke burden may lead to significant abdominal symptoms. Incidence of echinostomiasis is higher in Southeast Asian countries, and this report is an unusual case in India. We discuss general desirability of awareness of this kind of infection and training for better detection.

KEYWORDS: echinostome, retropositive, hookworm

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## Introduction

Echinostomes can cause severe abdominal pain accompanied by diarrhea, malnutrition, and fatigue and hence are of significant medical importance.<sup>1</sup> Around 20 species belonging to 8 genera (*Echinostoma, Echinochasmus, Acanthoparyphium, Artyfechinostomum, Episthmium, Himasthla, Hypoderaeum*, and *Isthmiophora*) are implicated in the causation of human disease worldwide.<sup>1,2</sup> Source of infections and life cycles are different for each echinostome species. Therefore, the specific diagnosis of adult echinostome infections should be considered. Diagnosis is mainly established by the demonstration of eggs in the fecal sample.

*Echinostoma ilocanum* was first described as a parasite of man in the Philippines<sup>3</sup> and was later reported in Indonesia.<sup>4</sup> The parasite was also found in animals: *Rattus norvegicus* Berkenhout, 1769, in the Philippines<sup>5</sup> and native dogs in Canton, China.<sup>6</sup> A few human cases of *Episthmium caninum*, which have microscopically indistinguishable eggs, have been documented to occur in northeastern Thailand and the dog serve as its natural definitive host. The infection is apparently acquired through the consumption of infected fish.

Incidence of echinostome infection is less in India, although reports from other Southeast Asian Countries are relatively common. The present case report is an unusual case of echinostomiasis with hookworm coinfection in a retropositive patient who is a resident of India.

**Case report.** A 35-year-old known retropositive male presented with swelling, pain, and purulent discharge over back

and right scapular region for 10 days. On evaluation, the patient was found to be retropositive. There was no history of diarrhea or abdominal pain and also no history of consumption of sea food.

Physical examination revealed that patient was conscious and cooperative. No pallor, icterus, clubbing, cyanosis, lymphadenopathy, or edema was present. Local examination revealed swelling over the back of around 4 cm  $\times$  3 cm. Fullness over right scapular region with diffuse fluctuation present and pus discharge present from swelling.

The level of hemoglobin was 10.9 g/dL and hematocrit was 33.6%; the total leukocyte count was  $8.9 \times 10^{3}/\mu$ L and was reactive for HIV 1 antibodies. The ratio of CD4/CD8 was 0.37. Absolute CD4 count and CD8 counts were 114 and 310 cells/ $\mu$ L respectively.

Stool examination revealed the presence of eggs of hookworm and echinostome (Fig. 1). The eggs of echinostome were approximately 94  $\mu$ m long (89–99  $\mu$ m) and 55  $\mu$ m wide (52–58  $\mu$ m) with a wide and inconspicuous operculum, a thin and refractile shell, and abopercular wrinkles terminally.

As the microscopy stool examination revealed the presence of eggs of hookworm, the patient was administered oral tinidazole 500 mg twice daily for 15 days. Treatment for echinostome with praziquantel was not considered due to the lack of abdominal symptoms.

### Discussion

Echinostomiasis is a zoonotic disease caused by *Echinos-toma*, which is an intestinal fluke belonging to the family





Figure 1. Egg of echinostome (light microscope: 40 × magnification).

Echinostomatidae. The family Echinostomatidae has 40 genera including 400 species. Echinostomes usually infect birds and mammals. Among 400 species, only 23 species are implicated in the causation of human disease.<sup>7</sup> Mild infections due to this parasite in humans are of little significance, but heavy fluke burden may lead to significant abdominal symptoms.<sup>8</sup>

Many animals may serve as definitive hosts for various echinostome species, including aquatic birds, carnivores, rodents, and humans. Unembryonated eggs are passed in feces and developed in the water. The miracidium takes an average of 10 days to mature before hatching and penetrating the first intermediate host, a snail. Several genera of snails may serve as the first intermediate host. The intramolluscan stages include a sporocyst, one or two generations of rediae, and cercariae. The cercariae may encyst as metacercariae within the same first intermediate host or leave the host and penetrate a new second intermediate host. Depending on the species, several animals may serve as the second intermediate host, including other snails, bivalves, fish, and tadpoles. The definitive host becomes infected after eating the infected second intermediate hosts. Metacercariae excyst in the duodenum and adults reside in the small intestine.

Echinostomiasis is distributed throughout the world, but the incidence is higher in Southeast Asian countries owing to their food habits. A study from Seoul Paik Hospital reported egg positivity rate of *Echinostoma* sp. to be 0.03%.<sup>9</sup> An epidemiological survey from Korea in 169 inhabitants and 473 junior high school students revealed 3 (0.5%) positive cases of echinostomiasis.<sup>10</sup> In another study from northeastern Thailand, in posttreatment, the fecal examination showed *Echinostoma malayanum* (8.3%), *E. ilocanum* (8.1%), and *Echinostoma revolutum* (0.8%).<sup>11</sup> A study from Thailand showed 18% of positive stool samples, infected with one or more parasites, of which 0.1% was *Echinostoma* species.<sup>12</sup>

Public health interventions to prevent infections with food-borne trematodes and to reduce the prevalence and intensity of infections, and, hence, morbidity and mortality, include chemotherapy, improved access to adequate sanitation, and the use of chemical fertilizers, food inspections, and information, education, and communication campaigns. The ultimate aim is to change human behavior, because the consumption of raw or undercooked freshwater fish and other aquatic products is the key risk factor for acquiring a foodborne trematode infection.

Generally, a single dose of 10 mg/kg praziquantel is recommended as the regimen for the treatment of intestinal fluke infections. The patient in the present report was not treated, probably due to the absence of significant clinical symptoms, which may have been due to the relatively low fluke burden. However, given the clinical history of HIV infection, perhaps it would have been advisable treating the echinostome infection in order to avoid any future complication. Furthermore, higher fluke load may lead to significant clinical manifestations.<sup>13</sup> In cases with heavy burden of the echinostome species, *Artyfechinostomum malayanum*, mortality due to intestinal perforation or marked malnutrition and anemia has been reported.<sup>1</sup>

The diagnosis of echinostomiasis by fecal examination is challenging. The number of eggs produced per worm per day varies by species and is relatively small in certain species<sup>1,2</sup> compared with other helminth parasites, such as *Ascaris lumbricoides*. Moreover, eggs of echinostome species are not routinely encountered by the laboratory personnel and clinicians and can go unnoticed, particularly in Kato-Katz fecal smears. The eggs of *Echinostoma* have close resemblance to the eggs of *Fasciola hepatica* and *Fasciolopsis buski* in shape, color, and contents but are smaller in size. To deal with this problem, training of microscopists to detect echinostome eggs is needed.

#### **Author Contributions**

Conceived and designed the experiments: VK. Analyzed the data: KT. Wrote the first draft of the manuscript: KT. Contributed to the writing of the manuscript: YM. Agree with manuscript results and conclusions: RK. Jointly developed the structure and arguments for the paper: KT. Made critical revisions and approved final version: VK. All authors reviewed and approved of the final manuscript.

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