

An unusual case of urinary sparganosis in the Indian subcontinent

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ABSTRACT

Spirometra is a genus of pseudophyllidean cestode that reproduces in canines and felines but can cause pathology in humans. When humans harbour plerocercoids of these tapeworms outside the intestine, it can cause sparganosis. We report a case of urinary sparganosis in a young woman, passing multiple spargana worms in her urine. The worm was identified as the plerocercoid larvae of *Spirometra* spp., and the case was managed successfully.

INTRODUCTION

Sparganosis is a rare parasitic infection caused by the plerocercoid larva (sparganum) of the genus *Spirometra*, a pseudophyllidean tapeworm.^[1-4] It was first described by Patrick Manson from China in 1882, and the first human case was reported by Charles Wardell Stiles from Florida in 1908. The adult *Spirometra* lives in the small intestines of canines and felines. Canines and felines act as the definitive hosts for the cestodes. Crustaceans act as the first intermediate hosts while reptiles, amphibians, and pisces act as the second intermediate hosts. Humans' acts as their accidental or paratenic hosts.^[3] Cases of sparganosis have been sporadically reported throughout the world including India. Here, we report an unusual and interesting case of urinary sparganosis, probably the first of its kind, in which the patient presented with shedding of multiple spargana worms in her urine.^[1]

CASE REPORT

A 30-year-old unmarried, female presented to the urology outpatient department of our superspeciality

center. She brought her urine sample along that contained two white, ribbon-like, motile worms measuring about 1.5–2 cm in length. The patient was a vegetarian, a biology student (as well as teaching in local coaching institutes), resident of Chattisgarh state and had a history of traveling to multiple places and residing in hostels within the state of Madhya Pradesh (including Gwalior, Bhopal, and Indore) over the period of past 6 months. Otherwise healthy, she complained of the intermittent shedding of 2–3 such worms in the urine for the past 1 month. She also complained about the mild irritative lower urinary tract symptoms 10 days back which were managed by consuming excess water and fluids. Before presenting to the clinician, she had already tried 2–3 doses of over-the-counter antiparasitic medications without any remarkable improvement. There was no history similar complaints in her family or locality.

The urine, containing worms, was received in our microbiology laboratory for examination and identification. Routine laboratory tests were unremarkable, there was no pyuria or hematuria. The urine culture was sterile following 48 h of incubation.

Both the worms measured 15–16 mm in length and about 1.5 mm in width [Figure 1]. They were whitish, glistening,

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Figure 1: *Spirometra* w.m

wrinkled, and ribbon shaped. Anterior end of the worm was flattened and grooved vertically. Therefore, the worm was identified as the larva of pseudophyllidean tapeworm belonging to the genus *Spirometra*. These grooves were the bothrium that divides the two lips on leaves and hence called as *Diphyllobothrium*. The bothrium establishes antero-posterior polarity of the worm. The worm had no scolex or internal organs. Its tegument was ridged at irregular interval, which is known as pseudosegmentation. They were showing slow, undulating movements.^[5] Therefore, the worm was identified as the larva of pseudophyllidean tapeworm belonging to the genus *Spirometra* [Figures 2a and b]. The patient was prescribed Praziquantel at a dose of 120–150 mg/Kg body weight over 2 days.^[2] The patient did not return herself and had to be telephonically reminded after 3 months to come for follow up. She revealed that she started feeling better after 10–12 days of treatment and shedding of worms in urine stopped. Because of her poor financial status, she refused further evaluation at the time of follow-up, however, she has been asked by the urologist to visit again in case of any discomfort.

DISCUSSION

Human Sparganosis is an extremely rare disease. It is sporadically reported globally with higher prevalence in the eastern and southeast Asian countries.^[3] The parasite is transmitted to humans either by drinking the water contaminated with copepods housing *Spirometra* larvae or by consuming the raw flesh of fish, snakes, or frogs. It may also be transmitted by placing the raw poultices of infected second intermediate hosts on the open wounds or eyes for medicinal or ritualistic reasons.^[1-4] The incubation period of the organism is from 20 days to ~3 years. The adult *Spirometra* lives in the small intestine of cats and dogs. The unembryonated eggs pass in their feces and hatch into coracidia that are ingested by copepods and develop into proceroid forms. These infected crustaceans are ingested by second intermediate host and the proceroid released from them develops into plerocercoid larval forms.^[3]

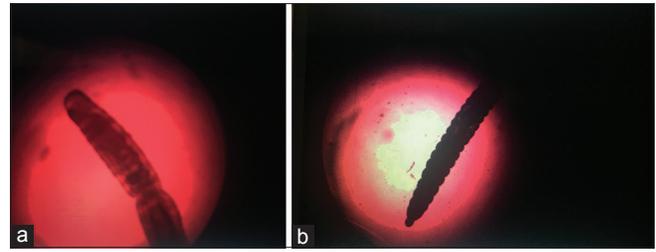


Figure 2: (a) Microscopic anterior view of *Spirometra*. (b) Microscopic posterior view of *Spirometra*

The most common cause of Sparganosis in the western hemisphere including India,^[3] and particularly, in our case is consumption of contaminated drinking water. This can be correlated with the history provided by our patient as she was a vegetarian and had traveled to various places throughout the state during the past 6 months.

The clinical manifestation of Sparganosis is diverse and often follows after larvae have migrated to the subcutaneous locations. The destination of the larvae is often a tissue or a muscle of the chest, abdominal walls or extremities. However, they may also migrate to the sites such as the eyes, brain, pleura, pericardium, spinal canals, breast, heart, lungs, and abdominal cavity.^[2,3] It also affects the genitor-urinary system involving epididymis, spermatic cord, penis, groin, testis, scrotum, retroperitoneum, labia, ureter, and urinary bladder.^[1,3] The larval worms usually grow into 1–2 cm irregular, painless nodules. They may persist for months or even years without any symptoms and then may suddenly become painful.^[3] The patient suffers from nonspecific discomforts, vague pain, palpable mass, headache, or is asymptomatic depending on the involved organs.^[1,4] Since our patient was shedding the worms in her urine, the most probable site of colonisation of the worms was a nodule somewhere in the urinary bladder. Also, our patient was asymptomatic at the time of presentation. The nodules that may have been present in the urinary bladder would have required ultrasonography and cystoscopy to be visualised. Had she not been shedding worms in her urine, we could have missed the case of vesicle sparganosis, because cystoscopy is unnecessary for a patient with mild irritative lower urinary tract symptoms.

Human sparganosis is a surgical disease that depends entirely on the detection of larvae in the lesions. Surgery has been the treatment of choice in all the cases.^[2,3] Visceral larva migrans due to this parasite has high morbidity and considerable mortality.^[3] Sparganosis is typically diagnosed by surgical excision of the worm. It can also be diagnosed by identification of eosinophilia or identification of worm in the tissue specimen. In case biopsy and excision procedures are not feasible, the antisparganum enzyme-linked immunosorbent assay (ELISA) test may be performed. Our patient neither showed definite eosinophilia nor the positive ELISA

test. This may be because of the involvement of the urinary tract. Preoperative diagnosis is rare and can be normally made by obtaining exposure history along with presence of painful, and migratory subcutaneous nodule.^[2,4] In our case, preoperative diagnosis was possible through identification of worms passing in patient's urine. Treatment of sparganosis is praziquantel but with limited success. Because the incidence of disease is extremely low, even in endemic areas, it is difficult for surgeons to suspect sparganosis based only on the preoperative data.^[2]

Because it is a rare infection, public health strategies have not made its prevention a priority. Public health strategies focusing on providing access to clean water, especially in areas where ponds or ditches provide peak potential habitats for infected copepods may help to reduce further sparganosis infection.^[3,6] Therefore, to conclude, clinicians should consider vesical sparganosis as one of the differential diagnosis in case of patients with suspicious history. Especially, parasitic infections (extraintestinal; due to visceral larva migrans) that are transmitted through consumption of contaminated water should not be ignored in countries like India. Recently, there is an urgent need to look for the copepods in water sources since some of the parasites complete their life cycle through these crustaceans.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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